1 – Method

Study aims

The overarching aims of this study were to:
- Review the quality of care provided to children and young people with a chronic neurodisability, using the cerebral palsies as exemplar conditions
- Examine the interface between care settings; and
- Assess the transition of care from paediatric to adult services.

Data were collated from a number of sources to allow the aims to be met. These are described below.

Method overview

Participation
For the organisational and clinical reviews National Health Service hospitals in England, Scotland, Wales and Northern Ireland were expected to participate as well as public hospitals in the Isle of Man, Guernsey and Jersey. Within each hospital, a named contact, referred to as the NCEPOD Local Reporter, acted as a link between NCEPOD and the hospital staff, facilitating case identification, dissemination of questionnaires and data collation.

Organisational survey
An organisational questionnaire was divided into 10 parts with the aim of collecting data from many different providers of care.

Patient and parent carer survey
Short questionnaires were made available on the NCEPOD website to enable children and young people with chronic neurodisabilities, and parent carers, to give their experience of the services they had encountered. Patient and carer support organisations were contacted to promote the survey. Local Reporters in hospitals were asked to display posters encouraging participation in the survey. Small cards were distributed with a brief explanation of the survey and the link, to be handed to patients and parent carers.

Clinical review using questionnaires and case notes
At a local level, questionnaires were sent to lead clinicians involved in a patient's care and copies of case note data were requested. These questionnaires and case notes were anonymised and put to a multidisciplinary group of clinicians to peer review the quality of care provided.

Review of routine national datasets and data linkage
At a national level, and by UK country, datasets were collated that included secondary healthcare data from England, Northern Ireland, Scotland and Wales. The Clinical Practice Research Datalink (CPRD) provided a 6.9% sample of primary care data from all four countries and linked secondary care data for a sample of GP practices in England.[12] In Wales linked primary and secondary healthcare data were also available. Data from the only remaining national cerebral palsies register and intensive care were also included where available.

Where possible anonymised data linkage was performed between datasets for individual children and young people. Data were analysed for the time period 2004-2014. The CPRD dataset was cleaned, analysed and accessed at Cardiff University. All other datasets were housed in the Secure Anonymised Information Linkage (SAIL) databank at Swansea University where the datasets were cleaned and prepared for analysis which then took place at Swansea and Cardiff University via a secure link.

All analysis relating to these data will be displayed on a grey background throughout the report.

Study Advisory Group

To help design the study and to act as a study steering group for all data collections and analysis, a Study Advisory Group (SAG) was formed. This group comprised a multidisciplinary group of clinicians as well as a family liaison officer and a carer. The clinicians represented physiotherapy, community and hospital paediatrics,

anaesthetics, neurosurgery, nursing, endocrinology, orthopaedics, palliative care medicine, rehabilitation, and general practice. The SAG identified the objectives that would be used to address the aims of the study. These are summarised under the detailed method sections below:

Method detail - organisational survey

Objectives
- To review access to healthcare services, including pathways of care and clinical leadership
- To review how healthcare services were delivered, including uni/multidisciplinary care, outreach clinics and co-location of services.

At the start of the study, a short questionnaire was sent to every trust/health board to identify which services were provided there and the lead clinician who would be responsible for completing an organisational questionnaire. The links to complete the questionnaire were then sent to the identified clinical leads for completion.

An organisational questionnaire was sent to all hospital trusts/boards where children and young people with a cerebral palsy may have been cared for. Data collected included information around pathways of care, transition, policies and protocols in place, and communication. Data were collected both electronically, and using hard copy questionnaires. Due to the complexity of the service structure, the organisational questionnaire was split into 10 sections:

1. The emergency department
2. Inpatient care - paediatrics
3. Outpatient care - paediatrics
4. Community paediatric care
5. Inpatient care - young adults
6. Outpatient care - young adults
7. Allied health professionals - paediatric inpatient care
8. Allied health professionals - paediatric clinics
9. Allied health professionals - young adult clinics
10. Allied health professionals - young adult inpatient care

Method detail - patient and parent carer survey

Objective
- To understand the views of the service users, so as not to second guess what their experiences had been.

A short patient questionnaire was circulated electronically via NCEPOD's network of Local Reporters and via patient networks to gather data on young people and carers' views on the services they used. This questionnaire was also made available on the NCEPOD website.

Method detail - clinical peer review using questionnaires and case notes

Objective
- To gain an in-depth view of the care received by patients, to highlight where improvements could be made as well as examples of good care.

On a case by case basis the following areas were assessed:
- Clinical services; including access to professionals with the required expertise, procedures and interventions, and access to equipment
- Symptom management; including pain, posture and movement, associated conditions, communication support and technology dependencies
- Support services; including family support and support at transition to adulthood
- Communication; at diagnosis and in preparation for adulthood
- Training for children and young people with cerebral palsies, families, and professionals (for those providing direct care and those across workforce sectors)
- Safeguarding and social care
- Transition to adult services
- Decision making with children, young people and families; including capacity and best interest decision making.

Table 1.1 ICD10 codes for a cerebral palsy used as inclusion codes

G80.0	Spastic quadriplegic cerebral palsy	G81.9	Hemiplegia, unspecified
G80.1	Spastic diplegic cerebral palsy	G82.3	Flaccid tetraplegia
G80.2	Spastic hemiplegic cerebral palsy	G82.4	Spastic tetraplegia
G80.3	Dyskinetic cerebral palsy	G82.5	Tetraplegia, unspecified
G80.4	Ataxic cerebral palsy	G83.0	Diplegia of upper limbs
G80.8	Other cerebral palsy	G83.1	Monoplegia of lower limb
G80.9	Cerebral palsy, unspecified	G83.2	Monoplegia of upper limb
G81.0	Flaccid hemiplegia	G83.3	Monoplegia, unspecified
G81.1	Spastic hemiplegia		

Study population and case ascertainment

Patients aged 0-25 years with an ICD10 code for a cerebral palsy (Table 1.1), who were admitted to hospital between Monday 7th September and Sunday 18th October 2015 inclusive were included in the study.

Case identification

The NCEPOD Local Reporter, based in each hospital was asked to populate a spreadsheet which detailed all patients who were admitted to the hospital during the study period with one of the included ICD10 codes. The spreadsheet included patient identifiers (hospital and NHS/CHI number, date of birth, gender), date of admission, ICD10 code for that admission, date of discharge, discharge destination and the details of the clinicians who were involved in the care of the patient. Details of any previous admissions in the four weeks prior to the study period were also requested.

Once uploaded to the secure study database, a maximum of ten cases per hospital were sampled for inclusion in the questionnaire and peer review process. Sampling was based on:
- A maximum of two day case patients per hospital
- At least two patients with multiple admissions (prior to and during the study period)
- At least three surgical patients with any length duration of stay
- At least three medical patients who had an admission for ≥48 hours.

Although the sample was identified based on a hospital admission, where possible, details were also collected on the community care the patient had received in the three year period prior to the hospital admission.

Clinical questionnaires and case notes

Three clinical questionnaires were used to collect data for this study:

1. Admitting clinician
This questionnaire collated data on the care provided during the patients identified admission. This questionnaire also captured whether the patient had a 'usual' lead for neurodisability care, or whether 'overall neurodisability care' was provided through the general practitioner.

2. Lead clinician for neurodisability care
Where the details of this clinician could be identified, a questionnaire was sent. This questionnaire collated information on the ongoing care provided to the patient in the community, in the three year period prior to the identified admission.

3. General practitioner (GP)
This questionnaire collated information on the last three years of primary care provided. It was sent to the GP if they were known to be the 'usual lead' for the patient's ongoing neurodisability care, or if the 'usual lead' was not known as it could not be ascertained from either the admission questionnaire or the case notes, in which case the GP was asked to indicate who the relevant clinician was.

Case notes

Extracts of patient case notes were requested for each included case.

Acute care notes
These case note extracts were requested, where applicable, from the time of the patient's arrival in hospital until the time of their discharge, day 30 or death:
- Emergency department records
- Clinical notes, both paper and electronic
- Operation/procedure notes and consent forms
- Nursing notes
- Any separate orthopaedic notes
- Emergency Health Care Plans /Emergency Care Summary
- Passports of care
- Discharge summary
- Community therapy notes
- Outpatient appointment correspondence
- The most recent community discharge summary
- Copies of GP letters
- Clinical notes from any previous admissions (including discharge summaries) (between the 10th August – 18th October 2015)

In addition to the extracts for the admission at the time of inclusion into the study, previous notes for the three years prior to the study admission were requested which included
- Clinic letters
- Discharge summaries for any previous hospital admissions

Community care notes
These were requested for the three years prior to the included admission:
- Community multidisciplinary summaries
- Relevant allied health professional notes
- Clinic letters

Clinical peer review process
A multidisciplinary group of case reviewers was recruited to peer review the case notes and associated clinician questionnaires. The group comprised: paediatric surgery, anaesthetics, orthopaedic surgery, paediatrics, physiotherapy, speech and language therapy, neurology, occupational therapy, intensive care and nursing.

All patient identifiers were removed prior to review. Neither the Clinical Co-ordinators at NCEPOD, nor the case reviewers had access to patient identifiable information.

After being anonymised, each case was reviewed by at least one reviewer within the multidisciplinary group. At regular intervals throughout the meeting the Chair allowed a period of discussion for each reviewer to summarise their cases and ask for opinions from other specialties or raise aspects of the case for further discussion.

To standardise the peer reviews, case reviewers used a semi structured electronic questionnaire and were encouraged to enter free text commentary at multiple points.

The overall quality of care of each case was summarised using the NCEPOD grading system:

> **Good practice:** A standard that you would accept from yourself, your trainees and your institution.
> **Room for improvement:** Aspects of **clinical** care that could have been better.
> **Room for improvement:** Aspects of **organisational** care that could have been better.
> **Room for improvement:** Aspects of both **clinical and organisational** care that could have been better.
> **Less than satisfactory:** Several aspects of clinical and/or organisational care that were well below that you would accept from yourself, your trainees and your institution.
> **Insufficient data:** Insufficient information submitted to NCEPOD to assess the quality of care.

Quality and confidentiality

Each case was given a unique NCEPOD number. Data from all questionnaires received were electronically scanned into a database. Prior to any analysis taking place, the data were cleaned to ensure that there were no duplicate records and that erroneous data had not been entered during scanning. Any fields that contained data that could not be validated were removed.

Data analysis

Following cleaning of the quantitative data, descriptive data summaries were produced. The qualitative data collected from the case reviewers' opinions and free text answers in the clinician questionnaires were coded by themes where possible to allow quantitative analysis. The data were reviewed by NCEPOD Clinical Co-ordinators, a Clinical Researcher and Researcher Assistant to identify the nature and frequency of recurring themes. All data were analysed using Microsoft Access™ and Excel™ by the research staff at NCEPOD.

Case studies have been used throughout this report to illustrate particular themes.

Method detail - review of routine national datasets

Objective
Routinely collected national datasets in this project were used to determine the extent to which they could contribute to an assessment of the health needs and the quality of care that children and young people with a cerebral palsy receive.

A four month project scoping period (July-October 2015) was completed, which included a literature search and consultation with data providers, project advisory group and the study advisory group to identify:
- Potential data sources in England, Wales, Northern Ireland, Scotland, the Channel Islands and Isle of Man. *(Data from the Channel Islands or Isle of Man could not be identified as the data were either 'not collected or would have to be obtained from a wide range of sources, making its reliability questionable')*
- Potential questions that could be addressed from the available datasets
- The approaches to data linkage that had the potential to address these questions
- The facilitators and barriers to data linkage between routinely collected datasets
- The process for gaining permission to access datasets
- Implications from data scoping for the methodological approach
- Revisions and finalisation of project protocol.

A series of descriptive cross sectional analyses of the datasets were designed to address the key questions. All had the potential to be addressed but the results were limited by data availability and factors such as data completeness, availability within the time frame of the project and the cost of the data.

The study population included children and young people aged 0-25 years who had a cerebral palsy, were resident in England, Wales, Scotland, Northern Ireland over an eleven year period (2004-2014) compared to children without a cerebral palsy over the same time period. All analyses were stratified in five year age bands (0-4, 5-9, 10-14, 15-19, 20-24 completed years) and results were compared between children and young people with and without a cerebral palsy and between participating countries, where possible (Figure 1.1).
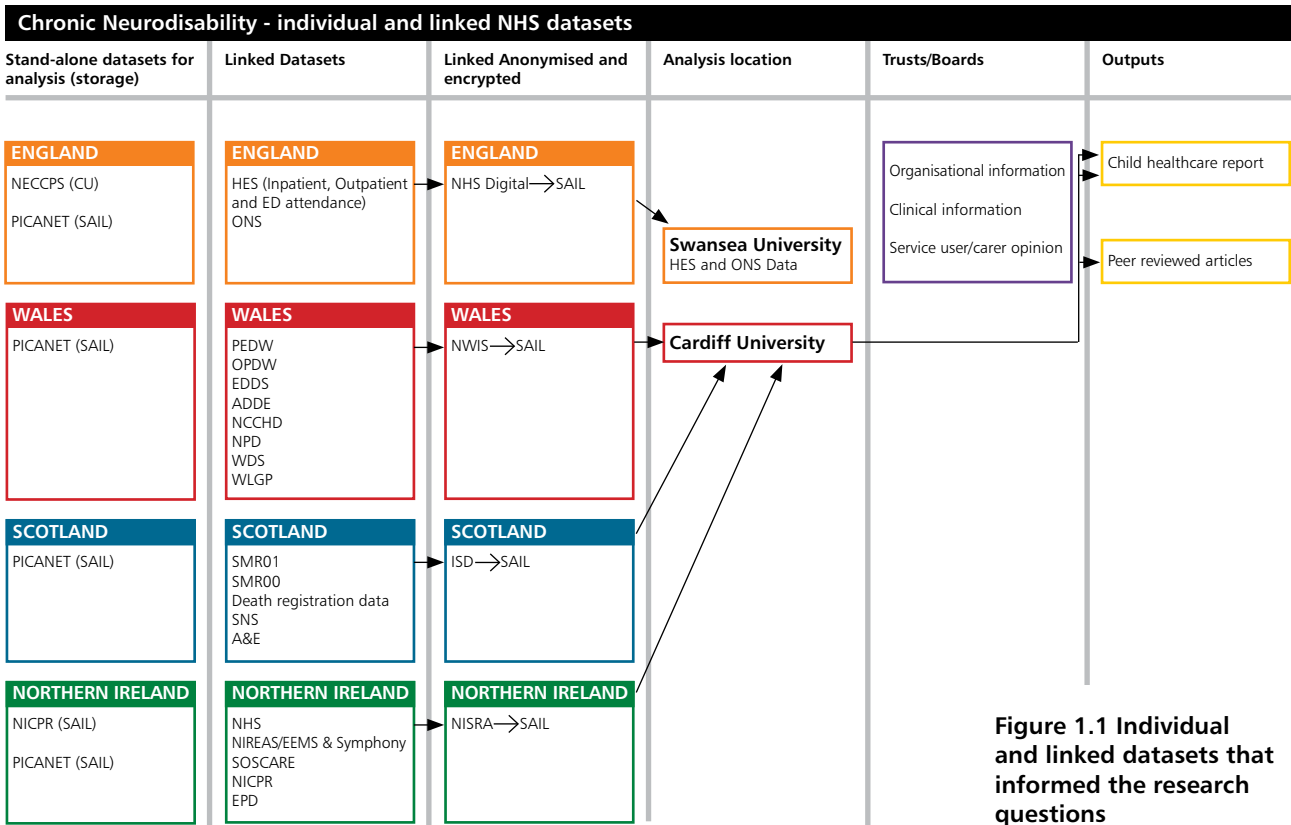
Chronic Neurodisability - individual and linked NHS datasets

Stand-alone datasets for analysis (storage)	Linked Datasets	Linked Anonymised and encrypted	Analysis location	Trusts/Boards	Outputs
ENGLAND NECCPS (CU) PICANET (SAIL)	**ENGLAND** HES (Inpatient, Outpatient and ED attendance) ONS	**ENGLAND** NHS Digital→SAIL	**Swansea University** HES and ONS Data	Organisational information Clinical information Service user/carer opinion	Child healthcare report Peer reviewed articles
WALES PICANET (SAIL)	**WALES** PEDW OPDW EDDS ADDE NCCHD NPD WDS WLGP	**WALES** NWIS→SAIL	**Cardiff University**		
SCOTLAND PICANET (SAIL)	**SCOTLAND** SMR01 SMR00 Death registration data SNS A&E	**SCOTLAND** ISD→SAIL			
NORTHERN IRELAND NICPR (SAIL) PICANET (SAIL)	**NORTHERN IRELAND** NHS NIREAS/EEMS & Symphony SOSCARE NICPR EPD	**NORTHERN IRELAND** NISRA→SAIL			

Figure 1.1 Individual and linked datasets that informed the research questions

Chronic Neurodisability - primary/secondary care interface

Primary care datasets	Linkage between primary and secondary care datasets	Anonymised and encrypted for linkage	Analysis location	Trusts/Boards	Outputs
ENGLAND CPRD	**ENGLAND** CPRD HES (Inpatients, outpatients and ED attendance ONS	**ENGLAND** NHS Digital and CPRD	**Cardiff University**	Organisational information Clinical information Service user/carer opinion	Child health review report Peer reviewed articles
WALES WLGP	**WALES** WLGP PEDW OPDW ADDE	**WALES** NWIS and SAIL			
UK CPRD					

*CPRD data linked to a sample of HES data and ONS data.
Recieved by CPRD as encrypted and anonymous from NHS Digital.
An eight step process is used to match patients in CPRD GOLD and HES using some or all of the following: NHS number, date of birth, sex and postcade.
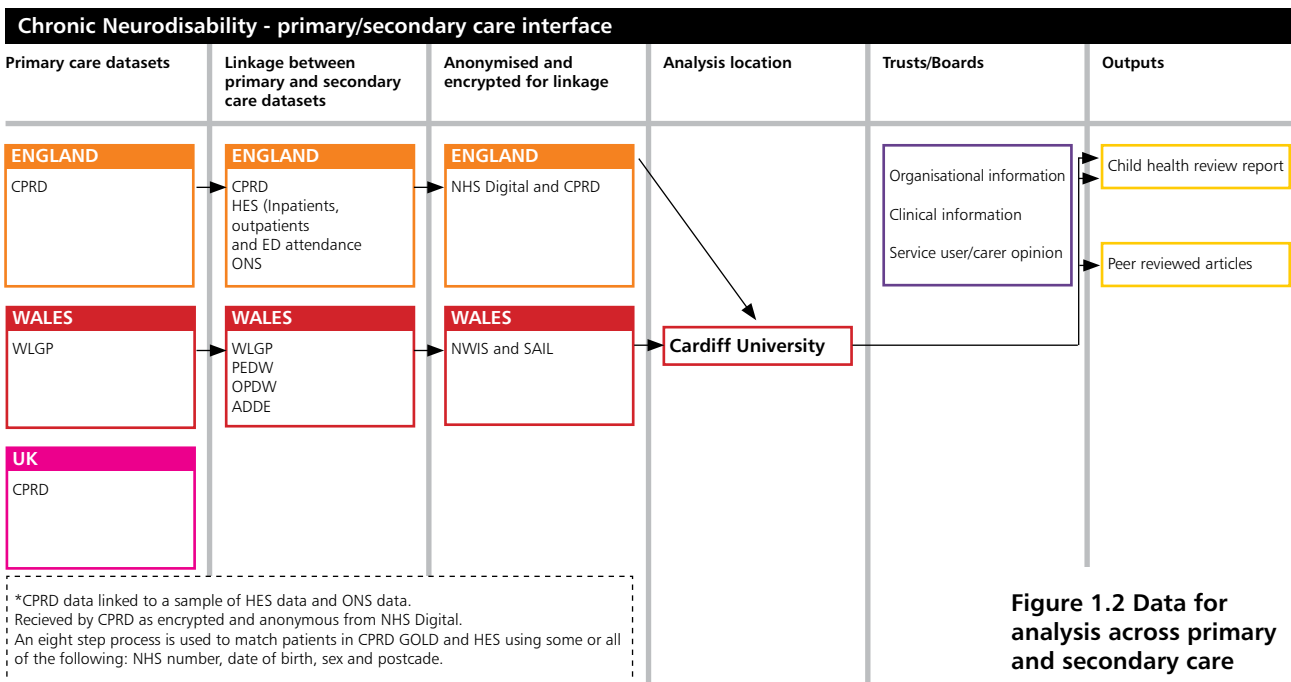
Figure 1.2 Data for analysis across primary and secondary care

Research questions

Hospital admissions and outpatient attendance

The following questions were addressed from secondary care datasets in England, Northern Ireland, Scotland and Wales: (2004-2014) and CPRD HES linked data for England. Analyses were compared between children and young people with a cerebral palsy and undertaken by age group, year of admission or attendance and deprivation of area of residence where available.

- What was the rate of hospital admissions, outpatient attendances (per 100 person years at risk) for children and young people with and without one of the cerebral palsies?
- What proportion of hospital admission episodes/outpatient attendances were attributed to children and young people with one of the cerebral palsies?

The following features were described and compared between children and young people with and without a cerebral palsy:

- Median length of stay by age group
- Median number of outpatient/inpatient attendances per year
- Type of hospital admission (emergency, elective)
- Reason for hospital admission/outpatient attendance by clinical specialty/disease type/procedure undertaken (where possible)

Intensive care admissions

The following questions were addressed from the PICANet dataset, a clinical audit that collects critical care data across all 34 paediatric intensive care units (PICUs) in the UK and Ireland and six specialist transport organisations. PICANet data were analysed for all admissions (2008-2014):

- How many children and young people with a cerebral palsy were admitted to PICU's across the UK?
- What proportion of PICU admissions were for children and young people with a cerebral palsy?
- Age distribution for those admitted to a PICU
- Clinical diagnosis (reason for admission defined post admission)
- Length of stay
- Place of discharge

Primary care attendances

The following questions were addressed in England, Wales, Scotland and Northern Ireland separately from the CPRD dataset and, for Wales, from Wales Primary Care GP dataset (2004-2014). Data were compared between children and young people with and without a cerebral palsy and undertaken by age group, year of attendance and deprivation of area of residence where available.

- What was the rate (per 100 person years at risk) of primary care consultation for children and young people with a cerebral palsy (by age and deprivation of area of residence)
  - Reasons for primary care attendances
  - Referral patterns to secondary care
  - Median length of stay in days

Transition

- What was the pattern of utilisation of adult and paediatric inpatient and outpatient healthcare facilities for children and young people with and without one of the cerebral palsies during transition?
- What were the reasons for outpatient attendance and inpatient admissions by age group during transition?

Cerebral palsy register analyses

It was originally planned that a cohort of children with a cerebral palsy could be identified in each nation and data-linked into routinely collected data. However the North of England Collaborative Cerebral Palsy Survey (NECCPS) dataset was disbanded during the study period and so this was not possible. The data linkage was pursued for the Northern Ireland Cerebral Palsy Register, however issues that arose surrounding the accurate linkage of individuals' data were not resolved within the timescale of the project; access to the individual CP registers was available and included relevant data to address the following key questions for the five age groups and included information on Gross Motor Function Classification System (GMFCS) severity and Index of Multiple Deprivation (IMD) where possible.

- How many children in each age group received an MRI scan at diagnosis?
- What were the associated functional impairments (analysed with respect to GMFCS level where possible)?
  - Vision
  - Seizures
  - GMFCS level
  - Type of cerebral palsy
  - Communication
  - Hearing
  - IQ
  - Feeding

Data sources

The data sources, to address the key questions are described in Table 1.2.

Table 1.2 Routinely collected healthcare data across NHS in England, Wales, Scotland and Northern Ireland sources and other useful data sources.

	England	Scotland	Wales	Northern Ireland	United Kingdom
Inpatients					
Name	*HES APC	SMR01	PEDW	PAS	
Source	NHS Digital	ISD	SAIL	HBS	
Data timescale	2004-2014	2004-2014	1979-2014	2004-2014	
Coverage	Total population	Total population	Total population	Total population	
Outpatients					
Name	*HES Outpatients	SMR00	OPDW	Outpatients Dataset	
Source	NHS Digital	ISD	SAIL	HBS	
Data timescale	2004-2014	2004-2014	1979-2014	2004-2014	
Coverage	Total population	Total population	Total population	Total population	
Primary care					
Name			WLGP	EPD	*CPRD
Source			SAIL	BSO	CPRD
Data timescale			1979-2014	2010-2014	1979-2014
Coverage			348 (73%) GP practices	Primary care prescriptions sent to BSO for total populations	> 11.3 million patients from 674 practices in the UK-approximately 6.9% of the UK population

Table 1.2 Routinely collected healthcare data across NHS in England, Wales, Scotland and Northern Ireland sources and other useful data sources. (continued)

	England	Scotland	Wales	Northern Ireland	United Kingdom
Emergency department					
Name	HES Accident and Emergency	A&E Datamart	EDDS	Symphony-Belfast, Northern & Western Trusts	
Source	NHS Digital	ISD	SAIL	HBS	
Data timescale	2007-2014	2007-2014	2009-2014	2011-2014	
Coverage	Total population	Total population	Total population from 2012-Prior to 2012, only major (24 hour, emergency led) A&Es submitted data	Symphony covers Belfast, Northern & Western Trusts EEMS covers Eastern & Southern Trusts	
Intensive care					
Name					PICANet
Source					PICANet
Data timescale					2008-2014
Coverage					Total UK population
Mortality					
Name	*ONS Mortality	Death Registration Data	ADDE	Death Registration Data	
Source	ONS	ISD	SAIL	Northern Ireland Statistics and Research Agency	
Data timescale	2004-2014	2004-2014	2004-2014	2004-2014	
Coverage	Population linked to HES	Population registered with a GP	Total population	Population in the GP Patients Registration Index	
Cerebral Palsy or Special Needs Registers					
Name	NECCPS	SNS		NICPR	
Source	Regional Maternity Survey Office	ISD		Queens University, Belfast	
Time Scale	Those born 2004-2014	2004-2014		Those born 1981-2011	
Coverage	North East and North Cumbria children and young people with cerebral palsy	Implemented at different times and with different completion rates in 12 NHS Boards		Northern Ireland population of children and young people with cerebral palsy	

CPRD provided data linkage between primary and secondary healthcare for an estimated 5.34% of the population of England.

Key to acronyms

A&E	Accident and Emergency
ADDE	Annual District Death Extract
BSO	Business Services Organisation
CPRD	Clinical Practice Research Database
EDDS	Emergency Department Dataset
EPD	Enhanced Prescribing Dataset
HES APC	Hospital Episode Statistics Admitted Patient Care
HBS	Honest Broker Service
ISD	Information Services Scotland
NECCPS	North of England Collaborative Cerebral Palsy Survey

NICPR	Northern Ireland Cerebral Palsy Register
OPDW	Outpatients Dataset Wales
PEDW	Patient Episode Database for Wales
SAIL	Secure Anonymised Information Linkage
SMR00	Scottish Morbidity Records - Outpatients Attendances and Appointments
SMR01	Scottish Morbidity Records - General Acute Inpatient and Day Case
SNS	Support Needs System
WLGP	Wales Primary Care GP Dataset

Data acquisition

Detailed application forms were completed and submitted to each data host stating the purpose for which the data would be used, the variables required, the datasets to be linked, and explaining how the data would be stored securely.

The duration between sending the application and receiving the data varied widely across data providers due to different procedures for assessing applications. There was a continuous need to update and address information governance throughout the project for the timeline appertaining to the application submission, approval dates, dates when data were received and costs (Appendix 2). The duration from first contact to receipt of data was longest for NHS Digital data for England. Special negotiations with the Northern Ireland Cerebral Palsy Register were approved and data were received June 2017.

Data linkage

The process for linking data is summarised in Figure 1.3. Once applied for and permissions to access data were granted, datasets were linked remotely (NHS Digital in England, SAIL Wales, ISD Scotland, HBS Northern Ireland) and provided to the Secure Anonymised Information Linkage (SAIL) Databank for data cleaning. The typical process for data linkage relied upon National Health Service number
for England, Wales and Northern Ireland and the Community Health Index: (CHI) in Scotland. A matching algorithm of combinations of potential patient identifiable fields accounted for individuals with missing NHS numbers (estimated at 17% of the population).[13] A description of data linkage process within SAIL for the Wales datasets can be found in Appendix 3.
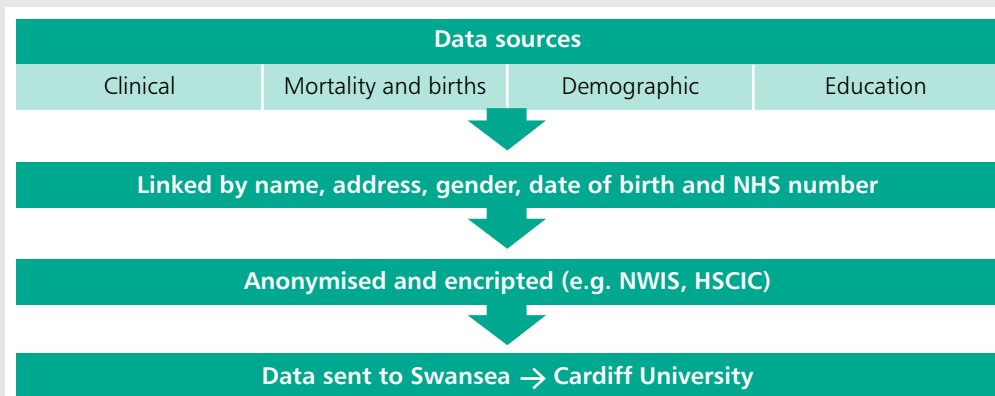

Figure 1.3 The process of data linkage

CPRD provided GP data that was linked to HES and ONS data for 77% of subscribing GP practices in England (an estimated 5.34% of the population of England). CPRD received HES data as encrypted and anonymised from NHS Digital. CPRD use an eight step process to match individual patients in CPRD GOLD and HES using some or all of the following; NHS number, date of birth, sex and postcode. Each individual was included in the study for a period dependent on the patient's dates of birth, death (if relevant) and registration with a GP, and the dates of the last collection of data from the GP where the data met CPRD's quality standard.

Data cleaning and preparation

Time-scales to prepare these large datasets for analysis varied from 6-10 months per dataset. Several analysts were employed in Swansea to undertake this process (for all datasets other than those from CPRD, PICANet and the NECCPS). Such data cleaning and preparation involved:

- De-duping based on encrypted codes, dates of health episodes, multiple admissions on the same date for the same individual, diagnostic codes, age, data that fell outside age range or time period of interest, incorrectly linked cases etc.
- Designing and creating a cohort of children and young people with a cerebral palsy in Wales taken from multiple datasets and ensuring consistent treatment of variables e.g. prioritisation of gender/week of birth/date of death from across various datasets in which they are found.
- Creating a list of clinical code groups of interest – diagnostic (ICD-10 and READ v2), operational (OPCS 4), product (READ v3), prescribing (BNF Chapter codes) and treatment specialty (specialty codes within HES, PEDW, OPDW, PAS, SMR00/01)
- Familiarisation and data quality assessment on datasets received
- Agreeing the handling of data anomalies/data quality issues identified
- Identification and flagging of children with one of the cerebral palsies

- Flagging morbidity codes (Codes are available on request)
- Defining and creating four nations person spells (hospital admissions) to enable comparison of hospital admissions across countries
- Calculation of denominators.

Identification of children with one of the cerebral palsies

Children with one of the cerebral palsies were identified from routine datasets using a disease diagnostic coding algorithm adapted from Meeraus et al.[14]

ICD-10 codes G80-83 (in any coding position at least once) were used to identify children and young people with a cerebral palsy within secondary care data sets.

Read Codes v2 and v3 were used to identify children with a cerebral palsy in Primary Care datasets and PICANet. Where relevant, in order to explore the interface between datasets that use Read codes and those that use ICD10), Read codes were mapped to ICD-10 codes (Available on request).

The cerebral palsies are chronic conditions, however they are not coded consistently at every contact point with NHS services. The CPRD and Welsh data were searched for data between 1st January 1979 to 31st December 2014 to identify patients in the older age groups at the start of the study period who may not have had a cerebral palsy code recorded for a number of years.

Within English (HES), Scottish, and Northern Irish data, only the date range 1st January 2004 to 31st December 2014 were searched as data were not provided for earlier years and relevant cases were only identified from hospital related data as no primary care datasets were available. Disease codes were poorly recorded in outpatient and emergency department datasets therefore case ascertainment was primarily from inpatient datasets which is likely to be biased towards the more severely affected children and young people with one of the cerebral palsies. Case ascertainment therefore varies across datasets.

A suite of disease related codes (ICD-10 and Read codes) were used to identify morbidity:

- the common causes or reasons behind hospital admissions e.g. respiratory disorders, epilepsy and neurological, cardiovascular, endocrine and metabolic, gastro intestinal conditions, infections and injuries. (Read codes mapped onto ICD-10 Chapter codes)
- procedures undertaken (e.g. gastrostomy, botulinum toxin, tendon release) adapted from Meeraus et al[14]
- medications prescribed (e.g. anticonvulsants, laxatives, neuromuscular relaxants) adapted from Meeraus et al.[14]

Validation of codes

It is not possible to validate the case ascertainment from individual large datasets. However the case ascertainment for the case note review provided some insight about the accuracy of coding for a cerebral palsy based upon confirmation of diagnosis from cases identified for case note review.

Definition of hospital admissions (hospital spells)

CPRD generate hospital spell numbers from HES Admitted Patient Care (APC) data to identify a continuous inpatient stay in a single hospital. A transfer from one hospital to another will lead to the creation of a new spell number. Thus, CPRD spells will reflect the number of hospital admissions correctly but counting the spells will overstate the number of 'person spells', i.e. continuous inpatient spells of care within the NHS, regardless of any inter-hospital transfers which may take place. On the other hand, the calculation of the length of a person's stay in a hospital will, for those patients transferred from one hospital to another, underestimate their total length of stay under hospital care.

Analysis of the four nations' inpatients (non-CPRD) data has used a different derivation of hospital spells developed at Swansea University and named the four nation person spell (4N person spell), aiming to approximate 'person spells'. (Available on request)

Throughout the report the term 'hospital admission' has been used to equate to hospital spells as defined above and identified the data source. Care needs to be taken,

therefore, when comparing statistics based on CPRD hospital spells or admissions with statistics based on the person hospital spells of admissions defined for the four nations' inpatients data.

Calculation of denominators

For CPRD, the basis for the calculation of person years at risk was CPRD's anonymised list of patients who had data of an acceptable standard for research purposes who were aged 0 up to 25 years at any point during the study period of 1 January 2004 to 31 December 2014. An individual's total time at risk within the study was then broken down by year and age band.

Denominators used for linked English data included only those individuals (within CPRD) marked as eligible for linkage.

For the 'All Wales' datasets a file of [anonymised] patient identifiers comprised the cohort of patients aged 0 up to 25 resident in Wales at any point during the study period of 1st January 2004 to 31st December 2014. Not all GP practices in Wales contributed data to SAIL but SAIL's coverage of NHS secondary care outpatient and inpatient activity is complete. Calculation of person years at risk was broadly similar to the approach taken with CPRD. For GP denominators, patients in the overall cohort were only included for those time periods when they were registered with a GP practice contributing to SAIL.

Data analysis

Data are presented for key questions in simple graphical form for trends across age groups, gender, time and IMD (where possible). Population rates according to person years at risk were calculated for key outcomes and compared by age group, year of event and IMD (utilising CPRD and Wales SAIL data). Reasons for attendance are described by proportion of attendances by diagnosis or treatment specialty where relevant confidence intervals were calculated to enable statistical comparisons. When interpreting the results, consideration must be given to the possible effects of the size and nature of the datasets, the variation in definitions, case ascertainment rates and methods and variation of case mix within and between datasets.

Information governance

All data received and handled by NCEPOD and Cardiff University complied with all relevant national requirements, including the Information Commissioners Office (NCEPOD Z5442652), the NHS Act 2006 (15/CAG/0210), the NHS Code of Practice and Public Benefit and Privacy Panel for Health and Social Care (for NHS Scotland). As anonymous data were requested ethical approvals were not required, but approvals from the data providers for each country was. 'Approved researcher status' for each member of the data linkage team was sought and granted in order to access data from the Office for National Statistics (ONS). Each member of the team completed Medical Research Council (MRC) Research Data and Confidentiality e-module training.

The findings of the report were reviewed by the Study Advisory Group, Reviewers, NCEPOD Steering Group including Clinical Co-ordinators, Trustees and Lay Representatives on four occasions prior to publication.

2 – Study limitations

Case note review and questionnaires

Part of the reason for doing this study was the concern that pathways of care for this group of patients were not clear and somewhat fragmented. This appeared to be confirmed quite early on as it was harder to identify leads to ask questions of and case notes did not tell the whole story as they were not linked across healthcare providers and it was challenging to glean the extra sections needed.

Ideally this study would have been conducted by identifying patients in the community and following their various pathways including access to healthcare. However, it was not possible to identify patients this way due to the complexity of identifying community links or contacting general practitioners. A pragmatic approach was therefore taken to identify patients though hospital coding and trace their pathways out into the community. Although this was a compromise as a study method, it should be borne in mind that this is what should be achievable, as a patient attending a hospital will not be carrying their notes with them.

There were some specific issues encountered:
- Not all NHS healthcare providers participated in this study – although it was ensured that all countries were represented and provided a representative sample
- Case notes received were not all complete (e.g. acute care notes were not always supplemented by the community care notes and vice versa)
- Although NCEPOD did request electronic medical records as well as those on paper, it was not always easy for the reviewers to work out what information would have been accessible to the clinician at the point of presentation of the patient to the hospital.
- Response rates from General Practitioners were lower than we would have hoped for as were response rates from the parent carer and patient surveys, but data from other sources was used to enhance what was available
- Responses to the community care part of the case reviewer assessment form were sometimes based on limited information from the case notes, as not available or not documented
- Organisational leads for the different areas of care were difficult to identify.

Routine national data

- The processes around obtaining data for the data linkage elements of the study, data cleaning for analysis proved to be complex and time consuming
- The various organisations that hold the data required different application processes and different governance requirements. Further applications for updated data were required and data application systems changed within the time frame of applying for datasets
- After the considerable time that was required to clean and prepare data for analysis, there were strict criteria to destroy datasets. The time frame available for detailed analysis was limited by the conditions of the data sharing agreements
- UK countries differed in the extent and type of data availability, whilst standard ICD-10, READ codes v2 and v3 are used, the variables that were collected differed between countries and different definitions and coding systems were used (e.g. for admission, discharge, transfer, A&E). The data quality and types and definitions of data fields included also differed. All contributed to making comparative analyses difficult
- Some of the data obtained lacked the level of detail necessary to get a full understanding of the range of needs and service utilisation of children and young people with cerebral palsies
- The extent to which data sources could be linked and the nature of the questions that could be addressed from each set of linked data varied and limited the ability to make comparisons across the UK. However different data linkage in different regions had the potential to reflect different components of healthcare

- The consistency, timeliness and accuracy of coding varied and affected the quality of data analysis. Completion of data fields (missing data) affected the potential for detailed analysis
- Children with cerebral palsies are largely managed within the community and outpatient settings. Routine data collection in these settings was poor and the amount of NHS involvement is likely to be under estimated
- Hospital case records are coded and data entered into routine healthcare datasets by operators who are not clinically trained. Coding will therefore be affected by the quality of data recorded within healthcare records, and the vigilance and interpretation of the data by the coder
- Cerebral palsy is associated with varying levels of severity both in terms of motor and cognitive impairment. These data are not currently collected routinely and confound detailed analysis of service utilisation and quality of care according to clinical need.

3 – Data returns and study populations

Organisational survey

Where a service was provided, the Local Reporter at the hospital was asked to provide the name of the service lead, and contact details so that an organisational questionnaire could be sent for completion. Table 3.1 shows the number of questionnaires included in the analysis.

Clinical review using questionnaires and case notes

For the study period 3,483 patients were identified as meeting the study inclusion criteria. Figure 3.1 details the return of the cases included.

Of particular note were the 148 patients who were subsequently excluded. In most instances this was because despite having had one of the included ICD10 codes applied, clinical review of the available information revealed that a cerebral palsy was not the correct diagnosis. Of the 634 sets of admission case notes, some included community notes and 242 sets of separate community case notes were

Table 3.1 Number of questionnaires included in the analysis

	n=
Emergency department care	92
Paediatric inpatient care	90
Paediatric outpatient care	84
Paediatric community care	81
Adult inpatient care	66
Adult outpatient care	53
Allied health professionals paediatric inpatient	63
Allied health professionals paediatric outpatient care	67
Allied health professionals adult outpatient care	41
Allied health professionals adult inpatient care	52

returned giving 350 sets of community notes; although not all were of good enough quality to assess. For 199 patients a complete set of case notes and questionnaires were received.
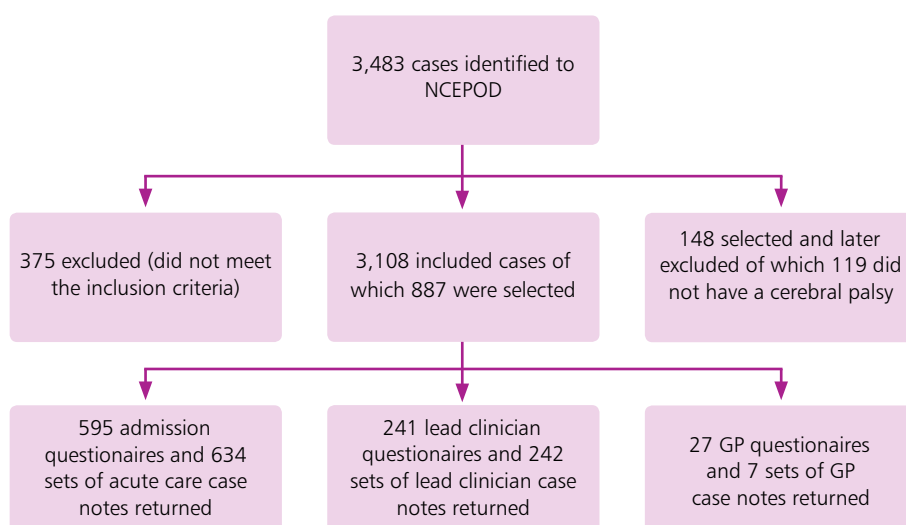

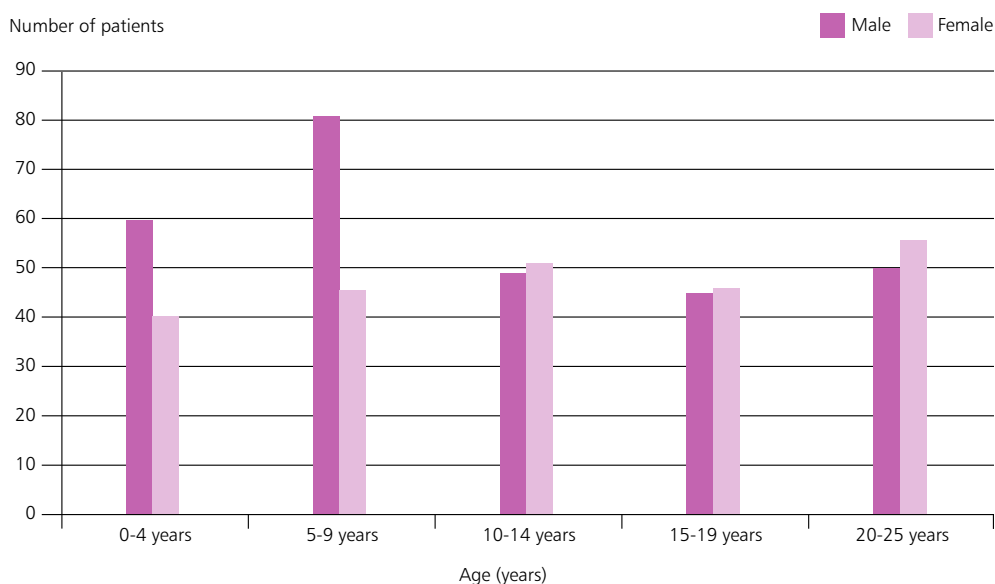**Figure 3.1 Data returns**

Number of patients

Figure 3.2 Age and gender of the study population

Please note that the denominators throughout the report will reflect the number of different data sources that have have been used, such as the various questionnaires, or case notes. The text around the data will provide context to numbers that have been used.

Study population

From the questionnaire, 290/531 (54.6%) patients were male; the age range was five months to 25 years, with a mean age of 11.8 years (Figure 3.2).

One third of the included sample had been admitted to district general hospitals with fewer than 500 beds, a third to larger district general hospitals (>500 beds) and a third to university teaching hospitals and specialist tertiary centres (Table 3.2).

Two thirds of the patients in the study sample were admitted as an emergency (including urgent) admission 337/509 (66.2%). One third (172/509; 33.8%) were elective (including planned) admissions. These admissions were generally for surgical procedures or a short procedure to

Table 3.2 Type of hospital the patient was admitted to

	n=	%
District general hospitals >500 beds	165	31.5
District general hospitals ≤500 beds	153	29.3
University teaching hospital	136	26.0
Specialist tertiary paediatric centre	54	10.3
Other specialty hospital	15	2.9
Subtotal	**523**	
Not answered	13	
Total	**536**	

be undertaken (Appendix 1).[15] The sample for this study deliberately included a proportion of children and young people undergoing a planned procedure or surgery, so the pattern of the admissions in this study was expected. The majority of patients arrived at hospital during standard working hours (08.00-17.59) with just over a third arriving 'out of hours' (18.00-07.59). Admissions occurred on all days of the week with a slight reduction at weekends, likely related to a lower number of patients undergoing elective/ planned surgery and procedures. 11

On arrival at hospital and considering the pathway of admission, the time to initial hospital assessment was reported by clinical case reviewers as delayed in 20/317 (6.3%) patients and in 17 patients a delay in management of their health condition (Table 3.3).

Table 3.3 Delay in initial assessment on arrival in hospital

	n=	%
Yes	20	6.3
No	297	93.7
Subtotal	**317**	
Unable to answer	35	
Total	**352**	

The majority of patients had a comprehensive set of basic physiological variables recorded with the exception of blood pressure recorded in only 77.3% of patients (367/475), and an early warning score (EWS) in only 76.8% (341/444). These data were for all admissions (elective and emergency).

For emergency admissions, delays in initiating specific treatment were also felt to be seen in very few patients and clinicians stated that this occurred very infrequently in only 8/311 (2.6%) patients.

Analysis of routine national datasets

Case ascertainment

Table 3.4 shows the number (proportion) of children and young people aged 0-25 years identified as having one of the cerebral palsies from routinely collected healthcare data within each country. (Please note that case ascertainment sources differed across all countries).

Table 3.4 Case ascertainment

Data Population And datasets used for ascertainment	Patients with a cerebral palsy Number (%)	Patients without a cerebral palsy Number (%)	Total population
CPRD			
England	6,170 (0.2)	2,726,461 (99.8)	2,732,631
England :HES Linked (HES APC, HES OPD, ONS Mortality and CPRD)	7,472 (0.4)	2,115,442 (99.6)	2,122,914
Wales	632 (0.2)	268,198 (99.8)	268,830
Northern Ireland	188 (0.2)	92,995 (99.8)	93,183
Scotland	794 (0.2)	325,612 (99.8)	326,406
Data linked in each of the four countries			
England NHS Digital (HES APC, Outpatients and ONS mortality	53,409 (0.5)	10,067,341 (99.5)	10,120,750
Wales (PEDW,OPDW, WLGP, ADDE)	5,397 (0.3)	1,630,855 (99.7)	1,636,252
Northern Ireland (PAS Inpatients and Death Registration Data)	1,744 (0.3)	510,607 (99.7)	512,348
Scotland (SMR01 and Death Registration Data)	4,183 (0.6)	690,231 (99.4)	694,414

Prevalence of children and young people with a cerebral palsy who access the NHS

Prevalence figures were derived from two regional datasets that linked routinely collected data from primary and secondary care

- CPRD (HES linked England) representing 5.34% of GP practices in England
- WLGP linked to PEDW representing 70% of GP practices in Wales

Cerebral palsy is a chronic condition, yet it is not coded consistently at every contact point with NHS services. CPRD and the Welsh dataset were searched from 1st January 1979 to 31st December 2014 to ensure that the cases in the older age groups were identified within the study period. Figure 3.3 illustrates the source of case ascertainment within HES linked English CPRD dataset.

Figure 3.3 Venn diagram illustrating the number of patients within each of the datasets where patients with a neurodisabling condition were identified within the HES Linked English CPRD dataset

There were 7,472 patients with a neurodisabling condition identified from a total of 2,122,914 cases within the HES Linked English CPRD dataset. Of these a cerebral palsy was recorded at least once in 2,736 (36.6%) of cases in HES inpatient data only, 1,541 (20.6%) in CPRD GP data only and 3,136 (42%) were identified from both sources. A small proportion 53 (0.7%) were identified from only the outpatient datasets where the completion of diagnostic coding was poor (Figure 3.3).

The prevalence of the cerebral palsies for children and young people 0-25 years of age (2004-2014) is shown in Figure 3.4.
- 3.5 (95% CI 3.4-3.6) per 1000 for England and 2.8 (95% CI 2.7-2.9) per 1000 for Wales
- There were significantly more males 55.4% (95% CI 54.3 - 56.5) with one of the cerebral palsies in comparison to 49% (95%CI: 48.9 - 49.0) of males within the population of children and young people without one of the cerebral palsies (England HES linked dataset).

Prevalence figures for children and young people aged 10-24 years recorded to have one of the cerebral palsies remained relatively constant across the 11 years of the study, the prevalence figures for 0-9 year olds decreased over time. This is particularly true for the 0-4 year olds and is likely to be due to the fact that 40% of cases do not have a cerebral palsy code recorded in NHS records until after their 5th birthday. The recognition of a cerebral palsy within the youngest age group may not have been confirmed or recorded in case notes. Clinical coding is undertaken by a third party of individuals who are not clinically trained and may not recognise or detect the diagnosis within clinical records. Furthermore case ascertainment was retrospective from 1979, extending back to the date of birth for those aged 20-24 years in 2004 to optimise case recognition. **5**

The prevalence of cerebral palsies increased significantly in England across the index of multiple deprivation (IMD) quintiles from 3.1 (95% CI: 3.0-3.3) per 1000 in the least deprived to 4.0 (95% CI 3.8-4.2) per 1000 in the most deprived quintile (Figure 3.5).

The overall mortality rate in England was 26 times higher for children and young people with one of the cerebral palsies than for those without (5.3 vs 0.2 per 1000 at risk) for 0-25 year olds. The mortality rate was greatest in those younger than five years of age (Figure 3.6).
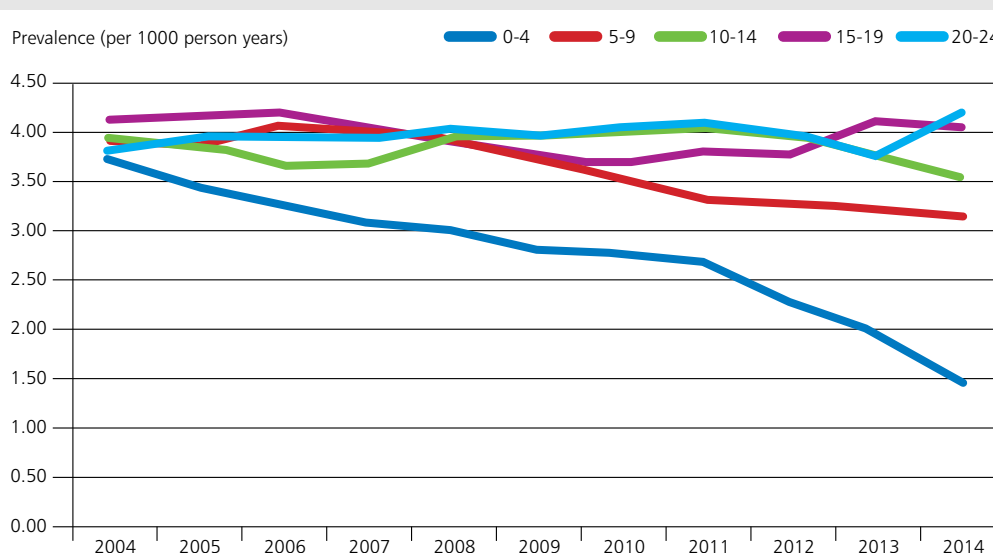

Figure 3.4 Prevalence of cerebral palsies by year and age group in CPRD (England HES Linked)
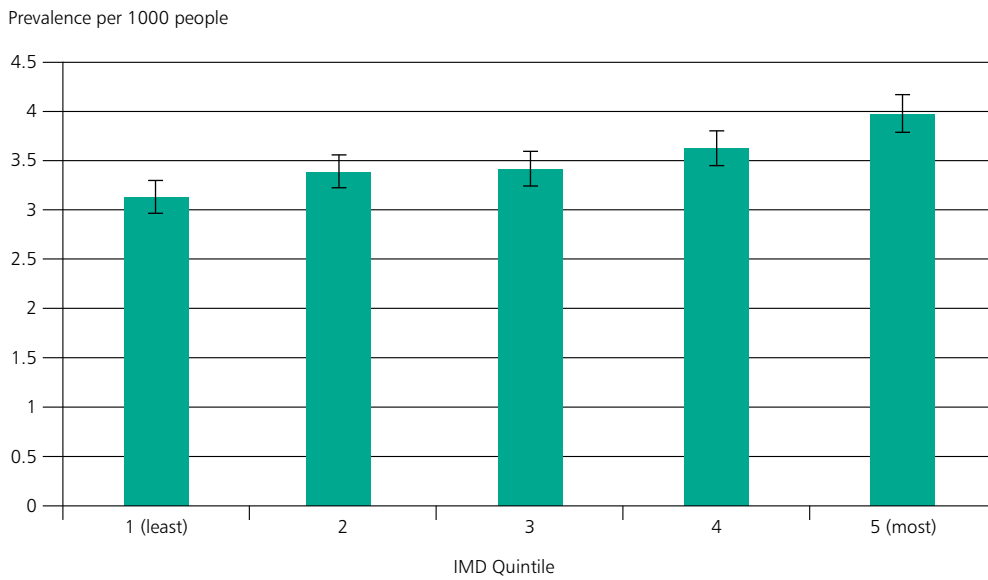
Prevalence per 1000 people

Figure 3.5 The prevalence of cerebral palsies in children and young people within each Index of Multiple Deprivation (IMD) quintile (CPRD: England HES Linked)

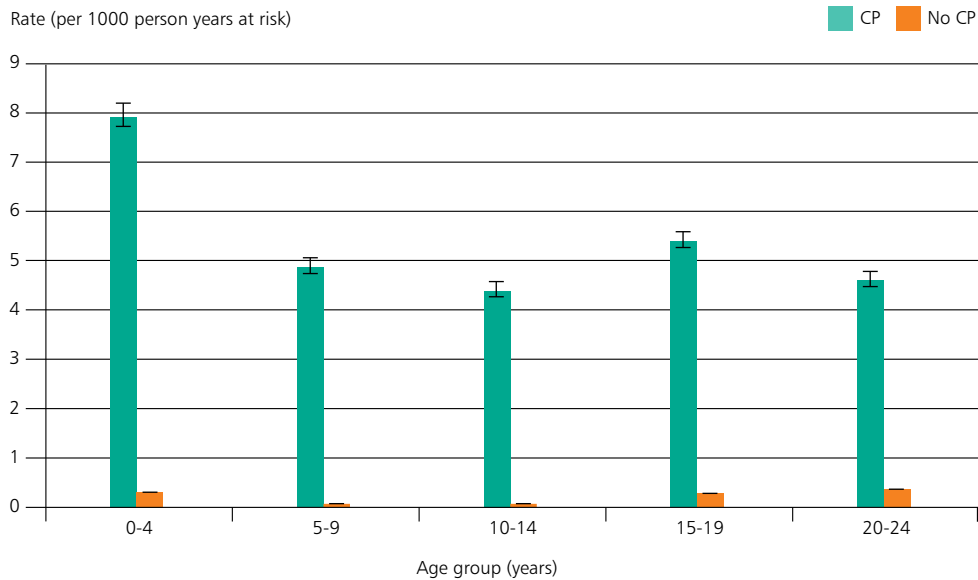Rate (per 1000 person years at risk)

CP   No CP

Figure 3.6 Mortality rate (per 1000 person years at risk) among children and young people with and without a cerebral palsy between 2004 and 2014 by age group (CPRD: England HES and OPD Linked)

Within the Wales WLGP/PEDW linked data, the mortality rate for those with one of the cerebral palsies was five per 1000 person years at risk across all age groups and 0.3 per 1000 person years at risk for those without a cerebral palsy.

The profile of recorded primary causes of death were very different between the two populations studied. By far the most commonly recorded primary causes of death for children and young people with a cerebral palsy were respiratory causes in 51% of cases (Figure 3.7). Similar results were seen across the four countries.
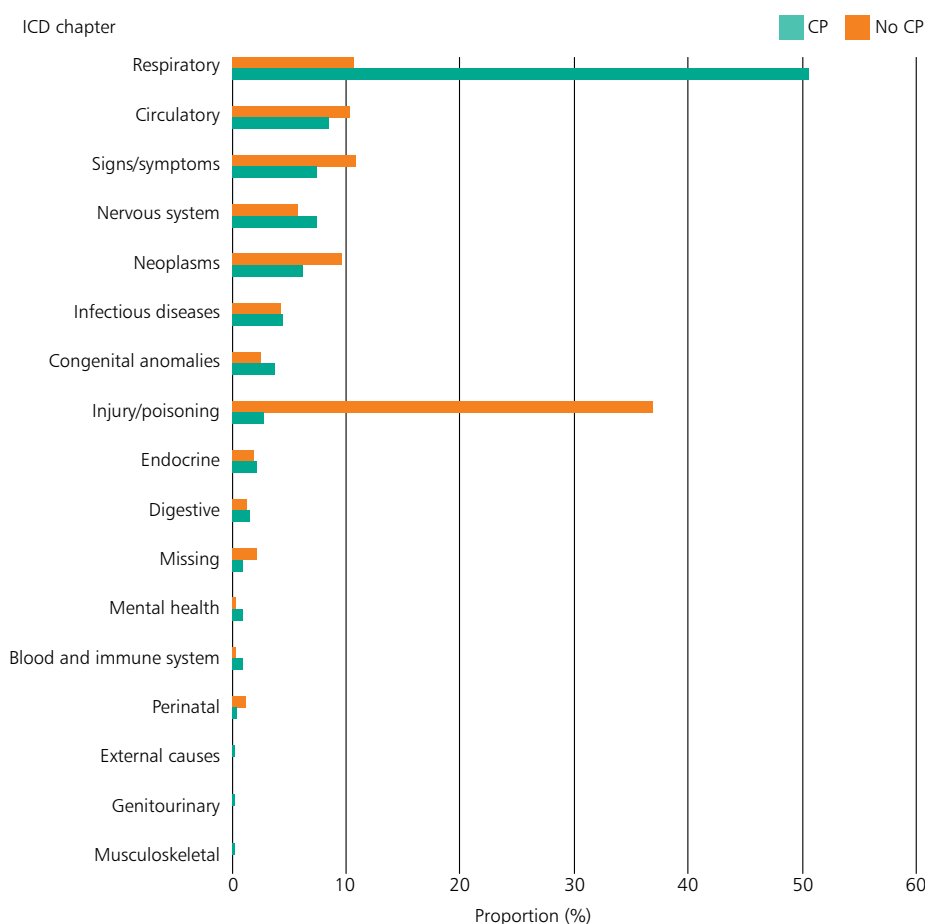
Figure 3.7 Primary cause of death for children and young people with (n=174) and without a cerebral palsy (n=2,026) aged 0-24 years between 2004 and 2014 as a proportion of total deaths(CPRD: England HES Linked)

It was not possible to determine the mortality rate according to population at risk of a cerebral palsy for Scotland or Northern Ireland. However, between 2005-2014 in Northern Ireland, 91/1,850 deaths were for children and young people with a cerebral palsy, accounting for 4.91% of all deaths in the dataset. For Scotland 2004-2014 there were 9.2% (335) of a total of 3,635 deaths for children and young people with a cerebral palsy.

Key Findings – routine national data

- The prevalence of the cerebral palsies identified within two datasets that represent cross sections of the population (0-25 years) in England and Wales give figures of 3.5 and 2.8 per 1000 respectively. There were a greater number of males identified and an increase in the prevalence with respect to increased social deprivation. Whilst there was a significant difference between the prevalence figures between the two countries, they are consistent with the estimated population prevalence of 2-3/1000.[16] This suggests that the case ascertainment for this study was reasonably comprehensive

- The inconsistent and variable codes used, and the failure to record cerebral palsies at every presentation to the NHS and the delay in recording cerebral palsies within NHS datasets may have lead us to under-estimate the number of younger children with the condition in the study sample. For similar reasons some conditions that are not one of the cerebral palsies but individuals with similar motor impairment may have been included **4**

- Respiratory conditions prevailed as the most common diagnostic group in mortality, PICU, emergency hospital admissions and primary healthcare consultations.

SEE RECOMMENDATIONS
1·2