Healthcare use by children and young adults with cerebral palsy

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ABBREVIATION

NICPR	Northern Ireland Cerebral Palsy Register

AIM To link routinely collected health data to a cerebral palsy (CP) register in order to enable analysis of healthcare use by severity of CP.

METHOD The Northern Ireland Cerebral Palsy Register was linked to hospital data. Data for those on the CP register born between 1st January 1981 and 31st December 2009 and alive in 2004 were extracted, forming a CP cohort (*n*=1684; 57% males, 43% females; aged 0–24y). Frequencies of healthcare events, and the reasons for them, were reported according to CP severity and compared with those without CP who had had at least one hospital attendance in Northern Ireland within the study period.

RESULTS Cases of CP represented 0.3% of the Northern Ireland population aged 0 to 24 years but accounted for 1.6% of hospital admissions and 1.6% of outpatient appointments. They had higher rates of elective admissions and multi-day hospital stays than the general population. Respiratory conditions were the most common reason for emergency admissions. Those with most severe CP were 10 times more likely to be admitted, and four times more likely to attend outpatients, than those with mild CP.

INTERPRETATION Linkage between a register and routinely collected healthcare data provided a confirmed cohort of cases of CP that was sufficiently detailed to analyse healthcare use by disease severity.

Cerebral palsy (CP) is a chronic condition associated with extensive comorbidity and healthcare needs.[1] An increasing population is likely to lead to an increase in the number of children and young adults living with CP, which will significantly impact the planning of health and social care.[1] There have been attempts[2] to use routinely collected healthcare data to identify the extent and nature of health care use in the UK for children and young adults with CP. However, the clinical details collected within these data sets were limited and thus reduced the value of analyses.[3]

Researchers in Victoria, Australia, have linked the regional CP register to hospital data[4] and described the rate and reasons for hospital admissions for children and young adults with CP of differing severity, according to the Gross Motor Function Classification System (GMFCS).[5] Compared with the general population, hospital admissions for children and young adults with CP accounted for 1.5% of all admissions, were more likely to be surgical, and were more likely to be elective than emergency.

Within the UK, the Northern Ireland Cerebral Palsy Register (NICPR) held by Queen's University Belfast is the last remaining CP register commissioned by the National Health Service owing to loss of funding for the four other registers and the UKCP database.[6] The NICPR is a confidential, opt-in register of children and young adults with CP in Northern Ireland born since 1977 or living in the area from 1992. It aims to use a systematic approach to monitor and provide surveillance of CP over time, and to support research into the condition.

Healthcare professionals involved in the care of children and young adults with CP (and sometimes parents) inform the NICPR monthly about any new cases with the condition. The NICPR collects patient information at notification including demographics, type and severity of movement problems (GMFCS), other problems (e.g. seizures, learning, speech and language, vision and hearing), school attended, and professionals seen. This information provides a snapshot of the diagnosis and the child's impairments at the age of approximately 5 years.[7]

Northern Ireland is the smallest of the four UK countries. The mid-year population estimate of 0- to 24-year-olds in Northern Ireland in 2009 was 606 042.[8] Healthcare data are routinely collected from hospitals across the five Health and Social Care Trusts in Northern Ireland: Belfast, Northern, South Eastern, Southern, and Western.[9]

This study aimed to link NICPR and routinely collected healthcare data to conduct an exploratory analysis of healthcare use according to severity of CP, to identify factors that influence CP hospital admissions and outpatient attendances, and to compare events to those without CP.

METHOD

Study design and setting

This retrospective data linkage study compared the healthcare use of children and young adults aged 0 to 24 years with a diagnosis of CP between 1st January 2004 and 31st December 2014 with those aged 0 to 24 years without a diagnosis of CP who had had at least one hospital episode in Northern Ireland within the same time period. This study was conducted at Cardiff University in partnership with the Secure Anonymous Information Linkage Databank at Swansea University and was ethically approved as part of a wider project.[3] Data received complied with relevant requirements, including the Information Commissioners Office (NCEPOD Z5442652) and the NHS Act 2006 (15/CAG/0210). As anonymous data were requested, ethical approvals were not required, but approvals from Northern Ireland Statistics and Research Agency were obtained, and 'approved researcher status' for each member of the data linkage team was sought and granted to access data from the Office for National Statistics. The NICPR has ongoing ethical consent, and data are recorded and used in line with GDPR regulations, thus safeguarding the privacy and safety of participants.

Data sources and data received

The NICPR

The NICPR data set received included children and young adults born between 1st January 1981 and 31st December 2009 and alive in 2004 so that children and young adults would be aged 0 to 24 years during the study period and a diagnosis of CP would have been confirmed. The encrypted Health and Care Number, a unique identifier for an individual allocated at birth, enabled linkage to the healthcare data. After voluntary notification of children with a diagnosis of CP to the NICPR by healthcare professionals or parents, a standardized assessment form collecting patient demographic and clinical information is completed by a clinician responsible for the child's care, usually a community paediatrician. In line with Surveillance of Cerebral Palsy in Europe guidance,[10] cases are not confirmed on the NICPR until a formal diagnosis of CP has been made, usually around the child's fifth birthday. The only exceptions to this are children who died before the diagnosis could be confirmed but who received a diagnosis after the age of 2 years, and cases who moved out of the catchment area before diagnosis confirmation but who received a diagnosis after the age of 3 years. These criteria are consistent with those of other CP registers in Europe to avoid underestimation of the prevalence of CP.[11]

Admissions and discharges inpatients

The admissions and discharges inpatients data set from the patient administration system includes recorded information for patients admitted to acute hospitals as inpatients or day cases. Each record relates to an individual episode within hospital. (Patients may have multiple episodes if they are transferred to the care of a consultant in a different specialty or hospital). To explore service use, hospital episodes were combined into whole spells, leaving one record (the first) per health event. Hospital spells were created using the method described in the 'Each and Every Need' report.[3] All admissions and discharges records from hospitals across the Northern Ireland trusts, including independent hospitals, between the years 2004 and 2014, were received. Maternity and other admissions, including births and patient transfers (see Business Services Organisation A&D Metadata: http://www.hscbusiness.hscni.net/services/2512.htm), were excluded from analyses of healthcare use as they were not admissions due to ill health or injury.

What this paper adds

- Children and young adults with cerebral palsy (CP) had higher rates of hospital admissions than the general population.
- CP outpatient appointments were more likely therapeutic and for younger children.
- Hospital admission and appointment rates increased with CP severity and over time.

Outpatients

The outpatient data set extract from the patient administration system in Northern Ireland details information on new and review attendances, ward attendances, missed appointments, cancellations (both hospital and patient), and consultant specialty. The data set includes outpatient activity at integrated clinical assessment and treatment services, and activity undertaken in the independent sector. All data recorded for 0- to 24-year-olds between 2004 and 2014 were requested; however, before 2010 they appeared to be incomplete so only data for the years 2010 to 2014 were analysed.

Participants

All children and young adults confirmed on the NICPR born between 1st January 1981 and 31st December 2009 and alive in 2004 were regarded as true CP cases. Children and young adults born after 2009 were excluded as their diagnosis of CP may not have been confirmed before their fifth birthday. CP severity was categorized according to GMFCS level.

Data linkage, access, and cleaning methods

Data linkage of the NICPR to healthcare data was undertaken by the Honest Broker Service for Health and Social Care, which is part of the Business Services Organisation in Northern Ireland. Data linkage was deterministic and based on the Health and Care Number. Thus, data for any individuals without a Health and Care Number (e.g. visitors to Northern Ireland) could not be included in the linkage. Once linked, data tables were sent securely to the Secure Anonymous Information Linkage databank. Data consistency checks were conducted by Secure Anonymous Information Linkage analysts and analysis of the data was

conducted by researchers at Cardiff University through remote desktop.

Data analysis

Population prevalence of CP was estimated using the number of confirmed NICPR cases aged between 0 and 24 years between the years 2004 and 2014 as the numerator, and dividing it by the average of the Office for National Statistics national population mid-year estimates between 2004 and 2014 for 0- to 24-year-olds. Healthcare use for the cohort of cases of CP identified from the NICPR was described according to patient age, frequency and type of admission, primary diagnosis, length of stay, and GMFCS level. The primary diagnoses, which describe the main reason for each admission, were categorized according to the International Statistical Classification of Diseases and Related Health Problems, 10th revision standard disease groupings.[12] Descriptive statistics were used to compare NICPR and non-NICPR admissions and outpatient appointments, and differences in healthcare use by CP severity. Incident rate ratios were calculated for the NICPR cases by dividing events through by the person-years; rates for those without CP were calculated by dividing events through by the sum of Office for National Statistics mid-year estimates between 2004 and 2014, which was 6 666 459 persons. The χ^2 test was used to determine whether admissions for the NICPR population (n=12 191) significantly differed from those without CP (n=732 449) in proportions of different age groups, sex, admission type, admission method, length of stay, specialty, and primary diagnosis. Rates of admissions and outpatient appointments for those with a diagnosis of CP were calculated using the number of events divided by the number of person-years on the NICPR. All analyses were conducted using Stata 14 (StataCorp, College Station, TX, USA) and R (R Core Team; R Foundation for Statistical Computing, Vienna, Austria).

RESULTS

The NICPR cohort

The NICPR cohort consisted of 1684 children and young adults with CP (57% males, 43% females) aged 0 to 24 years between the years 2004 and 2014, with a total exposure time of 13544.7 person-years. This gave an estimated CP prevalence of 2.7 (95% confidence interval 2.7–2.9) per 1000 persons within the general population from 2004 to 2014. The number of cases born in each year is presented in Figure S1 (online supporting information). Most (n=1528, 91%) cases had either spastic unilateral (n=841, 50%) or spastic bilateral (n=687, 41%) CP, 83 (5%) had dystonic CP, 46 (3%) had athetotic CP, and fewer than 10 (0.5%) had ataxic CP. CP type was missing for 19 (1.0%) children and young adults. The most severe level of motor impairment (GMFCS levels IV and V) was present in 445 (26.4%) cases, 277 had a moderate level of motor impairment (GMFCS level III), 943 had mild motor impairment (GMFCS levels I and II), and 19 had no recorded GMFCS level.

Hospital admissions (2004–2014)

Of the 1684 children and young adults on the NICPR, 1152 (68.4%) had at least one hospital admission, totalling 12 191 hospital admissions over the study period which accounted for 1.6% of the total 744 640 hospital admissions in the general population for the same age group. The non-CP sample of 333 812 children and young adults had 732 449 admissions. The hospital admission rate for NICPR cases was 79 per 100 person-years during the study period. The rate of hospital admissions increased with increasing severity of CP and was 10 times greater for children and young adults classified as functioning in GMFCS level V than for those functioning in GMFCS level I. Rates of hospital admissions decreased with increasing age (Table 1), males had a higher rate of admissions than females (Table 1), and rates increased over the study period, particularly for the children in the 0- to 4-year and 5- to 9-year age groups, but remained relatively stable for young people (15–19y) and young adults (20–24y; Fig. 1).

Children and young adults with CP had a significantly greater proportion of elective rather than emergency admissions (72.0% of CP admissions compared with 53.4% for general population; p<0.001). This was partly due to children and young adults with CP having many more planned 'holiday relief care' admissions (coded within 'other' category). Reasons for admissions were significantly different (p<0.001) between the two groups; respiratory or neurological conditions were more frequent in children and young adults with CP. Patients with CP had a significantly greater (p<0.001) proportion of multi-day admissions and fewer day-case admissions than the general population; multi-day admissions accounted for 65.2% of total CP admissions versus 47.7% for the general population. Children and young adults with CP had a greater proportion of admissions to psychiatry specialties and were admitted under the 'mental handicap' specialty.

Outpatient appointments (2010–2014)

Children and young adults with CP had a total of 13 414 outpatient appointments during the 5-year period (person-years=6182.4), accounting for 1.6% of the total 822 271 outpatient appointments giving an estimated outpatient appointment rate of 226 per 100 person-years for children and young adults with CP. Appointment rates by age group and year show that rates have remained stable or declined for all age groups except 0- to 4-year-olds (Fig. 1).

The rate of outpatient attendances increased with GMFCS severity and was four times greater for those classified as functioning in GMFCS level V than those functioning in GMFCS level I (Table 1). The rate of appointments decreased with increasing age and was greater for males than females (Table 1).

Table 1: The number, percentage, and rates (per 100 person-years) of children and young people with cerebral palsy (CP) who had an admission or outpatient appointment

	Children and young adults, *n*	Children and young adults admitted to hospital at least once, *n* (%)	Rate of hospital admissions for children and young adults with CP per 100 person-years (2004–2014)	Children and young adults with at least one outpatient appointment, *n* (%)	Rate of outpatient appointments per 100 person-years (2010–2014)
GMFCS level					
I	285	166 (58.2)	21.9	80 (28.3)	95.1
II	658	425 (64.6)	27.6	202 (30.7)	138.2
III	277	201 (72.6)	58.4	90 (32.5)	211.9
IV	105	84 (80.0)	69.2	34 (32.4)	483.1
V	340	259 (76.2)	240.6	136 (40.0)	464.1
Sex					
Male	960	672 (70.0)	90.6	266 (27.8)	237.1
Female	724	480 (66.3)	64.1	359 (49.6)	212.4

Data are by severity (Gross Motor Function Classification System [GMFCS] level) and sex for children and young adults on the Northern Ireland Cerebral Palsy Register (NICPR). Seventeen cases on the NICPR had no GMFCS level recorded.

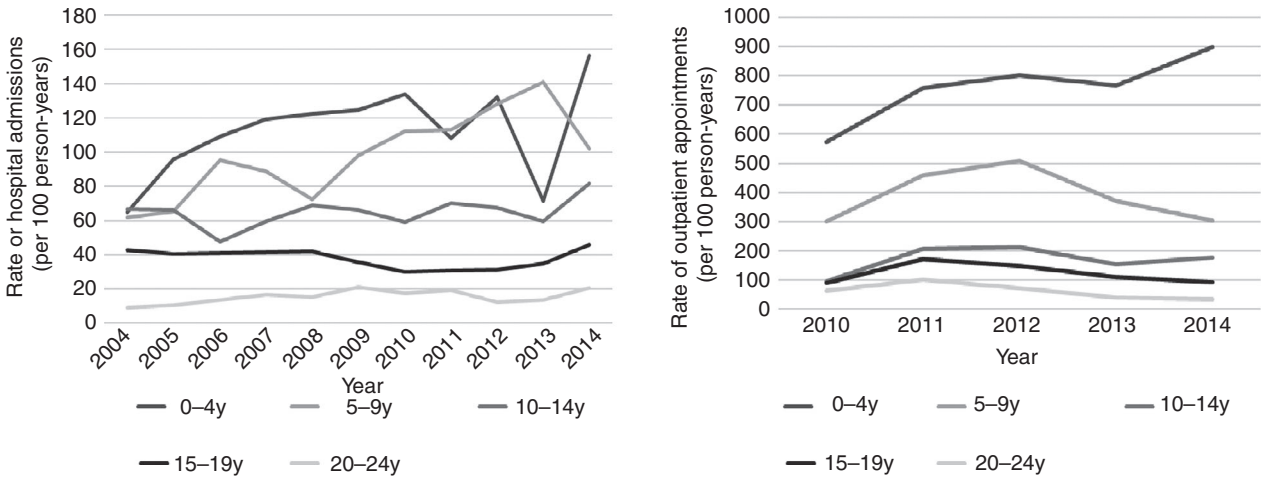

Figure 1: Rate of hospital admissions (per 100 person-years) for those on the Northern Ireland Cerebral Palsy Register (NICPR) by age group and year, and rate of outpatient appointments (per 100 person-years) for those on the NICPR by age group and year.

The patterns and type of outpatient appointments for children and young adults with CP compared with the general population was significantly different (Table S1, online supporting information). Children and young adults with CP had a significantly greater proportion of urgent appointments than the general population (16.1% of CP admissions compared with 14.9% for general population; $p<0.01$). Children and young adults with CP had a greater proportion of appointments to therapy specialties than the general population (37.8% of CP appointments and 9.6% of appointments for general population).

DISCUSSION

To our knowledge, this is the first study to explore healthcare use of children and young adults with CP by severity as measured by GMFCS level, recorded within CP register data within the UK. It has followed from the National Confidential Enquiry into Patient Outcomes and Deaths[3] 'Each and Every Need' project, which did not include an analysis of healthcare use by disease severity.

Using a population-based, specialist CP register linked to routinely collected hospital data has allowed service use and morbidity to be described in more detail using measures of CP severity. During the 11-year study period, NICPR cases represented 0.3% of the Northern Ireland population of 0- to 24-year-olds, yet they experienced 1.6% of total hospital admissions, 1.6% of total outpatient appointments, and accounted for 3.2% of deaths for this age group. Admissions and outpatient appointments have also generally increased over time for children and young adults with a diagnosis of CP.

Children and young adults with CP had more elective admissions and more multi-day stays in hospital than those without CP. Emergency admissions and cause of death were most likely to be for respiratory conditions followed by neurological conditions. The frequency of admissions

was found to vary by CP severity. Those with the most severe physical disabilities (GMFCS levels IV and V) were 10 times more likely to be admitted and four times more likely to attend outpatients than those classified with the least severe impairment (GMFCS levels I or II).

These findings are consistent with those of Meehan et al.,[4] who also linked CP register data with routinely collected hospital data. They found that, overall, 80% of cases on the Victoria CP register had at least one inpatient admission between 2007 and 2014, accounting for 1.5% of all admissions within the study age group. Compared with general population admissions, CP admissions were more likely to be longer, elective, medical, and for respiratory conditions. CP severity and complexity were associated with increased admissions and a higher proportion of admissions attributable to respiratory illness.

While the NICPR relies on information provided by practitioners and hence may underestimate cases, it nevertheless provides the best opportunity to study a cohort of confirmed cases of CP. Use of UK routinely collected healthcare data alone to identify children and young adults with CP has been shown to be severely limited by quality of coding.[13] However, use of the NICPR has allowed exploration of service use in a well-defined population of children and young adults with CP, compared with those without. Additionally, this method has enabled in-depth analysis of the effect of CP severity on morbidity and healthcare service use, which is not possible using routinely collected data alone.

Coding of reasons for outpatient appointments was incomplete, and outpatient data collection in general appeared to be poor in the earlier years of the study period. This prevented exploration of trends over the full 11-year period and understanding the reasons for appointments for children and young adults with CP. There were over 100 cases on the NICPR that could not be linked to the healthcare data because of data errors. These missing data could have biased results and possibly led to underestimation of the extent of service use for NICPR cases. Furthermore, the reliance on healthcare care meant that our comparisons were only with those who had had a hospital admission within Northern Ireland rather than the total Northern Ireland population of the same age within the given time period. This population of children and young adults are likely to have poorer health than those without CP and without a hospital admission; thus, the results presented should be considered conservative estimates of the differences in healthcare use between children and young adults with and without CP.

Regardless, policy makers and service planners can use these results to estimate and organize provision of services to meet the needs of this population. Registers including the NICPR can play a key role in planning care by providing surveillance and collecting in-depth information about severity and complexity of the CP. Understanding the reasons for healthcare attendances among this population will help to identify patterns of unnecessary use of services and preventable admissions and highlight areas for intervention.

Inclusion of primary care data and community data would complete the picture of healthcare use for children and young adults with CP and enable calculation of more accurate prevalence rates. Future UK research may benefit from the recent introduction of the Children and Young People's Health Services Data Set in England, a community care data set, as children and young adults with CP are likely to be managed predominantly within community teams. Furthermore, the NHS plans for universal Systematized Nomenclature of Medicine Clinical Terms (SNOMED CT) codes to be rolled out across the UK soon. These codes will record disease severity, and if successfully implemented may obviate the need to link to register data. Nonetheless, register data will continue to be an excellent source of rich data and enable CP surveillance.

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SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Age plot of cases on the NICPR by year of birth.

Table S1: Frequencies, proportions, and incident rate ratios of hospital admissions and outpatient appointments for those on the NICPR and the non-CP population

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