Children and Young People with Cerebral Palsy in Northern Ireland (1981 – 2008)

A Comprehensive Report from the Northern Ireland Cerebral Palsy Register

Queen's University Belfast

HSC Public Health Agency

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Introduction

Cerebral Palsy CP

Cerebral Palsy (CP) is an umbrella term which covers conditions affecting a child's ability to move or control their movement due to problems with development or injury to the immature brain. CP can also be associated with impaired vision, hearing, intellectual ability and seizures (SCPE, 2000). This term encompasses a collection of conditions varying in aetiology, severity and pathology (Mutch et al., 1992). Therefore, it is important to reach an agreement on the common characteristics displayed by these conditions. There are several definitions of CP in the literature (Balf et al., 1955; McKeith et al., 1959; Bax, 1964; Ingram et al., 1984; Mutch et al., 1992). Although these definitions may vary to some extent, they can be broadly summarised as follows:

> *"Cerebral Palsy is a group of permanent, but not unchanging, disorders of movement and/or posture and of motor function, which are due to a non-progressive interference, lesion, or abnormality of the developing/immature brain. This definition specifically excludes progressive disorders of motor function, defined as loss of previously acquired skills in the first 5 years of life."*

Although CP does not affect a large proportion of the general population, it is regarded as the main cause of physical disability in childhood (Stanley et al., 2000). The overall rate of CP in Europe for the period between 1980 and 1990 is 2.08 per 1,000 live births (SCPE, 2002). The prevalence in NI over a similar period is 2.24 per 1,000 live births (Parkes et al., 2001). Other countries in the world have reported prevalence rates varying between 1 and 4 per 1,000 live births (Maenner et al., 2012). In recent years, there have been increasing rates of preterm births: improved care provided to these infants means that more of them are surviving (Platt et al., 2007). At the same time, there has been an increase in multiple pregnancies and births (Surma et al., 2006). These infants are at greater risk of CP . The overall prevalence of CP in Europe between 1981 and 1996 is 50.6 per 1,000 live births of very low birth weight babies (Platt et al., 2007).

CP determines a great human and financial impact on families and services. Litigation costs might also be added to these costs. The majority of claims for compensation and settlements in the UK relate to adverse events around birth and involve children with CP (Surma et al., 2006).

The probability of survival has increased among children with a severe level of disability, meaning that appropriate services will need to be provided for this population beyond childhood and into adolescence and adulthood (SCPE, 2000). Much of the research in the last 30 years has focused on preterm and very low birth weight babies as well as the care given to mothers and babies around the time of birth (SCPE, 2000). However, improvements in the survival rate of these children and a shift in the focus towards quality of life mean that there is a need to improve the dialogue between families, service commissioners and providers as well as researchers (Surma et al., 2006). Data linkage with other administrative datasets such as Census, death notifications and prescribing data could potentially provide valuable and cost-effective information concerning the relationship between survival and severity of CP (Hemming et al., 2005), CP children's participation and integration into society, only to mention a few examples.

Although CP is the commonest cause for disability among children and is particularly common among very small babies, the overall number of children affected by the condition is not large. This has implications for the study of causes and prognosis of these children, as very few individuals can be identified at a given time in a given area. For this reason, it is important to collect specific information on a large number of births. Specialised registers and collaborations (Surma et al., 2006) at national and international level, such as the collaborative network Surveillance of CP in Europe (SCPE) can help meet this goal. As well as providing a valuable resource by pooling data collected across different countries for long periods, the work of the SCPE work has also helped in the harmonisation of descriptions for patterns, levels of severity and function (SCPE, 2000; Surma et al., 2006).

Long term planning is essential for disease specific registers more so in the case of CP. This is due to the need for sufficient time to ascertain cases as inclusion of children with this condition can only be confirmed around the ages of 5 years to allow for a robust diagnosis. Thus, continuity and funding are crucial to ensure the quality of a disease register (SCPE, 2000; Surma et al., 2006).

The Northern Ireland Cerebral Palsy Register

The Northern Ireland Cerebral Palsy Register (NICPR) commenced its work in 1992 and it is one of the longest standing registers in Europe. The two aims of the NICPR are to establish a systematic approach to the surveillance of CP among children in NI. Secondly the Register supports research and audits into the condition, covering aetiology, treatment and assessment as well as health and social care services.

The Register is funded by the Public Health Agency. The Register has been approved by the Queen's University Ethics Committee and more recently, in October 2013, by the Office for Research Ethics Committee in Northern Ireland (ORECNI). There are two consultants attached to the Register who provide clinical and epidemiological advice. The Register has an Advisory Committee to guide and advise on its management and its future direction. This committee includes a lay member of public and a parent representative. The Register has also a Research Sub-committee which reviews data requests linked to audits and research projects.

The Register has surveyed CP for a 37 birth year span (1977-2014) and currently counts on the support of 110 clinicians reporting cases of CP. We have received 7,551 notifications and completed 2,871 assessment forms identifying over 2,000 children with CP.

The scope of this report

This report summarises the work of the NICPR. It presents findings of the cumulative work done by the Register since 1992, which involved collecting data, monitoring and surveillance activities as well as research conducted in collaborations national and internationally. The work of the Research team would not have been possible without the support of the Public Health Agency, to which we are grateful. We also wish to acknowledge the precious ongoing support and collaboration of members in the Advisory Committee, Research Subcommittee, clinicians and families across NI.

Method

Case Definition

The definition of CP used to identify cases included in the Register was that provided by Mutch and colleagues (1992), whereby CP is defined as an "umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (p. 549).

Capitalising on the work of Surveillance of CP in Europe (SCPE), the NICPR has also used a decision-making tree to help professionals identify children that may be included in or excluded from the NICPR (See Appendix 1).

Children with so-called 'acquired' CP have been included in the NICPR. These are participants whose diagnosis of CP is due to lesions or anomalies which occurred 28 days after birth but before their 5th year. Children with acquired CP are, however, excluded in some analyses concerning birth prevalence and aetiology. The number of cases with acquired CP is reported in Figure 1 Case ascertainment.

Data collection was both retrospective and prospective during 1991-94. Since 1994, a mainly prospective notification system has been established for the identification of newly diagnosed and suspected cases, although periodic retrospective searches of hospital and special needs school records as well as the Child Health System also take place. Prospective notifications are encouraged using a report card (Appendix 2) sent monthly to an established network of clinicians including paediatric neurologists, acute and community paediatricians, neonatologists, orthopaedic consultants, paediatric physiotherapists, a voluntary agency and parents. Multiple and overlapping sources of case notification were used to provide confidence on case ascertainment.

Researchers recommend age of ascertainment for CP diagnosis should be 5 years (Stanley, Blair and Alberman, 2000). Nevertheless, this age may vary across registers. In the NICPR we followed the criteria agreed in the framework of the Surveillance of Cerebral Palsy in Europe (SCPE, 2000, NICPR Management Protocol, 2013). Cases reported to NICPR are considered to have a definite diagnosis of CP if they fulfil clinical criteria after their 4th birthday. Children whose diagnosis has not been confirmed until this age, are in the database on a preliminary basis until the diagnosis is confirmed.

Some of the children who are signalled as possible cases with CP in infancy, toddlerhood, or early childhood, pass away before their diagnosis can be confirmed at age four but after the age of two. The SCPE framework has agreed that exclusion of these children might lead to underestimation of CP prevalence. For this reason, the NICPR retains information on these children. Nonetheless, these cases are "flagged" and can be excluded from some analyses.

Furthermore, some cases might be lost to follow up (e.g. if the family moves out of Northern Ireland), which could also prevent from confirming the diagnosis after the child is four. Once again, exclusion of these cases might result in underestimation of prevalence of CP. In accordance with criteria agreed within the SCPE framework, children lost to follow up and for whom a diagnosis cannot be confirmed after age 4 are retained in the NICPR if they have received an unambiguous diagnosis after the age of 3 years, according to the clinical judgment of the consultant paediatrician attached to the NICPR.

Children's Assessment

Information about children notified to the Register is collected by means of a Standard Assessment Form. This form has been developed over the years based on work by Evans and colleagues (1989), advice from epidemiologists, clinicians and the work of SCPE. Clinicians and health records are consulted to complete the standard assessment forms. The information collected on this form includes name and address, sex, date of birth, motor impairment and functional level, subtype of CP and possible causes, MRI findings, associated impairment (e.g. intellectual impairment or epilepsy), birth details (e.g. gestational age, birth weight) and other relevant information (e.g. whether the child has a sibling with CP).

The consultant paediatrician attached to the Register checks each standardised assessment form for consistency and validates it. Since young children can outgrow early signs of CP (Nelson & Ellenberg, 1982), standardised assessment forms that have been completed before the age 4 are followed up and re-assessed after the 4th birthday to confirm eligibility for inclusion in the NICPR as well as clinical presentation.

Classification of CP

The classification of CP into different subtypes has attracted debate over the years. However, the work conducted by the SCPE network has produced a consensus on a classification scheme that can be used primarily for epidemiological purposes. Nonetheless, this classification scheme can also have clinical utility. The decision tree for CP classification is reported in Appendix 3. The main subtypes of CP in this scheme are:

- Spastic Unilateral: one side of the body is affected by velocity-related increase in muscle tone.
- Bilateral Spastic: both sides of the body are affected by velocity-related increase in muscle tone, which could in turn involve two, three or four limb dominated forms.
- Dyskinesia: this includes dystonia and choreo-athetosis; disorganised patterning with fluctuating tone and slow and writhing movements are observed.
- Ataxic: low tone is associated with short and jerky movements.

More information on the prevalence of these subtypes and associated impairments are included in the Results section of this report.

Gross Motor Function Classification System (GMFCS)

Functional impairment severity is described using the Gross Motor Function Classification System (GMFCS). This system includes five levels of functional and motor abilities specifically designed to capture the severity of motor functional impairment in children with CP (Palisano et al., 1997). Clinicians that complete the standardised assessment form are asked to score children using the GMFCS. For ease of completion, a description of each of these levels is provided in both text and pictorial form (see Appendix 4). However, information concerning children's trunk control, leg function (with and without use of aids), and arm function is used to determine a GMFCS score when this information is not provided by clinicians completing the form. Information on the prevalence and factors associated with different functional levels are reported in the Results section.

Motor and associated impairments

Clinicians who completed the standardised assessment form are also requested to provide information on motor impairments such as muscular tone, abnormal unwanted movement and contractures. Furthermore, they are asked to provide details on the presence and severity of oromotor function and feeding problems as well as seizures, intellectual, communication, vision and hearing impairments. Details requested include the presence of the impairment as well as its severity and, in the case of intellectual problems, the method of assessment.

Deaths, immigration and emigration

Deaths of participants before the establishment of the NICPR (1992) were ascertained from death certificates. However, this is known to be a potentially incomplete form of information. Deaths since 1991 have been ascertained by flagging all cases with the Business Service Organisation. Further information on survival rates is provided in the Results section.

Socio-Economic Deprivation

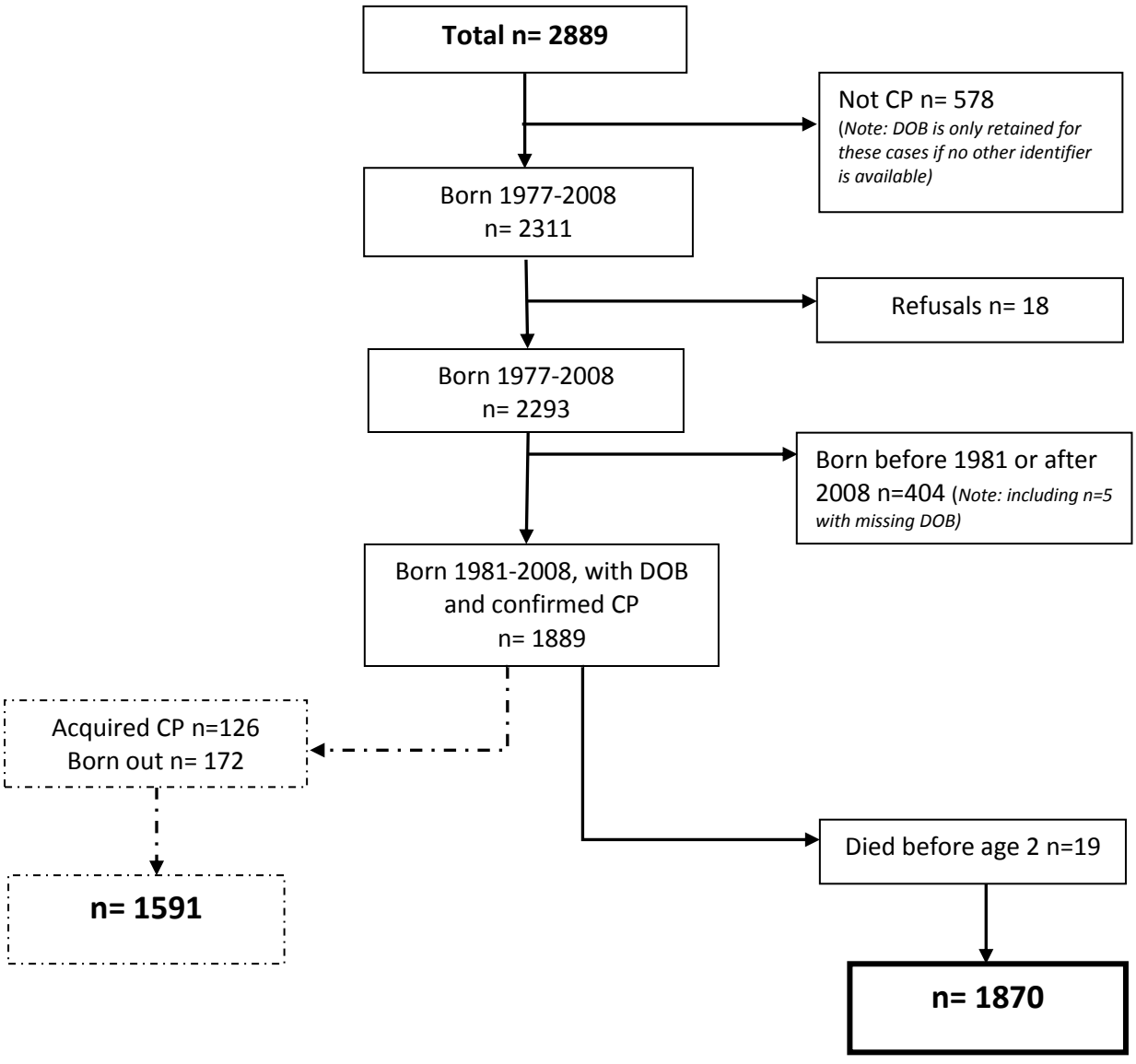
Information on exposure to socio-economic deprivation was collected by linking each individual's address of residence to the Northern Ireland Deprivation Measure (NISRA, 2005). This measure provides the basis on which different Super Output Areas (SOAs) are scored in a continuum from the most to the least deprived areas. In data analyses, we considered quintiles of the multiple deprivation score and derived a dichotomous indicator whereby participants living in areas falling into the most deprived quintile were compared to participants living in areas falling in the other, less deprived quintiles of multiple deprivation.

RESULTS

Participants

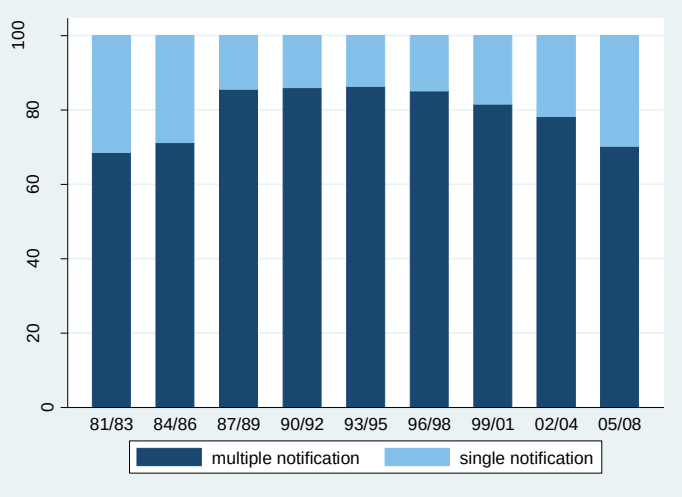
Overall, 1,870 participants were considered for analyses reported here. These are all the children recorded in the NICPR by December 2014 who are aged 4 or older. Children born between 1977 and 1980 have been excluded from analyses due to incompleteness of ascertainment for those birth years. These children were leaving school and/or reducing contact with health services at the time the NICPR had begun its work in 1992, and it was therefore difficult to collect information on them and complete ascertainment. Participants born after 2009 were also excluded from analyses as this cohort had not been fully ascertained at the time of this report. Additionally, 578 cases who had been initially notified to the NICPR, were excluded from the Register as their medical diagnosis was later confirmed to be inconsistent with CP. Finally, children who had deceased before the age of 2 have been excluded from some of the analyses (see Figure 1).

Figure 1. Flowchart of exclusion from NICPR dataset



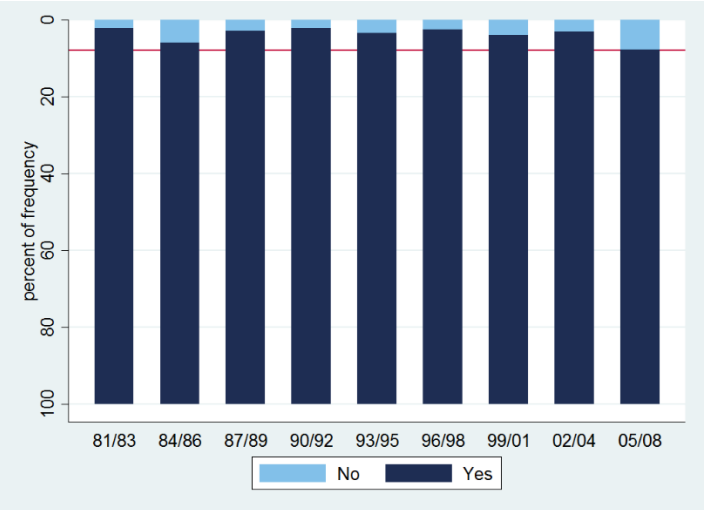
Of the 1870 cases included in the analysis (Figure 1) 1477 (79%) were multiply notified. Figure 2 reports the percentages of multiple or single notifications by period of birth.

Figure 2. Percentages of participants notified by multiple or single sources by period of birth.



Overall, considering the n=1,870 children with diagnosis of CP born between 1981 and 2008 who were not deceased before age 2 years, and excluding 127 cases that have moved out of Northern Ireland and 12 that have been lost to follow up, a total of 1,665 out of 1,731 (96%) had been assessed using a standardised assessment form. Percentages of cases with completed standardised assessment by period of birth are reported in Figure 3. In each period of birth considered, completion of standardised assessment is within the target of 92% ascertainment rate.

Figure 3. Percentages of cases with completed standardised assessment form by period of birth. The 92% threshold of cases assessed using the standardised assessment form is indicated by the red line across bars.

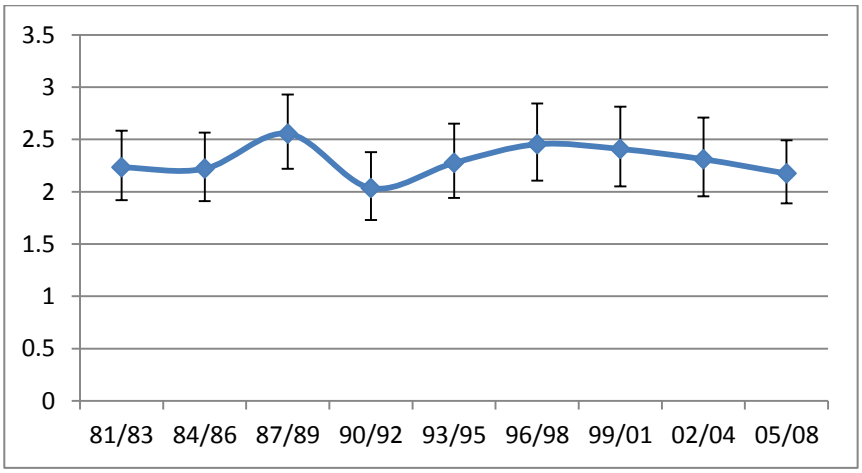


CP among children born in Northern Ireland: Overall prevalence

Prevalence of CP has been calculated considering the number of cases diagnosed with congenital CP and the total of live births in NI during the same period. Data on the number of live births in NI are provided by the Northern Ireland Statistics and Research Agency (NISRA, 2015). The total of CP cases born in the period 1981 – 2008, excluding n=126 with acquired CP and n=172 who were born outside NI was 1,591 (see Figure 1). Considering a total of 694,688 live births in the same period, this provides a prevalence of 2.29 CP cases per 1,000 live births (*95% CI* 2.17 to 2.41). Among these 1,591 cases with CP are also included n=19 cases who died before reaching age 2 years and 78 cases whose diagnosis had not yet being confirmed by age 5. Therefore their inclusion may result in a slight overestimation of CP prevalence.

In Figure 4 we report the prevalence of CP by birth period. Despite small variations across periods, the prevalence of CP over time does not change remarkably. On average, there were approximately 57 cases of congenital CP diagnosed every year during the period considered.

Figure 4. Prevalence of CP per 1,000 live births by birth period. Estimates and 95% CI intervals are reported (Poisson exact estimates).



In Table 1 we also report the prevalence of CP by Health and Social Care Trust (HSCT) at birth. Infants whose affiliation to one HSCT at the time of birth was unknown are excluded from these statistics: Health and Social Care Trust at birth was unknown for 64 out of 1,591children (roughly 4%). Overall prevalence of CP did not vary largely across Trusts.

Table 1. Prevalence (Pr) of CP per 1,000 live births by Health and Social Care Trust at birth

Birth period	Health and Social Care Trust										Total
	BHSCT		SEHSCT		SHSCT		NHSCT		WHSCT		
	Pr	Freq	Pr	Freq	Pr	Freq	Pr	Freq	Pr	Freq	
81/83	2.43	40	2.33	31	1.63	26	1.97	38	2.74	44	**181**
84/86	2.17	38	2.18	30	2.16	35	2.16	42	2.13	34	**184**
87/89	2.44	42	2.28	31	2.97	47	2.66	51	2.18	33	**207**
90/92	2.29	38	1.84	25	1.72	26	1.91	35	2.22	31	**158**
93/95	2.15	32	2.43	31	2.33	33	2.1	37	2.05	27	**165**
96/98	2.44	35	2.46	32	2.14	30	2.22	39	2.81	37	**177**
99/01	2.45	31	2.06	25	1.8	24	2.45	40	2.6	31	**160**
02/04	2.89	35	2.72	32	1.55	21	1.82	30	1.83	21	**151**
05/08*	2.13	37	2.3	40	1.83	38	2.06	49	1.41	23	**208**
Total	**328**		**277**		**280**		**361**		**281**		**1,591**

Note: BHSCT=Belfast; SEHSCT=South East; SHSCT=South; NHSCT=North; WHSCT=West.

** Four years birth period.*

Sex of child

Overall, among children with congenital CP there was a majority of males (57%). A similar prevalence was also observed among children with acquired CP, whereby males accounted for 61% of the total of children with CP. Increased risk of CP for males is consistent with research conducted by other registers (e.g. Australian CP Register, 2013).

Gestational age and low-birth weight

Prematurity is associated with increased risk of CP (Tronnes, Wilcox, Lie, Markestad, Moster, 2014). Gestational age was known for 1,652 (88%) cases out of the 1,870 born between 1981 and 2008 included in analyses. In Table 2 we report the number and percentage of non-premature children (born at 37 weeks or more), moderately premature (born between 32 and 36 weeks) and very to extremely premature (born at less than 32 weeks). Considering that the rate of premature births is approximately 7% in England and Wales in 2005 (ONS, 2007), and assuming a similar rate of premature births would be observed in NI, results reported in Table 2 suggest a higher prevalence of premature children in children with CP compared to the general population.

Table 2. Frequencies and percentages of children with CP born 1981/2008 by gestational age.*

Gestational Age	Frequency (%)
37 weeks or more	959 (58.05)
32-36 weeks	278 (16.83)
less than 32 weeks	415 (25.12)
Total	**1,652**

**Percentages are calculated based on cases with valid information n= 1,652 (88.34% of the total 1,870). There were n=218 (11.66%) excluded because of unavailable information.*

Low birth weight (birth weight below 2,500 grams) is also a significant risk factor for CP. The proportion of low birth weight new-borns in more developed countries was estimated to be 7% in the year 2000 (UNICEF, 2004). Birth weight was known for 1,546 (82.67%) of the 1,870 included in analyses. The frequency of children in different categories of birth weight is reported in Table 3. Results indicate that approximately 43% of children with CP were born weighting less than 2,500 grams.

Table 3. Frequencies and percentages of children with CP born 1981/2008 by gestational age.*

Gestational Age	Frequency (%)
2,500 grams or more	878 (56.79)
1,500 – 2,499 grams	341 (22.06)
Less than 1,500 grams	327 (21.15)
Total	**1,546**

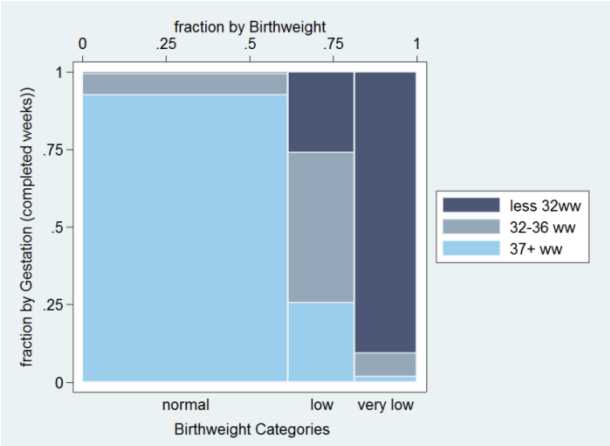
**Percentages are calculated based on cases with valid information n= 1, 546 (82.67% of the total 1,870). There were n=324 (17.33%) cases excluded because of unavailable information.*

Multiple births

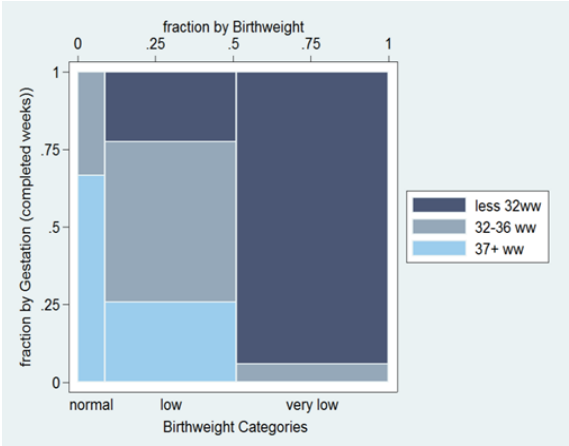
Multiple births have a higher risk of CP, although this increased risk may be related to higher risk of birth before term of multiple births (Topp et al., 2004). In the NICPR, information on multiple or single births is available for 1,691 (90.43%) cases out of 1,870 included in analyses. Overall, single births were the majority, with 1,535 (90.77%) out of 1,691 children being singleton while 156 (9.23%) were multiple births. The clustering of risk factors such as prematurity and low birth weight in single and multiple births is illustrated in Figure 5, showing the proportions of children in different categories of birth weight and gestational age by single or multiple births.

Figure 5. Proportions of children in categories of birth weight and preterm birth by single or multiple births. The areas represent respective prevalence of birth weight categories in singletons and multiple births.

Single births



Multiple births



Clinical subtypes

Clinicians provided information concerning the CP subtype of 1,745 cases (93.32%) from the 1,870 cases included in analyses. In Table 4 we report the prevalence of CP clinical subtypes. Overall, there was a higher number of children who fitted the definition of Spastic Bilateral CP subtype followed by Spastic Unilateral subtype. These two CP subtypes together represented 91.92% of the 1,745 cases considered in analyses.

Table 4: Frequencies and percentages of children by CP clinical subtypes*

CP subtype	Frequency (%)
Spastic Bilateral	901 (51.63)
Spastic Unilateral	703 (40.29)
Dyskinetic	83 (4.76)
Ataxic	46 (2.64)
Unclassifiable	12 (0.69)
Total	**1,745**

**Percentages are calculated based on cases with valid information n=1,745 (93.32% of the total 1,870). There were n=125 (6.68%) cases excluded because of unavailable information.*

In Table 5 we report the prevalence of CP subtypes by Health and Social Care Trust (HSCT) at time of birth. Overall, despite some variations, there are no significant differences in prevalence of CP subtypes across Trusts.

Table 5: Frequencies and percentages of CP clinical subtypes by Health and Social Care Trust*

	CP subtype					
HSCT	**Spastic bilateral**	**Spastic unilateral**	**Dyskinetic**	**Ataxic**	**Unclassifiable**	**Total**
BHSCT	173 (50)	145 (41.91)	17 (4.91)	9 (2.6)	2 (0.58)	**346**
SEHSCT	154 (54.42)	106 (37.46)	16 (5.65)	7 (2.47)	0 (0)	**283**
SHSCT	132 (45.83)	136 (47.22)	15 (5.21)	5 (1.74)	0 (0)	**288**
NHSCT	185 (51.25)	146 (40.44)	(3.88)	13 (3.6)	3 (0.83)	**361**
WHSCT	159 (53.36)	115 (38.59)	13 (4.36)	8 (2.68)	3 (1.01)	**298**
Born out	90 (57.32)	53 (33.76)	8 (5.1)	2 (1.27)	4 (2.55)	**157**
Total	**893 (51.53)**	**701 (40.45)**	**83 (4.79)**	**44 (2.54)**	**12 (0.69)**	**1,733**

Note: BHSCT=Belfast; SEHSCT=South East; SHSCT=South; NHSCT=North; WHSCT=West.

**Percentages are calculated based on cases with valid information n= 1, 733 (92.67% of the total 1,870). There were n=137 (7.33%) cases excluded because of unavailable information.*

Functional severity (GMFCS)

Among the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2, information concerning the level of functional ability as measured using the GMFCS was available for 1,764 cases (94.34%). In Table 6 we report the frequency and percentage of the five levels of functional severity for children in the NICPR. Most children (61% approximately) were considered to be in the first two GMFCS levels, which indicate less severe forms of impairment. Overall, approximately 29% of children with CP displayed more severe forms of functional impairment (Level IV and Level V).

*Table 6: Frequencies and percentages*of children by GMFCS level*

GMFCS	Freq (%)
Level I	330 (18.7)
Level II	750 (42.52)
Level III	168 (9.52)
Level IV	125 (7.09)
Level V	391 (22.17)
Total	**1,764**

**Percentages are calculated based on cases with valid information n= 1, 764 (94.34% of the total 1,870). There were n=106 (5.66%) cases excluded because of unavailable information.*

In Table 7 we also report GMFCS levels by Health and Social Trust at birth. Despite some variations across Trusts, the results reported do not indicate substantial differences in functional severity across Trusts, χ^2 (20) = 23.03, p = .29.

Table 7: Percentages of GMFCS level by Health and Social Trust of birth*

	GMFCS					
HSCT	**Level I**	**Level II**	**Level III**	**Level IV**	**Level V**	**Total**
BHSCT	69 (19.77)	145 (41.55)	25 (7.16)	24 (6.88)	86 (24.64)	**349**
SEHSCT	57 (20.07)	111 (39.08)	25 (8.80)	17 (5.99)	74 (26.06)	**284**
SHSCT	51 (17.35)	134 (45.58)	28 (9.52)	15 (5.1)	66 (22.45)	**294**
NHSCT	68 (18.99)	155 (43.3)	29 (8.10)	28 (7.82)	78 (21.79)	**358**
WHSCT	52 (17.51)	130 (43.77)	40 (13.47)	25 (8.42)	50 (16.84)	**297**
Born out	25 (15.82)	67 (42.41)	20 (12.66)	13 (8.23)	33 (20.89)	**158**
Total	**322 (18.51)**	**742 (42.64)**	**167 (9.6)**	**122 (7.01)**	**387 (22.24)**	**1,740**

Note: BHSCT=Belfast; SEHSCT=South East; SHSCT=South; NHSCT=North; WHSCT=West.

** Percentages are calculated based on cases with valid information n= 1, 740 (93.04% of the total 1,870). There were n=130 (6.96%) cases excluded because of unavailable information.*

In the next table (Table 8) we also report the functional severity (GMFCS) by primary informant of the case. There were some noticeable differences. In particular, paediatricians are more likely to report more severe cases of CP (GMFCS Level V).

*Table 8. Percentage of GMFCS by notifier***.**

	GMFCS						
Notifier	**Level I**	**Level II**	**Level III**	**Level IV**	**Level V**	**Missing**	**TOTAL**
Paediatrician	102 (15.05)	285 (42.04)	59 (8.7)	42 (6.19)	171(25.22)	19 (2.8)	**678**
Neonatologist	19 (17.43)	35 (32.11)	6 (5.5)	11 (10.09)	31(28.45)	7 (6.42)	**109**
Ortho /physio	82 (23.84)	124 (36.04)	27 (7.85)	26 (7.56)	69 (20.06)	16 (4.65)	**344**
S/CMOs	42 (12)	156 (44.57)	39 (11.14)	25 (7.14)	78 (22.28)	10 (2.86)	**350**
Other	85 (21.85)	150 (38.56)	37 (9.51)	21 (5.4)	42 (10.8)	54 (13.88)	**389**
Total	**330 (17.65)**	**750 (40.11)**	**168 (8.98)**	**125 (6.6)**	**391 (20.91)**	**106 (5.67)**	**1,870**

Note: Ortho= Orthopaedic surgeons; physio= physiotherapists; S/CMOS= senior medical officers

Association between clinical subtype and functional severity (GMFCS)

A cross-tabulation of CP subtypes and levels of function (GMFCS) in Table 9 indicates that the Spastic Bilateral subtype is associated with more severe forms of impairment. Conversely, children with Spastic Unilateral CP displayed less severe levels of functional impairment, with most of these children classified in Level I or Level II of the GMFCS. Children with Dyskinesia also displayed high levels of functional impairment, with approximately 63% of children in this clinical category classified at the highest level of GMFCS impairment. Compared to children with a diagnosis of Bilateral Spastic CP sub-type, children with a diagnosis of Spastic Unilateral CP displayed a 40-fold increase in the odds of being classified at the less severe levels of impairment, Level I or Level 2 of the GMFCS (OR=40.7, 95%CI 27.7 to 60.0).

Table 9: Frequencies and percentages of GMFCS by CP subtype*

	GMFCS					
CP Subtype	**Level I**	**Level II**	**Level III**	**Level IV**	**Level V**	**Total**
Spastic Bilateral	67 (7.46)	264 (29.4)	134 (14.92)	107 (11.92)	326 (36.3)	**898**
Spastic Unilateral	241 (34.48)	434 (62.09)	16 (2.29)	4 (0.57)	4 (0.57)	**699**
Dyskinetic	1 (1.27)	10 (12.66)	9 (11.39)	9 (11.39)	50 (63.29)	**79**
Ataxic	12 (26.09)	24 (52.17)	4 (8.70)	3 (6.52)	3 (6.52)	**46**
Unclassifiable	2 (20.00)	4 (40.00)	2 (20.00)	1 (10.00)	1 (10.00)	**10**
Total	**323 (18.65)**	**736 (42.49)**	**165 (9.53)**	**124 (7.16)**	**384 (22.17)**	**1,732**

**Percentages are calculated based on cases with valid information n= 1, 732 (92.62% of the total 1,870). There were n=138 (7.38%) cases excluded because of unavailable information.*

Survival

Among the 1,889 children with CP born between 1981 and 2008 (including those that passed away before age 2), 184 (9.74%) had been reported to be deceased. In Table 9 we report the percentages of children that have died by Health and Social Care Trust. Overall, the survival rate of children with CP did not vary to a significant degree across Trusts, $\chi^2(6) = 6.89$, $p = .33$.

Table 10: Frequencies and percentages of deceased children with CP by Health and Social Care Trust of Birth*

HSCT Trust	Died (%)	Total
BHSCT	41 (11.42)	**359**
SEHSCT	34 (11.49)	**296**
SHSCT	32 (10.49)	**305**
NHSCT	37 (9.76)	**379**
WHSCT	25 (8.22)	**304**
Born out	11 (6.04)	**182**
Total	**180 (9.86)**	**1,825**

**Percentages are calculated based on cases with valid information n= 1, 825 (96.61% of the total 1,889). There were n=64 (3.39%) cases excluded because of unavailable information.*

In Table 11 we report the percentage of children who died by CP subtype. The results indicate that a higher percentage of those children who had deceased had a diagnosis of Spastic Bilateral CP subtype. Overall, approximately 84% of children in the Spastic Bilateral subtype survived by December 2014, compared to approximately 99% of those in the Spastic Unilateral, 89% of those in the Dyskinetic CP subtype and 96% of those in the Ataxic subtype. The difference in frequencies of deceased between the Spastic Bilateral and Spastic Unilateral subtypes was significant, $\chi^2(1) = 106$, $p < .001$.

Table 11. Percentages of children who survived and died by CP subtype*

CP subtype	Alive (%)	Died (%)	Total
Spastic Bilateral	764 (83.5)	151 (16.50)	**915**
Spastic Unilateral	696 (98.86)	8 (1.14)	**704**
Dyskinetic	74 (89.16)	9 (10.84)	**83**
Ataxic	44 (95.65)	2 (4.35)	**46**
Unclassifiable	12 (92.31)	1 (7.69)	**13**
Total	**1,590 (90.29)**	**171 (9.71)**	**1,761**

**Percentages are calculated based on cases with valid information n= 1, 761 (93.22% of the total of 1,889). There were n=128 (6.78%) cases excluded because of unavailable information.*

In Table 12 we also report the percentage of children alive and who have died by functional severity level (GMFCS). Overall, children in the most severe level of functional impairment (Level V) displayed a reduced likelihood of surviving compared to children at less severe levels.

Table 12. Percentages of children who survived and died by GMFCS level*

GMFCS	Alive (%)	Died (%)	Total
Level I	329 (99.4)	2 (0.6)	**331**
Level II	742 (98.67)	10 (1.33)	**752**
Level III	168 (100)	0 (0)	**168**
Level IV	120 (96)	5 (4)	**125**
Level V	249 (61.79)	154 (38.21)	**403**
Total	**1.638 (92.07)**	**141 (7.93)**	**1,779**

**Percentages are calculated based on cases with valid information n= 1, 779 (94.18% of the total of 1,889). There were n= 110 (5.82%) cases excluded because of unavailable information.*

Whenever information about the cause of the death cited "Cerebral Palsy", it was often referred to as the principal cause of death. However, information on the cause of death is missing for most of the cases that had been reported to have deceased (62%).

Acquired CP

Among the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2, a total of 126 (6.7%) were reported to have acquired CP. Clinicians and other informants are also requested to provide information about the causes of acquired CP. Informants reported several different causes, but in approximately 12% of cases, the cause of acquired CP is not known or has not been identified. In approximately 17% of the cases with acquired CP, the main cause reported is some form of head injury. In 15% of the cases, the principal cause of acquired CP reported is meningitis, and a further 5% whereby the principal cause of acquired CP reported is herpes infection.

Other Diagnoses

Information is also collected on other diagnosis of children with CP. The most commonly reported diagnoses together with CP are listed in Table 13. Information on further diagnoses is qualitative (i.e. open field for practitioners), therefore results reported here need to be considered with caution as they represent a first screening and might need further, more in-depth analyses.

Table 13. Other diagnosis reported (N=1,870). Please note these categories are not mutually exclusive

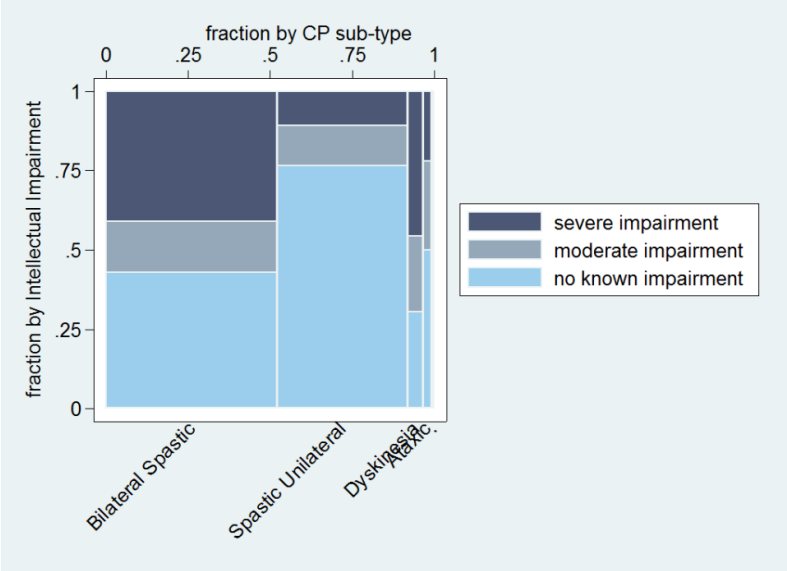
Diagnosis	Frequency (%)
Aggression or conduct problems	53 (2.83)
General Developmental Delay	17 (0.91)
Autism	19 (1.02)
Attention deficit and/or hyperactivity	12 (0.64)
Kidney problems	9 (0.48)
Pneumonia or pulmonary problems	38 (2.03)
Asthma	54 (2.89)

Intellectual impairment

Information on intellectual function was available for 1,747 children (93.42%) of the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2. Overall, 764 of these children (43.73% of n=1,747) were reported to display some level of intellectual impairment, of whom 493 (64% of those with some intellectual impairment) were reported to display severe intellectual delay.

In Figure 6 we report the proportions of children with moderate and severe intellectual impairment by CP subtype. The results suggest that severe intellectual impairment is more prevalent in the Spastic Bilateral and the Dyskinetic subtypes compared to Spastic Unilateral CP.

Figure 6. Proportion of intellectual impairment level by CP subtype: CP subtype area represents the prevalence of each subtype respectively.



In Table 14 we also report the percentage of children with different levels of intellectual impairment by functional severity (GMFCS). The results indicate a strong association between functional impairment severity and intellectual impairment.

Table 14. Frequencies and percentages of children with different levels of intellectual impairment by GMFCS level*

	Intellectual impairment			
GMFCS	**No**	**Moderate**	**Severe**	**Total**
Level I	249 (78.3)	51 (16.04)	18 (5.66)	**318**
Level II	522 (72)	103 (14.21)	100 (13.79)	**725**
Level III	101 (60.12)	42 (25)	25 (14.88)	**168**
Level IV	47 (38.52)	27 (22.13)	48 (39.34)	**122**
Level V	39 (10.16)	48 (12.5)	297 (77.34)	**384**
Total	**958 (55.77)**	**271 (15.78)**	**488 (28.42)**	**1,717**

**Percentages are calculated based on cases with valid information n= 1,717 (91.82% of the total of 1,870). There were n= 153 (8.18%) cases excluded because of unavailable information.*

Epilepsy

Information on seizures was reported for 1,592 children (85.13%) of the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2. Overall, 681 (42.78% of n= 1,592) were reported to have seizures in the last year or in the past. The presence of seizures was more prevalent in Spastic Bilateral and Dyskinetic subtypes compared to Spastic Unilateral CP, as indicated in Table 15. Reports of seizures were also more common among children in the more severe levels of functional impairment (Level IV and Level V), compared to other levels of the GMFCS.

Table 15. Frequencies and percentages of children with report of seizures (recent or ever) by CP subtype and GMFCS.*

CP subtype	Ever seizures		Total
	Yes (%)	No (%)	
Spastic Bilateral	407 (49.76)	411 (50.24)	**818**
Spastic Unilateral	200 (32.05)	424 (67.95)	**624**
Dyskinetic	45 (59.21)	31(40.79)	**76**
Ataxic	16 (38.10)	26 (61.9)	**42**
Unclassifiable	2 (20)	8 (80)	**10**
Total	**670 (42.68)**	**488 (28.42)**	**1,570**

**Percentages are calculated based on cases with valid information n= 1, 570 (83.96% of the total of 1,870). There were n=300 (16.04%) cases excluded because of information unavailable.*

GMFCS	Ever seizures		Total
	Yes (%)	No (%)	
Level I	83 (29.02)	203 (70.98)	**286**
Level II	199 (30.11)	462 (69.89)	**661**
Level III	52 (33.99)	101 (66.01)	**153**
Level IV	51 (45.95)	60 (54.05)	**111**
Level V	292 (79.13)	77 (20.87)	**369**
Total	**677 (42.680**	**903 (57.15)**	**1,580**

**Percentages are calculated based on cases with valid information n= 1, 580 (84.49% of the total of 1,870). There were n=290 (15.51%) cases excluded because of unavailable information.*

Visual impairment

There was information on visual impairments for 1,849 (98.88%) of the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2. In total 513 children (27.74% of n= 1,849) were reported to display some visual impairment. Visual impairments were more prevalent among children with Bilateral and Dyskinetic CP, as indicated in Table 16. Visual impairments were also more prevalent in children with more severe levels of functional impairment (GMFCS), as indicated also in Table 16.

Table 16. Frequencies and percentages of children with a report of visual impairment by CP subtype and GMFCS.*

CP subtype	Visual impairment		Total
	Yes (%)	No (%)	
Spastic Bilateral	340 (37.82)	559 (62.18)	**899**
Spastic Unilateral	126 (17.97)	575 (82.03)	**701**
Dyskinetic	26 (31.71)	56 (68.29)	**82**
Ataxic	8 (17.39)	38 (82.61)	**46**
Unclassifiable	3 (25)	9 (75)	**12**
Total	**503 (28.91)**	**1,237 (71.09)**	**1,740**

**Percentages are calculated based on cases with valid information n= 1, 740 (93.05% of the total of 1,870). There were n=130 (6.95%) cases excluded because of unavailable information.*

GMFCS	Visual impairment		Total
	Yes (%)	No (%)	
Level I	47 (14.42)	279 (85.58)	**326**
Level II	164 (22.07)	579 (77.93)	**743**
Level III	55 (32.93)	112 (67.07)	**167**
Level IV	44 (35.2)	81 (64.8)	**125**
Level V	199 (50.9)	192 (49.1)	**391**
Total	**509 (29.05)**	**1,243 (70.95)**	**1,752**

**Percentages are calculated based on cases with valid information n= 1, 752 (93.69% of the total of 1,870). There were n=118 (6.31%) cases excluded because of unavailable information.*

Hearing impairments

Information on hearing impairments was available for 1,734 (92.73%) of the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2. In total, 134 children (7.73% of n= 1,734) were reported to present some hearing impairment. Hearing impairments were reported more frequently among children with Dyskinetic CP, as illustrated in Table 17. Hearing impairments were more frequent for children in the most severe level of GMFCS functional impairment, as also illustrated in Table 16.

Table 17. Frequencies and percentages of children with hearing impairment by CP subtype and GMFCS.*

CP subtype	Hearing Impairment		Total
	Yes (%)	No (%)	
Spastic Bilateral	77 (8.7)	808 (91.3)	**885**
Spastic Unilateral	32 (4.68)	652 (95.32)	**684**
Dyskinetic	13 (15.85)	69 (84.15)	**82**
Ataxic	5 (11.63)	38 (88.37)	**43**
Unclassifiable	2 (16.67)	10 (83.33)	**12**
Total	**129 (7.56)**	**1,577 (92.44)**	**1,706**

**Percentages are calculated based on cases with valid information n= 1, 706 (91.23% of the total of 1,870). There were n=164 (8.77%) cases excluded because of unavailable information.*

GMFCS	Hearing Impairment		Total
	Yes (%)	No (%)	
Level I	17 (5.38)	299 (94.62)	**316**
Level II	48 (6.63)	676 (93.37)	**724**
Level III	5 (2.98)	163 (97.02)	**168**
Level IV	9 (7.32)	114 (92.68)	**123**
Level V	53 (13.77)	332 (86.29)	**385**
Total	**132(7.69)**	**1,584 (92.31)**	**1,716**

**Percentages are calculated based on cases with valid information n= 1, 716 (91.76% of the total of 1,870). There were n=154 (8.24%) cases excluded because of unavailable information.*

Communication impairments

Clinicians provided information on communication impairments of 1,745 (93%) out of 1,870 included in analyses. In total, 783 children (42% of n=1,870) were reported to display some communication impairment. Communication impairments were reported more frequently among children with Dyskinetic CP, as illustrated in Table 17. Similar problems were however more frequent among children with Spastic Bilateral CP compared to children with Spastic Unilateral CP. Problems in communication capacities were more prevalent among children at the most severe level of GMFCS functional impairment, as illustrated also in Table 18.

Table 18. Frequencies and percentages of children with communication impairments by CP subtype and GMFCS.*

CP subtype	Communication impairments		Total
	Yes (%)	No (%)	
Spastic Bilateral	513 (57.77)	375 (42.23)	**888**
Spastic Unilateral	154 (22.32)	536 (77.68)	**690**
Dyskinetic	72 (88.89)	9 (11.11)	**81**
Ataxic	22 (50)	22 (50)	**44**
Unclassifiable	7 (58.33)	5 (41.67)	**12**
Total	**768 (44.78)**	**947 (55.22)**	**1,715**

**Percentages are calculated based on cases with valid information n= 1,715 (91.71% of the total of 1,870). There were n=155 (8.29%) cases excluded because of information unavailable.*

GMFCS	Communication impairments		Total
	Yes (%)	No (%)	
Level I	64 (19.88)	258 (80.12)	**322**
Level II	206 (28.14)	526 (71.86)	**732**
Level III	67 (40.61)	98 (59.39)	**165**
Level IV	76 (62.3)	46 (37.7)	**122**
Level V	364(94.3)	22 (5.7)	**386**
Total	**777 (44.99)**	**950 (55.01)**	**1,727**

**Percentages are calculated based on cases with valid information n= 1, 727 (92.35% of the total of 1,870). There were n=143 (7.65%) cases excluded because of information unavailable.*

Feeding problems

There was information available on feeding problems of 1,629 (87.12%) of the 1,870 children with CP born between 1981 and 2008 and who had not passed away before reaching age 2. In total, 399 children (24.49% of n= 1,629) were reported to display different feeding problems. Feeding problems were reported more frequently among children with Dyskinetic CP, as illustrated in Table 19. However, feeding problems were more frequent among children with Spastic Bilateral CP compared to those with Spastic Unilateral CP. Feeding problems were more frequent for children at the most severe level of GMFCS functional impairment, as also illustrated in Table 19.

Table 19. Frequencies and percentages of children with feeding problems by CP subtype and GMFCS.*

CP subtype	Feeding problems		Total
	Yes (%)	No (%)	
Spastic Bilateral	301 (35.83)	539 (64.17)	**840**
Spastic Unilateral	37 (5.79)	602 (94.21)	**639**
Dyskinetic	50 (69.44)	22 (30.56)	**72**
Ataxic	3 (7.14)	39 (92.86)	**42**
Unclassifiable	2 (18.18)	9(81.82)	**11**
Total	**393 (24.5)**	**1,211 (75.5)**	**1,604**

**Percentages are calculated based on cases with valid information n= 1, 604 (85.77% of the total of 1,870). There were n=266 (14.23%) cases excluded because of information unavailable.*

GMFCS	Feeding problems		Total
	Yes (%)	No (%)	
Level I	18 (6)	287 (94)	**300**
Level II	55 (8.1)	624 (91.9)	**679**
Level III	20 (12.27)	143 (87.73)	**163**
Level IV	35 (29.66)	83 (70.34)	**118**
Level V	268 (75.71)	86 (24.29)	**354**
Total	**396 (24.54)**	**1,218 (75.46)**	**1,614**

**Percentages are calculated based on cases with valid information n= 1, 614 (86.31% of the total of 1,870). There were n= 256 (13.69%) cases excluded because of information unavailable.*

Conclusions

The NICPR provides valuable information on children with CP in Northern Ireland, their impairments and their needs. The NICPR has surveyed the country population for one of the longest periods compared to many other CP registers in Europe. Furthermore, this register is one of the only two CP registers still active in the whole of the UK. For these reasons, the NICPR is in an excellent position to meet the needs of surveillance and public health activities, and to serve as the basis for aetiological, epidemiological and clinical research.

In this report, we have provided evidence concerning the rate of CP children in Northern Ireland. While the overall rate of CP has not changed dramatically over time, the results suggest associations between CP and known risk factors such as gender, pre-term birth and multiple births. These risk factors for CP have been highlighted by previous research using data collected by the NICPR, as well as data collected by other registers.

The results reported in this report also provide an initial picture of the functional impairment of children with CP. Furthermore, they also provide information on other impairments affecting children with CP, such as intellectual, visual, and communication impairments. These impairments seem to be more prevalent and more severe in some types of CP and more severe levels of functional ability impairment (GMFCS).

We hope that the results reported here will help provide an idea of the wealth and depth of data collected by the NICPR and their possible uses for surveillance, public health and social care, and research.

Research and Collaborations

The NICPR is committed to facilitating high quality research and producing quality outputs (see Appendix 4). In this section, we highlight some of the recent and on-going collaborations and research contributions of the NICPR. These collaborations are expected to lead to high-quality outputs, which have the potential of significant impact. With the help of professionals, parents and end-users in the Advisory Committee, we will also ensure that pathways to dissemination and impact of research outputs are identified.

Surveillance of Cerebral Palsy in Europe (SCPE)

SCPE is a collaboration of registers of children with Cerebral Palsy (CP) that formed in 1998 and since then has undertaken successive programmes of work funded by the European Union (EU).The work has brought together paediatricians, paediatric neurologists, epidemiologists and therapists from across Europe.

The aim of the SCPE network is to:

1. Disseminate knowledge about cerebral palsy through epidemiological data;

2. Develop best practice in monitoring trends in cerebral palsy;

3. Raise standards of care for children with cerebral palsy.

CP occurs in about two babies per 1000 live births across Europe. It is important to measure trends in prevalence over time and to understand better the causes of the Cerebral Palsy. Registers of all children born in a region with CP can help with this. They can also help with planning services and allow parents of children with Cerebral Palsy to find more information about research on this condition.

An SCPE common database has now been set up which has more than 15,000 CP children born from 1975 to 2004. The SCPE common database contains information on demographic characteristics (although anonymous), description of the CP type, associated impairments, and also information on associated co-morbidity such as congenital anomalies and perinatal characteristics. Overall 63 variables with an agreed standard definition are regularly collected from participating registers across Europe. In each centre, data have been collected on CP children as well as on denominators (population statistics).

One of the contributions of this network lies in providing a consensus on definitions, classifications and inclusion criteria of CP. Classification systems based on clinical findings are currently the most widely used. Drawing on published work, SCPE has classified CP into three main groups, spastic, ataxic and dyskinetic CP. Agreement has been reached on the clinical findings associated with each classification sub-group as follows:

Spastic CP is characterised by at least two of:

- Abnormal pattern of posture and/or movement;
- Increased tone (not necessarily constantly);
- Pathological reflexes (hyper-reflexia or pyramidal signs e.g. Babinski response);
- It may be unilateral (hemiplegia) or bilateral.

Ataxic CP is characterised by both of:

- Pattern of posture and/or movement;
- Loss of orderly muscular co-ordination so that movements are performed with abnormal force, rhythm and accuracy.

Dyskinetic CP is characterised by both of:

- Abnormal pattern of posture and/or movement;
- Involuntary, uncontrolled, recurring, occasionally stereotyped movements of affected body parts;
- Dyskinetic CP may be either ;
- Dystonic CP, dominated by both hypokinesia and hypertonia;
- Choreo-athetotic CP, dominated by both hyperkinesia and hypotonia.

The SCPE network has also contributed to a Reference and Training manual aimed at a vast audience. The aim of the SCPE Reference and Training manual is to promote a shared understanding of the words and phrases used to describe the clinical, functional and neurological features of CP. Text and video material are used to illustrate these features and discuss pitfalls in diagnosis and classification. The Reference and Training manual is for health professionals who are compiling CP registers and those wishing to enter data onto the SCPE database and for training purposes for health professionals interested in children with CP. This can be accessed online at http://www.scpenetwork.eu/en/rtm

Finally, the SCPE network has also contributed to a number of research papers, to which the NICPR and its staff have provided a pivotal contribution (see Appendix 4 for a list of publications).

Study of Participation of Children with Cerebral Palsy Living in Europe (SPARCLE)

SPARCLE 1 investigated the influence of the environment on the participation and quality of life of children with CP in seven EU countries. It was funded by the European Commission Research Framework 5 Programme. It started on 1st October 2002 and continued until December 2006.

SPARCLE 1 investigated those aspects of the environment that play a major role in determining the participation and quality of life of children with CP. The information has contributed to inform EU policies.

Building on the success of SPARCLE 1, the research team was able to obtain funding for SPARCLE 2, a study to investigate the factors that promote the quality of life and participation of young people with CP. In particular the study examined whether there are aspects of the lives of the children aged 8-12 years and their families in SPARCLE 1 which influenced quality of life at age 13-17. For example,

did stress in a family persist and still influence quality of life? Did pain persist and still influence quality of life?

SPARCLE 1 and 2 have been co-ordinated by the study centre at Newcastle University in collaboration with six further European countries (Denmark, France, Ireland, Italy, Germany and Sweden).

The study has produced a number of papers published in high-impact journals. Reports that concern more specifically data collected in Northern Ireland include (see Appendix 4):

- Madden A & Parkes J. Impact of learning disability on the health, participation and quality of life of children with cerebral palsy. *Learning Disability Practice, 2010, December, Vol 13*, No 10, p28-33.
- Parkes J, McCullough N & Madden A. Participation of children with cerebral palsy: a population-based study. *Health & Social Care in the Community, 2010, May;18 (3):*304-15.
- Parkes J, McCullough N, Madden A & McCahey E. The health of children with cerebral palsy and stress in their parents: a population-based survey. *Journal of Advanced Nursing, 2009, Nov, 65 (11)*: 2311-23. Epub 2009 Sep 8.

Ongoing research

Transition Study

Transition is a programme of research that aims to address this question: How can health services contribute most effectively to facilitating successful transition of young people with complex health needs from childhood to adulthood?

Northumbria Healthcare NHS Foundation Trust and Newcastle University have developed a research programme to answer this question. The programme involves further NHS Trusts and the voluntary sector.

The Transition Research Programme is independent research funded by the National Institute for Health Research (NIHR) under its Programme Grants for Applied Research scheme (RP-PG-0610-10112). NICPR is involved in this study in a collaborator role and by providing a sampling framework for the inclusion of children with CP in the study.

The overall purpose of the Research Programme is to provide evidence to help NHS Commissioners and Trusts allocate resources to facilitate successful transfer of young people with complex health needs from child to adult services. The Programme will identify a small number of key components of transition arrangements for which there is an evidence base, and for which there are outcome measures against which implementation of these components can be judged, using an audit tool, in NHS environments.

'Transition' is the purposeful, planned process that addresses the medical, psychosocial and educational/vocational needs of adolescents and young adults with chronic physical and medical conditions as they move from child-centred to adult oriented health care systems.

Young people with "complex health needs" are those with a physical, mental or health impairment that has the potential for a substantial and long-term adverse effect on their ability to carry out

normal day-to-day activities. Importance of the topic Transition is a challenge and a government priority area.

The National Service Framework recognises that some disabled young people are transferred to adult services with inadequate care plans. Another report shows that transition contributes to poorer health outcomes because young people lose simultaneously the continuity of children's health services and school support.

The Kennedy Report, September 2010, in Recommendation 32 stated: "Arrangements must be agreed, regarding funding and other matters, to address the changing needs of children and young people as they mature, including greater continuity of care into adulthood. Ensuring a smooth transition between children's and adults' services should be a priority for local commissioners."

The Programme has three objectives:

1) To work with young people with complex health needs to determine what successful transition means to them and what is important in their transitional care;

2) To identify the features of transitional care which are effective and efficient;

3) To determine how transitional care should be organised, provided and commissioned.

Three broad groups of young people are being addressed, those with Neurodevelopmental disorders, such as Autism Spectrum Disorder; Chronic illness such as diabetes; Complex physical problems such as CP.

More information can be found at: http://research.ncl.ac.uk/transition/

Collaborative Cerebral Palsy (CP) and Congenital Anomalies (CAs) project: Europe & Australia

This is a spin-off ongoing collaboration from the SCPE (described above). The purpose is to work collaboratively on analyses using data from registers in Europe and Australia to investigate the following research questions:

1. What is the frequency and proportion of non-cerebral CAs found in CP?

2. How accurately can the timing of non-cerebral CAs in CP be identified?

3. Can a relationship between the timing of non-cerebral CAs and the timing of injury responsible for CP be identified?

4. What are the clinical outcomes (type of CP and severity) of CP cases with non-cerebral CAs? Do outcomes vary by type of non-cerebral CA?

5. What is the risk of CP in infants with different types of non-cerebral CAs?

A series of meetings have taken place which have contributed to refining the research questions and methods.

A comparison of cerebral palsy rate in singleton, twin and multiple births; an internal register-based study

This is a collaboration between Oliver Perra (QUB; NICPR), Guiomar Garcia Jalon (QUB; NICPR) and staff from the University of Newcastle (Prof Judith Rankin, Prof Allan Colver, D r Karen Horridge) to conduct analyses on data collected by SCPE. The aims of the proposed study are: (1) to describe changes in CP risk between singleton and multiple births; (b) to investigate the pre- and peri-natal factors that moderate CP risk in singletons and multiple births; (c) to compare CP type and severity between singletons and multiple births and whether these have changed over time.

Exploring the problem of pain in the Cerebral Palsy population: Piloting a big data approach

This is a project funded by the Improving Children's Lives small grant scheme, Queen's University Belfast. This is a collaboration between Guiomar Garcia Jalon (QUB; NICPR), Oliver Perra (QUB; NICPR), and other QUB staff (Dermot O'Reilly, Centre for Public Health; Allen Thurston, School of Education; Anna Gavin, NI Cancer Registry).

The aim of this case control study is to explore the potential of data linkage between NICPR and Business Service Organisation-Honest Brokers Service (BSO-HBS) to investigate the prevalence and significance of pain in the CP population. Method: a case control study where data will be obtained by linking the NICPR database and information held by BSO-HBS including Enhanced Prescribing database, Index deprivation scores, Settlement bands, the Family Practitioner Services and Health and Care Index. The sample will include cases in the NICPR born. Information for age and sex matched samples in the general population will also be requested for comparison.

Principal research question:
Is data linkage a viable and reliable method to assess the prevalence of pain in the CP population using information on prescribed pain medication as a proxy?
The principal hypothesis is that people with CP are more affected by pain than the general population. Therefore we want to assess if there are differences in prescription of pain medication between people with CP and others while controlling for potentially confounding factors, for example GP practice.

Secondary research questions:

- Can the results obtained from linking data between NICPR and the BSO-HBS be compared to the results of the SPARCLE studies regarding prevalence of pain in the NI sample?
- What is the prevalence of pain medication use in the CP population in comparison to age and sex matched controls from the general population?
- If data linkage between NICPR and BSO-HBS Enhanced Prescription Database is a viable and reliable method to assess the prevalence of pain in the CP population:
    a. What is the prevalence of pain in the CP population in comparison to age and sex matched controls from the general population?
    b. Is there an association between pain and CP clinical presentation, e.g. severity, CP subtype, associated impairments?
- Are there differences in prescription of pain medication that can be ascribed to differences across GP practices, geographical areas, settlement band, distance from services, deprivation measure or other.

Recent Collaborations with local clinicians - Audits

The NICPR also plays a pivotal role in facilitating audits. The NICPR provides the sample frame for audits involving children, young people and adults with Cerebral Palsy (CP) using internationally agreed inclusion criteria and definitions of CP. This facilitates and enables the work of practitioners that require information on persons with a diagnosis of CP by ensuring that cases with CP have been screened and ascertained using robust criteria. The fact that these criteria are internationally recognised also facilitates the task of comparing data and information collected across Trusts in Northern Ireland, and provides information that could be compared with that collected in other countries in the UK and beyond.

Furthermore, the NICPR provides information on the abilities and level of impairment of this population. This also represents an important contribution to the work of practitioners, insofar as information pivotal to assessment and evaluation of functional abilities and quality of life of this population is available in a single repository that updates, monitors and validates it. Information on functional abilities of children with CP is validated using clinical tools that have been designed and developed internationally, and that therefore facilitate comparisons across countries. The NICPR has also contributed to the design and development of these instruments, for example through its established collaboration with the Surveillance of CP in Europe (SCPE) Network.

The NICPR also provides background information on persons with a diagnosis of CP (e.g. gestational age). The fact that this information is collated and validated in a single repository can also facilitate the completion of audits, for example by allowing selection of cases based on criteria.

The NICPR staff provides consultation on different aspects of an audit, such as sampling frame and criteria, revision, provision of clinically-relevant information, and so on. We hope in this way to support one of the key aims of the NICPR, which involves public health activities.

In what follows, we report some information on recent audits.

Audit considering the medical management of spasticity with Cerebral Palsy

Chief investigator: Dr Claire Watterson, ST4 Paediatrics Western Health and Social Care Trust

Other investigators: Dr Jenny Fairfield, Consultant Paediatrician Western Health and Social Care Trust

Protocol

Aim: This retrospective audit aims to assess current medical management, specifically oral drugs, of spasticity in a random sample of children with a diagnosis of CP in the Western Health and Social Care Trust

Background: CP is the most common condition associated with spasticity in children. The prevalence in the UK is 186 per 100,000 population, with a total of 110,000 people affected. The impact of spasticity and co-existing motor disorders and their early MSK complications varies.

Common problems affecting this population are impaired motor function affecting the person's ability to participate in society; muscle spasms causing pain; motor development delay; complications secondary to spasticity such as contractures leading to difficulties with daily care.

Methods.

Sample: We performed a search of children from Bridgeview House with a diagnosis of CP using the Northern Ireland Cerebral Palsy Register (NICPR). Twenty two children were selected randomly (<18yrs old) and a data collection form was completed for each patient.

Criteria/standards being used: NICE Clinical Guideline CG145; Spasticity in children and young people with non-progressive brain disorders: Management of spasticity and co-existing motor disorders and their early musculoskeletal complications (2012).The NICE clinical audit tool specifically looking at oral drugs was used as a framework.

Results: Within the group affected by spasticity, 40% had documented functional disability only without pain or muscle spasms. It was unclear from the majority of the notes if Baclofen was considered necessary in these cases. Some of the cases had documented GMFCS 1 and therefore not commencing Baclofen was the appropriate decision.

Was oral Diazepam considered? Oral Diazepam was not documented as being considered appropriate first line therapy for any patient with spasticity. If oral Diazepam was used initially, was Baclofen considered for long-term treatment? N/A: Oral Diazepam was never used initially.

Was Baclofen introduced using a step-wise approach? Yes in 100% of cases.

Was review of spasticity agent adequate? The patients taking Benzhexol were reviewed in 100% of cases <6months. One patient's treatment had started in RBHSC. When stopping Diazepam/Baclofen was the dose reduced in stages? No, patients included in this audit had ever had Baclofen discontinued either in the past or present.

Conclusions: Questions regarding whether the NICPR has a complete record of the patients diagnosed with CP in the Western Trust. It was unclear from the notes whether patients suffered from pain, muscle spasms or functional disability as it was not recorded. These problems should be routinely assessed in OPD. Sleep and posturing behaviours should also be documented. It is also recommended using the paediatric pain profile. Regarding oral medication treatment for spasticity, the advantages and disadvantages of using Diazepam as recommended by NICE should be considered. Finally, patients who started taking Baclofen were not reviewed within the time limit the NICE guidelines recommend.

Audit on the use of cranial ultrasound and magnetic resonance imaging in children with Cerebral Palsy

Chief investigator: Dr Janice Bothwell, Consultant Paediatrician Belfast Health and Social Care Trust

Other investigators: Dr Moira Steward, Dr Steven McIstry, Dr Paul Burns, Dr BenWatson, Belfast Health and Social Care Trust

Protocol

Aim and objective: To assess the use of neuroimaging with a focus on cranial ultrasound (US) cerebral magnetic resonance imaging (MRI) in children with Cerebral Palsy (CP) in the Belfast Trust.

Objectives:

1. Identify children born between 01/01/00 and 31/12/08 inclusive in the Belfast Trust with a diagnosis of CP.

2. Identify which of these children had serial cranial US and cerebral MRI performed.

3. Identify timing/age of cranial US and cerebral MRI performance.

4. Classification of cranial US and cerebral MRI findings.

Background: Neuroimaging has expanded exponentially over the last number of years with magnetic resonance scanning becoming more widely available. The use of MRI to assist in identifying aetiology is recommended as part of the assessment and management of children with CP. (Ashwal et al 2004). The worldwide incidence in CP is 2 – 2.5 per 1000 live births rising to 40 – 100 per 1000 in infants born very early or with very low birth weight. This translates to 52 births of children with CP in Northern Ireland each year. CP is the commonest cause of physical disability in early childhood. Recommendations regarding type and timing of neuroimaging in neonates, to identify those at risk of a poor neuro-developmental outcome, were published in 2002 by Ment et al. More recently the Surveillance of CP in Europe group (SCPE) has suggested a simple classification system which can be used for neuroimaging of both the neonate and the older child with CP (http://www.scpenetwork.eu/).

This audit was undertaken to assess the use of neuroimaging specifically cranial ultrasound (USS) and cerebral magnetic resonance imaging (MRI) in children with CP in Belfast HSCT.

Methods.

Selection criteria: Children born between 01/01/2000 and 31/12/2008 born or who received neonatal care in the Belfast HSCT catchment area with a diagnosis of CP were identified using the Northern Ireland Register Northern Ireland (NICPR).

Data collection: Neonatal records were accessed to identify whether cranial ultrasound had been completed. Cross reference with the Belfast HSCT picture archiving and communication system identified those who had a cerebral MRI scan(s) performed. A proforma was completed which included gestational age, birth weight, type of CP and associated impairment alongside timing and findings of cranial USS and MRI scan. Neonatal care records were reviewed by a junior doctor and a Consultant Paediatrician to collect data regarding cranial US. MRI scans were reviewed

independently by two neuroradiologists using the SCPE classification system. Current practice was audited against published recommendations. The NICPR provided data on HSC Trust at birth, gestational age, birth weight, whether the birth was multiple or not, subtype of CP, motor impairment and function, including the Gross Motor Functional Scale and associated problems (intellectual impairment, communication, vision and hearing problems, seizures)

Guidelines: The criteria being used has been published by the Surveillance of Cerebral Palsy in Europe network (SCPE-NET, 2013). These recommendations are based on the most up-to-date evidence in the use of neuroimaging techniques. Regarding cranial US authors state that infants born at less than 32 weeks gestational age should undergo cranial US shortly after birth, between the 3rd and 7th day of life and weekly thereafter until discharge or at term age equivalent. For those born at 32 weeks gestation or after and clinically healthy, cranial US should be performed on the 3rd day of life and weekly thereafter until discharged. For those infants born at term but who present adverse clinical signs, congenital malformations or neurological symptoms, cranial US should be conducted shortly after birth. The frequency of cranial US should be increased in case of clinical deterioration, sepsis, necrotising enterocolitis, apneas and bradycardias, decrease in Hb level, neurological symptoms, pre and post major surgery and/or previously detected ventricular dilatation (VD).SCPE recommendations also refer to cerebral MRI. For preterm infants cerebral MRI should be conducted in two situations: when serial cranial US conducted during the 1st week of life of the preterm infant show periventricular leukomalacia (PVL), intraventricular haemorrhage (IVH) and further serial cranial US confirm the findings. MRI should also be conducted when the initial cranial US does not show PVL/IVH but successive low frequency serial cranial US show complications of brain growth and maturation. In both situations MRI should be carried out at term equivalent age. For term infants cerebral MRI should be conducted between 1st and 2nd week of life in cases of perinatal brain injury. Earlier examination should always include diffusion-weighted imaging (DWI). Since some of the cases in the sample of the proposed audit would have been born before the above guidelines were published, the audit will also measure practice against criteria published by the American Academy of Neurology and the Child Neurology Society (Ment et al., 2002). These guidelines recommend that infants born at less than 30 weeks gestation should undergo cranial US between their 7th and 14th day of life and be repeated at 36 to 40 weeks postmenstrual age. Regarding cerebral MRI authors reported that at the time the guidelines were written there was insufficient evidence for routine MRI in all very low birth weight infants with abnormal cranial US. However, cerebral MRI was recommended if CT results were unclear for term infants with a neonatal history of encephalopathy, significant birth trauma and evidence of low hematocrit or coagulation. MRI was also recommended for term infants with acute encephalopathy and it should be done at age 2 to 8 days, it should include magnetic resonance spectroscopy (MRS) single-voxel if available and DWI should also be conducted at the time of MRI if available.

Status: Data regarding MRI has been collected and entered in the database for audit. Collecting final Cranial US data.

Audit of the current practice of hip surveillance in children aged 5 -18 years with cerebral palsy within the Belfast Health and Social Care Trust

Chief investigator: Dr Joseph Clarke, MB BCh BAO MRCPCH; Neurology Department, Royal Belfast Hospital for Sick Children

Other investigators: Dr Claire Lundy, MB BCh BAO MRCPCH; Neurology Department, Royal Belfast Hospital for Sick Children

Protocol

Aims and objectives: This retrospective audit aims to identify surveillance practice and orthopaedic management of the hips of children with cerebral palsy who reside within the Belfast Health and Social Care Trust. Specifically it will identify:

- the number of children who have an identified hip pathology
- frequency of use of orthopaedic interventions for the hips (surgery, botulinum toxin A injection)
- the extent of use of standard radiological investigations and measurements
- the most commonly reported elements of clinical examination of the hip, the clinical use of a standard measure of severity of motor function (GMFCS)

Background: Cerebral palsy (CP) has a prevalence of 2.2 cases per 1,000 live births (Parkes et al, 2005) and it is the commonest form of physical disability. Children with CP are at higher risk of lateral displacement of the femoral head and hip dislocation compared with typically developing children. Population-based studies suggest that the natural risk of hip dislocation for children with CP is 15-20% (Soo et al., 2006; Hägglund et al., 2007a), with younger age, greater severity of the condition, and migration percentage (a radiographical measurement) greater than 30% being identified as risk factors (Hägglund and colleagues 2005, 2007a,b, NICE 2012). In Northern Ireland, children with CP requiring orthopaedic management are reviewed by a Consultant Paediatric Orthopaedic Surgeon (POS), and x-rayed at the surgeon's request. Local research with children and young people with severe CP (Donnelly et al., 2007) revealed that 67% (82/123 children) had a hip x-ray in the previous year. Of these, 61% (50/82 children) had a migration percentage greater than 30% in one or both hips, thus would be considered 'at risk' and should be monitored, however frequency and outcome of the current monitoring system has not been documented.

Hip displacement or dislocation can result in significant morbidity in terms of pain and deformity, inability to sit, functional restrictions and daily living problems, and skin ulceration. Hip surveillance to ensure timely access to orthopaedic surgery and to avoid preventable complications is considered a priority action area internationally due to the recent publication of NICE guidelines (Hägglund et al., 2005, 2007a,b, 2009; Wynter et al., 2008; NICE, 2012). Reported benefits of implementation of hip surveillance programmes include an increased number of preventative treatment measures, earlier age of receipt of these treatments, reduction in overall radiation exposure for patients, and reduction in health costs (Dobson et al., 2002; Hägglund et al., 2005, 2007a, 2009; Soo et al., 2006; Gordon and Simkiss et al 2006; Elkamil et al., 2011).

Previous research in Northern Ireland investigated orthopaedic problems experienced by children with severe CP (Donnelly et al, 2007) however did not audit service arrangements, nor include

children with less severe impairments. Examination of regional practice using a representative sample of children is warranted as this will determine how service arrangements and clinical outcomes for all children with CP in Northern Ireland compare with other developed countries. Benchmarking regional surveillance practices with published standardised surveillance recommendations will facilitate identification of potential strengths and weaknesses in current practice in Northern Ireland; in doing so it will be possible to ascertain if current orthopaedic management of children with CP in Northern Ireland is optimal. The proposed audit will provide information on current service arrangements for monitoring the hips of children with CP in Northern Ireland (orthopaedic and x-ray review), and will report on the interventions received and outcomes of these children, relating to their hips. This will facilitate robust service evaluation and allow evidence-based recommendations for service improvements to be brought forward, if appropriate.

Sample: The sample will be identified through the children and young people registered with the Northern Ireland Cerebral Palsy Register (NICPR).

Criteria for inclusion in the audit will be: current resident of the Belfast Health and Social Care Trust; diagnosis of CP and aged 5 years to 18 years

A sample exploration in September 2014 by Dr Garcia-Jalon, NICPR Research Fellow, identified 805 children aged between 5 and 18 years in Northern Ireland with CP. Of these, 149 children live within the Belfast Health and Social Care Trust hence will be the subject of this audit.

Criteria/standards being used: This audit will use the NICE Clinical Guideline CG145; Spasticity in children and young people (2012). The NICE Clinical guideline recommends a hip x-ray to assess for hip displacement:

- at 24 months in children with bilateral CP
- if there are clinical concerns about possible hip displacement. These include pain arising from the hips, clinically important leg length difference, deterioration in range of hip movement, increased hip muscle tone, deterioration in sitting or standing or increasing difficulty with perineal care or hygiene.
- annually in children or young people who are at Gross Motor Function Classification System level III, IV or V
- after 6 months in children and young people where the initial hip migration is greater than 30% and then repeated 6 monthly if hip migration is increasing by more than 10 percentage points per year.

Data Collection: Data collection will be performed using case note review and the two electronic databases: Northern Irelands Radiology system (NIPACS) and Northern Ireland Electronic Health Care Record (NIECR).

Data collected will include:

- Gender
- Age
- Gestation at birth
- CP classification
- Use of Gross Motor Function Classification System (GMFCS)
- Number of contacts with Orthopaedics
- Findings of clinical assessment
- Number of hip x-rays
- Interval hip x-rays
- Intervals of radiographs
- Interventions after hip x-ray

Data analysis: Descriptive analysis of adherence of hip surveillance in children aged 5 to 18 years with CP in the Belfast Health and Social Care Trust with National Guidelines.

Data Management: A password protected list of cases with CP who fulfil the audit criteria will be supplied by the NICPR. This list will be transferred to the Belfast Health and Social Care Trust account of Dr Joseph Clarke using an Iron Key and a computer held in the Royal Belfast Hospital for Sick Children. The file with the list of cases will only remain on Dr Joseph Clarke's staff account and will remain password protected. Only Dr Joseph Clarke will have access to the Electronic Master List and therefore be the custodian of the data. Mr Fintan McErlean from the Audit Department in the Belfast Heath and Social Care Trust has been made aware of this and this is in keeping with the Trust's Data protection and confidentiality policy.

Once the data are collected the forms as well as the electronic database will be anonymised and each case will be identified by an audit number e.g. BHSCT 001. Mr Fintan McErlean of the Audit Department in the Belfast Health and Social Care states that the Trust's Audit Department only keeps data collection sheets for up to 6 months after the audit is completed. If storage of the data is required for a longer period by the NICPR, he suggests that both paper and electronic copies of the data may need to be stored by NICPR.

Reporting of results: Local conference presentation

Status: Ongoing; following up with the Standards, Quality and Audit department in the Belfast Health and Social Care Trust.

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Appendices

Appendix 1: Decision tree for Cerebral Palsy

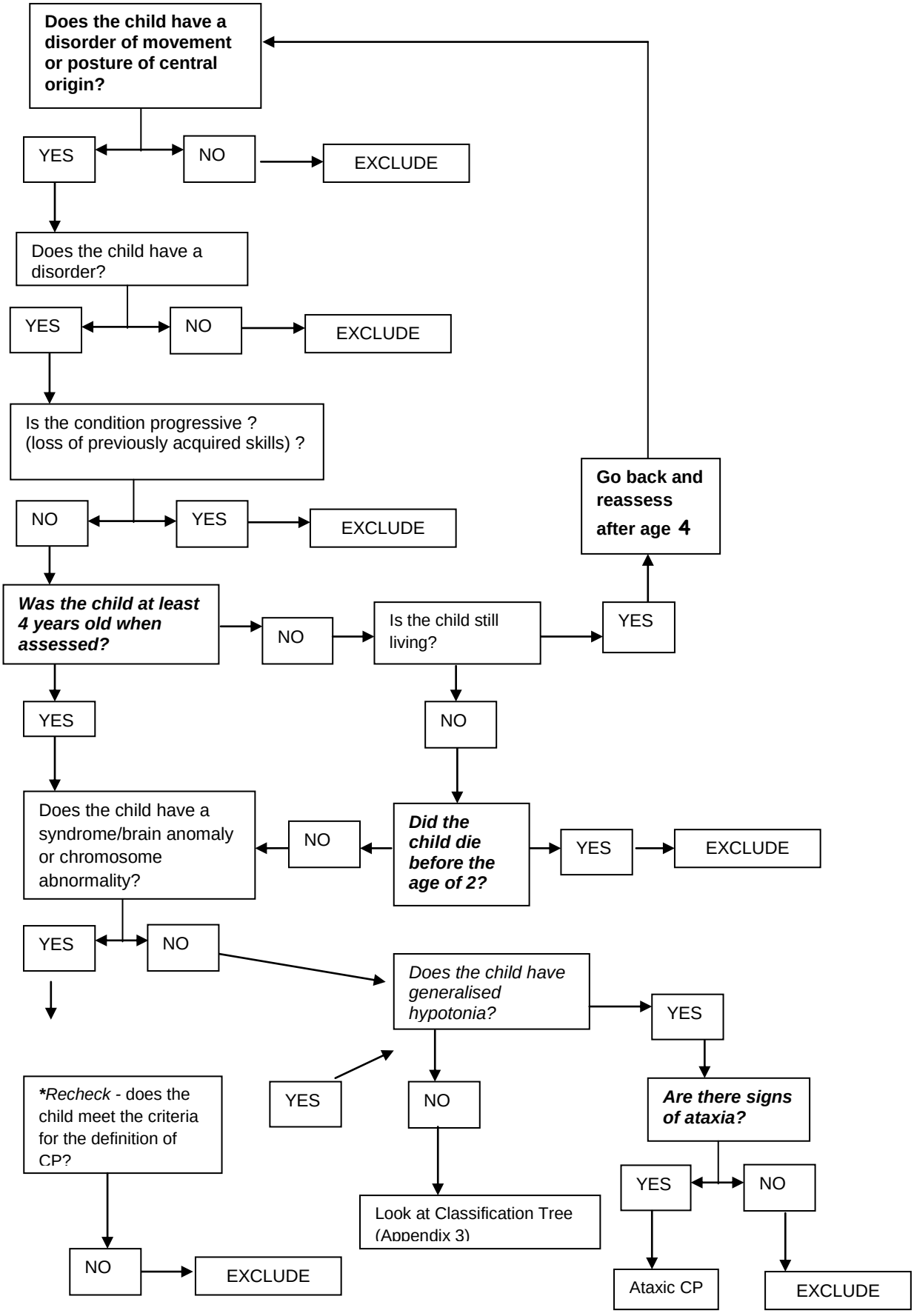
Appendix 2: Report card for notifiers

Appendix 3: Classification tree for CP subtypes

Appendix 4: GMFCS descriptors and illustrations for notifiers

Appendix 5: Abstracts of publications from 2005 onwards in which the NICPR has been involved

Appendix 1: Decision tree for Cerebral Palsy



Appendix 2: Report card for notifiers

Guidelines for completion

Please record the number of 'new' cases or suspected cases of cerebral palsy seen in this month (i.e. newly diagnosed or new to you). If you have seen no new cases please tick 'nothing to report' and return the card.

If you have seen a child please complete this form in the space provided (see over) and you will be sent the appropriate number of booklets to complete but only if this information has not already been collected. Please note that this information will not be used to contact families directly at this stage. Families may be followed up in the future but only with your prior consent.

Address: Northern Ireland Cerebral Palsy Register
Room 1.36
Mulhouse Building
Institute of Clinical Science
Royal Victoria Hospital
Grosvenor Road
BELFAST BT12 6BJ

Email: j.parkes@qub.ac.uk Telephone 028 9063 5045

January 2014

Northern Ireland Cerebral Palsy Register
Report Card

CONFIDENTIAL

See back cover for guidelines

Northern Ireland Cerebral Palsy Register **January 2014**

Professional's Name_____

In the last month have you seen a 'new' child with suspected or definite cerebral palsy (i.e. newly diagnosed or newly referred to you)?

☐ Yes If yes, how many? _____

Please complete the notification form below

☐ Nothing to report

Notification form

This card will serve as a preliminary notification of children with confirmed or suspected cerebral palsy. If a booklet is not already available then you will be sent a form and asked to complete the information for each child notified.

	Name	DOB	Address
1.			
2.			
3.			
4.			
5.			
6.			

CDS 96226

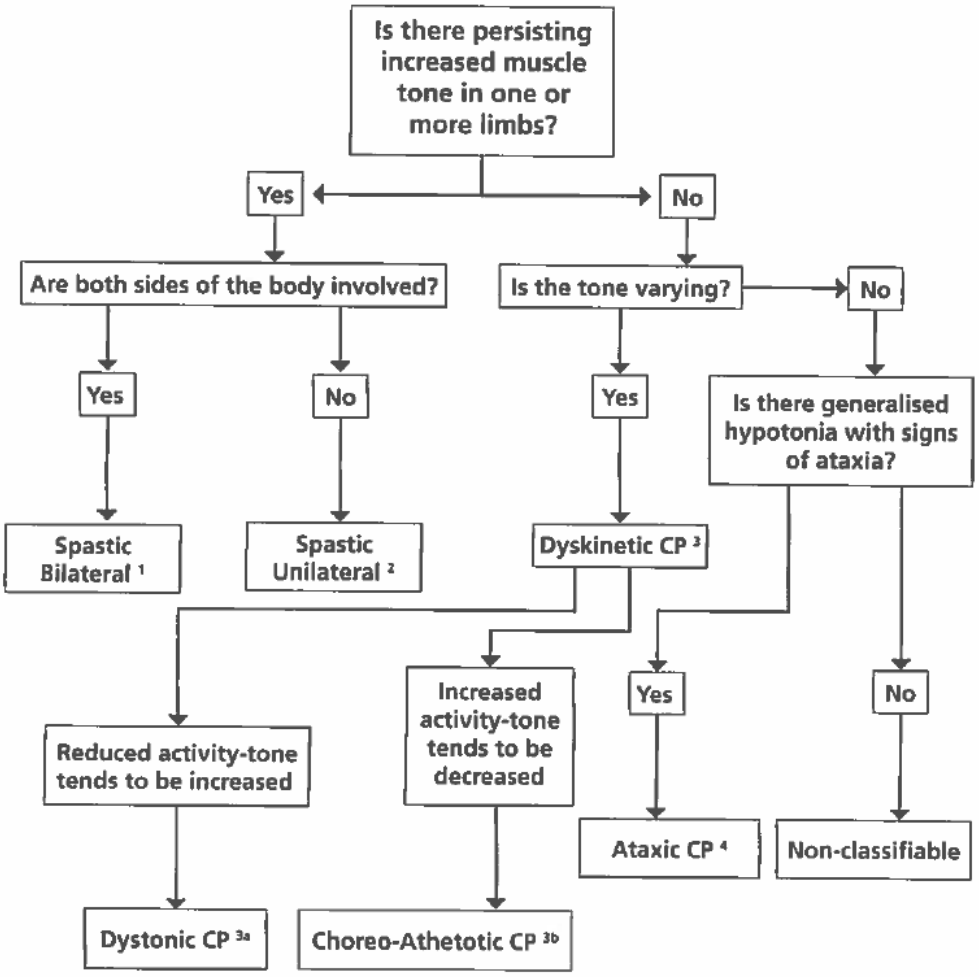
Appendix 3: Classification tree for subtypes of CP

Guidelines For Completion

This form is intended as an epidemiological tool for collecting data on impairment and disability in children with a motor deficit of central origin (subsumed under the umbrella term 'cerebral palsy' or CP). It will provide the data necessary to identify groups of children with similar clinical profiles over a period of time as a basis for aetiological and health services research.

CLASSIFICATION TREE FOR subtypes of cerebral palsy

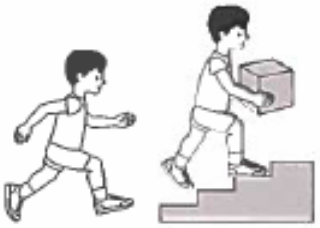
Use the following Classification Tree to help determine the type of cerebral palsy[1]



1 SCPE Collaborative Group. Surveillance of cerebral palsy in Europe: A collaboration of cerebral palsy surveys and registers. Dev Med Child Neuro. 2000; 42:816-824

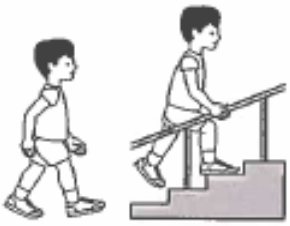
Appendix 4: GMFCS descriptors and illustrations for notifiers

GMFCS E & R between 6th and 12th birthday: Descriptors and illustrations



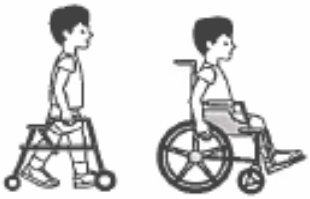
GMFCS Level I

Children walk at home, school, outdoors and in the community. They can climb stairs without the use of a railing. Children perform gross motor skills such as running and jumping, but speed, balance and coordination are limited



GMFCS Level II

Children walk in most settings and climb stairs holding onto a railing. They may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas or confined spaces. Children may walk with physical assistance, a hand-held mobility device or used wheeled mobility over long distances. Children have only minimal ability to perform gross motor skills such as running and jumping.



GMFCS Level III

Children walk using a hand-held mobility device in most indoor settings. They may climb stairs holding onto a railing with supervision or assistance. Children use wheeled mobility when traveling long distances and may self-propel for shorter distances.

GMFCS Level IV

Children use methods of mobility that require physical assistance or powered mobility in most settings. They may walk for short distances at home with physical assistance or use powered mobility or a body support walker when positioned. At school, outdoors and in the community children are transported in a manual wheelchair or use powered mobility.

GMFCS Level V

Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements.

GMFCS descriptors: Palisano et al. (1997) Dev Med Child Neurol 39:214-23

Appendix 5: Abstracts of publications from 2005 onwards in which the NICPR has been involved

Validity of a 1 minute walk test for children with cerebral palsy.

Brona C McDowell BSc PhD; Claire Kerr BSc; Jackie Parkes BNurs PhD; Aidan Cosgrove MD FRCS, Gait Analysis Laboratory*

*Developmental Medicine and Child Neurology, **2005**, Vol. 47(11), pp.744-748.*

The concurrent validity of a 1 minute walk test at a child's maximum walking speed was assessed in children with bilateral spastic cerebral palsy (BSCP). The distance covered during the 1 minute walk test was compared with the children's gross motor function as assessed by the Gross Motor Function Measure (GMFM). Twenty-four male and 10 female children with CP (mean age 11y, range 4 to 16y) participated in the study. Gross Motor Function Classification System (GMFCS) levels were; level I (***n***=3), level II (***n***=17), level III (***n***=10), and level IV (***n***=4). Participants had clinical diagnoses of symmetrical diplegia (***n***=19), asymmetrical diplegia (***n***=14), and quadriplegia (***n***=1). Results showed a significant correlation between GMFM score and the distance covered during the 1 minute walk (***r***=0.92; ***p***<0.001). There was also a significant decrease in the distance walked with increasing GMFCS level (***p***<0.001). We concluded that the 1 minute walk test is a valid measure for assessing functional ability in children with ambulatory BSCP. Its cost effectiveness and user friendliness make it a potentially useful tool in the clinical setting. Further study needs to address its reliability and ability to detect change over time.

Trends in the prevalence of cerebral palsy in Northern Ireland, 1981–1997.

Helen Dolk Jackie Parkes Nan Hill*

*Developmental Medicine & Child Neurology **2006**, 48: 406–412*

We describe trends in the prevalence of cerebral palsy (CP) by birth weight group, and in the severity of motor impairments and presence of associated intellectual impairment, in Northern Ireland from 1981 to 1997 (***n***=909; 510 males, 399 females; total population 415 936 live births) using data from a population based register of CP. Children with suspected CP or who died before 1 year of age and those with CP of post neonatal origin were excluded. Prevalence of CP was 2.2 per 1000 live births without significant change over time. Among very-low birthweight (<1500g) live births, prevalence was 44.5 per 1000 (95% confidence interval 32.3–59.8) from 1994 to 1997, with evidence of a statistically significant decline in prevalence since the mid- to late 1980s accompanied by a decrease in the severity of motor impairment and likelihood of intellectual impairment. Among moderately-low-birthweight (1500–2499g) children there was weaker evidence of a peak prevalence in the late 1980s. Prevalence among normal-birthweight infants did not change significantly, but outcome in terms of severity of motor impairment and intellectual impairment improved in the 1990s. Occurrence of bilateral spasticity from 1994 to 1997 was associated with greater severity and likelihood of intellectual impairment for normal-birthweight individuals than for low- or Study protocol

UKCP: a collaborative network of cerebral palsy registers in the United Kingdom.
Geraldine Surman, Sandra Bonellie, James Chalmers, Allan Colver, Helen Dolk, Karla Hemming, Andy King, Jennifer J. Kurinczuk, Jackie Parkes, Mary Jane Platt
*Public Health **2006**, Vol, 28, pp.148-156.*
Cerebral palsy (CP) is a relatively rare condition with enormous social and financial impact. Information about CP is not routinely collected in the United Kingdom. We have pooled non-identifiable data from the five currently active UK CP registers to form the UKCP database: birth years 1960–1997. This article describes the rationale behind this collaboration and the creation of the database. Data about 6910 children with CP are currently held. The mean annual prevalence rate was 2.0 per 1000 live births for birth years 1986–1996. Where type is known, 91 per cent have spastic CP. Where data are available, nearly one-third of children have severely impaired lower limb function, and nearly a quarter have severely impaired upper limb function. As well as describing the range and complexity of motor and associated impairments, the pooled data from the UKCP database provide a platform for studies of aetiology, long-term outcomes, participation and service needs. The UKCP database is an important national resource for the surveillance of CP and the study of its epidemiology in the United Kingdom.

Accommodative Dysfunction in Children with Cerebral Palsy: A Population-Based Study.
Julie F. McClelland, Jackie Parkes, Nan Hill, A. Jonathan Jackson and Kathryn J. Saunders
*Investigative Ophthalmology & Visual Science**, 2006**, Vol. 47(5), pp. 1824-1830.*
PURPOSE. To determine the prevalence, nature, and degree of accommodative dysfunction among children with different types and severities of cerebral palsy (CP) in Northern Ireland.
METHODS. Ninety subjects with CP (aged 4–15 years) were recruited through the Northern Ireland CP Register (NICPR).Modified Nott dynamic retinoscopy was used to measure lag and lead of accommodation at three test distances: 25 cm (4D), 16.7 cm (6 D), and 10 cm (10 D) with the distance correction in place. Accommodative function was also assessed in an age-matched control group (*n* _ 125) for comparison. Each subject's neurologic status was derived from the NICPR.
RESULTS. Children with CP demonstrate significantly reduced accommodative responses compared with their neurologically normal peers. Of the subjects with CP, 57.6% demonstrated an accommodative lag outside normal limits at one or more distances Reduced accommodative responses were significantly associated with more severe motor and intellectual impairments
CONCLUSIONS. Brain injury such as that present in CP has a significant impact on accommodative function. These findings have implications for the optometric care of children with CP and inform our understanding of the impact of early brain injury on visual development. (*Invest Ophthalmol Vis Sci.* 2006;47:1824–1830) DOI:10.1167/iovs.05-0825

Assessment of data quality in a multi-centre cross-sectional study of participation and quality of life of children with cerebral palsy.

Heather Dickinson, Kathryn Parkinson, Vicki McManus,Catherine Arnaud, Eva Beckung, Jérôme Fauconnier, Susan I Michelsen, Jackie Parkes, Giorgio Schirripa, Ute Thyen and Allan Colver

*BMC Public Health, **2006**, Vol. 6, pp. 273*

Background: SPARCLE is a cross-sectional survey in nine European regions, examining the relationship of the environment of children with cerebral palsy to their participation and quality of life. The objective of this report is to assess data quality, in particular heterogeneity between regions, family and item non-response and potential for bias.

Methods: 1,174 children aged 8–12 years were selected from eight population-based registers of children with cerebral palsy; one further centre recruited 75 children from multiple sources. Families were visited by trained researchers who administered psychometric questionnaires. Logistic regression was used to assess factors related to family non-response and self-completion of questionnaires by children.

Results: 431/1,174 (37%) families identified from registers did not respond: 146 (12%) were not traced; of the 1,028 traced families, 250 (24%) declined to participate and 35 (3%) were not approached. Families whose disabled children could walk unaided were more likely to decline to participate. 818 children entered the study of which 500 (61%) self-reported their quality of life; children with low IQ, seizures or inability to walk were less likely to self-report. There was substantial heterogeneity between regions in response rates and socio-demographic characteristics of families but not in age or gender of children. Item non-response was 2% for children and ranged from 0.4% to 5% for questionnaires completed by parents.

Conclusion: While the proportion of untraced families was higher than in similar surveys, the refusal rate was comparable. To reduce bias, all analyses should allow for region, walking ability, age and socio-demographic characteristics. The 75 children in the region without a population based register are unlikely to introduce bias.

Study protocol: SPARCLE – a multi-centre European study of the relationship of environment to participation and quality of life in children with cerebral palsy.

Allan Colver and the SPARCLE* group*

*BMC Public Health **2006***

Background: SPARCLE is a nine-centre European epidemiological research study examining the relationship of participation and quality of life to impairment and environment (physical, social and attitudinal) in 8–12 year old children with cerebral palsy. Concepts are adopted from the International Classification of Functioning, Disability and Health which bridges the medical and social models of disability.

Methods/Design: A cross sectional study of children with cerebral palsy sampled from total population databases in 9 European regions. Children were visited by research associates in each country who had been trained together. The main instruments used were KIDSCREEN, Life-H, Strength and Difficulties Questionnaire, Parenting Stress Index. A measure of environment was developed within the study. All instruments were translated according to international guidelines. The potential for bias due to non response and missing data will be examined. After initial analysis using multivariate regression of how the data captured by each instrument relate to impairment and socio-economic characteristics, relationships between the latent traits captured by the instruments will then be analysed using structural equation modelling.

Discussion: This study is original in its methods by directly engaging children themselves, ensuring those with learning or communication difficulty are not excluded, and by studying in quantitative terms the crucial outcomes of participation and quality of life. Specification and publication of this protocol prior to analysis, which is not common in epidemiology but well established for randomised controlled trials and systematic reviews, should avoid the pitfalls of data dredging and post hoc analyses.

Interobserver agreement of the Gross Motor Function Classification System in an ambulant population of children with cerebral palsy.

Brona C McDowell BSc Physio PhD; Claire Kerr BSc Physio PhD, Gait Analysis Laboratory, Musgrave Park Hospital; Jackie Parkes BNurs PhD, School of Nursing and Midwifery,*
*Developmental Medicine and Child Neurology, **2007**, Vol. 49 (7), pp. 528-533.*

Gross Motor Function Classification System (GMFCS) level was reported by three independent assessors in a population of children with cerebral palsy (CP) aged between 4 and 18 years (***n***=184; 112 males, 72 females; mean age 10y 10mo [SD 3y 7mo]). A software algorithm also provided a computed GMFCS level from a regional CP registry. Participants had clinical diagnoses of unilateral (***n***=94) and bilateral (***n***=84) spastic CP, ataxia (***n***=4), dyskinesia (***n***=1), and hypotonia (***n***=1), and could walk independently with or without the use of an aid (GMFCS Levels I–IV). Research physiotherapist (***n***=184) and parent/guardian data (***n***=178) were collected in a research environment. Data from the child's community physiotherapist (***n***=143) were obtained by postal questionnaire. Results, using the kappa statistic with linear weighting (κlw), showed good agreement between the parent/guardian and research physiotherapist (κlw=0.75) with more moderate levels of agreement between the clinical physiotherapist and researcher (κlw=0.64) and the clinical physiotherapist and parent/guardian (κlw=0.57). Agreement was consistently better for older children (>2y). This study has shown that agreement with parent report increases with therapists' experience of the GMFCS and knowledge of the child at the time of grading. Substantial agreement between a computed GMFCS and an experienced therapist (κlw=0.74) also demonstrates the potential for extrapolation of GMFCS rating from an existing CP registry, providing the latter has sufficient data on locomotor ability.

Lifestyle limitations of children and young people with severe cerebral palsy: a population study protocol.

Collette Donnelly, Jackie Parkes, Brona McDowell & Catherine Duffy
*Journal of Advanced Nursing 61(5), 557–569 **2007***

Aim. This paper is a presentation of a study protocol to establish the prevalence of orthopaedic problems (hip dislocation, pelvic obliquity, spinal deformity and contractures) and their impact on pain, function, participation and health in a population of children and young people with severe cerebral palsy.

Background. Cerebral palsy is the commonest cause of motor impairment in childhood and is associated with life-long disability. An estimated 30% of people with cerebral palsy have severe forms and are non-ambulant. Although the underlying neurological damage is not amenable to correction, many health services are dedicated to providing therapeutic and adaptive support to help people with the condition reach their potential.

Method. A cross-sectional survey of children and young people, aged 4–25 years with severe, non-ambulant cerebral palsy as defined using the Gross Motor Function Classification System (Levels IV

and V). Study participants will be identified from a pre-existing, geographically defined case register and recruited via a healthcare professional known to them. Two assessments will be undertaken: one involving parents/carers at home and using questionnaires; the other involving the child/young person ideally in one of three settings and including X-rays if clinically indicated.
Discussion. This study will contribute to our knowledge of the history and epidemiology of orthopaedic problems in children and young people with cerebral palsy and how these problems accumulate and impact on participation, health and wellbeing. The study will also identify unmet need and make recommendations for good practice in relation to the orthopaedic care and management for people with severe cerebral palsy.

Self-reported quality of life of 8–12-year-old children with cerebral palsy: a cross-sectional European study.
Heather O Dickinson, Kathryn N Parkinson, Ulrike Ravens-Sieberer, Giorgio Schirripa, Ute Thyen, Catherine Arnaud, Eva Beckung, Jérôme Fauconnier, Vicki McManus, Susan I Michelsen, Jackie Parkes, Allan F Colver*
*Lancet **2007**; 369: 2171–78*
Background Little is known about the quality of life (QoL) of disabled children. We describe self-reported QoL of children with cerebral palsy, factors that influence it, and how it compares with QoL of the general population.
Methods 1174 children aged 8–12 years were randomly selected from eight population-based registers of children with cerebral palsy in six European countries and 743 (63%) agreed to participate; one further region recruited
75 children from multiple sources. Researchers visited these 818 children. 318 (39%) with severe intellectual impairment could not self-report; 500 (61%) reported their QoL using KIDSCREEN, an instrument with scores in ten domains, each with SD=10. Multivariable regression was used to relate QoL to impairments, pain, and sociodemographic characteristics. Comparisons were made with QoL data from the general population.
Findings Impairments were not significantly associated with six KIDSCREEN domains. Comparison of least and most able groups showed that severely limited self-mobility was significantly associated with reduced mean score for physical wellbeing (7·6, 95% CI 2·7–12·4); intellectual impairment with reduced mean for moods and emotions (3·7, 1·5–5·9) and autonomy (3·3, 0·9–5·7); and speech difficulties with reduced mean for relationships with parents (4·5, 1·9–7·1). Pain was common and associated with lower QoL on all domains. Impairments and pain explained upto 3% and 7%, respectively, of variation in QoL. Children with cerebral palsy had similar QoL to children in the general population in all domains except schooling, in which evidence was equivocal, and physical wellbeing, in which comparison was not possible.
Interpretation Parents can be reassured that most children aged 8–12 years with cerebral palsy will have similar QoL to other children. This finding should guide social and educational policy to ensure that disabled children participate fully in society. Because of its association with QoL, children's pain should be carefully assessed.
Determinants of Child-Parent Agreement in Quality-of-Life Reports: A European Study of Children With Cerebral Palsy.
Melanie White-Koning, PhDa,b, Catherine Arnaud, MDa,b,c, Heather O. Dickinson, PhDd, Ute Thyen, MDe, Eva Beckung, PhDf, Jerome Fauconnier, MDg, Vicki McManus, BAh, Susan I. Michelsen, PhDi,

*Jackie Parkes, PhDj, Kathryn Parkinson, PhDk, Giorgio Schirripa, MDl†, Allan Colver, MDm Pediatrics, **2007**, Vol. 120(4), pp. e804-e814.*

Objectives: The differences between child self-reports and parent proxy reports of quality of life in a large population of children with cerebral palsy were studied. We examined whether child characteristics, severity of impairment, socioeconomic factors, and parental stress were associated with parent proxy reports being respectively higher or lower than child self-reports of quality of life.

Methods: This study was conducted in 2004–2005 and assessed child quality of life (using the Kidscreen questionnaire, 10 domains, each scored 0–100) through self-reports and parent proxy reports of 500 children aged 8 to 12 years who had cerebral palsy and were living in 7 countries in Europe.

Results: The mean child-reported scores of quality of life were significantly higher than the parent proxy reports in 8 domains, significantly lower for the finances domain, and similar for the emotions domain. The average frequency of disagreement (child-parent difference greater than half an SD of child scores) over all domains was 64%, with parents rating their child's quality of life lower than the children themselves in 29% to 57% of child-parent pairs. We found that high levels of stress in parenting negatively influenced parents' perception of their child's quality of life, whereas the main factor explaining parents' ratings of children's quality of life higher than the children themselves is self-reported severe child pain.

Conclusions: This study shows that the factors associated with disagreement are different according to the direction of disagreement. In particular, parental wellbeing and child pain should be taken into account in the interpretation of parent proxy reports, especially when no child self-report of quality of life is available. In the latter cases, it may be advisable to obtain additional proxy reports (from caregivers, teachers, or clinicians) to obtain complementary information on the child's quality of life.

Use of the Child Health Questionnaire in Children with Cerebral Palsy: A Systematic Review and Evaluation of the Psychometric Properties.

*Nichola McCullough, BSC, PHD and Jackie Parkes, BNURS, PHD Journal of Paediatric Psychology, **2008**, Vol. 33, pp. 80-90*

Objective: To review the psychometric performance of the Child Health Questionnaire (CHQ) in samples of children with cerebral palsy (CP). Method Four search terms were applied to five databases in a search for papers published between 1993 and January 2007. Results A total of 13 papers were identified, providing data on 1229 unique children aged 2–18 years old. Three studies reported on the reliability of the CHQ (internal consistency), whilst six studies provided evidence on various dimensions of validity (concurrent; discriminant and item discriminant validity). **Conclusions** This review identified a number of psychometric issues that need to be addressed. These include the assessment of additional types of reliability; an examination of the factor structure of the CHQ within the CP population; and the development of normative data using substantial representative samples, particularly in Europe. Until these issues are addressed, researchers utilizing the CHQ in children with CP should be cautious about its interpretation

Psychological problems in children with cerebral palsy: a cross-sectional European study.

Jackie Parkes Melanie White-Koning, Heather O Dickinson, Ute Thyen, Catherine Arnaud, Eva Beckung, Jerome Fauconnier, Marco Marcelli, Vicki McManus, Susan I. Michelsen, Kathryn Parkinson and Allan Colver

*Journal of Child Psychology and Psychiatry, **2008**, Vol. 49, pp. 405-413*

Objectives: To describe psychological symptoms in 8–12-year-old children with cerebral palsy; to investigate predictors of these symptoms and their impact on the child and family.
Design: cross sectional multi-centre survey. Participants: Eight hundred and eighteen children with cerebral palsy, aged 8–12 years, identified from population-based registers of cerebral palsy in eight European regions and from multiple sources in one further region. Main outcome measures: The Strengths and Difficulties Questionnaire (SDQ) P4)16 and the Total Difficulties Score (TDS) dichotomised into normal/borderline (TDS £ 16) versus abnormal (TDS > 16). Statistical analysis: Multilevel, multivariable logistic regression to relate the presence of psychological symptoms to child and family characteristics.
Results: About a quarter of the children had TDS > 16 indicating significant psychological symptoms, most commonly in the domain Peer Problems. Better gross motor function, poorer intellect, more pain, having a disabled or ill sibling and living in a town were independently associated with TDS > 16. The risk of TDS > 16 was odds ratio (OR) ¼ .2 (95% CI: .1 to .3) comparing children with the most and least severe functional limitations; OR ¼ 3.2 (95%CI: 2.1 to 4.8) comparing children with IQ < 70 and others; OR ¼ 2.7 (95% CI: 1.5 to 4.6) comparing children in severe pain and others; OR ¼ 2.7 (95% CI:1.6 to 4.6) comparing children with another disabled sibling or OR ¼ 1.8 (95%CI: 1.2 to 2.8) no siblings and others; OR ¼ 1.8 (95% CI: 1.1 to 2.8) comparing children resident in a town and others. Among parents who reported their child to have psychological problems, 95% said they had lasted over a year, 37% said they distressed their child and 42% said they burdened the family at least 'quite a lot'. Conclusions: A significant proportion of children with cerebral palsy have psychological symptoms or social impairment sufficiently severe to warrant referral to specialist services. Care must be taken in the assessment and management of children with cerebral palsy to ensure psychological problems are not overlooked and potentially preventable risk factors like pain are treated effectively. The validity of the SDQ for children with severe disability warrants further assessment.

Energy efficiency in gait, activity, participation, and health status in children with cerebral palsy.
Claire Kerr Jackie Parkes Mike Stevenson BSc (Hons) PGCHET Aidan P Cosgrove MD, FRCS (Orth); Brona C McDowell BSc (Hons) Physio PhD*
*Developmental Medicine & Child Neurology, **2008**. Vol. 50(3), pp. 204-210.*
The aim of the study was to establish if a relationship exists between the energy efficiency of gait, and measures of activity limitation, participation restriction, and health status in a representative sample of children with cerebral palsy (CP). Secondary aims were to investigate potential differences between clinical subtypes and gross motor classification, and to explore other relationships between the measures under investigation. A longitudinal study of a representative sample of 184 children with ambulant CP was conducted (112 males, 72 females; 94 had unilateral spastic CP, 84 had bilateral spastic CP, and six had non-spastic forms; age range 4–17y; Gross Motor Function Classification System Level I, ***n***=57; Level II, ***n***=91; Level III, ***n***=22; and Level IV, ***n***=14); energy efficiency (oxygen cost) during gait, activity limitation, participation restriction, and health status were recorded. Energy efficiency during gait was shown to correlate significantly with activity limitations; no relationship between energy efficiency during gait was found with either participation restriction or health status. With the exception of psychosocial health, all other measures showed significant differences by clinical subtype and gross motor classification. The energy efficiency of walking is not reflective of participation restriction or health status. Thus, therapies leading to improved energy efficiency may not necessarily lead to improved participation or general health.

Children with cerebral palsy (CP) have an increased energy cost of walking compared with typically-developing peers.1, 2 Within the spectrum of the condition, increasing energy costs are associated with an increase in the severity of functional involvement.3 Functional and community ambulation issues associated with this increased cost of walking may have a direct effect on participation and social integration of the child at home, at school, and in the community setting. These considerations are particularly relevant given the World Health Organization's *International Classification of Functioning, Disability and Health* (ICF).4 This classification has provided a useful framework for the comprehensive assessment of the individual: 'impairments' are defined as problems in body structures and functions; 'activity limitations' as difficulties an individual may have in the execution of a task or action; and 'participation restrictions' as the problems an individual may experience in involvement in life situations.4 Although increasing levels of impairment would appear to be associated with increasing levels of activity limitations, there are conflicting reports in the literature with regard to the strength of this relationship.5–8 More recently, the relationship between measures of activity limitations and participation restrictions and/or health status have been explored in an attempt to understand the day-to-day implications of living with functional limitations.9–16 Three studies to date15,17,18 have investigated the relationship between energy efficiency of gait and other aspects of health and lifestyle. Tervo et al.17 reported moderate correlations between energy efficiency and four of the five Pediatric Orthopedic Society of North America subscales: global function and comfort; upper extremity function; transfers and mobility; and physical function and sports. Oeffinger and colleagues, 15 in a retrospective review, also detected moderate correlations between oxygen cost (O2 cost) and dimensions D and E of the Gross Motor Function Measure (GMFM). Recently published prospective work from the same research group18 confirmed these findings; however, no relationship was found between O2 cost and the emotional and social subscales of the Pediatric Quality of Life Inventory. To describe the consequences of CP over time, a longitudinal study investigating the locomotor abilities of children within a representative sample of the population was undertaken. The primary hypothesis at baseline was to establish if a relationship existed between energy efficiency (as defined by the O2 cost of walking) and (1) activity limitations, (2) participation restrictions, and (3) health status. Secondary aims were to establish if the measures used could detect significant differences between children of differing clinical subtypes and Gross Motor Function Classification System (GMFCS) levels;19 and to explore the relationships between the measures used.

Health status of children with cerebral palsy living in Europe: a multi-centre study.
E. Beckung, M. White-Koning, M. Marcelli, V. McManus, S. Michelsen, J. Parkes, K. Parkinson, U. Thyen, C. Arnaud, J. Fauconnier and A. Colver
*Child: Care, Health & Development, **2008**, Vol. 34, pp. 806-814*
Aim: The aim of this report is to describe the health status of 8–12-year-old children with cerebral palsy (CP) of all severities in Europe using the Child Health Questionnaire (CHQ).
Method: A total of 818 children with CP from nine centres in defined geographical areas participated. CP type, gross and fine motor function, additional impairments were classified and family data were obtained. The CHQ was used to measure the parent's perception of their child's physical (PHY) and psychosocial (PSY) health.
Results: PHY scores were lower than the reference samples with a median of 46.The severity of gross motor function influenced the CHQ scores significantly in the PHY scale with the lowest scores for children with least gross motor function. There were significant differences between the CP types

in PHY with the higher scores for children with unilateral spastic and the lowest scores for children with bilateral spastic and dyskinetic CP type. Fine motor function severity significantly affected both the PHY and PSY scales.The severity of intellectual impairment was significantly associated with CHQ scores in most dimensions with higher scores for higher IQ level in PHY and PSY. Children with seizures during the last year had a significantly lower health compared with children without seizures. The results of the multivariate regression analyses (forward stepwise regression) of CHQ scores on CP subtype, gross and fine motor function, cognitive function, additional impairments, seizures, parental education and employment revealed gross motor function, cognitive level and type of school attended were significant prognostic factors.
Conclusion: This report is based on the largest sample to date of children with CP. Health status as measured using the CHQ was affected in all children and was highly variable. Gross motor function level correlates with health from the PHY well-being perspective but the PSY and emotional aspects do not appear to follow the same pattern.

Frequency of participation of 8–12-year-old children with cerebral palsy: A multi-centre cross-sectional European study.
Susan I. Michelsena, Esben M. Flachsa, Peter Uldallb, Eva L. Eriksenc, Vicki McManusd, Jackie Parkese, Kathryn N. Parkinsonf, Ute Thyeng, Catherine Arnaudh, Eva Beckungi,j, Heather O. Dickinsonk, Je´rome Fauconnierl, Marco Marcellim, Allan Colvern
*European Paediatric Neurology Society **2008***
Participation in home, school and community is important for all children; and little is known about the frequency of participation of disabled children. Frequency of participation is a valuable outcome measure for evaluating habilitation programmes for disabled children and for planning social and health services. We investigated how frequency of participation varied between children with cerebral palsy and the general population; and examined variation across countries to understand better how the environmental factors such as legislation, public attitudes and regulation in different countries might influence participation. We undertook a multi-centre, population-based study in children with and without cerebral palsy. Working from the Life-H instrument, we developed a questionnaire to capture frequency of participation in 8–12-year-old children. In nine regions of seven European countries, parents of 813 children with cerebral palsy and 2939 children from the general populations completed the questionnaire. Frequency of participation for each question was dichotomised about the median; multivariable logistic regressions were carried out. In the general population, frequency of participation varied between countries. Children with cerebral palsy participated less frequently in many but not all areas of everyday life, compared with children from the general population. There was regional variation in the domains with reduced participation and in the magnitude of the differences. We discuss how this regional variation might be explained by the different environments in which children live. Attending a special school or class was not associated with further reduction in participation in most areas of everyday life. 2008 European Paediatric Neurology Society. Published by Elsevier Ltd. All rights reserved.

Parent-Reported Quality of Life of Children With Cerebral Palsy in Europe.
Catherine Arnaud, MDa,b, Melanie White-Koning, PhDa, Susan Ishoy Michelsen, MD, PhDc, Jackie Parkes, PhD, BNursd, Kathryn Parkinson, PhDe, Ute Thyen, MDf, Eva Beckung, PhDg, Heather O. Dickinson, BSc, PhDh, Jerome Fauconnier, MDi, Marco Marcelli, MDj, Vicki McManus, BA, RGN, RCNk, Allan Colver, MD, FRCPCHl

Pediatrics, Vol. 121, pp. 54-64

Objective: The goal was to determine whether the type and severity of the child's impairments and the family's psychosocial, social, and economic characteristics influence parent-reported child quality of life across the spectrum of severity of cerebral palsy.

Methods: Our population-based, cross-sectional survey conducted in 2004 to 2005 involved 818 children with cerebral palsy, 8 to 12 years of age, from 7 countries (9 regions) in Europe. Child quality of life was assessed through parent reports by using the Kidscreen questionnaire, and data were analyzed separately for each of its 10 domains.

Results: The parental response rates were _93% for all domains except one. Gross motor function and IQ level were found to be associated independently with quality of life in most domains. However, greater severity of impairment was not always associated with poorer quality of life; in the moods and emotions, self-perception, social acceptance, and school environment domains, less severely impaired children were more likely to have poor quality of life. Pain was associated with poor quality of life in the physical and psychological well-being and self-perception domains. Parents with higher levels of stress were more likely to report poor quality of life in all domains, which suggests that factors other than the severity of the child's impairment may influence the way in which parents report quality of life.

Conclusions: The parent-reported quality of life for children with cerebral palsy is associated strongly with impairment. However, depending on the areas of life, the most severely impaired children (in terms of motor functioning or intellectual ability) do not always have the poorest quality of life.

Frequency of participation of 8–12-year-old children with cerebral palsy: A multi-centre cross-sectional European study.

Susan I. Michelsena, Esben M. Flachsa, Peter Uldallb, Eva L. Eriksenc, Vicki McManusd, Jackie Parkese, Kathryn N. Parkinsonf, Ute Thyeng, Catherine Arnaudh, Eva Beckungi,j, Heather O. Dickinsonk, Jerome Fauconnierl, Marco Marcellim, Allan Colvern

*European Journal of Paediatric Neurology, **2009**, Vol. 13, pp. 165-177.*

Participation in home, school and community is important for all children; and little is known about the frequency of participation of disabled children. Frequency of participation is a valuable outcome measure for evaluating habilitation programmes for disabled children and for planning social and health services. We investigated how frequency of participation varied between children with cerebral palsy and the general population; and examined variation across countries to understand better how the environmental factors such as legislation, public attitudes and regulation in different countries might influence participation. We undertook a multi-centre, population-based study in children with and without cerebral palsy. Working from the Life-H instrument, we developed a questionnaire to capture frequency of participation in 8–12-year-old children. In nine regions of seven European countries, parents of 813 children with cerebral palsy and 2939 children from the general populations completed the questionnaire.

Reliability and Validity of the Child Health QuestionnairePF-50 for European Children with Cerebral Palsy.

Nichola McCullough, PHD, Jackie Parkes, PHD, BNURS, Melanie White-Koning, PHD, Eva Beckung, PT, PHD, and Allan Colver MD, FRCPH

*Journal of Pediatric Psychology **2009,** Vol. 34, pp. 41-50.*

Objective: to evaluate the psychometric performance of the Child Health Questionnaire (CHQ) in children with cerebral palsy (CP). **Method** 818 parents of children with CP, aged 8–12 from nine regions of Europe completed the CHQ (parent form 50 items). Functional abilities were classified using the five-level Gross Motor Function Classification Scheme (Levels I–III as ambulant; Level IV–V as non ambulant CP). **Results** Ceiling effects were observed for a number of subscales and summary scores across all Gross Motor Function Classification System levels, whilst floor effects occurred only in the physical functioning scale (Level V CP). Reliability was satisfactory overall. Confirmatory factor analysis (CFA) revealed a seven-factor structure for the total sample of children with CP but with different factor structures for ambulant and non ambulant children. **Conclusion** The CHQ has limited applicability in children with CP, although with judicious use of certain domains for ambulant and non ambulant children can provide useful and comparable data about child health status for descriptive purposes.

The health of children with cerebral palsy and stress in their parents.

Jackie Parkes, Nichola McCullough, Ann Madden & Elaine McCahey

*Journal of Advanced Nursing**, 2009**, Vol. 65, pp. 2311-2323.*

Title: The health of children with cerebral palsy and stress in their parents.

Aim: This paper is a report of a study conducted to describe the health of children with cerebral palsy and investigate predictors of stress in their parents.

Background: Children with severe cerebral palsy tend to have poorer health than their able-bodied peers, and their parents are more likely to be stressed and have poorer health.

Method: A cross-sectional survey with home visits using standard questionnaires was administered to parents in 2004–05. A total of 102/199 (51%) children and parents participated. The children were compared with a normative sample.

Results: Children with cerebral palsy had poorer physical health, and 79% of parents reported that their child had moderate to severe pain. Their poorer health, in comparison with the normal sample and measured by the Child Health Questionnaire, was related to feeding problems and seizures, general health perceptions to intellectual and feeding impairment, and family activities with severe motor, intellectual and feeding impairment. Poorer psychological wellbeing on the hyperactivity domain of the Strengths & Difficulties Questionnaire was related to feeding difficulties, on the pro social domain to more severe forms of all child impairments, and on the social impairment scale to intellectual impairment. Children with psychological problems had statistically significantly increased odds (OR = 7Æ2, 95% CIs 2Æ6–20Æ3) of having parents with high stress.

Conclusion: Children with cerebral palsy and associated impairments are at higher risk of poorer health and family well-being. A family-centred approach to the care of children with cerebral palsy and their families is essential to ensure both receive adequate care and support.

Psychological problems in children with hemiplegia: a European multicentre survey.

J Parkes, M White-Koning, N McCullough, A Colver

*Archives of Disease in Childhood, **2009**, Vol. 94, pp. 429-433.*

Objective: To describe the prevalence and determinants of psychological problems in European children with hemiplegia.
Design: Cross-sectional survey.
Setting: Home visits in nine European regions by research associates who administered standard questionnaires to parents.
Patients: 279 children with hemiplegia aged 8–12 years were recruited from population-based case registers. Outcome measure: Strengths and Difficulties Questionnaire comprising emotion, conduct, hyperactivity, peer problems and pro social domains. An ''impact score'' (IS) measures the social and psychological impact of the child's difficulties.
Results: Children with hemiplegia had higher mean scores on the total difficulties score (TDS) compared with a normative sample (p, 0.001). 48% and 57% of children, respectively, had borderline–abnormal TDS and IS. Significant, independent associations were observed between intellectual impairment and an increased risk for hyperactivity (odds ratio; OR 8.4, 95% CI 3.4 to 20.8), peer problems (OR 3.1, 95% CI 1.7 to 5.5), psychological and social impact (OR 3.0, 95% CI 1.6 to 5.6) when children with an intellectual quotient (IQ),50 were compared with those with an IQ .70. Boys had an increased risk for conduct (OR 2.1, 95% CI 1.2 to 3.7) and hyperactivity disorders (OR 2.5, 95% CI 1.4 to 4.6). Poor self-esteem was associated with an increased risk for peer problems (OR 5.8, 95% CI 2.5 to 13.4) and poor pro social skills (OR 7.5, 95% CI 2.4 to 23.2) compared with those with high self-esteem. Other determinants of psychological adjustment were impaired communication, severe pain and living with a single parent.
Conclusions: Many of the psychological problems identified are amenable to treatment. Special attention should be given to those at highest risk of developing psychological difficulties.

Children with cerebral palsy: severity and trends over time.
Geraldine Surmana, Karla Hemmingb, Mary J. Plattc, Jackie Parkesd, Abbi Greene, Jane Huttonb and Jennifer J. Kurinczuka
*Paediatric and Perinatal Epidemiology **2009**; 23: 513–521*
Increasingly, more very-low-birthweight infants in the developed world are now expected to survive the neonatal period than was previously the case. There are concerns that there may be a related increase in the number of infants developing severe sensorimotor impairments. Pooled data from five registers contributing to the UK Network of Cerebral Palsy Registers, Surveys and Databases were used to identify patterns of motor impairment in relation to additional impairments and to birthweight, and to assess whether prevalence of cerebral palsy (CP) by birthweight and by severity of motor impairment had changed over time. Low-birthweight infants are at greater risk of developing CP than larger-birthweight babies. The CP rate amongst children with birthweights <2500 g was significantly higher at 16 per 1000 livebirths [95% confidence interval (CI) 14.9, 16.2] than 1.2 per 1000 livebirths [95% CI 11, 1.2] for normal-birthweight children. Despite being at greater risk of developing CP, smaller-birthweight babies are proportionately less likely to develop the most severe forms of motor impairment. Of those born weighing _2500 g, 23% compared with 15% weighing <1000 g ($P < 0.001$) were in the most severely motor impaired group. Severe motor impairment is associated with higher levels of additional impairments. CP rates for each motor impairment group in the 1990s were similar to those in the late 1970s. Rates of CP among infants born below normal birthweight are high but have decreased over time. The CP rate for infants weighing 1000–1499 g at birth decreased from around 180 per 1000 livebirths in 1979 to around 50 per 1000 livebirths from the early 1990s onwards.

Determinants of participation and quality of life of adolescents with cerebral palsy: a longitudinal study (SPARCLE2).

Allan F Colver, Heather O Dickinson and SPARCLE group

*BMC Public Health **2010***

Background: Children and adults with impairments such as cerebral palsy have lower participation in life situations than able-bodied people. Less is known about their subjective perception of their lives, called their quality of life. During adolescence, rapid physical and psychological changes occur; although these may be more difficult for disabled than for able-bodied adolescents, little research has examined the lives of disabled adolescents. In 2003-4 a European Union funded project, SPARCLE, visited 818 children aged 8-12 years with cerebral palsy, sampled from population-based registers in nine European regions. The quality of life reported by these disabled children was similar to that of the general population but their participation was lower; levels of participation varied between countries even for children with similar severity of cerebral palsy. We are currently following up these children, now aged 13-17 years, to identify (i) to what extent contemporaneous factors (pain, impairment, psychological health and parental stress) predict their participation and quality of life, (ii) what factors modify how participation and quality of life at age 8-12 years are associated with participation and quality of life in adolescence, and (iii) whether differences between European countries in participation and quality of life can be explained by variations in environmental factors.

Methods/Design: Trained researchers will visit families to administer questionnaires to capture the adolescents' type and severity of impairment, socio-demographic characteristics, participation, quality of life, psychological health, pain, environmental access and parental stress. We will use multivariable models (linear, logistic or ordinal) to assess how adolescent participation, quality of life, psychological health, pain, environmental access and parental stress, vary with impairment and socio-demographic characteristics and, where possible, how these outcomes compare with general population data. For participation and quality of life, longitudinal analyses will assess to what extent these are predicted by corresponding levels in childhood and what factors modify this relationship. Structural equation modelling will be used to identify indirect relationships mediated by other factors.

Socio-economic inequalities in cerebral palsy prevalence in the United Kingdom: a register-based study.

Helen Dolka, Sam Pattendenc, Sandra Bonellied, Allan Colvere, Andy Kingf, Jennifer J Kurinczukf, Jackie Parkesb, Mary Jane Plattg and Geraldine Surman

*Paediatric & Perinatal Epidemiology, **2010**, Vol. 24, pp. 149-155*

Evidence is unclear as to whether there is a socio-economic gradient in cerebral palsy (CP) prevalence beyond what would be expected from the socio-economic gradient for low birthweight, a strong risk factor for CP. We conducted a population-based study in five regions of the UK with CP registers, to investigate the relationship between CP prevalence and socio-economic deprivation, and how it varies by region, by birthweight and by severity and type of CP. The total study population was 1 657 569 live births, born between 1984 and 1997. Wards of residence were classified into five quintiles according to a census-based deprivation index, from Q1 (least deprived) to Q5 (most deprived). Socio-economic gradients were modelled by Poisson regression, and region-specific estimates combined by meta-analysis.

The prevalence of post neonatally acquired CP was 0.14 per 1000 live births overall. The mean deprivation gradient, expressed as the relative risk in the most deprived vs. the least deprived quintile, was 1.86 (95% confidence interval [95% CI 1.19, 2.88]). The prevalence of non-acquired CP was 2.22 per 1000 live births. For non-acquired CP the gradient was 1.16 [95% CI 1.00, 1.35]. Evidence for a socio-economic gradient was strongest for spastic bilateral cases (1.32 [95% CI 1.09, 1.59]) and cases with severe intellectual impairment (1.59 [95% CI 1.06, 2.39]). There was evidence for differences in gradient between regions. The gradient of risk of CP among normal birthweight births was not statistically significant overall (1.21 [95% CI 0.95, 1.54]), but was significant in two regions. There was non-significant evidence of a reduction in gradients over time.
The reduction of the higher rates of post neonatally acquired CP in the more socioeconomically deprived areas is a clear goal for prevention. While we found evidence for a socio-economic gradient for non-acquired CP of antenatal or perinatal origin, the picture was not consistent across regions, and there was some evidence of a decline in inequalities over time. The steeper gradients in some regions for normal birth weight cases and cases with severe intellectual impairment require further investigation

The impact of intellectual impairment on the quality of life of children with cerebral palsy.
Ann Madden and Jackie Parkes
*Learning Disability Practice, **2010**, Vol. 13, pp. 28-33*
Cerebral palsy is the most common cause of physical disability in childhood and half of these children will have an intellectual impairment. This article reports on the quality of life of children with cerebral palsy and explores the impact of intellectual impairment. Learning disability nurses have a critical role to play in improving the quality of life for this patient group, particularly in relation to their physical health and creating opportunities to promote social skill development and social inclusion.

Nursing babies at risk of cerebral palsy in the neonatal period.
Sharon Nurse, Jackie Parkes
*Journal of Neonatal Nursing, **2010**, Vol. 16, pp. 215-220*
Abstract Cerebral palsy (CP) is a leading cause of physical disability in childhood with evidence that 90% of children with the condition sustain damage or malformation to their developing brain during the antenatal period. With half of all cases of children with CP being born prematurely many need extra help and support in the neonatal period. The aims of neonatal nursing for this high risk group include prevention of further neurological complications as well as working maintain stable infant physiology and provide information and support to parents. While a diagnosis of CP is seldom welcome there is now evidence that most children with CP are mildly affected, most have a normal life expectancy, most are well adjusted and most are happy, reporting a quality of life similar to children without CP. Neonatal nurses are ideally placed to communicate and prepare parents of children at high risk of developing CP about more positive future likely outcomes than previously thought.

Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study.

Jackie Parkes Nan Hill Mary Jane Platt Caroline Donnelly

*Developmental Medicine & Child Neurology, **2010**, Vol. 52 pp. 1113-1119*

Aim: To report the prevalence, clinical associations, and trends over time of oromotor dysfunction and communication impairments in children with cerebral palsy (CP).

Method: Multiple sources of ascertainment were used and children followed up with a standardized assessment including motor speech problems, swallowing ⁄ chewing difficulties, excessive drooling, and communication impairments at age 5 years.

Results: A total of 1357 children born between 1980 and 2001 were studied (781 males, 576 females; median age 5y 11mo, interquartile range 3–9y; unilateral spastic CP, n=447; bilateral spastic CP, n=496; other, n=112; Gross Motor Function Classification System [GMFCS] level: I, 181; II, 563; III, 123; IV, 82; IV, 276). Of those with 'early-onset' CP (n=1268), 36% had motor speech problems, 21% had swallowing ⁄ chewing difficulties, 22% had excessive drooling, and 42% had communication impairments (excluding articulation defects). All impairments were significantly related to poorer gross motor function and intellectual impairment. In addition, motor speech problems were related to clinical subtype; swallowing ⁄ chewing problems and communication impairments to early mortality; and communication impairments to the presence of seizures. Of those with CP in GMFCS levels IV to V, a significant proportion showed a decline in the rate of motor speech impairment (p=0.008) and excessive drooling (p=0.009) over time.

Interpretation: These impairments are common in children with CP and are associated with poorer gross motor function and intellectual impairment.

Age-related changes in energy efficiency of gait, activity, and participation in children with cerebral palsy.

Kerr, C; McDowell, B; Parkes, JL; Stevenson, M; Cosgrove, A.

*Developmental Medicine and Child Neurology, **2011**, Vol. 53, pp. 61-67.*

Aim: The aim of this study was to use a prospective longitudinal study to describe age-related trends in energy efficiency during gait, activity, and participation in ambulatory children with cerebral palsy (CP).

Method: Gross Motor Function Measure (GMFM), Paediatric Evaluation of Disability Inventory (PEDI), and Lifestyle Assessment Questionnaire-Cerebral Palsy (LAQ-CP) scores, and energy efficiency (oxygen cost) during gait were assessed in representative sample of 184 children (112 male; 72 female; mean age 10y 9mo; range 4–16y) with CP. Ninety-four children had unilateral spastic CP, 84 bilateral spastic CP, and six had other forms of CP. Fifty-seven were classified as Gross Motor Function Classification System(GMFCS) level I, 91 as level II, 22 as level III, and 14 as level IV). Assessments were carried out on two occasions (visit 1 and visit 2) separated by an interval of 2 years and 7 months. A total of 157 participants returned for reassessment.

Results: Significant improvements in mean raw scores for GMFM, PEDI, and LAQ-CP were recorded; however, mean raw oxygen cost deteriorated over time. Age-related trends revealed gait to be most inefficient at the age of 12 years, but GMFM scores continued to improve until the age of 13 years, and two PEDI subscales to age 14 years, before deteriorating ($p<0.05$). Baseline score was consistently the single greatest predictor of visit 2 score. Substantial agreement in **GMFCS** ratings over time was achieved (jlw =0.74–0.76).

Interpretation: These findings have implications in terms of optimal provision and delivery of services for young people with CP to maximize physical capabilities and maintain functional skills into adulthood.

The health of children and young people with cerebral palsy: A longitudinal, population-based study.

Nichola McCullough, Jackie Parkes α, Claire Kerr, Brona C. McDowell
International Journal of Nursing Studies, ***2011*** *Feb 15.*

Background: Cerebral palsy (CP) is a chronic condition about which little is known in relation to the long term stability of and factors influencing health.
Objectives: To describe the health status of 4–17 year olds with ambulant CP, compare with the general population and identify factors predicting change in health over time.
Design: A longitudinal, clinical survey.
Setting: A regional hospital-based Gait Analysis Laboratory.
Participants: Those aged 4–17 years and able to walk at least 10 m independently were identified from a case register of people with CP. A total of 184 subjects took part (38% of all eligibles in the region); 154 (84%) returned for a second assessment on average 2.5 years later.
Methods: The Child Health Questionnaire (Parent-form-50) was completed by 184 parentsat time 1, and 156 at time 2.
Results: Children and young people with CP have significantly poorer health across a number of domains when compared to children in the general child population. Over time improvements occurred in behaviour ($p = 0.01$), family activities ($p < 0.001$) and physical functioning ($p = 0.05$). Linear regression showed that gross motor function ($p < 0.001$) and cerebral palsy subtype ($p < 0.05$) were associated with changes in physical functioning; age was associated with changes in behaviour ($p = 0.007$) and family activities ($p = 0.01$); and communication ability was significantly associated with changes in family activities ($p = 0.005$).
Conclusions: Children and young people with CP have poorer health than their able bodied peers but relatively stable health over 2.5 years. Where change occurred, it was for the better.

Parenting stress and children with cerebral palsy: a European cross-sectional survey.

Jackie Parke, Barbara Caravale, Marco Marcelli, Francesco Franco, Allan Colver
Developmental Medicine & Child Neurology, ***2011*** *Sep; 53(9): 815-21*

Aim: The aim of this study was to describe stress in the parents of children with cerebral palsy and investigate associations with very high stress.
Method: A cross-sectional survey was conducted of parents of 818 children aged 8 to 12 years from nine regions in Europe. Families were eligible to participate if they were living in one of the specified geographic areas. Parental stress was captured using the Parenting Stress Index Short Form, which has 36 items and takes 10 minutes to complete. Parents rate items on a 5-point Likert scale, with higher scores indicating higher stress. The Short Form yields scores on three subscales and a Total Stress score. A trained research associate administered the questionnaire in the child's home and visits lasted 90 to 120 minutes. All data collected were reported by parents unless otherwise stated.
Results: The Total Stress score on the Parenting Stress Index was dichotomized into scores of less than 99 or 99 or more, the latter indicating 'very high' stress. Most respondents were mothers (94%), and 26%reported very high stress levels. The parents of children with communication impairment had higher odds for very high stress (odds ratio [OR] 1.9; 95% confidence interval [CI]

1.2–3.0) than those whose child had no such impairment; the parents of children with moderate or severe pain had higher odds for very high stress (OR 1.7 [95%CI 1.1–2.4] and 2.5 [95% CI 1.5–4.3] respectively)than those whose child had no pain; and the parents of children with an intellectual impairment had higher odds for very high stress (OR 1.8; 95%CI 1.2–2.9) than those whose child had none. There was no association between very high stress and motor impairment. The subscales 'parent–child dysfunctional interaction' and 'difficult child' contributed most to the Total Stress score.
Interpretation: Parents of children with communication difficulties, intellectual impairment, or pain are at very high risk of stress. The final model explained 12% of the observed variation in very high stress.

Accommodative Dysfunction in Children with Cerebral Palsy: A Population-Based Study.
Julie F. McClelland, Jackie Parkes, Nan Hill, A. Jonathan Jackson and Kathryn J. Saunders
*Archives of Physical Medicine and Rehabilitation, **2012**.*
PURPOSE. To determine the prevalence, nature, and degree of accommodative dysfunction among children with different types and severities of cerebral palsy (CP) in Northern Ireland. **METHODS.** Ninety subjects with CP (aged 4–15 years) were recruited through the Northern Ireland CP Register (NICPR). Modified Nott dynamic retinoscopy was used to measure lag and lead of accommodation at three test distances: 25 cm (4 D), 16.7 cm (6 D), and 10 cm (10 D) with the distance correction in place. Accommodative function was also assessed in an age-matched control group (*n* _ 125) for comparison. Each subject's neurologic status was derived from the NICPR. **RESULTS.** Children with CP demonstrate significantly reduced accommodative responses compared with their neurologically normal peers. Of the subjects with CP, 57.6% demonstrated an accommodative lag outside normal limits at one or more distances. Reduced accommodative responses were significantly associated with more severe motor and intellectual impairments (ANOVA *P* _ 0.001, *P* _ 0.01, respectively). **CONCLUSIONS.** Brain injury such as that present in CP has a significant impact on accommodative function. These findings have implications for the optometric care of children with CP and inform our understanding of the impact of early brain injury on visual development.

Association Between Participation in Life Situations of Children With Cerebral Palsy and Their Physical, Social, and Attitudinal Environment: A Cross-Sectional Multicenter European Study.
Allan Colver, MD, Ute Thyen, MD, Catherine Arnaud, MD, Eva Beckung, PhD, Jerome Fauconnier, MD, Marco Marcelli, MD, Vicki McManus, MSc, Susan I. Michelsen, MD, Jackie Parkes, PhD, Kathryn Parkinson, PhD, Heather O. Dickinson, PhD
*Archives of Physical Medicine and Rehabilitation, **2012.***
Abstract. Association between participation in life situations of children with cerebral palsy and their physical, social, and attitudinal environment: a cross-sectional multicenter European study.
Objective: To evaluate how participation of children with cerebral palsy (CP) varied with their environment.
Design: Home visits to children. Administration of Assessment of Life Habits and European Child Environment Questionnaires. Structural equation modeling of putative associations between specific domains of participation and environment, while allowing for severity of child's impairments and pain.
Setting: European regions with population-based registries of children with CP.

Participants: Children (n_1174) aged 8 to 12 years were randomly selected from 8 population-based registries of children with CP in 6 European countries. Of these, 743 (63%) agreed to participate; 1 further region recruited 75 children from multiple sources. Thus, there were 818 children in the study.
Interventions: Not applicable.
Main Outcome Measure: Participation in life situations.
Results: For the hypothesized associations, the models confirmed that higher participation was associated with better availability of environmental items. Higher participation in daily activities—mealtimes, health hygiene, personal care, and home life—was significantly associated with a better physical environment at home (*P*_.01). Mobility was associated with transport and physical environment in the community. Participation in social roles (responsibilities, relationships, recreation) was associated with attitudes of classmates and social support at home. School participation was associated with attitudes of teachers and therapists. Environment explained between 14% and 52% of the variation in participation.
Conclusions: The findings confirmed the social model of disability. The physical, social, and attitudinal environment of disabled children influences their participation in everyday activities and social roles.

Predictors of drop-out in a multi-centre longitudinal study of participation and quality of life of children with cerebral palsy.
Dickinson H; Rapp M; Arnaud C; Carlsson M; Colver AF; Fauconnier J; Lyons A; Marcelli M; Michelsen SI; Parkes, JL; Parkinson K.
*BMC research notes, **2012**.*
Background: SPARCLE is a study across nine European regions which examines the predictors of participation and quality of life of children with cerebral palsy. Children and their families were initially interviewed in 2004/2005when the children were aged 8–12 years (SPARCLE1); they were approached again in 2009/2010 at age 13–17 years (SPARCLE2). The objective of this report is to assess potential for bias due to family non-response in SPARCLE2.
Logistic regression was used to assess whether socio-demographic factors, parental stress and child impairment were related to non-response, both overall and by category (failure to trace families, death of child, traced families declining to participate).
Results: Of the 818 families who participated in SPARCLE1, 224/818 (27%) did not participate in SPARCLE2. 51/818 (6%) were not traced. Among the 767 traced families, 32/767 (4%) children with cerebral palsy had died, seven children had been incorrectly diagnosed as having cerebral palsy, thirteen families had moved out of the region and one family had language problems. Of the remaining 714 families, 120/714 (17%) declined to participate.
Drop-out between SPARCLE1 and SPARCLE2 varied significantly between regions; families were more difficult to trace and more likely to decline to participate if the parents' educational qualifications, as recorded in SPARCLE1, were lower; they were also more likely to decline to participate if SPARCLE1 recorded that they were more stressed or if they had not completed a SPARCLE1 stress questionnaire.
Conclusions: To reduce the risk of bias, all SPARCLE2 analyses should allow for factors (region and walking ability) which determined the sampling strategy, either by adjusting for these factors or by using sampling weights. Further analyses should be performed, adjusting for additional factors that were associated with non-response: parents' educational qualifications, family structure and

parental stress. To allow for differential non-response in studies which sample from population registers, such registers should routinely record socio-demographic information.

Recruitment bias and characteristics of participants with severe cerebral palsy in a cross-sectional survey.

Jackie Parkes, Collette Donnelly, Brona McDowell & Catherine Duffy

*Journal of Advanced Nursing, **2012** Feb; 68(2):368-78.*

Aim. This article is a report of recruitment bias in a sample of 5–25-year-oldpatients with severe cerebral palsy.

Background. The way in which study participants are recruited into research can be a source of bias.

Method. A cross-sectional survey of 5–25-year-old patients with severe cerebral palsy using standardized questionnaires with parents/carers was undertaken in 2007/2008. A case register was used as the sampling frame, and 260 families were approached: 178/260 (68%) responded and 82/260 families never replied (non respondents).

Among responders: 127/178 (71%) opted in to the study, but only 123/127 were assessed, and 82/178 were opted out (or refused). Multivariable logistic regression giving odds ratios was used to study the association between participant characteristics and study outcomes (responders vs. non-responders; opting in vs. opting out; assessed vs. eligible, but not assessed).

Results. Responders (compared with non-responders) were significantly more likely to have a family member with cerebral palsy who was male and resident in more affluent areas. Families who opted in (compared with those opting out and refusing) were more likely to have a family member with cerebral palsy and intellectual impairment and to reside in certain geographical areas. Families who were actually assessed (compared with all eligible, but not assessed) were more likely to have a family member with cerebral palsy and intellectual impairment.

Conclusion. Several sources of bias were identified during recruitment for this study. This has implications for the interpretation and conclusions of surveys of people with disabilities and complex needs.

Stability of motor function and associated impairments between childhood and adolescence in young people with cerebral palsy in Europe.

Malin Nystrand; Eva Beckung; Heather Dickinson; Allan Colver

*Developmental Medicine and Child Neurology **2014***

AIM The aim of the study was to investigate whether impairments associated with cerebral palsy were stable between childhood and adolescence. **METHOD** The Study of Participation of Children with Cerebral Palsy Living in Europe (SPARCLE) longitudinal study was conducted in nine European regions. In total, 818 children aged 8 to 12 years were randomly selected from population-based registers; 594 (73%) were followed up at the age of 13 to 17 years (344 males, 250 females; median age 10y 4mo) Research associates visited them in their homes and recorded their motor function and additional impairments. Stability of impairment was assessed using the weighted kappa coefficient. **RESULTS** The proportion of participants whose level of impairment remained unchanged varied from 63% for fine motor function to 98% for hearing. For gross motor function, communication, and cognitive level, the kappa and the lower bound of its 95% confidence interval (CI) were above 0.75, indicating stability between childhood and adolescence; for fine motor function and feeding, the kappa was above 0.75 but the lower bound of the 95% CI was below 0.75, indicating probable stability; for seizures and vision, the kappa was below 0.75, although the upper

bound of the 95% CI was above 0.75, indicating possible change; for hearing the kappa and its entire CI were below 0.75, indicating change. Overall, 81% of participants had no seizures in childhood, of whom 93% were seizure-free in adolescence. **INTERPRETATION** Motor function and additional impairments were generally stable between childhood and adolescence.

Predictors of participation of adolescents with cerebral palsy: A European multi-centre longitudinal study.

*Van Moˆ Dang , Allan Colver , Heather O. Dickinson , Marco Marcelli , Susan I.Michelsen , Jackie Parkes , Kathryn Parkinson , Marion Rapp ;Catherine Arnaud, Malin Nystrand, Jerome Fauconnier Research in Developmental Disabilities 36 (**2015**) 551–564*

We investigated whether childhood factors that are amenable to intervention(parenting stress, child psychological problems and pain) predicted participation in daily activities and social roles of adolescents with cerebral; palsy(CP).We randomly selected 1174 children aged 8–12 years from eight population-based registers of children with CP in six European countries;743(63%) agreed to participate. One further region recruited 75 children from multiple sources. These 818 children were visited at home at age 8–12 years, 594(73%) agreed to follow-up at age13–17years. We used the following measures: parent reported stress(Parenting Stress Index Short Form),their child's psychological difficulties (Strength and Difficulties Questionnaire)and frequency and severity of pain; either child or parent reported the child's participation (LIFE Habits questionnaire).We fitted a structural equation model to each of the participation domains, regressing participation in childhood and adolescence on parenting stress, child psychological problems and pain, and regressing adolescent factors on the corresponding childhood factors; models were adjusted for impairment, region, age and gender. Pain in childhood predicted restricted adolescent participation in all domains except Mealtimes and Communication (standardized total indirect effects b 0.05 to 0.18, $0.01 < p < 0.05$ to $p < 0.001$, depending on domain). Psychological problems in childhood predicted restricted adolescent participation in all domains of social roles, and in Personal Care and Communication (b 0.07 to 0.17, $0.001 < p < 0.01$ to $p < 0.001$). Parenting stress * in childhood predicted restricted adolescent participation in Health Hygiene, Mobility and Relationships (b 0.07 to 0.18, $0.001 < p < 0.01$ to $p < 0.001$). These childhood factors predicted adolescent participation largely via their effects on childhood participation; though in some domains early psychological problems and parenting stress in childhood predicted adolescent participation largely through their persistence into adolescence. We conclude that participation of adolescents with CP was predicted by early modifiable factors related to the child and family .Interventions for reduction of pain, psychological difficulties and parenting stress in childhood are justified not only for their intrinsic value, but also for probable benefits to childhood and adolescent participation.

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