Access to Hospital Outpatient Services for Children and Young People with Cerebral Palsy

A thesis submitted to fulfil requirements for the degree of Doctor of Philosophy

By

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I am grateful to have had this opportunity to develop knowledge and skills that have made me a better researcher and research end user. Many people have contributed to the work that this thesis contains, and I am grateful to you all.

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A special thanks too to my amazing coauthors and collaborators on the studies in this thesis. I've sincerely appreciated the depth and breadth of knowledge that you've shared with me during this time, your kindness and encouragement.

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I have no confidence that words I write here will do justice to Bec Nogajski's role in my life. I am incredibly grateful for her love and partnership, her patience and understanding and the many sacrifices that she, and our children, Ted, Tilly and Will, made to support me during this thesis. All my love.

Statement of Originality

I certify that the intellectual content of this thesis is the product of my own work and that all the assistance received in preparing this thesis and sources have been acknowledged.

Dr Simon Paget		Date:

Authorship Attribution Statement

This thesis contains material previously published as peer-reviewed journal articles. The relevant articles and included in **Chapter 10 Appendix B**, along with statements of the relative contributions of each co-author.

This thesis is primarily the work of Dr Simon Paget. Dr Paget is the sole author of the introduction and literature review (**Chapter 1**), methods introduction (**Chapter 3**) and summary and conclusions (**Chapter 8**). He was the lead author for the five studies presented in **Chapters 2,4-7**. He held primary responsibility for the conceptualisation and design of the studies, data cleaning, statistical analysis and interpreting results. He was primarily responsible for writing and revising the studies manuscripts, and for creating tables and figures for the manuscripts and in this thesis.

Adjunct Prof Sarah McIntyre and Prof Natasha Nassar provided substantial support for the work this thesis comprises, including with conceptualisation and design of the included studies, interpretation of results and critical review of manuscripts and chapters.

Dr Simon Paget		Date
Dr Sarah McIntyre		Date
Prof Natasha Nassar		Date

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Thesis Abstract

BACKGROUND: People with cerebral palsy (CP) experience increased risk of adverse health outcomes related to their disability and associated comorbidities. As a result, they need to access hospital health services more frequently than the general population. The aim of this thesis is to examine access, and equity of access, to hospital-based outpatient services that are intended to support children and young people with CP.

METHODS: Existing research about access to and utilisation of health services by children with CP was synthesised in a systematic review. A series of population-based studies using the New South Wales (NSW) / Australian Capital Territory (ACT) CP Register linked to administrative health data were conducted. Ascertainment of children with CP using administrative health data was compared to the NSW/ACT CP Register. Frequency of attendance, factors associated with non-attendance and use of telemedicine in outpatient services before, during and after the COVID-19 pandemic were examined at two specialist children's hospitals. For the whole NSW population, access to outpatient health services, continuity of care and unplanned health care use in NSW were investigated.

RESULTS: The systematic review identified associations between CP severity and comorbidities and increased hospital health service use. Hospital admission data identified many children with CP (~70%), but they had more comorbidities and higher risk of death compared with the NSW/ACT CP Register. In NSW, non-attendance at specialist outpatient clinics was associated with (older) age, socioeconomic disadvantage and previous non-attendance. Telemedicine use in outpatient clinics increased during the COVID-19 pandemic (peak 61.4%), but then declined. Children living in regional and remote areas had lower rates of telemedicine than children in major cities. Access to outpatient services for Aboriginal and/or Torres Strait Islander children with CP was broadly similar to the whole population. Better continuity of outpatient care was associated with less urgent hospital care for children with CP.

CONCLUSION: Improving equity of access to outpatient services is important to improve health outcomes for children and young people with CP. Multiple interventions are likely to be required to support this improvement, and research to examine these interventions should be a priority.

Thesis Roadmap

Thesis Aims

Cerebral palsy (CP) is the most common cause of physical disability in childhood, and a common cause of disability in adults. For many people CP is also associated with chronic health conditions, and an increased use of health services. The primary aim of this thesis is to examine access, and equity of access, to hospital-based outpatient services that are intended to support children and young people with CP.

Structure of the thesis

This thesis has a total of eight chapters, explained below and represented in **Figure i**. The thesis begins with an introduction to the topic and supportive literature review (**Chapter 1**) and a systematic review of factors associated with health service utilisation in cerebral palsy (**Chapter 2**). **Chapter 3** provides an overview of study methods which involve the use of population data and series of data linkage studies, involving the use of the NSW/ACT CP Register linked with health administrative data to support four distinct studies. These include an analysis of differences in study populations derived from CP registers and health administrative data (**Chapter 4**), three studies exploring different

dimensions of outpatient health service access (**Chapters 5**, **6** and **7**). **Chapter 8** presents a summary and discussion of the results of the thesis, implications of findings and recommendations for future research. The final chapters (**Chapter 9** and **10**) contain the appendices which include supplemental data from published studies, published versions of the manuscripts and other supporting information.

Chapter 1 – Introduction.

This chapter provides the background on CP, health service utilisation, social determinants of health and health equity and access to health services for children and young people with CP.

Chapter 2 – A systematic review of health service utilisation in cerebral palsy.

This chapter provides a systematic review of factors associated with hospital health service utilisation in people with CP. This chapter was peer reviewed and published in *Archives of Physical Medicine and Rehabilitation* as Paget S, Ostojic K, Goldsmith S, Nassar N, McIntyre S. Determinants of Hospital-Based Health Service Utilization in Cerebral Palsy: a Systematic Review. Arch Phys Med Rehabil. 2022;103(8):1628-1637. doi:10.1016/j.apmr.2021.12.003.

Chapter 3 – Study Methods.

This chapter provides an outline of the major data sources used for the studies included in this thesis and an introduction to the formal data linkage processes that were used in some studies.

Chapter 4 - Comparison of cohorts of children with cerebral palsy from a population register and administrative health data: a data linkage study.

The literature review presented in this thesis drew attention to two common methodologies to define study populations in CP epidemiological studies: CP population registers and using International Classification of Diseases (ICD) codes in health administrative datasets. This study explores the consequences of this methodological choice by comparing the clinical and sociodemographic characteristics of these populations. This chapter was peer reviewed and published in *Paediatric and Perinatal Epidemiology* as Paget SP, McIntyre S, Lain S, Goldsmith S, Nassar N. A comparison of cohorts of children with cerebral palsy from a population register and hospital admission data: A data linkage study. Paediatr Perinat Epidemiol. 2024;38(1):22-30. doi:10.1111/ppe.13024.

Chapter 5 - Non-attendance at outpatient clinic appointments by children with cerebral palsy.

This study examines factors associated with non-attendance at outpatient clinics at two specialised children's hospitals in NSW. This chapter was peer reviewed and published in *Developmental Medicine and Child Neurology* as Paget SP, McIntyre S, Goldsmith S, et al. Non-attendance at outpatient clinic appointments by children with cerebral palsy. Dev Med Child Neurol. 2022;64(9):1106-1113. doi:10.1111/dmcn.15197.

Chapter 6 – Telemedicine for children with cerebral palsy before, during and after the COVID-19 pandemic.

The work that this thesis represents occurred during the COVID-19 pandemic, a time which saw momentous changes in health service access and the development of new models of care, many reliant on telehealth. This study explores how telehealth use changed for children and young people with CP at two specialised children's hospitals in NSW. This manuscript is currently in peer view with *Journal of Child Neurology*.

Chapter 7 - Outpatient encounters, continuity of care and unplanned hospital care for children and young people with cerebral palsy.

This study explores outpatient service utilisation across New South Wales (NSW) for children and young people with CP, exploring equity of access to services, including children and young people who are Aboriginal and/or Torres Strait Islanders. The study examines the association of outpatient encounters and continuity of care of outpatient care with unplanned hospital care comprising emergency department attendances and urgent hospital admissions. This chapter was peer reviewed and published in *Developmental Medicine and Child Neurology* as Paget SP, Mcintyre S, Schneuer FJ, et al. Outpatient encounters, continuity of care, and unplanned hospital care for children and young people with cerebral palsy. Dev Med Child Neurol. 2024;66(6):733-743. doi:10.1111/dmcn.15800.

Chapter 8 – Summary and conclusions.

This chapter provides a synthesis of the results of the preceding chapters and provides discussion about the implications for service delivery and potential directions for future research.

Figure i. Concept map for this thesis.

	Knowledge Gap	Objective	Chapter	Publication	Key Findings
Introduction	What is already known about health service access in CP?	Background and literature review	**Chapter 1**		
		(I) To systematically review the existing literature regarding determinants of hospital-based health service access among people with CP.	**Chapter 2**	Determinants of Hospital-Based Health Service Utilisation in CP: a Systematic Review. *doi:10.1016/j.apmr.2021.12.003*	• Hospital health service utilisation associated with age, severity and comorbidities.
Study Methods 	How do study populations differ between a CP population register and hospital admission data?	Introduce the NSW/ACT CP Register, administrative data sets and data linkage.	**Chapter 3**		
		(II) To compare the sociodemographic and clinical characteristics of children with CP identified from a CP population register and hospital admission data.	**Chapter 4**	A comparison of cohorts of children with CP from a population register and hospital admission data. *doi:10.1111/ppe.13024*	• Hospital admission data has sensitivity of 0.7 for a diagnosis of CP. • Children with CP from hospital admission data are older, live in major cities, more comorbidities and early death.
Dimensions of outpatient health service access and CP 	What factors influence non-attendance at specialty outpatient clinics?	(III) To explore the factors associated with non-attendance by children with CP at specialty outpatient clinics.	**Chapter 5**	Non-attendance at outpatient clinic appointments by children with CP. *doi:10.1111/dmcn.15197*	• Non-attendance associated with older age, socioeconomic disadvantage and previous non-attendance.
	How does telemedicine influence equity of access to outpatient care?	(IV) To determine how telemedicine supports equity of access to specialist outpatient care for children with CP.	**Chapter 6**	Telemedicine for children with CP before, during and after the COVID-19 pandemic. *Under review.*	• Telemedicine use lower in children living in regional and remote areas.
	How have telemedicine rates have changed since the COVID-19 pandemic?	(V) To determine the impact of the COVID-19 pandemic on telemedicine use in children with CP.			• Telemedicine use increased substantially during COVID-19 pandemic and since has declined to near baseline.
	What factors influence access to outpatient services?	(VI) To determine how clinical and sociodemographic determinants influence hospital outpatient service access in CP.	**Chapter 7**	Outpatient encounters, continuity of care and unplanned hospital care for children and young people with CP. *doi:10.1111/dmcn.15800*	• Outpatient service use associated with metropolitan areas, GMFCS IV-V, epilepsy, intellectual disability.
	Are CP outpatient services accessible for Aboriginal and/or Torres Strait Islander children?	(VII) To determine hospital outpatient service utilisation for Aboriginal and/or Torres Strait Islander children and young people with CP.			• Rates of outpatient service use for Aboriginal and/or Torres Strait Islander children are similar to non-indigenous population.
	How does outpatient continuity of care impact unplanned hospital care?	(VIII) To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.			• Decreased continuity of care is associated with increased unplanned hospital care.
Summary and Conclusions		Summary of key findings and implications for clinical practice, policy and research.	**Chapter 8**		

Preface

First Author Publications Related to Thesis

Paget SP, Ostojic K, Goldsmith S, Nassar N, McIntyre S. Determinants of Hospital-Based Health Service Utilization in Cerebral Palsy: a Systematic Review. *Archives of Physical Medicine and Rehabilitation*. 2022; *103*(8): 1628–1637. https://doi.org/10.1016/j.apmr.2021.12.003

Paget SP, McIntyre S, Goldsmith S, Ostojic K, Shrapnel J, Schneuer F, Waugh MC, Kyriagis M, Nassar N. Non-attendance at outpatient clinic appointments by children with cerebral palsy. *Dev Med Child Neurol*. 2022; 64: 1106–1113. https://doi.org/10.1111/dmcn.15197

Paget SP, Mcintyre S, Schneuer FJ, Martin T, Sellars L, Burnett H, Price S, Nassar N. Outpatient encounters, continuity of care, and unplanned hospital care for children and young people with cerebral palsy. *Dev Med Child Neurol*. 2024; 66: 733-743. https://doi.org/10.1111/dmcn.15800

Paget SP, McIntyre S, Lain S, Goldsmith S, Nassar N. A comparison of cohorts of children with cerebral palsy from a population register and hospital admission data: A data linkage study. *Paediatr Perinat Epidemiol*. 2024; 38: 22-30. https://doi.org/10.1111/ppe.13024

First Author Paper Presentations Related to Thesis

Paget SP, McIntyre S, Nassar N. Towards personalised medicine in cerebral palsy: factors that influence specialist outpatient service access and utilisation in children with cerebral palsy in New South Wales. University of Sydney Higher Degree by Research (HDR) conference 9 August 2019

Paget SP, Ostojic K, Goldsmith S, Nassar N, McIntyre S. Determinants of hospital-based health service utilisation in cerebral palsy: a systematic review. Australasian Academy of Cerebral Palsy and Developmental Medicine / International Alliance of Academies of Childhood Disability Conference 1-5 March 2022.

Paget SP, McIntyre S, Schneuer F, Nassar N. Factors associated with non-attendance at scheduled outpatient clinics in children with cerebral palsy. Australasian Academy of Cerebral Palsy and Developmental Medicine / International Alliance of Academies of Childhood Disability Conference 1-5 March 2022.

Paget SP, McIntyre S, Nassar N. Outpatient, continuity of care and unplanned health care use among children and young people with cerebral palsy. NSW Agency for Clinical Innovation Annual Education Forum 3 August 2022.

Paget SP, McIntyre S, Schneuer F, Martin T, Sellars L, Burnett H, Price S, Nassar N. Outpatient Encounters, Continuity of Care and Unplanned Hospital Care for Children and Young People with Cerebral Palsy in New South Wales, Australia. Perinatal Society of Australia and New Zealand 5-8 March 2023. (Abstract chosen for 'Best of the Best' session).

Paget SP, McIntyre S, Schneuer F, Martin T, Sellars L, Burnett H, Price S, Nassar N. Outpatient Encounters, Continuity of Care and Unplanned Hospital Care for Children

and Young People with Cerebral Palsy in New South Wales, Australia. American Academy of Cerebral Palsy and Developmental Medicine 77th Annual Meeting 10-13 September 2023. (Nominated for 2023 Gayle G. Arnold Award for Excellence).

Paget SP, McIntyre S, von Huben A, Nassar N. Telemedicine for children with cerebral palsy after the COVID-19 pandemic: a cohort study. 18th World Congress of the International Society of Physical and Rehabilitation (ISPRM) 1-6 June 2024.

Paget SP, McIntyre S, von Huben A, Stewart K, Williams T, Maly E, Ford K, Woolfenden S, Nassar N. Telemedicine for children with cerebral palsy before, during and after the COVID-19 pandemic: a cohort study. 12th Australasian Academy of Cerebral Palsy and Developmental Medicine (AusACPDM) Biennial Conference 31 July-3 August 2024.

First Author Poster Presentations Related to Thesis

Paget SP, McIntyre S, Goldsmith S, Ostojic K, Shrapnel J, Schneuer F, Waugh M-C, Kyriagis M, Nassar N. Non-attendance at outpatient clinic appointments by children with cerebral palsy. Rehabilitation Medicine Society of Australia and New Zealand 5th Annual Scientific Meeting (poster presentation) 7-10 June 2022.

First Author Awards Related to Thesis

Paget SP. Recipient of Career Development Grant from the Research Foundation of Cerebral Palsy Alliance 2019; Sydney.

Paget SP. Recipient of Research Entry Scholarship from the Royal Australasian College of Physicians Australasian Faculty of Rehabilitation Medicine 2020; Sydney.

Paget SP. Certificate of Achievement – 2nd place 'Developing Brain' Clinical Research Colloquium, Research Publication Awards 2023; Sydney.

List of Abbreviations

ACHI	Australian Classification of Health Interventions
ACPR	Australian Cerebral Palsy Register
ACT	Australian Capital Territory
APDC	Admitted Patient Data Collection
ARIA+	Accessibility/Remoteness Index of Australia
ASGS	Australian Statistical Geography Standard
BDM	(New South Wales Registry of) Births, Deaths and Marriages
CI	Confidence Interval
CFCS	Communication Function Classification System
CHeReL	Centre for Health Record Linkage
CP	Cerebral Palsy
CPR	Cerebral Palsy Register
DSM-5	Diagnostic and Statistical Manual of Mental Disorders, 5th Edition
ED	Emergency Department
EDACS	Eating and Drinking Classification System
EDDC	Emergency Department Data Collection
EMR	Electronic Medical Record
FACS	Family and Community Services
GMFCS	Gross Motor Function Classification System
HREC	Human Research Ethics Committee
HRIP	Health Records and Information Privacy Act

ICD	International Classification of Diseases
ID	Intellectual Disability
IQR	Interquartile Range
IRR	Incident Rate Ratio
IRSD	Index of Relative Socioeconomic Disadvantage
LHS	Learning Health System
MACS	Manual Abilities Classification System
MDT	Multidisciplinary Team
NAP	Non-Admitted Patient
NDIS	National Disability Insurance Scheme
NSW	New South Wales
NSW/ACT CPR	New South Wales / Australian Capital Territory Cerebral Palsy Register
OR	Odds Ratio
PDC	Perinatal Data Collection
PIPP	Privacy and Personal Information Protection Act
PPV	Positive Predictive Value
PROSPERO	Prospective Register of Systematic Reviews
RBDM	NSW Registry of Births, Deaths and Marriages Death Registrations
SCHN	Sydney Children's Hospitals Network
SCPE	Surveillance of Cerebral Palsy in Europe
SDH	Social Determinants of Health
SDPR	Single Digital Patient Record
SEIFA	Socio-Economic Indexes for Areas
SMD	Standardised Mean Difference
UPCI	Usual Provider of Care Index

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Chapter 1 Introduction

1.1 Overview

This thesis is presented as a series of papers that have been published in peer-reviewed journals, that explore access, and equity of access, to hospital-based outpatient services for children and young people with cerebral palsy (CP). This chapter will introduce the key concepts and provide a rationale for the thesis research aim and objectives. The concept map for this thesis is shown in **Figure 1.1**.

1.2 Context

At the time I commenced this doctorate, I worked as a paediatric rehabilitation medicine physician at the Children's Hospital at Westmead, New South Wales, Australia. My role included leadership of the Cerebral Palsy & Movement Disorder service, a primarily outpatient, multidisciplinary service that provides assessment, consultation and a range of rehabilitation interventions aimed at improving health and wellbeing for approximately 900 children and young people with CP and related conditions. Working in this role I was struck by the challenges that some families needed to overcome to access these

services, including travel, transport and competing priorities of work and family responsibilities. Many of the children I saw in my clinical work needed to attend multiple appointments with different specialty groups to help better manage their health, meaning that they were required to face these challenges multiple times each year. As CP is lifelong, for many families these challenges were ongoing. These challenges increased in complexity during the COVID-19 pandemic. This unprecedented event resulted in huge disruptions to the health services provided by my team and influenced system-wide changes such as increased use of telemedicine. The work this thesis represents is driven by a desire to better understand how accessible health care is for children and young people with CP and their families, to ensure they have the opportunity to live healthy lives.

Figure 1.1. Thesis Concept Map for Chapters 1 and 2.

	Knowledge Gap	Objective	Chapter	Publication	Key Findings
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Study Methods	How do study populations differ between a CP population register and hospital admission data?	Introduce the NSW/ACT CP Register, administrative data sets and data linkage.	**Chapter 3**		
		(II) To compare the sociodemographic and clinical characteristics of children with CP identified from a CP population register and hospital admission data.	**Chapter 4**	A comparison of cohorts of children with CP from a population register and hospital admission data. *doi:10.1111/ppe.13024*	• Hospital admission data has sensitivity of 0.7 for a diagnosis of CP. • Children with CP from hospital admission data are older, live in major cities, more comorbidities and early death.
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	How does outpatient continuity of care impact unplanned hospital care?	(VIII) To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.			• Decreased continuity of care is associated with increased unplanned hospital care.
Summary and Conclusions		Summary of key findings and implications for clinical practice, policy and research.	**Chapter 8**		

1.3 Introduction to Cerebral Palsy

CP is a neurological disorder that is lifelong and is often described as the most common physical disability of childhood. In recent years, the birth prevalence of CP has decreased in high income countries,(1) including in Australia, where CP occurs in 1.6 per 1000 live births (birth years 2013-2014).(2) CP is often described as 'an umbrella diagnosis', acknowledging that is a clinical description that is agnostic to underlying aetiology.(3)

Commonly used definitions of CP focus on five components: (i) that it is a(n) ('umbrella') group of conditions, (ii) that are all characterised by disorders of movement and posture which causes activity limitation (disability), (iii) that occur as the result of a process / processes that disrupt typical brain development, (iv) which occurs early in life (foetal and infancy) and (v) that the process / processes are static over time with no ongoing degenerative disease.(2, 4, 5)

CP is commonly described in terms of predominant motor disorder, topography and gross motor function; the next sections will briefly discuss these concepts.

1.3.1 Predominant motor disorder

CP is often described and categorised based on predominant motor disorder and distribution of motor involvement. Common motor disorder types include spasticity, dyskinesia (dystonia, chorea, athetosis), and less commonly ataxia and hypotonia.

- **Spasticity** is a form of hypertonia ('abnormally increased resistance to externally imposed movement about a joint')(6) defined as 'hypertonia in which one or both of the following signs are present: (i) resistance to externally imposed movement increases with increasing speed of stretch and varies with the direction of joint movement, and/or (ii) resistance to externally imposed movement rises rapidly above a threshold speed or joint angle.'(6)
- **Dyskinesia** is characterised by an excess of unwanted movements. Common forms of dyskinesia include **dystonia (**'involuntary sustained or intermittent muscle contractions cause twisting and repetitive movements, abnormal postures, or both'),(6) **chorea** ('ongoing random-appearing sequence of one or more discrete involuntary movements or movement fragments.'),(7) and **athetosis** ('slow, continuous, involuntary writhing movement that prevents maintenance of a stable posture').(7)
- **Ataxia** has been defined as 'an inability to generate a normal or expected voluntary movement trajectory that cannot be attributed to weakness or involuntary muscle activity about the affected joints'.(8)

- **Hypotonia** ('decrease in skeletal muscle tone leading to decreased resistance to passive stretching')(9) represents a more hotly discussed subtype of CP, in that hypotonia is often poorly characterised(10) and is a feature of many other neurodevelopmental disorders. Some CP classification systems (e.g., The Surveillance of Cerebral Palsy in Europe (SCPE)) do not recognise hypotonic CP as a distinct subtype.(11)

Whilst people with CP are commonly classified by predominant motor type, individuals exhibit multiple motor types more often than not.(12)

1.3.2 Topography

Topography of motor disorder in CP has been historically described using terms 'hemiplegia', 'diplegia' and 'quadriplegia' (and occasionally 'monoplegia' and 'triplegia').

- **Hemiplegia** describes motor disorder assessed/observed in an arm and leg on either left or right sides of the body.
- **Diplegia** describes motor disorder predominantly of both lower limbs (acknowledging that mild impairments in upper limbs are noted in most children with diplegia).
- **Quadriplegia** describes motor involvement of both arms and both legs (and frequently the trunk).

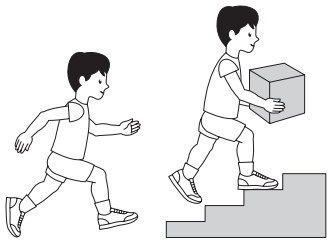
Registries, including the Australian CP Register (ACPR), apply topographical descriptions to the spastic motor type (i.e., spastic hemiplegic, spastic diplegic, spastic quadriplegic), with other motor types (dyskinetic, ataxic, hypotonic) not classified topographically.(2) The SCPE have adopted a simplified approach describing patterns of spasticity as either unilateral (involving an arm and/or leg on one side of the body) or bilateral (involving arms and/or legs on both sides of the body).(11) Motor disorders in CP are often more complex and can include substantial asymmetry in diplegia/quadriplegia. The distinction between diplegia and quadriplegia is also open to interpretation, which may account for the varying proportions of the topographies seen across different registries.(13)

1.3.3 Gross Motor Function Classification System (GMFCS)

While, historically, severity in CP was described as 'mild', 'moderate' and 'severe', functional abilities in CP are now classified through a range of classification systems. The most established of these is the Gross Motor Function Classification System (GMFCS).(14) The GMFCS and the GMFCS (Extended and Revised) (2007) classify gross motor function of people with CP (e.g., sitting, crawling, walking and use of mobility technologies) over five levels, with an emphasis on everyday performance rather than capacity in an idealised environment. Acknowledging the impact of age and development on gross motor function, the GMFCS adopts descriptions for age groups (0-2 years, 2-4 years, 4-6 years, 6-12 years, 12-18 years). Commonly used descriptors for GMFCS classification for 6–12-year-olds are shown in **Figure 1.2**.

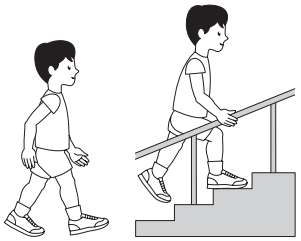
Figure 1.2. Gross Motor Function Classification System Descriptors (adapted from Palisano 1997). (14)

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



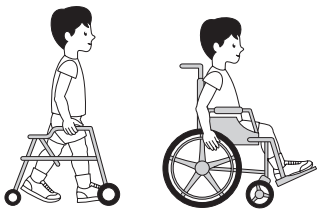
GMFCS Level I

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.



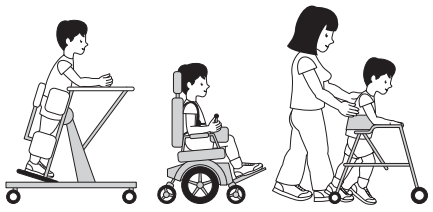
GMFCS Level II

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.



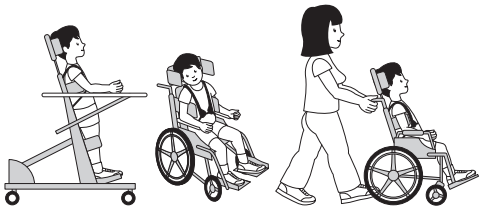
GMFCS Level III

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
CanChild: www.canchild.ca

GMFCS has been shown to be stable over time (except for 0-2 years), with a minority of children moving between levels, even with major interventions.(15) GMFCS has been adopted widely in clinical usage, where it is used, for example, to support frequency of hip surveillance.(16)

Since the development of the GMFCS, other classification systems have been developed that support classification of functional performance across other domains including manual abilities (Manual Abilities Classification System (MACS)),(17) communication (Communication Function Classification System (CFCS))(18) and eating and drinking (Eating and Drinking Ability Classification System (EDACS)).(19) The classification systems all share the same five point ordinal structure, and assist simplification and standardisation of classifying these different aspects of function to support better communication between clinicians, and standardisation for research.(20)

1.4 Common comorbidities in cerebral palsy

People with CP have higher rates of other comorbidities than the general population including disorders also caused by disruptions in the developing brain, complications of CP and diseases unrelated to CP aetiology.(21) Almost all (95%) children and young people with CP have one or more comorbidities,(21) more common comorbidities

include epilepsy, intellectual disability and respiratory and gastrointestinal diseases as outlined below. These comorbidities require ongoing management and treatment and are a common reason for use of health services (see **Section 1.6.4**).

1.4.1 Epilepsy

Epilepsy is a disease of the brain defined by (i) at least two unprovoked (or reflex) seizures occurring > 24 hours apart and/or (ii) one unprovoked (or reflex) seizure and a probability of further seizures similar to the general recurrence risk (> 60% over 10 years) and/or (iii) diagnosis of an epilepsy syndrome.(22) The lifetime prevalence of epilepsy in the general population is 7.60 per 1,000 persons (95% Confidence Interval 6.17–9.38),(23) and 3.2–5.5 per 1,000 children in high income countries.(24) The prevalence of epilepsy in CP is higher than the general population, estimated between 28% and 43% in high income countries.(25-28) In children with CP, epilepsy has been associated with dyskinetic and bilateral spastic motor types, GMFCS levels IV-V, severe intellectual disability and sensory impairments in univariable analyses,(28) and with MACS levels IV-V, intellectual disability and vision impairment in multivariable analyses.(26)

1.4.2 Intellectual disability

Intellectual disability (ID) is defined in the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5) as deficits in intellectual functioning (Full Scale

Intelligence Quotient (IQ) < 70), with associated deficits in adaptive functioning (conceptual, social and practical skills required for everyday functioning), and onset during childhood. Approximately 1.8 per cent of the Australian population have an ID.(29) The prevalence of ID amongst children with CP is much higher, estimated to be between 28-49% in different jurisdictions.(25, 30-33) Some of this variation is likely influenced by the difficulties in assessing cognition in children with motor impairments where the assessment is reliant on fine motor function and speech/communication, and intellectual function in children functioning at GMFCS level V is often assumed rather than formally assessed.(34) In children with CP, ID is associated with increasing GMFCS classification, dyskinetic and hypotonic motor types (compared with spastic), and concomitant epilepsy, and/or severe vision and/or hearing impairment.(31) Amongst children with spastic CP, severe ID (IQ<50) is more prevalent in those with bilateral motor impairment (compared with unilateral motor impairment) and those born at term (compared with those born moderately or very preterm).(35)

1.4.3 Other common comorbidities in cerebral palsy

1.4.3.1 Respiratory disease

Respiratory failure and disease are the most common causes of death in people with CP,(36) and a leading cause of hospital admissions (see **Section 1.6.4)**. Respiratory illnesses have been associated with children functioning at GMFCS level V, EDACS levels

III-V, epilepsy and gastro-oesophageal reflux disease, as well as previous respiratory disease and/or symptoms.(37)

1.4.3.2 Gastrointestinal diseases

Gastrointestinal diseases are also common in CP, including oropharyngeal dysphagia (impaired swallow), gastro-oesophageal reflux disease, and use of gastrostomy tube for feeding. Oropharyngeal dysphagia is common in CP, found in 85% of preschool-aged children, associated with GMFCS level.(38) The incidence of gastro-oesophageal reflux in children with CP has been reported to be between 31-75%,(39) and the incidence of gastrostomy tube feeds in a European study was 11% (95% confidence interval 9-12), with variation noted between different countries.(40) Gastrostomy tube feeding is associated with GMFCS levels IV-V, bilateral spastic and dyskinetic motor types.(40)

1.5 Health equity, health inequities and the social determinants of health

Health equity means that everyone has a fair and just opportunity to be healthy.(41) In practice, promoting health equity means reducing or eliminating health inequities - differences in health that are unnecessary, avoidable, unfair and unjust.(42) While the

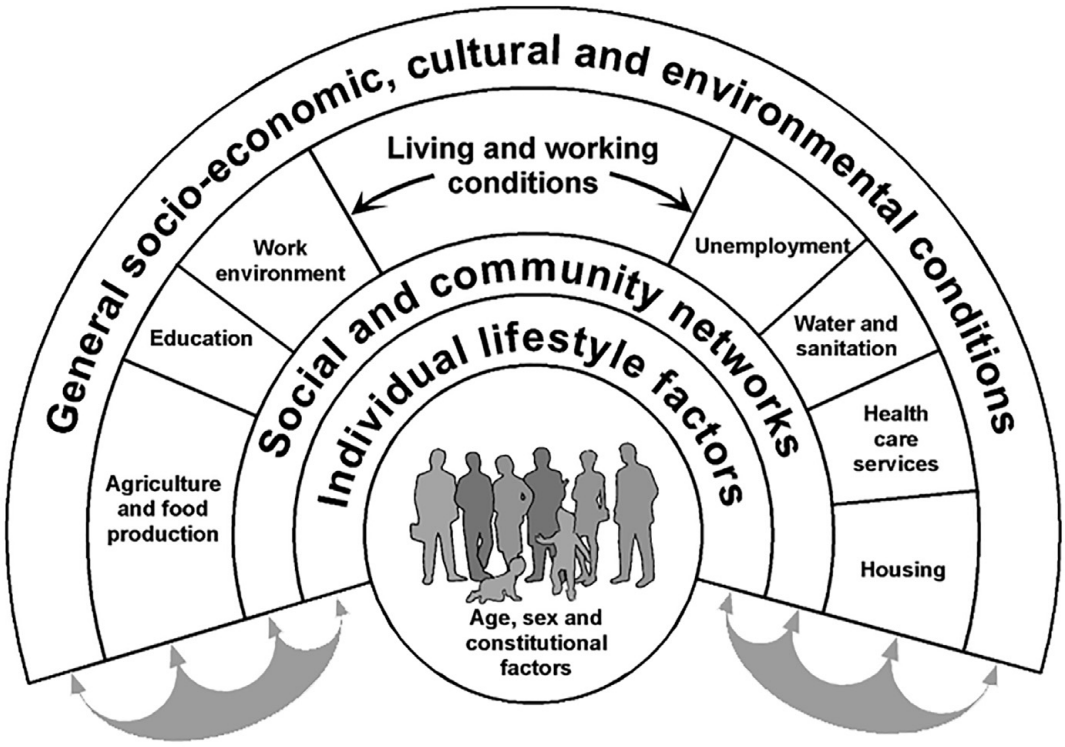
terms are often applied to health and health outcomes, they are also applied to differences in access to health care.

The existence of social gradient of health inequities in outcomes such as life expectancy and disability-free life is now well described.(43) There are now also examples of child health inequities in multiple settings and across a range of physical and developmental health outcomes, including overweight/obesity,(44, 45) chronic diseases,(45, 46) and socioemotional and behavioural problems.(45, 46) Health inequities are also apparent in CP.(47-49) Addressing child health inequities is a priority,(43, 50-52) not least because disadvantages in early-life can influence health across the lifetime, and potentially across generations.(50, 51)

The primary cause of health inequities are believed to be differences experienced in the Social Determinants of Health (SDH) – the "conditions in which people are born, grow, live, work, and age."(43) The social determinants of health are often described visually using the Dahlgren-Whitehead 'Rainbow Model' (**Figure 1.3**) which emphasizes the interplay between influences at an individual, local community and societal levels.(53) The mechanisms through which the SDH mediate differences in health outcomes are complex and poorly understood,(54) and models differentiate between material living conditions (e.g., safe housing, nutrition), psychosocial factors (e.g., the experience of

stress), and structural factors (e.g., socioeconomic and political environment, accessibility of health services).(54)

Figure 1.3. The Dahlgren-Whitehead model of health determinants. (53)



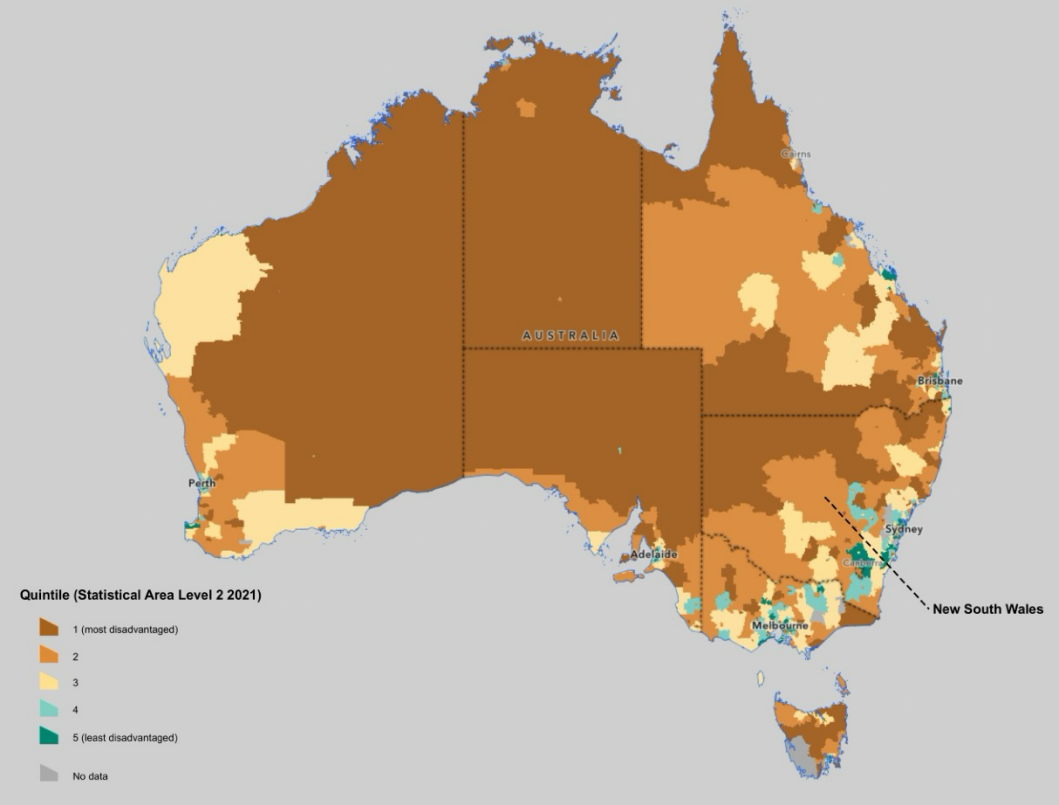
Two major SDH that are explored in this thesis (socioeconomic disadvantage, residential remoteness) sit at the level of "General socio-economic, cultural and environmental conditions" in the Rainbow Model. A further cultural determinant, that of being Aboriginal and/or Torres Strait Islander is explored in **Chapter 7**. It is important to acknowledge that being Aboriginal and/or Torres Strait Islander is not considered itself as a risk factor; rather the focus in this thesis is the recognized systemic health inequities that Aboriginal

and/or Torres Strait Islander people face. As described briefly below, there is substantial intersectionality between socioeconomic disadvantage and residential remoteness, and Aboriginal and/or Torres Strait Islander people experience both at high rates within the Australian population.

1.5.1 Socioeconomic disadvantage

Socioeconomic position is typically described related to income, educational attainment and level of occupation.(55) The Australian Bureau of Statistics derives Socio-Economic Indexes for Areas (SEIFA) for Australia, including ranking areas based on relative socio-economic disadvantage (e.g., high proportions of low household incomes, people without qualifications, people in low skilled occupations) using the Index of Relative Socioeconomic Disadvantage (IRSD).(56) Socioeconomic disadvantage is unevenly spread across Australia with, generally, more areas of less disadvantage in major cities and more areas of disadvantage in regional and remote communities (**Figure 1.4**). Further information about SEIFA and IRSD is provided in **Chapter 3**.

Figure 1.4. Index of Relative Socioeconomic Disadvantage (IRSD) by Statistical Area Level 2, Australia (2021).



Adapted from Australian Bureau of Statistics Index of Relative Socio-economic Disadvantage map (Available at https://experience.arcgis.com/experience/32dcbb18c1d24f4aa89caf680413c741/page/IRSD/)

In Australia, area-level socioeconomic disadvantage is associated with increased prevalence of obesity, chronic diseases (e.g., diabetes, ischaemic heart disease and cancer), and increased burden of disease (disability adjusted life years) for several common chronic health conditions.(57) Socioeconomic disadvantage (both individual(58, 59) and area level(48, 60)) has been found to be associated with increased prevalence of CP in some countries,(47) including studies that adjusted for confounding

variables (e.g., birthweight, gestational age).(48) In Australia, socioeconomic disadvantage has also been shown to influence CP severity, including GMFCS level,(61, 62) spastic bilateral CP(61) and severe comorbidities.(62)

1.5.2 Residential remoteness

Over one quarter of people in Australia live outside major cities in regional and remote areas.(63, 64) These people experience inequitable health outcomes compared to people living in major cities, with lower life expectancy, higher rates of hospitalisations and injuries,(63) and a higher burden from common chronic diseases.(64) Children living in regional and remote areas also have poorer health outcomes, poorer developmental outcomes and are more likely to be developmentally vulnerable than children living in major cities.(65) For many children living in regional and remote areas, remoteness is compounded by socioeconomic disadvantage, unemployment, and social isolation.(65) Almost 30% of children with CP in Australia live in regional or remote areas,(2) and only a small minority (approximately 9%) of families of children with CP move to less remote areas to access services during the first 5 years of life.(66) Further information about classification of areas is provided in **Chapter 3**.

1.5.3 Aboriginal and/or Torres Strait Islander Australians

Aboriginal and/or Torres Strait Islander people are a culturally and linguistically diverse population that represent 3.8% of the Australian population.(67) Aboriginal and/or Torres Strait Islander people show great resilience to the continuing legacy of colonisation which has contributed to the significant health inequities they experience.

Aboriginal and/or Torres Strait Islander people have lower life expectancy, double the burden of chronic disease, and are more likely to live with disability than the non-indigenous population.(68, 69) Until recently, there was limited knowledge about Aboriginal and/or Torres Strait Islander people living with CP.(70, 71) Aboriginal and/or Torres Strait Islander children born between 1996 and 2005 have been found to be at greater risk of CP.(71) More recent evidence suggests, pleasingly, a decline in the birth prevalence of CP in Aboriginal and/or Torres Strait Islander children since the mid 2000s, similar to that seen in the non-Indigenous population.(70) Two in five Aboriginal and/or Torres Strait Islander children with CP live in the most socioeconomically disadvantaged areas and one in five live in remote or very remote Australia.(70)

1.6 Access to and utilisation of hospital health care

Access to high quality health care is an important determinant of health outcomes, including for people with CP. Access to high quality health care however, is often least available to those with most need.(72) This section provides an overview of the Australian health system, focussing on the public health system and particularly specialist paediatric hospital outpatient services such as those providing medical rehabilitation treatments, and managing the health consequences of CP and associated comorbidities. **Chapter 2** describes a systematic review of determinants of health service utilisation for people with CP.

1.6.1 The Australian Health System

The Australian health system is complex and includes services funded by the Australian (Federal) Government, state and territory governments and through the private sector.(73, 74) Primary care includes services provided by general practitioners, community nurses and midwives, pharmacists, dentists, and allied health professionals. Public hospitals in Australia provide a range of services provided free at point of contact, managed largely by state and territory governments, and supported through funding provided by the Australian Government. Broadly, health care provided in hospitals is categorised as admitted patients, a patient who undergoes a hospital admission to

receive treatment and/or care; and non-admitted patients, a patient who receives hospital services that do not require a formal hospital admission.(75) Non-admitted patient encounters include those provided in emergency departments (EDs) and in outpatient clinics (provided by specialist medical practitioners, clinical nurse specialists and/or allied health professionals), diagnostic and procedural clinics.(75)

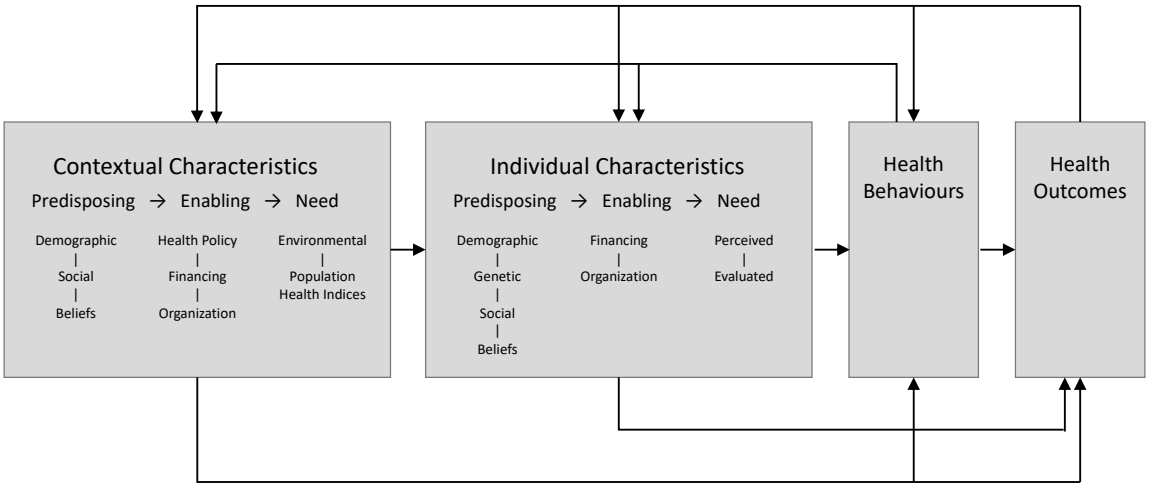
This thesis is focussed on health care provided in public hospitals, and particularly that provided in specialist paediatric hospital outpatient clinics. There are several reasons for this: specialist outpatient (paediatric rehabilitation medicine) clinics are my primary place of work and interaction with people with CP; children with CP frequently access health care in these outpatient settings;(76) and these services play a central role in the management of CP and comorbidities including prevention of and early intervention for adverse outcomes - particularly during childhood. Importantly, there are opportunities to explore whether inequities exist in access to outpatient care, and whether innovative models of care, including using digital health, can improve equity of access to outpatient services and improve health outcomes.

1.6.2 Models of health care access and utilisation

Access to health care services is a complex and multidimensional construct.(77) Definitions also vary, but most recognise access to health care as an interplay between

the characteristics of individual (health care) users, the characteristics of health systems and of the broader community.(77, 78) The Andersen Behavioural Model is a widely used conceptual framework that represents health service access as an interplay between individual and contextual (health organisation, provider and community) characteristics and the *predisposing*, *enabling* and *need* determinants that underpin these characteristics (**Figure 1.5**).(79, 80)

Figure 1.5. The Andersen behavioural model of health service access and use (modified from Andersen *et al*. (80))



At a contextual characteristic level:

- *Predisposing* factors include broad determinants of the community such as age and sex distribution, community attitudes and beliefs about health.
- *Enabling* factors include health policies, and financing and organisation of the health system.
- *Need* characteristics include general indicators of community health.

At an individual characteristic level:

- *Predisposing* factors include demographic, genetic and social factors that influence health service use.
- *Enabling* factors include the individual's capacity to attend health services (e.g., finance, travel).
- *Need* factors include perceived health status and health status evaluated by the health system.

A point of distinction highlighted in the Andersen Behavioural Model is the difference between *Potential Access* (i.e., the existence of accessible health services) and *Realised Access* (i.e., health care utilisation, actual use of health care services). **Chapter 2** uses

the Andersen Behavioural Model to classify determinants of *Realised Access* to hospital health services (utilisation) in CP.[1]

The Andersen Behavioural Model has gone through several iterations since its inception in the 1960s, with the later versions considering dimensions of health care access such as *Effective Access* (i.e., health care use resulting in positive health experience and health outcomes), and *Efficient Access* (i.e., minimising health care costs required to improve health outcomes).

1.6.3 Health care access and the social determinants of health

The inequities in health outcomes described above can be contributed to and compounded by inequities in health care access. Studies have supported Tudor Hart's assertion in 1970 that "the availability of good medical care tends to vary inversely with the need of the population served" (the 'Inverse Care Law').(72) Children living with socioeconomic disadvantage have been shown to access less GP, diagnostic and private specialist health care,(81-83) including in children who are developmentally vulnerable,(84) and to have higher use of ED and emergency inpatient admissions.(81-83) Children with chronic health conditions from areas with most socioeconomic

[1] A note about terminology: In the component studies of this thesis, health care access has been measured in terms of realised access (utilisation). The terms used in these studies 'access' and 'utilisation' usually refer to 'Realised Access'.

disadvantage also have a higher proportion of emergency hospital admissions.(85) Similarly, people in regional and remote Australia have substantial barriers to accessing health care, dubbed 'the tyranny of distance', which increases travel and/or costs associated for providers and families alike.(65, 86) This results in people living in regional and remote Australia having more hospitalisations and being less likely or waiting longer to see specialist providers.(63, 64) Children living in regional and remote Australia have also been identified as having poorer access to services, including paediatricians and allied health specialists.(65) Aboriginal and/or Torres Strait Islanders experience additional barriers to accessing health services related to availability, cost and cultural appropriateness. Aboriginal and/or Torres Strait Islander people access GP services less (when adjusted for burden of disease) and have a higher proportion of hospitalisations than non-indigenous Australians.(69) Aboriginal and/or Torres Strait Islander children access less paediatrician and/or general practitioner health services than non-indigenous Australians, but are more likely to be hospitalised.(87)

1.6.4 Health service access and utilisation in cerebral palsy

People living with CP are frequent users of hospital health services for medical rehabilitation treatments, and to manage the health consequences of CP and the frequently associated comorbidities. Studies have suggested that children(88-90) and adults(90, 91) with CP use inpatient hospital services proportionally more than the general population. Qualitative differences have also been noted in inpatient hospital

health service use, with children with CP having proportionally more elective (as opposed to emergency)(92, 93), and multiday admissions.(92) The reasons (diagnoses) for hospital admissions reflect the musculoskeletal impacts of the condition and the range of comorbidities seen in CP, including diseases of the nervous system, respiratory system and the gastrointestinal system.(88, 89, 93, 94)

A similar pattern is seen when looking at hospital emergency department (ED) and outpatient use. Children(90, 95) and adults(90, 91) with CP are more likely to attend ED, and children with CP are more likely to arrive by ambulance and/or be admitted to hospital from ED than the general population.(96) Children(90) and adults(90, 91) with CP also have more outpatient encounters than the general population, with a higher proportion of (allied health) therapy appointments,(92) although there is less research describing outpatient access from an epidemiological perspective. There is also limited research exploring how social determinants of health impact health care access for people with CP. Private health insurance, (parental) full time employment, and regional/remote location were associated with increased proportions of use of private specialist services in one Victorian study.(97)

Knowledge gaps: *There is a need for research that systematically reviews determinants of health service access for people with CP* (**Thesis Objective 1**); *better understanding of how the social determinants of health influence access to hospital*

outpatient services for children and young people with CP (**Thesis Objective 6**); and *understanding about health service access for Aboriginal and/or Torres Strait Islander people with CP* (**Thesis Objective 7**).

Existing population-level research about health service access in CP have generally used one of two sources to define their study population. Some studies have used CP population registers (e.g., (31, 92, 93, 96, 98)), whereas others have used administrative health data using case definitions based on individual or patterns of International Classification of Diseases (ICD-9, ICD-10) codes for CP (e.g., (94, 99-101)). Studies that have compared CP registers and administrative health data have suggested that administrative health data is only moderately sensitive to a diagnosis of CP, and less sensitive in those born term or with hemiplegia (potentially influenced by less routine follow-up and/or lesser severity).(102) A Norwegian study found administrative health data to misdiagnose CP in 14% of cases.(103)

Knowledge gap: *There is a need to determine how choice of CP data source, population register or administrative health data (based on ICD codes) influences the characteristics of the populations derived, particularly in the Australian context* (**Thesis Objective 2**).

1.7 Dimensions of health service access: non-attendance, continuity of care and telemedicine

Potential Access is insufficient to meet the health needs of the population. For health needs to be (better) met attendance (*Realised Access*) and high quality care (*Effective Access*) are also required.(80) This thesis examines dimensions of access to specialist hospital outpatient services including non-attendance, telemedicine and continuity of care. This section briefly reviews these dimensions.

1.7.1 Non-attendance

Non-attendance at scheduled outpatient appointments, defined as a missed appointment without prior notification, is recognised as a major issue across the health care system and health conditions. At a health service level, non-attendance is recognised to increase health care costs, decrease services' effective capacity, and adds to waiting times for consultations and procedures.(104) At a patient level, non-attendance may represent a missed opportunity for the early diagnosis of a health-related problem, or the initiation of an intervention to improve health or wellbeing.(105) In children with neurological conditions, non-attendance has been associated with increased unplanned health care use such as emergency department presentations.(104) There are multiple factors that contribute to non-attendance,

including individual (e.g., younger age, history of previous non-attendance), societal (e.g., socioeconomic disadvantage, ethnicity, insurance status) and those related to the clinical service (e.g., waiting times for appointments, administrative error).(105-107)

The reasons that families do not attend outpatient clinic appointments have also been the subject of recent qualitative studies.(108, 109) Common reasons reported included travel difficulties, competing priorities and administrative issues (e.g., not receiving an appointment, difficulties in changing an appointment) - highlighting the complexities that families need to manage to balance their child's health and other priorities.(110)

> **Knowledge Gap**: *There is a need for better understanding of determinants associated with non-attendance at outpatient appointments for children with CP to enable the identification of strategies to improve access and health outcomes* (**Thesis Objective 3**).

1.7.2 Continuity of care

Continuity of care is a key measure of the quality of outpatient services.(111, 112) Continuity of care describes the extent with which patients experience the health system as coherent and interconnected over time.(112) It can include informational continuity (using knowledge of past events and personal history to influence current management),

management continuity (consistency in management of a health condition across providers) and relational continuity (an ongoing therapeutic relationship between provider and patient).(113) Improved continuity of care has been associated with decreased hospital admissions and emergency department presentations in other childhood populations.(114-116) In one study of children with CP, continuity of care was associated with decreased hospital admission duration.(101)

Knowledge gap: *There is a need to better understand the impact of continuity of care on health outcomes in children with CP* (**Thesis Objective 8**).

1.7.3 Telemedicine

Telemedicine[2] describes the delivery of health care services, by healthcare professionals, using information and communications technologies for the exchange of information.(117) This is particularly pertinent when distance is a critical factor. Telemedicine includes outpatient encounters where consultation occurs through video or telephone, but the term is often used to include broader approaches including the sharing of medical images and remote monitoring of vital signs (e.g., heart rate, blood pressure, temperature).(118) Telemedicine is an important component of the Australian

[2] A note about terminology: terms used in telemedicine such as 'telemedicine' and 'telehealth' are often used in research and policy documentation interchangeably. NSW Health's preferred term is Virtual Care. The term telemedicine has been used predominantly in this thesis, consistent with the majority of current research literature.

National Digital Health Strategy,(119) and may improve equity of access to some health care,(120, 121) particularly for families living in regional and remote areas.(122, 123)

The COVID-19 pandemic was associated with a rapid and substantial increase in telemedicine as public health restrictions and the direct impacts of the virus limited access to in-person health care,(124) including for children with CP.(125) Studies conducted during this period, particularly among children with CP and other neurodevelopmental disorders, described increased telemedicine use for medical consultations.(125) They also explored the potential for telemedicine to provide rehabilitation programs to improve gross motor function(126) or manage common comorbidities such as epilepsy.(127) One study suggested that telemedicine did not suit all aspects of care for children with neurodevelopmental disabilities.(128) Other studies highlighted inequities in telemedicine access(127) that are also seen more broadly in society,(129) such as availability of information technologies (e.g., smartphones, broadband internet) and usability of digital platforms for those with less experience of technology.(130)

Knowledge gaps: *There is a need to better determine to what extent telemedicine supports equity of access to specialist outpatient care for children with CP* (**Thesis Objective 4**), and *how telemedicine use changed during and particularly since the COVID-19 pandemic* (**Thesis Objective 5**).

1.8 Thesis aim and objectives

This chapter has described the complexity of CP including the high frequency of comorbidities that determine the need for hospital health services. It has also described how non-medical factors, the Social Determinants of Health, such as socioeconomic disadvantage and residential remoteness can influence health outcomes, and health care access. As identified above, there remain significant knowledge gaps in how clinical and non-medical factors influence health service access in CP.

The overall aim of this thesis is to examine access, and equity of access to hospital-based outpatient services that are intended to support children and young people with CP.

In order to meet this aim, the thesis uses the New South Wales (NSW) / Australian Capital Territory (ACT) CP Register as the study population for a set of epidemiological health service studies to address the knowledge gaps identified above.

This thesis investigates 8 specific objectives:

Objective 1. To systematically review the existing literature regarding (clinical and sociodemographic) determinants of hospital-based health service access among people with CP.

Method: systematic review of hospital health service utilisation (**Chapter 2**).

Objective 2. To compare sociodemographic and clinical descriptors of children and young people identified as having CP in either a CP population register or hospital admission data.

Method: population-based data linkage study (**Chapter 4**).

Objective 3. To explore the association of clinical and sociodemographic descriptors with non-attendance at specialty children's hospital outpatient clinics.

Method: data linkage study of children's hospital outpatient services (**Chapter 5**).

Objective 4. To determine to what extent telemedicine supports equity of access to specialty children's hospital outpatient services for children with CP living with socioeconomic disadvantage and/or in regional and remote areas.

Objective 5. To determine the impact of the COVID-19 pandemic on telemedicine use at specialty children's hospital outpatient clinics.

Method: data linkage study of children's hospital outpatient services (**Chapter 6**).

Objective 6. To determine how clinical and sociodemographic determinants influence hospital outpatient service access and utilisation across NSW.

Objective 7. To determine hospital outpatient service utilisation for Aboriginal and Torres Strait Islander children and young people with CP.

Objective 8. To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.

Method: population-based data linkage study (**Chapter 7**).

1.9 Chapter Synopsis

In **Chapter 1**, the aim of this thesis was described, and literature on the topic was presented with key knowledge gaps identified. Specific thesis objectives were outlined, along with the methods proposed to meet these objectives. **Chapter 2** will explore the determinants of health service utilisation for people living with CP.

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Chapter 2 Determinants of hospital-based health service utilisation in cerebral palsy: a systematic review

2.1 Published Manuscript

This chapter has been reformatted for publication as part of this thesis.

The citation for the published manuscript is:

> Paget S, Ostojic K, Goldsmith S, Nassar N, McIntyre S. Determinants of Hospital-Based Health Service Utilization in Cerebral Palsy: a Systematic Review. Arch Phys Med Rehabil. 2022;103(8):1628-1637. doi:10.1016/j.apmr.2021.12.003

A copy of the published article as well as a statement of the specific contributions of the co-authors can be found in **Chapter 10 - Appendix B**.

2.2 Introduction

Cerebral palsy (CP) is the most common cause of physical disability in childhood, with a current estimated worldwide incidence of 2.0 per 1000 live births.(1) In addition to the motor disorder that defines the condition, almost all children with CP have comorbidities (e.g., epilepsy, intellectual disability) and/or complications (e.g., hip subluxation, scoliosis),(2) and in adulthood, people with CP experience higher rates of many non-communicable diseases compared with adults without CP.(3)

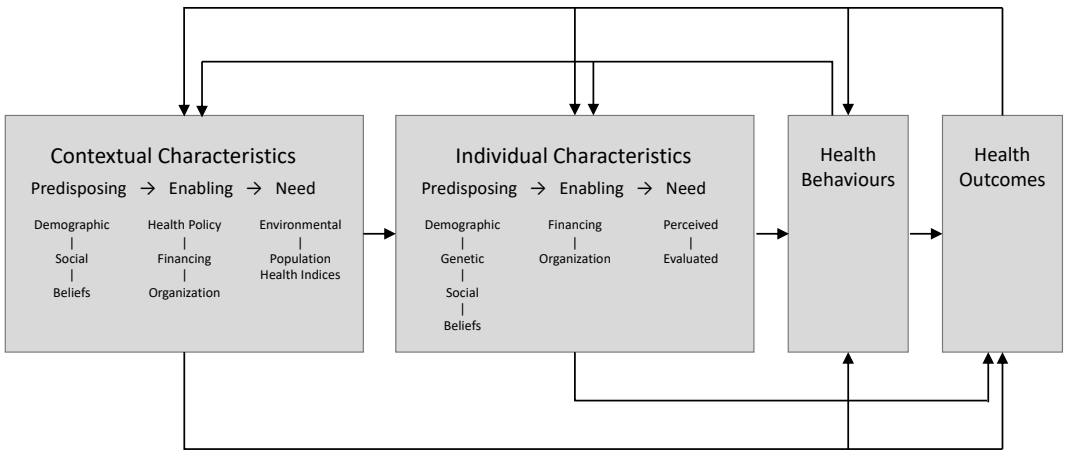
Surveillance for and treatment of these health problems is the primary role of health services. Ensuring that people with CP have access to "the right [health] service at the right time in the right place" (page 866)(4) can support improved health outcomes, decreased costs for the health system,(5) and reduced unplanned healthcare use.(6)

Studies exploring other health conditions (e.g., HIV infection,(7) common mental disorders(8)), suggest that health service utilisation is influenced not only by need. The Andersen behavioural model is a widely used framework that represents health service access and utilisation as an interplay between individual and contextual (health organisation, provider and community) characteristics and the predisposing, enabling and need determinants that underpin these characteristics (**Figure 2.1**).(9, 10) Examples

of individual factors incorporated in the model include age and sex (individual predisposing factors), personal finances and transport (individual enabling factors), and perceived health status, symptom severity and duration (individual need factors).

Achieving a better understanding of the factors that influence health service utilisation for people with CP is an important first step to improve equity of access. Use of the Andersen model can highlight where utilisation is driven by determinants other than need. The aim of this systematic review was to identify evidence of determinants associated with hospital-based health service use among people with CP using the Andersen model as a framework.

Figure 2.1. The Andersen behavioural model of health service access and use (modified from (10)).



2.3 Methods

The systematic review was conducted following PRISMA guidelines and protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) on 28/04/2020 (registration number CRD42020176271).

2.3.1 Search Strategy

We searched MEDLINE (January 2000 to March 25, 2020), Embase (January 2000 to March 25, 2020) and APA Psycinfo (January 2000 - March Week 4, 2020) through the Ovid platform (date of search 25/03/2020, updated 20/04/2020). The search strategies are shown in **Supplementary Table 9.1**, with the search adjusted as appropriate for each database. The references of included studies were reviewed for additional inclusions.

2.3.2 Eligibility Criteria

The inclusion criteria for studies were those that: (i) identified people with CP (of any age), including studies where data was available and attributed distinctly to people with CP amongst other health conditions or developmental disabilities; (ii) observational studies that reported quantitative measures of formal utilisation of medical and/or multidisciplinary hospital-based health services (hospital inpatient, emergency

department and outpatient services) (including those in private / clinic settings);(iii) reported determinants that show a relationship with rates and/or types of hospital-based health service utilisation; and (iv) were conducted in high income countries based on the World Bank classification of national economies.(11)

We excluded studies: (i) that were limited to a subset of people with CP not representative of the whole population (examples of this are studies limited to only those with severe intellectual disability, studies limited to those attending a specific medical subspecialty); and (ii) that only reported therapy service utilisation (hospital-based or community). We decided to exclude therapy services following piloting of the study selection process. This identified a wide range of settings for therapy services (hospital, community) in different countries which we felt would make any appropriate comparison and synthesis challenging. We also felt this approach would be more consistent with exclusion of studies reporting only on specific medical subspecialties. Further minor protocol amendments are documented in PROSPERO.

2.3.3 Study Selection

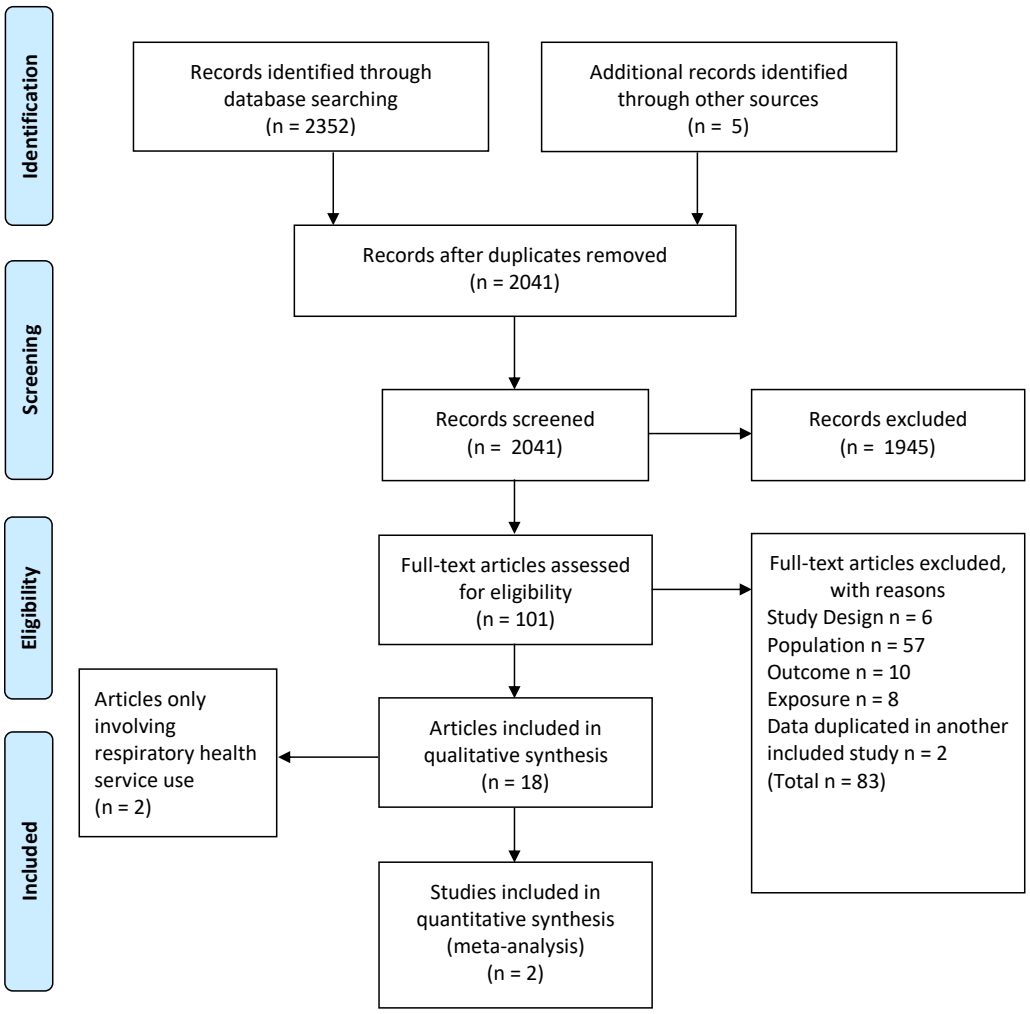
All references selected were imported into Endnote (Endnote X9; Thomson Reuters) and duplicates removed. One author (SP) independently screened the titles and abstracts of the selected references. Titles and/or abstracts not published in English were excluded.

Full texts were retrieved for all articles included after title/abstract screen. Studies that met title and abstract inclusion criteria with English-language title and abstract, but non-English full-text articles, were translated by native speakers of the language. Full texts were reviewed by two authors for inclusion (SP in all cases with either KO, SG or SM) and reasons for exclusion were recorded (**Figure 2.2**). Where disagreements arose, these were resolved through discussion between reviewers.

2.3.4 Risk of bias assessment

We evaluated the quality and risk of bias of included cohort studies using the Newcastle Ottawa Scale (12) and a modified Newcastle Ottawa Scale for cross-sectional studies. (13) We supplemented additional questions from two other risk of bias tools (14, 15) to meet the domains identified to be important in the methodological design of observational studies (**Supplementary Table 9.2**).(16) Consistent with advice,(16, 17) summary scores were not calculated, rather risk of bias assessment for each study reviewed was presented as a matrix with compliance to criteria noted (**Table 2.1**). For each study, risk of bias was independently assessed by two authors (SP in all cases with either KO, SG or SM) with any differences in scores resolved by discussion.

Figure 2.2. PRISMA flow diagram.



2.3.5 Data Extraction

The data extracted from the included studies were study design, country of origin, sample size and age range. Determinants of health service utilisation reported in included studies were classified consistent with the Andersen behavioural model into individual predisposing, enabling, need and contextual factors by two authors (SP in all cases with either KO, SG or SM).(10) Outcomes were defined as rates and/or proportions of populations using health services and classified by type: inpatient, emergency department or outpatient. One author (SP) extracted data from each of the included studies and accuracy of data extraction was checked by a second author (KO, SG, SM). Any differences were resolved through review and discussion.

Where possible, data were transformed into proportions, and odds ratios calculated to aid comparison and a measure of effect size. Meta-analysis using random effects models was performed to provide pooled odds ratios (OR) and 95% confidence intervals (CI) of health service utilisation from two or more comparable studies using Review Manager (RevMan) (Version 5.3. Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014).

Table 2.1. Characteristics and quality assessment of studies included in the systematic review.

Study details	Setting	Study period	Population (N)	Age range (years)	Outcome types	Data source (outcome)	Risk of Bias assessment scales and items									
							Newcastle Ottawa Scale								A	D
Cohort studies							**Representativeness**	**Non-exposed cohort**	**Exposure ascertainment**	**Outcome timing**	**Controls**	**Outcome assessment**	**Follow-up duration**	**Adequacy of follow-up**	**Missing outcomes**	**Statistics**
Abdullahi 2020 (18)	Australia	1983-2010	1663	0-27	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Blackmore 2018 & 2020 (19, 20)	Australia	2010-2016	482	1-26	IP, ED	Administrative health data	★	★	★	★	★	★	★	★	★	★
Carter 2020 (21)	UK	2004-2014 (IP) 2010-2014 (OP)	1684	0-24	IP, OP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Kim 2014 (22)	South Korea	2007	10659	1-18	IP, OP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Meehan 2017a (23)†	Australia	2007-2014	1748	0-18	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Meehan 2017b (24)†	Australia	2007-2014	1748	0-18	ED	Administrative health data	★	★	★	★	0	★	★	★	★	★
Morgan 2020 (25)	Australia	2013-2017	551	0-80	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Reid 2018 (26) †	Australia	2007-2014	1748	0-18	IP, ED	Administrative health data	★	★	★	★	★	★	★	★	★	★

Yang 2020 (27)	Taiwan	2000-2013	3234	0-18	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
							Newcastle Ottawa Scale									A
Cross-sectional studies							**Representativeness**	**Sample size**	**Response rate**	**Exposure ascertainment**	**Confounding controlled**	**Outcome assessment**	**Statistics**	**Missing outcomes**		
Boulet 2009 (28)	US	1997-2003	305	3-17	OP	Self-report	★	★	★	★	★	0	★	★		
Fortuna 2018 (29)	US	2011-2014	229	18 - >59 ‡	OP, ED	Medical records	★	★	★	★	0	★	★	★		
Meehan 2016 (30)	Australia	2014	350	5-19	OP	Self-report	★	★	★	0	0	★	★	★		
Pons 2017 (31)	France	2010-2012	282	18->39 ‡	OP	Self-report	★	★	★	★	0	0	★	★		
Roquet 2018 (32)	France	2010-2013	512	2->39 ‡	OP	Self-report	★	★	★	★	★	0	★	★		
Whitney 2019 (3)	US	2016	5555	18-64	IP, OP, ED	Administrative health data	★	★	★	★	★	★	★	★		
Young 2007 (33)††	Canada	1999-2002	1064	9-32	OP	Administrative health data	★	★	0	★	0	★	★	★		
Young 2011 (34)††	Canada	1999-2002	1064	9-32	IP	Administrative health data	★	★	0	★	0	★	★	★		

A: Agency for Healthcare Research and Quality (15); D: Downs and Black (14); ED: Emergency; IP: Inpatient; OP: Outpatient

* Blackmore 2018, 2020 limited to inpatient admissions, emergency department attendances due to respiratory causes

† Studies (23, 24, 26) derived from same population †† Studies (33, 34) derived from same population

‡ Upper age limit unspecified beyond detail given

2.4 Results

Figure 2.2 presents the PRISMA flowchart of study selection. We identified eighteen articles that described seventeen studies to be included in the review.(3, 18-34) The results of one study were reported in an original article(19) and follow-up letter;(20) both articles were included but described as one study for the purpose of this review. Of the seventeen studies, five reported different aspects of health service utilisation for the same population (Meehan 2017a, Meehan 2017b and Reid 2018;(23, 24, 26) Young 2007 and Young 2011(33, 34)). Articles were published between 2006 and 2020, with two-thirds (n=12 studies, 71%) published during or after 2017 (**Table 2.1**).

Studies reported on research conducted in Australia (n=7), the United States of America (n=3), Canada (n=2), France (n=2), United Kingdom (n=1), South Korea (n=1) and Taiwan (n=1). Half of the studies (n=9, 53%) were based on cohort studies and the remaining eight were research of cross-sectional design. Seven studies reported exclusively about research on children with CP; three reported exclusively about research on adults with CP and seven reported on research about both children and adults. The sample size in each study ranged between 229-10,659 individuals with CP. The quality of the included studies was generally high (**Table 2.1**).

Articles reported research examining health service utilisation in inpatient (n=10 studies), outpatient (n=9) and emergency department (n=5) settings. Research measured health service use over varied time periods from one year to 27 years; most (n=12) used administrative data / medical records to measure utilisation; and four studies used self-report. Sixteen studies reported on health service utilisation of any cause. One study(19, 20) included only respiratory causes of hospital admissions (over two time periods) and emergency department attendances and was considered separately.

2.4.1 Overall health service utilisation

2.4.1.1 Determinants

The sixteen studies described a total of 26 determinants related to health service use (**Table 2.2**, **Table 2.3**). The most common determinants examined were age (n=7 studies), sex (n=4), Gross Motor Function Classification System (GMFCS) level (35) (n=6) and epilepsy (n=4). Determinants were matched to Andersen behavioural model characteristics (individual predisposing / individual enabling / individual need and contextual), with most (n=15) determinants matched to “individual need” characteristics.

2.4.1.2 Individual Predisposing Factors

Younger children had more inpatient admissions(27) and outpatient attendances(22) than older children (**Table 2.2**). Younger children also represented a greater proportion of total inpatient admissions(23) and emergency department attendances(24) than older children (**Table 2.2**). Adults had 50% lower odds of inpatient admission than youth (OR 0.5; 95%CI 0.38-0.67)(33) and represented a lower proportion of inpatient admissions than children (**Table 2.3**).(25) Comparison of outpatient use between adults and children was mixed depending on specialty (**Table 2.3**).(32) Most studies identified no association between sex and inpatient admissions (**Table 2.2**).(3, 21, 25, 27, 33) One study reported lower rates of outpatient attendance in males (OR 0.39; 95%CI 0.32-0.48) (**Table 2.3**),(21) but rates of health service use between sexes was similar in another.(3)

Table 2.2. Determinants of health service utilisation evaluated and number of studies investigating and reporting an association.

Andersen Model characteristic	Determinant	Total articles (n)	Inpatient		Outpatient		Emergency	
			Number reporting association	Direction of association	Number reporting association	Direction of association	Number reporting association	Direction of association
Predisposing								
Demographic	Age	7						
	- Younger child v older child	3	1/1	↑	2/2	↑		
	- Child v adult	3	2/2	↑	1/1	Varied		
	- Younger adult v older adult	1			0/1	NA	0/1	NA
	Male sex	4	1/4	NA	1/1	↓		
Social Structure	Foreign-born mothers from high-income countries	1	1/1	↓				
	Two parent household	1			0/1	NA		
	Parental UG degree or higher	1			0/1	NA		
Enabling								
Assets	>=1 parent in FT employment	1			1/1	↑ (Pr)		
	Private Health Insurance	1			1/1	↑ (Pr)		
Need								
Perceived	Reported good health	1			0/1	NA		
Evaluated - Disability Severity	GMFCS IV-V	6	2/2	↑	3/4	↑	1/1	↑
	Bilateral topography	1	1/1	↑				
	Dyskinetic Motor Type	1	1/1	↑				

Evaluated - Comorbidity	ID	2	1/2	↑			1/1	↑
	DD	1			1/1	↑	1/1	↑
	Epilepsy	4	2/2	↑	1/1	↑	1/1	↑
	Gastrostomy	3	1/1	↑	1/1	↑	1/1	↑
	GORD	1	1/1	↑				
	Nasogastric tube use	1	1/1	↑				
	ITB Pump	2	1/1	↑			1/1	↑
	Asthma	1	0/1	NA				
	PTL & SGA	1	1/1	↓				
	Perinatal complication	1	0/1	NA				
	Pneumonia	1	1/1	↑				
Contextual								
Place of Residence	Place of Residence	2	1/1	Varied	1/1	↑ (Pr)		
	Urbanisation Level	1	0/1	NA				
Health Service Factors	High Continuity of Care	1	1/1	↓				
	Smaller hospital	1	1/1	↑				

↑ - Majority of studies report exposure is associated with increased HSU; ↓ - Majority of studies report exposure is associated with decreased HSU; NA Non-applicable – studies do not support association with HSU; Varied - studies suggest varied associations with HSU.

COB country of birth; DD developmental disability; FT full-time; GMFCS Gross Motor Function Classification System; GORD gastro-oesophageal reflux disease; ID intellectual disability; ITB intrathecal baclofen; Pr private; PTL pre-term labour; SGA small for gestational age; UG undergraduate

Table 2.3. Reported association between Andersen Model determinants and health service utilisation by health setting.

Determinant	Setting	Association (Odds Ratio, 95%CI)	Source
Predisposing			
Age 5-9 years (v older child)	OP	2.32 (1.32-4.07)	Meehan 2016 (30)
Age (Adult v Child)	IP	0.50 (0.38-0.67)	Young 2007 (33)
	OP (PMR)	0.29 (0.19-0.45)	Roquet 2018 (32)
	OP (Psych)	6.42 (3.31-12.47)	Roquet 2018 (32)
	OP (Neuro)	0.67 (0.46-0.97)	Roquet 2018 (32)
Male sex	OP	0.39 (0.32-0.48)	Carter 2020 (21)
Enabling			
>=1 parent in FT employment	OP (Private)	4.33 (2.68-6.99)	Meehan 2016 (30)
Private Health Insurance	OP (Private)	3.07 (1.94-4.87)	Meehan 2016 (30)
Need			
GMFCS IV-V	IP	1.82 (1.41-2.33)	Carter 2020 (21)
	IP	3.63 (2.51-5.25)	Meehan 2017a (23)
	OP	1.41 (1.12-1.77)	Carter 2020 (21)
	OP	2.34 (1.44-3.80)*	Meehan 2016 (30)
	OP (PMR)	1.75 (1.08-2.83)	Pons 2017 (31)
	ED	3.27 (2.57-4.17)	Meehan 2017b (24)
Bilateral topography	IP	1.63 (1.28-2.07)	Meehan 2017a (23)
Dyskinetic motor type	IP	2.20 (1.13-4.27)	Meehan 2017a (23)
Developmental Disability	OP	11.89 (6.77-20.87)	Boulet 2009 (28)
	ED	4.44 (1.87-10.51)	Boulet 2009 (28)
Epilepsy	IP	2.55 (1.85-3.52)	Meehan 2017a (23)
	OP	2.74 (1.51-4.96)	Meehan 2016 (30)
	ED	2.86 (2.27-3.59)	Meehan 2017b (24)

Gastrostomy	IP	6.65 (3.37-13.1)	Meehan 2017a (23)
	OP	3.99 (1.75-9.08)	Meehan 2016 (30)
	ED	4.52 (3.15-6.48)	Meehan 2017b (24)
ITB	IP	7.27 (0.99-53.53)	Meehan 2017a (23)
	ED	4.2 (1.6-11.03)	Meehan 2017b (24)
Contextual			
Region of residence (regional v metropolitan)	OP (Private)	1.64 (1-2.7)	Meehan 2016 (30)

ED Emergency Department; FT full-time; GMFCS Gross Motor Function Classification System; IP Inpatient; ITB Intrathecal Baclofen; OP Outpatient; Neuro Neurologist; PMR Physical Medicine & Rehabilitation Physician; Psych Psychiatrist.
* Meehan 2016 (27) is GMFCS III-V

2.4.1.3 Individual Enabling Factors

The impact of parental employment status and private health insurance on outpatient health care utilisation was explored in one Australian study (**Table 2.2**).(30) Full-time employment and access to private health insurance were both reported to be associated with increased use of private paediatric medical specialists.(30)

2.4.1.4 Individual Need Factors

A total of 15 individual need characteristics (perceived and evaluated) were identified. Persons who primarily used a wheelchair for mobility (GMFCS IV-V; compared with GMFCS I-III) had increased health service use across inpatient,(21, 23) outpatient(21, 30, 31) and emergency department(24) settings (**Table 2.2**, **Table 2.3**). Meta-analysis of 2483 children from two studies(21, 23) showed children functioning at GMFCS levels IV-V having increased odds of hospital admission (pooled OR 2.53; 95%CI 1.28-5.02) (**Supplementary Figure 9.1**). Children with a comorbidity of epilepsy had consistently higher odds of inpatient admissions (OR 2.55; 95%CI 1.85-3.52), (23, 27) outpatient (public) paediatric medical specialist appointments (OR 2.74; 95%CI 1.51-4.96)(30) and emergency department attendances (OR 2.86; 95%CI 2.27-3.59)(24) (**Table 2.3**). Children with a gastrostomy, (23, 24, 30) and intellectual (18, 26) and/or additional developmental disabilities(28) (e.g. autism, attention-deficit disorder) also had increased likelihood of health service use across settings (**Table 2.2**, **Table 2.3**).

2.4.1.5 Contextual Factors

Few studies examined the impact of contextual factors on health service utilisation (**Table 2.2**). Some regions of residence, larger hospital and better provision of continuity of care were reported to be associated with reduced inpatient admission rates in one Taiwanese study;(27) and regional versus metropolitan location was associated with increased likelihood of private outpatient use in one Australian study.(30)

2.4.2 Respiratory health service utilisation

Diseases of the respiratory system were identified as a leading cause of inpatient admissions across age ranges in multiple settings.(23, 25, 36) Determinants of respiratory inpatient admissions and emergency department attendances was the focus of one study (19, 20) based in Western Australia which included some determinants (n=8) not examined in other studies (frequent respiratory symptoms; smoking; scoliosis; oropharyngeal dysphagia; respiratory symptoms with meals; snoring; two or more courses of antibiotics in previous year; previous hospitalisation). This study found respiratory admissions over five years were associated with gross motor severity and similar comorbidities (e.g., epilepsy, gastro-oesophageal reflux disease) as (general) inpatient admissions and emergency department attendances. A higher rate of hospital admission was reported for GMFCS level V (compared with GMFCS level I) (Incident Rate Ratio (IRR) 80.0; 95%CI 28.4-192.5), oropharyngeal dysphagia (IRR 33.6; 95% CI 17.7-63.7) and epilepsy (IRR 25.6; 95% CI 13.3-49.4). Previous respiratory hospital admission

was also associated with higher rates of inpatient admission (IRR 29.4; 95% CI 11.2-77.5) and emergency department attendance (IRR 11.8; 95% CI 5.6-24.7).

2.5 Discussion

During the last decade there has been an increase in research examining health service utilisation in people with CP. To our knowledge, this is the first systematic review to examine the factors that influence rates of health service utilisation in this population. Our main findings were evidence of associations between CP severity, comorbidities and age with health service utilisation across settings. We found health service utilisation across inpatient, outpatient and emergency department settings to be increased in children with CP with more severe gross motor function limitations (GMFCS levels IV-V) and with associated comorbidities including intellectual disability, epilepsy and gastrostomy-use. We also found limited research investigating and reporting on socio-economic "individual enabling" factors and health system "contextual" determinants.

These findings are perhaps not surprising given that the management of comorbidities is a main focus of health services and the known association between CP severity and comorbidities.(2) They do, however, highlight the important role that outpatient health services play in the preventive management of complications and associated conditions

of severe CP. Furthermore, they emphasise the necessity of ensuring equitable access to services to minimise unplanned health care use(6) and poor health outcomes.

We found evidence that age across the lifespan influences health service utilisation for people with CP. Studies in our review suggested inpatient admissions reduce with age during childhood and adulthood, and outpatient health service utilisations changes in the rates and types of outpatient health services attended between children and adults. The reasons for this are likely to involve both individual and contextual factors, including differences between how paediatric and adult health systems are structured. This may be explained, at least in part, by differences in the goals of the paediatric and adult systems, with the paediatric health system guiding the developing child to optimal physical functioning and participation, compared with the adult health system focus on treating existing health conditions. Similar findings have been described in health service utilisation research exploring CP subtypes and severities .(37-40) Most children with CP now live into adulthood,(41) develop additional health problems as adults,(3, 42) and many report their health needs are not met during this time.(40, 43) There is a need for the health system to adapt to meet this need, with particular emphasis around the time of transition from paediatric to adult health services to ensure that young people with CP do not fall out of the system(39) and greater capacity for adults with CP throughout the lifespan. More research is needed to improve understanding of how health service utilisation changes as adults with CP grow older, and to support any changes to the health system.

We found less research that has explored the impact of individual enabling factors, such as socio-economic status, on health service utilisation, other than one study suggesting an association between indicators of socio-economic status with (self-reported) private outpatient service attendance.(30) Socio-economic disadvantage has been shown to be associated with both CP severity (non-ambulant status, moderate intellectual disability or greater and severe comorbidities)(44) and decreased used of health services in other childhood populations.(45, 46) Research in broader disability groups have supported inequities of access to health service based around income.(47, 48) Children who rely on public funding (Medicaid and Children's Health Insurance Program) may be denied access to some outpatient services or wait longer for treatment.(49) A greater understanding of the socio-economic factors that influence health service access in CP, and the consequences of this can support improvements in health service and support advocacy for structural reforms for funding of children's healthcare.(50) Studies have shown that simple measures such as continuity of care,(27) integration of clinical care and research data collection,(51) and telehealth services (52) can all improve access and/or health outcomes for children with CP.

Understanding the health service utilisation literature and applying its findings is challenging given the complexity of the data. This review draws attention to the influence that CP severity and comorbidities have on health service use and highlights the need for further research about predisposing and enabling factors that investigate equity of access.

2.5.1 Study Limitations

While the quality of the included studies was generally high, our review was limited in its restriction to high income countries in that differences in the structure and funding of health systems between countries may impact the generalisability of some of the findings. For example, in countries with 'user pay' or less universal or established health systems, availability and access to health care for people with CP may be reduced. Our eligibility criteria also excluded studies that focused on particular subgroups of the CP population that would be relevant when considering health service utilisation for that subgroup. After protocol amendment, we excluded studies that only reported therapy service utilisation; this type of health service provides an important source of support for children with CP; further research that explores determinants of therapy service utilisation would be valuable. Some included studies also used self-reported health service use or survey design that may lead to recall bias;(53) however, given this was only a minority of studies, we believe our findings are less likely to be affected by this. We have not reported all measures of health service use described in the studies we have reviewed such as inpatient admission duration (18, 22, 23, 25, 33, 34) and subtypes of inpatient admission (e.g. emergency, elective).(23, 26) These may also be important measures of health service utilisation to consider.

2.5.2 Conclusions

Understanding the determinants that influence health service utilisation is an important first step towards improving equity of access for people with CP. We identified evidence that health service utilisation is associated with age and "individual need", but less evidence to support how "individual predisposing" or "individual enabling" factors such as socio-economic status might influence access. Improving this understanding should be a research priority, to guide the development of new models of care that aim to provide equitable access to the increasing number of interventions aimed at improving the health and wellbeing of people living with CP.

2.6 Chapter Synopsis

Chapter 2 identified research that reported differences in health service utilisation in people with CP related to age, CP severity and comorbidities. One study reported a reduction in inpatient admissions related to continuity of care. There were limited studies that reported how the social determinants of health impact health service access in CP. The remaining studies in this thesis aim to explore this research gap. The review was published in *Archives of Physical Medicine and Rehabilitation*. **Chapter 3** will discuss the research methodologies used in the remaining studies.

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Chapter 3 Methods

3.1 Introduction

To meet the thesis aim and objectives, studies included in this thesis use data from the New South Wales (NSW) / Australian Capital Territory (ACT) Cerebral Palsy Register (NSW/ACT CPR), administrative health data and other datasets. This chapter outlines the setting; data sources used; provides an introduction to data linkage research and considerations; introduces the main measures of socioeconomic disadvantage and residential remoteness used in this thesis; outlines the structure and ethics approvals for the component studies and describes how variables from the NSW/ACT CPR were transformed for the component studies presented in this thesis. The concept map for the thesis is shown in **Figure 3.1**.

Figure 3.1. Thesis Concept Map for Chapters 3 and 4

	Knowledge Gap		Objective	Chapter	Publication	Key Findings
Introduction			Background and literature review	Chapter 1		
	What is already known about health service access in CP?	I	To systematically review the existing literature regarding determinants of hospital-based health service access among people with CP.	Chapter 2	Determinants of Hospital-Based Health Service Utilisation in CP: a Systematic Review. *doi:10.1016/j.apmr.2021.12.003*	• Hospital health service utilisation associated with age, severity and comorbidities.
Study Methods 			Introduce the NSW/ACT CP Register, administrative data sets and data linkage.	**Chapter 3**		
	How do study populations differ between a CP population register and hospital admission data?	II	To compare the sociodemographic and clinical characteristics of children with CP identified from a CP population register and hospital admission data.	**Chapter 4**	A comparison of cohorts of children with CP from a population register and hospital admission data. *doi:10.1111/ppe.13024*	• Hospital admission data has sensitivity of 0.7 for a diagnosis of CP. • Children with CP from hospital admission data are older, live in major cities, more comorbidities and early death.
Dimensions of outpatient health service access and CP	What factors influence non-attendance at specialty outpatient clinics?	III	To explore the factors associated with non-attendance by children with CP at specialty outpatient clinics.	Chapter 5	Non-attendance at outpatient clinic appointments by children with CP. *doi:10.1111/dmcn.15197*	• Non-attendance associated with older age, socioeconomic disadvantage and previous non-attendance.
	How does telemedicine influence equity of access to outpatient care?	IV	To determine how telemedicine supports equity of access to specialist outpatient care for children with CP.	Chapter 6	Telemedicine for children with CP before, during and after the COVID-19 pandemic. *Under review.*	• Telemedicine use lower in children living in regional and remote areas.
	How have telemedicine rates have changed since the COVID-19 pandemic?	V	To determine the impact of the COVID-19 pandemic on telemedicine use in children with CP.			• Telemedicine use increased substantially during COVID-19 pandemic and since has declined to near baseline.
	What factors influence access to outpatient services?	VI	To determine how clinical and sociodemographic determinants influence hospital outpatient service access in CP.	Chapter 7	Outpatient encounters, continuity of care and unplanned hospital care for children and young people with CP. *doi:10.1111/dmcn.15800*	• Outpatient service use associated with metropolitan areas, GMFCS IV-V, epilepsy, intellectual disability.
	Are CP outpatient services accessible for Aboriginal and/or Torres Strait Islander children?	VII	To determine hospital outpatient service utilisation for Aboriginal and/or Torres Strait Islander children and young people with CP.			• Rates of outpatient service use for Aboriginal and/or Torres Strait Islander children are similar to non-indigenous population.
	How does outpatient continuity of care impact unplanned hospital care?	VIII	To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.			• Decreased continuity of care is associated with increased unplanned hospital care.
Summary and Conclusions			Summary of key findings and implications for clinical practice, policy and research.	Chapter 8		

3.2 Setting

The component studies in this thesis were conducted in New South Wales, Australia (**Figure 3.2**). New South Wales (NSW) is Australia's most populous state, with a population of approximately 8.1 million (30 June 2022) and land area of over 800,000 square kilometres.(1) Sydney, the capital city, is located on the eastern coast of NSW. Two thirds of the NSW population live in the Greater Sydney.(1)

Figure 3.2. New South Wales

Source: NSW Government Spatial Collaboration Portal (https://portal.spatial.nsw.gov.au/portal/apps/sites/#/homepage) (accessed: 2024-09-09).

3.3 Data sources

3.3.1 New South Wales / Australian Capital Territory Cerebral Palsy Register

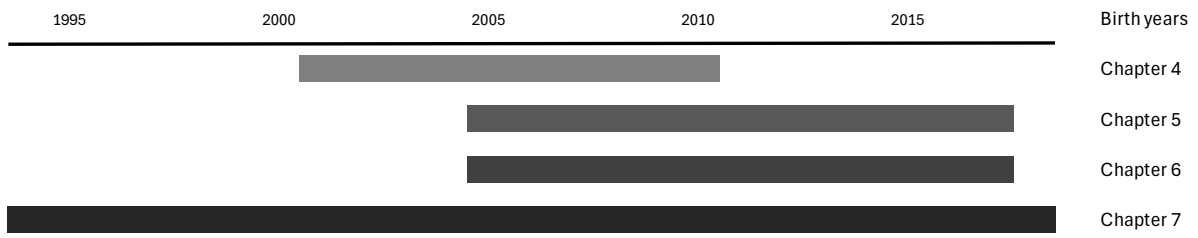
The New South Wales (NSW)/ Australian Capital Territory (ACT) Cerebral Palsy (CP) Register (NSW/ACT CPR) is a population-based register with multiple ascertainment strategies for individuals with CP who were born or live in NSW or ACT. The NSW/ACT CPR has collected data for these children since (birth cohort) 1993.(2) Families are given the option to opt out of the NSW/ACT CPR, and for those that do not opt out, a minimum data set of sociodemographic, aetiological, and clinical data are collected and used for reporting and research purposes. Data are collected by trained NSW/ACT CPR staff in a standardised way when children are first clinically diagnosed. Cases are verified and clinical information obtained from multiple sources, including tertiary children's hospitals, community-based services, and through cross reference to health professionals and medical records. A second data collection is performed for each participant at/after the age of five years to confirm diagnosis and comorbidities and where the diagnosis is still being questioned by treating health professionals, NSW/ACT CPR staff "keep following" until a definitive decision is made.

3.3.1.1 CP cohorts

For studies in this thesis, I used different subsets of the NSW/ACT CPR to define birth cohorts of children with CP: birth years 2001 to 2010 for **Chapter 4** (comparison of

populations between NSW/ACT CPR and hospital admission data); birth years 2005-2017 for **Chapters 5** (non-attendance to outpatient clinics) and **Chapter 6** (telemedicine); and birth years 1994 to 2018 for **Chapter 7** (outpatient continuity of care and unplanned hospital care) (**Figure 3.3**).

Figure 3.3. Birth years defining cohorts of children and young people with CP for studies included in this thesis.



3.3.2 Administrative Health Data

Administrative health data are data that are routinely generated during contact with the health system.(3) Data are typically collected by government bodies, primarily for the purposes of funding and/or service planning. Common information included in administrative health data include personal details (name, address, sex, date of birth), service events (e.g., type, date, location) and financial data (e.g. Australian Refined Diagnosis Related Groups, National Weighted Acivity Units). Some health administrative data sources include clinical / diagnostic information (e.g., diagnoses coded using the

International Classification of Diseases systems) and information on procedures (e.g., Australian Classification of Health Interventions) relevant to the episode of care.

Use of administrative health data have been identified as an attractive data source for use in research for several reasons:(4)

(i) less time-consuming and can be more cost-effective than collection of similar data (again) for research purposes.(5)

(ii) include broad and clinically meaningful patient populations therefore reducing risk of participation bias (e.g., recruitment to new research studies), and recall bias (e.g., if using different methodologies such as surveys).

(iii) population coverage means they are often able to capture infrequent outcomes that might be missed without access to a total population.

(iv) are often available over a prolonged time period, enabling analysis of trends over time.

(v) may contain a diversity of data than enables a broad range of research questions.

However, as administrative health data are collected primarily for non-research purposes (e.g., to support the administration of health programs) they are vulnerable to reduced quality control and missing data when compared with research registries.

In NSW, the privacy of administrative health data is safeguarded by two laws: the NSW Privacy and Personal Information Protection (PPIP) Act 1998 and the NSW Health Records and Information Privacy (HRIP) Act 2002.(6) However, administrative health data may be used for research where deidentified, or where exemption is sought and obtained for data linkage research (e.g., following the Statutory Guidelines on Research from the Information and Privacy Commission of NSW)(7) and appropriate Human Research Ethics Committee approval.

This thesis uses the following NSW administrative health data sources:

3.3.2.1 NSW Non-Admitted Patient (NAP) Data Collection

The Non-Admitted Patient (NAP) data collection is maintained by the New South Wales (NSW) Ministry of Health and is available for research from July 2015. The NAP data collection includes patient-level data for all non-admitted patient services with clinical and/or therapeutic content of sufficient significance to warrant a note being made in the patient's medical record from NSW Health facilities.(8) Variables included in the NAP data collection include dates of episode, facility identifiers, service contact mode (e.g., in person, telephone, videoconference, email, no client contact), (Tier 2) clinical group (e.g., diagnostic, procedural, medical, allied health / nursing) and class (i.e. specialty).(8) **Chapters 5 and 6** use NAP data specific to the Sydney Children's Hospitals Network (SCHN) (comprising the two specialised children's hospitals of Sydney, the Children's

Hospital at Westmead and Sydney Children's Hospital Randwick); **Chapters 4 and 7** use NAP data from all NSW Health facilities.

3.3.2.2 NSW Admitted Patient Data Collection (APDC)

The Admitted Patient Data Collection (APDC) is maintained by the NSW Ministry of Health and is available for research from July 2001. The APDC includes all inpatient admissions in NSW public and private hospitals.(8) Variables include start and end dates of the care episode, area and facility identifiers and mode of separation (discharges, transfers and deaths). In addition, a principal and up to 50 additional diagnoses (based on the International Classification of Diseases Australian Modification (ICD10-AM)) and a principal and up to 50 additional procedures based on the Australian Classification of Health Interventions (ACHI) are coded for each episode.(8)

3.3.2.3 NSW Emergency Department Data Collection (EDDC)

The Emergency Department Data Collection (EDDC) is maintained by the NSW Ministry of Health and is available for research from July 2005. The EDDC includes data related to presentations to and activity in Emergency Departments (EDs) in public hospitals in NSW.(8) Variables include facility identifiers, arrival and departure dates, triage category and mode of separation (including admission to hospital).(8)

3.3.2.4 NSW Perinatal Data Collection (PDC)

The NSW Perinatal Data Collection (PDC) is maintained by the NSW Ministry of Health Centre for Epidemiology and Evidence and is available for research from January 1994. The PDC includes data of all births (live births and stillbirths) of at least 20 weeks gestation or at least 400 grams birthweight occurring in NSW public and private hospitals and homebirths. Variables include (baby's) date of birth, birthweight, plurality, gestation and sex, and maternal variables including demographics and details of pregnancy, labour and birth.(8)

3.3.2.5 NSW Family and Community Services (FACS) Disability Dataset

The FACS dataset includes basic diagnostic information from all publicly funded providers of disability services including case management, therapy and early intervention services in NSW. Variables include the individual's first date of service and primary and other significant disability type(s) selected from twelve broad disability groupings. FACS data were available from July 2003 until December 2017 at which time these services transitioned to the National Disability Insurance Scheme (NDIS).

3.3.2.6 NSW Registry of Births, Deaths and Marriages (BDM) Death Registrations

The NSW Registry of Births, Deaths and Marriages (BDM) Death Registrations is maintained by the NSW Ministry of Health Centre for Epidemiology and Evidence and is

available for research from January 1985. The BDM dataset records date of death for all deaths occurring in NSW. Deaths of NSW residents who die interstate are not available for linkage. A secondary dataset includes primary and contributing causes of death.(8)

3.4 Data linkage research

Data linkage has been defined as "the bringing together from two or more different sources, data that relate to the same individual, family, place or event".(5) Data linkage enables the development of rich and novel datasets that support answering of research questions that would not be answerable in any single data set. Australia has developed strong systems to enable data linkage research whilst preserving privacy.(9) The amount of data-linkage research (e.g., cancer, circulatory diseases, pregnancy etc) in Australia has increased substantially since 2010,(9) and as more data sources become available (e.g., genomic data, wearable sensors), data linkage techniques will be increasingly important to integrate these data sources into research.(10)

In NSW, the development of The Centre for Health Record Linkage (CHeReL) in 2006 has greatly supported the conduct of data linkage research in NSW and ACT through providing a mechanism for linkage of health-related data which meets ethical, legal and privacy and confidentiality requirements.(6) CHeReL is managed and funded by the NSW

Ministry of Health. Using linked data for research requires data custodian and Human Research Ethics Committee (HREC) approval.

Risks related to privacy preservation in data linkage research are greatly minimised by the process of separation. Separation (of linking data and data analysis) ensures that none of the three main stakeholder groups - data custodians (responsible for the datasets), researchers, and data linkage units ever see or hold the combined data that includes both personally identifiable demographic data and research data of interest (e.g., clinical data, health service utilisation data). Key stages of data linkage involve (i) data custodians providing the data linkage unit with personal identifiers (e.g., name, sex, address, date of birth, other identifiers) (ii) the data linkage unit identifies individuals across datasets (see below), and applies a unique project number to each individual (iii) researchers are supplied with datasets containing the unique project number in place of personally identifiable information and relevant clinical and/or health administrative data.(11)

The process of record linkage can be performed using one of two techniques:

Probabilistic linkage techniques which match identifying information (e.g., name, date of birth, address) across records using a range of matching algorithms to find common records. These overcome issues related to human error in data entry, incomplete or missing data. As such, probabilistic techniques may be more suitable for administrative health data in accommodating errors and data that may change over time (e.g.,

addresses).(12) CHeReL uses ChoiceMaker software for probabilistic linkage which uses an automated blocking algorithm and machine learning technique for 'scoring' or assigning weights to common records.(13) Blocking strategies make linkage more manageable by restricting comparison of records to those that are likely matches, reducing the clerical checks required.(12)

Deterministic linkage involves linkage where there is exact agreement on a unique identifier (e.g., a social security number) or on each element of a collection of identifiers. (12) Deterministic linkage reduce false matches, and can be useful when identifiers are stable and free of errors but can result in increased missed linkages.(14) This is not possible in Australia as there are not unique identifiers that are used consistently across multiple administrative sources.

3.5 Measuring the social determinants of health

3.5.1 Socioeconomic disadvantage

In this thesis, socioeconomic disadvantage is measured using the Index of Relative Socioeconomic Disadvantage (IRSD). IRSD is one of four measures collected in the Socio-Economic Indexes for Areas (SEIFA), constructed by the Australian Bureau of Statistics (ABS). (15) The SEIFA indexes are constructed using data from the Australian Census of Population and Housing, a five yearly survey of all households in Australia (e.g.,

in the 2016 census this included approximately 10 million households of more than 25 million people).(16) The SEIFA indexes define socioeconomic disadvantage as people's access to material and social resources, and their ability to participate in society. (15) The IRSD uses variables from the Census that are indicators of socioeconomic disadvantage (**Table 3.1**).

Like the other SEIFA indexes, IRSD is reported as an area-based index, indicating the collective socioeconomic characteristics of the people living in a particular area (and not at an individual level). SEIFA reports indexes down to Statistical Area Level 1 (SA1) geographical area levels. SA1s generally have an average population of 400 people. As the index scores derived use an arbitrary numerical scale, SEIFA indexes are typically reported using index rankings of areas (quintiles in this thesis, also deciles), for IRSD ranking areas from the most disadvantaged (quintile 1) to the least disadvantaged (quintile 5).

The IRSD is calculated as a weighted sum of these variables using principal component analysis to reduce (correlated) original variables into a set of principal components that approximate the variance of the original variables.(17)

Table 3.1. Indicators of disadvantage used in the Index of Relative Socioeconomic Disadvantage.

Variable name	Variable description
INC_LOW	Per cent of people with stated household equivalised income between $1 and $25,999 per year
CHILDJOBLESS	Per cent of families with children under 15 years of age who live with jobless parents
NOYEAR12ORHIGHER	Per cent of people aged 15 years and over whose highest level of education is Year 11 or lower (Includes Certificate I and II)
LOWRENT	Per cent of occupied private dwellings paying rent less than $250 per week (excluding $0 per week)
UNEMPLOYED	Per cent of people (in the labour force) who are unemployed
OCC_LABOUR	Per cent of employed people classified as Labourers
DISABILITYU70	Per cent of people aged under 70 who need assistance with core activities due to a long–term health condition, disability, or old age
ONEPARENT	Per cent of one parent families with dependent offspring only
OVERCROWD	Per cent of occupied private dwellings requiring one or more extra bedrooms.
OCC_DRIVERS	Per cent of employed people classified as Machinery Operators and Drivers
SEPDIVORCED	Per cent of people aged 15 years and over who are separated or divorced
NOEDU	Per cent of people aged 15 years and over who have no educational attainment
OCC_SERVICE_L	Per cent of employed people classified as low skill (skill level 4 and 5) Community and Personal Service workers
NOCAR	Per cent of occupied private dwellings with no cars
ENGLISHPOOR	Per cent of people who do not speak English well

Source: Australian Bureau of Statistics, Socio-Economic Indexes for Areas (SEIFA), Australia methodology 2021

While SEIFA indexes have been criticised for under-representing heterogeneity of populations, particularly when used for larger geographical areas (e.g., Local Government Areas),(18) they are widely used in (primary, specialist, hospital) health

service access research in Australia.(19) They therefore represent an appropriate measure for use in answering the objectives of this thesis.

3.5.2 Residential Remoteness

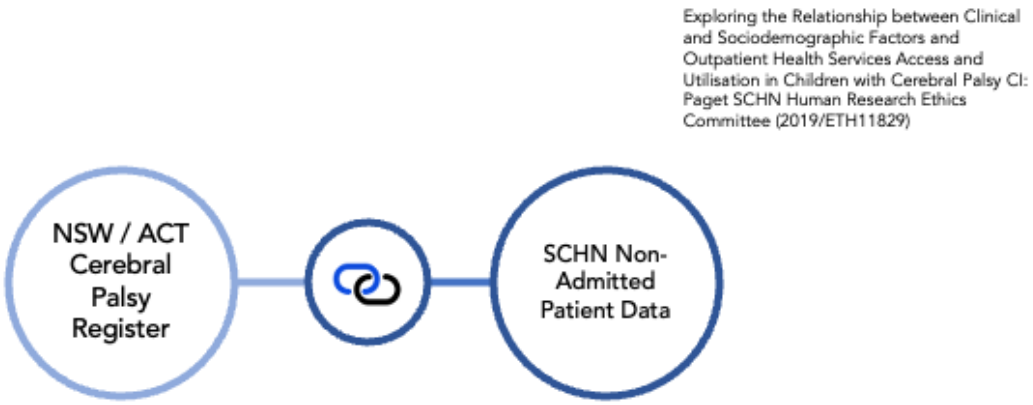
As described in **Chapter 1**, more than one quarter of people in Australia live outside major cities in regional and remote areas and these people are recognised to have barriers to accessing health care.(20) In this thesis, residential remoteness is measured using the Australian Statistical Geography Standard (ASGS) Remoteness Structure.(21) Remoteness is defined as relative geographic access to services. It is measured using the Accessibility/Remoteness Index of Australia (ARIA+) which is an index ranging from 0 (high accessibility) to 15 (high remoteness) derived by measuring distance via the road network to five levels of 'Service Centres' i.e., populations of increasing size.(22)

The ASGS remoteness structure classifies areas in five classes of relative geographical remoteness: Major Cities of Australia, Inner Regional Australia, Outer Regional Australia, Remote Australia, Very Remote Australia.

3.6 Details of studies using linked data in this thesis

Studies in this thesis used different linked datasets to conduct research and analyses. Two primary research studies (**Chapter 5, 6**) linked the NSW/ACT CPR to non-admitted patient datasets from the Sydney Children's Hospitals Network (SCHN) (comprising the Children's Hospital at Westmead and Sydney Children's Hospital, Randwick) (**Figure 3.4**). For these studies, ethics approval was obtained through Sydney Children's Hospitals Network Human Research Ethics Committee as "Exploring the Relationship between Clinical and Sociodemographic Factors and Outpatient Health Services Access and Utilisation in Children with Cerebral Palsy" (2019/ETH11829), with additional Governance approval through the Cerebral Palsy Alliance (2019_08_04). Formal data linkage processes were not used for these projects. Nonetheless, the process of using personal identifiers to identify children and young people with CP from the NSW/ACT CPR in SCHN NAP data was performed separately from data analysis, and at no time did researchers have access to personal identifiers for individuals included. Matching was performed by the SCHN data custodian, using fuzzy string matching in Python, using date of birth, full name and address.(23)

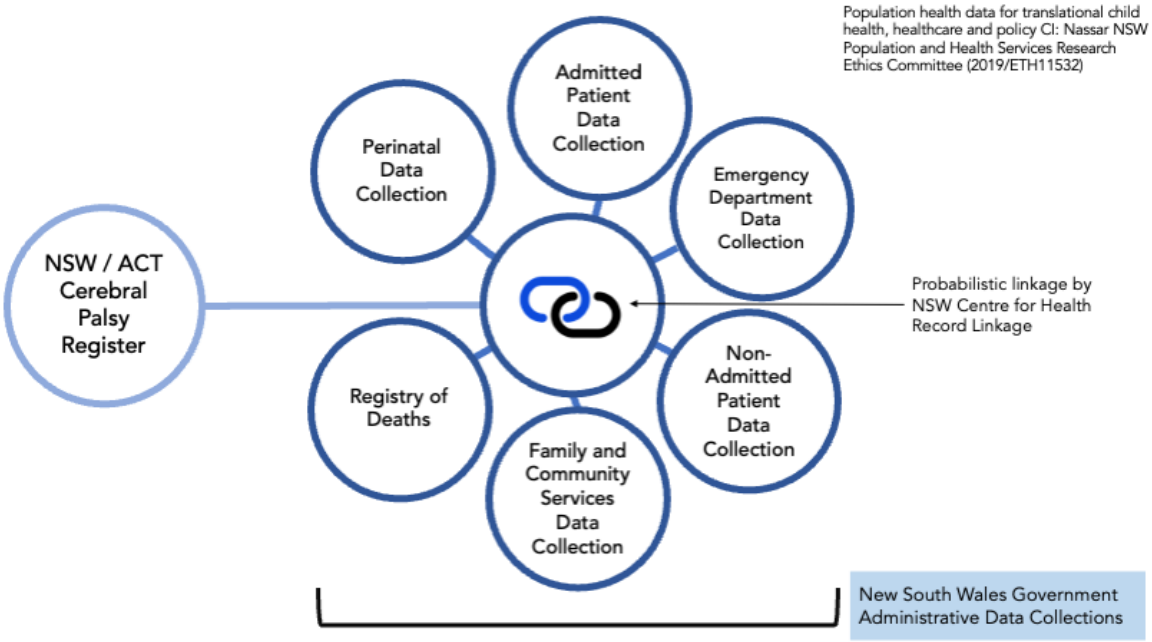
Figure 3.4. Data sources in studies described in Chapters 5,6. Completed under approval from Sydney Children's Hospitals Network Human Research Ethics committee (2019/ETH11829).



ACT Australian Capital Territory; NSW New South Wales; SCHN Sydney Children's Hospital's Network

A further two primary research studies included in this thesis (**Chapter 3**, **Chapter 7**) were undertaken using data from a large data linkage project "Population health data for translational child health, healthcare and policy" that linked the NSW/ACT CPR to multiple administrative health datasets (**Figure 3.5**). These studies received ethics approval as amendments to ethics agreement from the NSW Population and Health Services Research Ethics Committee (2019/ETH11532).

Figure 3.5. Data sources linked for studies described in Chapters 3, 7. Completed under agreement from the NSW Population and Health Services Research Ethics Committee (2019/ETH11532).



ACT Australian Capital Territory; NSW New South Wales

3.7 Data variables from the NSW/ACT CPR

For the purposes of the studies presented in this thesis (**Chapters 4-7**) major variables presented in the NSW/ACT CPR were categorised for ease of understanding. A summary of these variables is shown in **Table 3.2**.

Table 3.2. Categorisation of major variables from the New South Wales / Australian Capital Territory Cerebral Palsy Register used for studies in this thesis, including suggestions of combining variables for future data linkage research.

Variable in NSW/ACT CPR	Name in NSW/ACT CPR	Description	Codes in NSW/ACT CPR	Codes used for this thesis	Notes
Gestational Age at birth	GestationalAge	Gestational age at birth in completed weeks.	Numerical	Premature (<37 weeks)	
				Term (>=37 weeks)	
Maternal Country of Birth	Mth:BirthCountry	Based on Standard Australian Classification of Countries (SACC) (ABS 1269.0 1998).	Text based on SACC codes	Australia	Consider use of World Bank Classification by income level *.
				Overseas	
Motor type	InitialCPType	Combination of topography and predominant motor disorder at first entry in NSW/ACT CPR.	Left spastic hemiplegia/monoplegia	Spastic Unilateral	
			Right spastic hemiplegia/monoplegia	Spastic Unilateral	
			Monoplegia / Hemiplegia unspecified	Spastic Unilateral	
			Spastic Diplegia	Spastic Bilateral	
			Spastic Triplegia	Spastic Bilateral	Depending on research question, may want to combine Tri/Quad and keep Diplegia separated.
			Spastic Quadriplegia	Spastic Bilateral	
			Dyskinesia - mainly Athetosis	Dyskinetic	May also want to separate unilateral and bilateral dyskinesia.
			Dyskinesia - mainly Dystonia	Dyskinetic	

Variable in NSW/ACT CPR	Name in NSW/ACT CPR	Description	Codes in NSW/ACT CPR	Codes used for this thesis	Notes
			Ataxia	Ataxia	
			Hypotonia	Hypotonia	Depending on research question, may combine ataxia/hypotonia.
			Under Age 5 / 'At risk of CP'	Early at risk CP	If in this category the CP motor type is unclear at initial diagnosis.
			Unknown	Unknown	This term is used, when CP is confirmed, but no reference to motor type.
Motor type	Age5CPType	Combination of topography and predominant motor disorder at or near age 5 years.	Left spastic hemiplegia/monoplegia	Spastic Unilateral	Use in preference to initialcptype where available due to recency and stability in phenotype.
			Right spastic hemiplegia/monoplegia	Spastic Unilateral	
			Monoplegia / Hemiplegia unspecified	Spastic Unilateral	
			Spastic Diplegia	Spastic Bilateral	
			Spastic Triplegia	Spastic Bilateral	Depending on research question, may want to combine Tri/Quad and keep Diplegia separated.
			Spastic Quadriplegia	Spastic Bilateral	
			Dyskinesia - mainly Athetosis	Dyskinetic	May also want to separate unilateral and bilateral dyskinesia.
			Dyskinesia - mainly Dystonia	Dyskinetic	

Variable in NSW/ACT CPR	Name in NSW/ACT CPR	Description	Codes in NSW/ACT CPR	Codes used for this thesis	Notes
			Ataxia	Ataxia	
			Hypotonia	Hypotonia	Depending on research question, may combine ataxia/hypotonia.
			Under Age 5 / 'At risk of CP'	Early at risk CP	If in this category the CP motor type is unclear at initial diagnosis.
			Unknown	Unknown	This term is used, when CP is confirmed, but no reference to motor type.
Gross Motor Function Classification System	InitialCPSeverity	Gross Motor Function Classification System (GMFCS) classification at first entry in NSW/ACT CPR.	GMFCS - Level I	GMFCS I	Often collapsed to two groups: GMFCS I-III (i.e., ambulant, including with aid), GMFCS IV-V (i.e., mobility dependent on wheelchair).
			GMFCS - Level II	GMFCS II	
			GMFCS - Level III	GMFCS III	
			GMFCS - Level IV	GMFCS IV	
			GMFCS - Level V	GMFCS V	
			Unknown	Unknown	
Gross Motor Function Classification System	Age5CPSeverity	Gross Motor Function Classification System (GMFCS) classification at or near age 5 years.	GMFCS - Level I	GMFCS I	Use in preference to InitialCPSeverity where available due to recency and stability in phenotype. Often collapsed to two groups: GMFCS I-III, GMFCS IV-V.
			GMFCS - Level II	GMFCS II	
			GMFCS - Level III	GMFCS III	
			GMFCS - Level IV	GMFCS IV	
			GMFCS - Level V	GMFCS V	

Variable in NSW/ACT CPR	Name in NSW/ACT CPR	Description	Codes in NSW/ACT CPR	Codes used for this thesis	Notes
			Unknown	Unknown	
Epilepsy	Epilepsy	Presence of epilepsy at time of assessment.	Yes	Yes	
			No	None or resolved by age 5	
			Resolved by age 5		
			Unknown	Unknown	
Intellectual impairment	IntellectualImpairment	Presence of intellectual impairment at age 5 years.	No impairment	None	
			Probably no impairment	None	
			Probably some impairment	Yes	Also appropriate to combine Probably some and mild depending on research question.
			Mild (IQ50-69)	Yes	
			Moderate (IQ 35-49)	Yes	Also appropriate to combine moderate and severe depending on research question.
			Severe (IQ <35)	Yes	
			Unknown	Unknown	

ACT Australian Capital Territory; CPR Cerebral Palsy Register; GMFCS Gross Motor Function Classification System; IQ Intelligence Quotient; NSW New South Wales

* World Bank Classification available at https://datahelpdesk.worldbank.org/knowledgebase/articles/378834-how-does-the-world-bank-classify-countries [Accessed 22 August 2024].

3.8 Chapter Synopsis

In **Chapter 3**, the primary data sources used for the component studies in this thesis were introduced, data linkage methods were introduced, and measures of socioeconomic disadvantage and residential remoteness were explained. In **Chapter 4**, a strength of data linkage research is illustrated by comparing sociodemographic and clinical characteristics of cohorts of children with CP identified from administrative health data (hospital admission data) and the NSW/ACT CP Register.

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Chapter 4 Comparison of cohorts of children with cerebral palsy from a population register and hospital admission data: a data linkage study.

4.1 Published Manuscript

This chapter has been reformatted for publication as part of this thesis.

The citation for the published manuscript is:

> Paget SP, McIntyre S, Lain S, Goldsmith S, Nassar N. A comparison of cohorts of children with cerebral palsy from a population register and hospital admission data: A data linkage study. Paediatr Perinat Epidemiol. 2024;38(1):22-30. doi:10.1111/ppe.13024. https://doi.org/10.1111/ppe.13024

A copy of the published article as well as a statement of the specific contributions of the co-authors can be found in **Chapter 10 - Appendix B**.

4.2 Introduction

Cerebral palsy (CP) is a common childhood health condition and a leading cause of physical disability. CP represents an 'umbrella diagnosis' under which a broad range of aetiological pathways result in disruption of early brain development, and cause disorders of movement and posture that can differ in phenomenology, topography, and functional impact.(1) There are currently no specific treatments that target the underlying brain lesion that causes CP, and the condition is lifelong, with substantial health, social and economic impact, both at an individual and societal level.(2) No biomarkers exist to support the diagnosis of CP, and there are barriers to early diagnosis, with 50% cases diagnosed after 1 year of age and a false positive diagnosis rate of up to 5%.(3)

Epidemiological research has an important role in improving understanding of CP, including reporting trends in prevalence, aetiology, health outcomes and health care utilisation. Recent CP epidemiological research has tended to use one of two data sources to define CP populations. Firstly, an increasing number of countries and jurisdictions have developed CP population registers that collect (sociodemographic and clinical) data from people with CP.(4) Research driven by CP registers has been central in documenting the recent decline in birth prevalence of cerebral palsy in high income countries(5), and improved life expectancy.(6) Secondly, other research studies have used administrative health data i.e., data that are routinely generated during patient

encounters with the health system, often for financial or clinical purposes.(7) Types of administrative health data have included hospital admission data,(8-14) outpatient visits(8, 12-14) and insurance claims(10, 15) and research using this approach has described prevalence,(10, 15) patterns of health service utilisation(9, 11) and comorbidities.(14, 16) Administrative health data are an attractive data source for researchers where CP registers are not available,(10) as they can often be used to identify large cohorts in relatively timely and cost-effective ways, and with little loss to follow-up. However, because the data have been collected for administrative/financial purposes, there is a greater need to assess their validity for research.(17) In addition, administrative health data mostly do not include clinical descriptions (e.g., predominant motor disorder, topography, comorbidities) that are typically available in CP registers. People with CP are often identified in administrative health data using case definitions based on (individual or patterns of) International Classification of Diseases (ICD-9, ICD-10) codes for CP. Published studies have used a number of different case definitions; (8-15) the impact of chosen case definition on clinical differences in the population studied is unknown.

Studies that have compared CP registers and administrative health data have suggested that administrative health data is only moderately sensitive to a diagnosis of CP, and less sensitive in those born term or with hemiplegia (potentially influenced by less routine follow-up and/or lesser severity).(12) A Norwegian study found administrative health data to misdiagnose CP in 14% of cases.(18) We hypothesized that there are clinically relevant differences between populations of children with CP derived from administrative health

data and CP registers, and that these differences may influence research findings. The aims of this study were to compare the sociodemographic and clinical characteristics of cohorts of children with CP identified from administrative health data (hospital admission data) and a CP register and to determine differences between cohorts for a key health outcome (mortality).

4.3 Methods

4.3.1 Study population and data sources

We conducted this study in New South Wales (NSW), Australia's most populous state with an estimated population of 8.3 million inhabitants, using data from NSW and from neighbouring Australian Capital Territory (ACT) (population 460,000; geographically a small region within NSW borders).(19) There are an estimated 37,000 people with CP in Australia,(20) with a rate that is declining, from 2.1/1000 live births in the mid 1990s to a current low of 1.5/1000 live births (from birth year 2016).(21)

Children and young people with a diagnosis of CP were identified from two data sources (i) the New South Wales (NSW)/Australian Capital Territory (ACT) CP Register (NSW/ACT CPR) and/or (ii) the NSW Admitted Patient Data Collection (APDC). Children and young people were included if born between 1st January 2001 and 30th June 2010 to enable a

minimum 10 year follow up in available administrative health datasets. Children were aged between 2 months and 19 years 6 months at study end or time of death.

The NSW/ACT CPR is a population-based register with multiple ascertainment strategies of individuals with CP who were born or live in NSW or ACT. Families are given the option to opt out of the NSW/ACT CPR (we estimate 120 (7%) children in the age range of this study), and for those that do not opt out, a minimum data set of sociodemographic, aetiological, and clinical data are collected and used for reporting and research purposes. Data are collected by trained NSW/ACT CPR staff in a standardised way when children are first clinically diagnosed. Cases are verified and clinical information obtained from multiple sources, including tertiary children's hospitals, community-based services, and through cross reference to health professionals and medical records. A second data collection is performed for each participant at/after the age of five years to confirm diagnosis and comorbidities, and where the diagnosis is still being questioned by treating health professionals, register staff “keep following” until a definitive decision is made. The NSW/ACT CPR is considered to be complete (population-based) for birth years 2001 to 2016. The more recent birth years are generally not complete until five years after birth i.e., the 2018 birth year data will be complete in early 2024. The NSW/ACT CPR was designated as the gold standard for CP diagnosis/description for comparison to health administrative data.

The APDC contains data for all (inpatient, day or overnight) admissions to public and private hospitals in NSW and contains details of the admission, including dates, up to 50 diagnoses classified using the International Classification of Diseases Australian Modification (ICD10-AM) and up to 50 procedures classified using the Australian Classification of Health Interventions (ACHI) (8th edition). Many ACT residents use hospitals in NSW to support their health needs.

Additional clinical data were available for many of these children through data linkage with other datasets. Perinatal data were obtained from the NSW Perinatal Data Collection, which contains data relating to all births in public and private hospitals and homebirths in NSW. Information about developmental disabilities was obtained from the NSW Family and Community Services (FACS) Disability Dataset, which includes basic diagnostic information from all publicly funded providers of disability services (including case management, therapy and early intervention services). Deaths of individuals were identified through the NSW Registry of Births, Deaths and Marriages (BDM) Death Registrations and Cause of Death Unit Record File. Datasets were linked by the New South Wales (NSW) Centre for Health Record Linkage using a probabilistic approach to matching personal identifying information including an automated blocking algorithm and machine learning techniques for assigning weights to different information types.(22)

4.3.2 Comparison of CP cohorts derived from CP Register and APDC

Children and young people with CP in the APDC were identified using hospital discharge dates between 1st January 2001 and 30 June 2020 and applying case definitions based on relevant ICD10-AM code: G80 (**Table 4.2**). We also included a case definition that included other codes: G81 (hemiplegia), G82 (paraplegia and tetraplegia) and G83 (other paralytic symptoms) as this was used in one study identified (**Table 4.2**).(8) Various case definitions were compared based on those identified as used in recent studies (**Supplementary Table 9.3**).(8-15) This included the use of one or more ICD10-AM CP diagnosis codes, codes recorded in specific principal or additional fields, in single or multiple admissions, and with and without age restrictions. Children with only birth admissions were excluded from analysis.

Table 4.1. Coding of cerebral palsy using the International Statistical Classification of Diseases and Related Health Problems, Tenth Revision Australian Modification (ICD10-AM)

G80	Cerebral palsy
G80.0	Spastic cerebral palsy
G80.1	Spastic diplegia
G80.2	Infantile hemiplegia
G80.3	Dyskinetic cerebral palsy
G80.4	Ataxic cerebral palsy
G80.8	Other cerebral palsy
G80.9	Cerebral palsy, unspecified
G81	Hemiplegia
G82	Paraplegia and tetraplegia
G83	Other paralytic syndromes

We compared sociodemographic characteristics of children and young people with CP identified from either the NSW/ACT CPR or from the APDC, focussing on the most common case definition of CP in the literature, one or more admissions with G80 code (henceforth APDC-G80) (**Supplementary Figure 9.2**). Postcode of residence, and statistical areas were used to determine geographical remoteness and socioeconomic disadvantage. Socioeconomic disadvantage was derived from the Index of Relative Socioeconomic Disadvantage (IRSD) and grouped into quintiles (quintile 1 being the most disadvantaged, and quintile 5 the least disadvantaged).(23) Geographical

remoteness (metropolitan; regional; remote areas) was defined using Australian Statistical Geography Standard.(24)

We then compared clinical characteristics of the NSW/ACT CPR and APDC-G80 cohorts using variables obtained via data linkage (**Supplementary Figure 9.2**). Gestation at birth (preterm/term) was available in the NSW/ACT CPR and Perinatal Data Collection. Clinical data (predominant motor type, Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS)) were available from the NSW/ACT CPR. The diagnosis of epilepsy was available in the NSW/ACT CPR and APDC. Epilepsy in the APDC was coded as yes/no and defined as one or more admissions with a ICD10 G40 diagnosis. Gastrostomy use (as a marker of severe difficulties with eating and drinking) was derived from the APDC. Gastrostomy was coded as yes/no and defined as one or more admissions with ACHI codes for gastrostomy insertion or revision (30375-07, 30481-00, 30482-00, 30483-00, 92073-00, 90302-00). Neurodevelopmental disorders (intellectual disability, autism) were derived from the NSW/ACT CPR and/or FACS. Deaths, age at death and underlying cause of death were derived from the BDM. Some clinical characteristics were available in multiple data sources, for these the NSW/ACT CPR was defined as gold standard for epilepsy and intellectual disability and Perinatal Data Collection for gestation at birth.

4.3.3 Statistical analysis

Identification of children with CP was determined from the NSW/ACT CPR and using different case definitions from the APDC. We compared case definitions using the APDC with the NSW/ACT CPR as gold standard using sensitivity and positive predictive value. We compared sociodemographic details of children with CP in the NSW/ACT CPR and APDC-G80 cohorts using standardised mean difference. We then identified clinical characteristics of the same cohorts using data linkage of multiple data sources using standardised mean difference. Differences in data sources were considered when the absolute standardized mean difference was greater than 0.1.(25) Where variables occurred in multiple data sources agreement was tested using Cohen's kappa statistic. To examine the impact of different data sources on outcomes, we compared proportions of deaths between groups, age at death (medians, interquartile ranges) and top three causes of death (based on ICD-10 chapters). We also compared the clinical characteristics of children identified in both NSW/ACT CPR and APDC G80 cohorts with those in NSW/ACT CPR cohort only using standardised mean difference. SAS v9.4 was used to identify individuals with CP and perform analyses and R package tableone to calculate standardised mean difference. Ethics approval for the study was attained from the NSW Population and Health Services Research Ethics Committee (2019/ETH11532).

4.4 Results

A total of 1,598 children and young people with a confirmed diagnosis of CP on the NSW/ACT CPR were identified (42.8% female). Of these, 97.3% (n=1,554) had at least one (non-birth) hospital admission identified in the APDC during the study period. Of the children and young people who had no hospital admissions during the study period (n=44), 52% had spastic unilateral CP (n=23) and 75% were classified GMFCS levels I or II (n=33). All children who were identified as residing in ACT had at least one hospital admission during the study period.

We identified between 917 and 2,439 children and young people with a diagnosis of CP recorded in the APDC using a range of ICD10-AM case definitions (**Table 4.2**). Sensitivity ranged from 0.40 (95% confidence interval (95%CI) 0.37, 0.42) (at least one G80 code before 4 years of age) to 0.74 (95%CI 0.72, 0.76) (at least one G80, G81, G82 or G83 codes). Positive predictive value ranged from 0.47 (95%CI 0.45, 0.49) (at least one G80, G81, G82 or G83 codes) to 0.73 (95%CI 0.70, 0.75) (at least one G80 code, principal diagnosis) (**Table 4.2**).

Comparison of sociodemographic characteristics of children and young people with CP identified in either the NSW/ACT CPR (n=1,598) or APDC with common case definition of

one or more admissions with G80 code (APDC-G80) (n=1,748) is shown in **Table 4.3**. Compared with children identified in the NSW/ACT CPR, there was a higher proportion in the APDC-G80 cohort who were born in the earlier period (2001-2005, 56.2% v 50.5%) and who lived in major cities (65.0% v 54.8%) (SMD >0.1).

Table 4.2. Sensitivity and positive predictive value of a diagnosis of cerebral palsy from the Admitted Patient Data Collection (APDC) compared with the NSW / ACT Cerebral Palsy Register (CPR), using various ICD10-AM diagnostic code case definitions for cerebral palsy.

ICD10-AM Diagnosis code case definition used in APDC	CP diagnosis in CPR and APDC (n)	CP diagnosis in APDC, not in CPR* (n)	CP diagnosis in CPR, not in APDC (n)	CP diagnosis in APDC (total) (n)	Sensitivity (95% CI)	PPV (95% CI)
	a	b	c	a+b	a/(a+c)	a/(a+b)
1 or more G80 codes	1082	666	472	1748	0.70 (0.67, 0.72)	0.62 (0.60, 0.64)
1 or more G80 codes after 2y age	1049	568	505	1617	0.68 (0.62, 0.70)	0.65 (0.63, 0.67)
1 or more G80 codes before 4y age	614	303	940	917	0.40 (0.37, 0.42)	0.67 (0.64, 0.70)
1 or more G80 codes, principal field	790	295	764	1085	0.51 (0.48, 0.53)	0.73 (0.70, 0.76)
1 or more G80 codes (top 3 fields)	1041	592	513	1633	0.67 (0.65, 0.70)	0.64 (0.61, 0.66)
2 or more plus G80 codes	854	362	700	1216	0.55 (0.53, 0.57)	0.70 (0.68, 0.73)
1 or more G80 or G81 codes	1142	1154	412	2296	0.74 (0.71, 0.76)	0.50 (0.48, 0.52)
1 or more G80, 81, 82 or 83 codes	1153	1286	401	2439	0.74 (0.72, 0.76)	0.47 (0.45, 0.49)

*There are approximately 120 children that are “counted” in the CP Register prevalence analyses, that were not able to be linked, as they have “opted off” the CP Register.

CI confidence interval; CP cerebral palsy; ICD10-AM International Statistical Classification of Diseases and Related Health Problems, Tenth Revision, Australian Modification; PPV Positive Predictive Value; ICD10-AM codes: G80 cerebral palsy; G81 hemiplegia; G82 paraplegia, tetraplegia; G83 other paralytic syndromes

Table 4.3. Comparison of sociodemographic characteristics of children and young people identified as having cerebral palsy in either the CP Register (CPR) or hospital admission data (APDC).

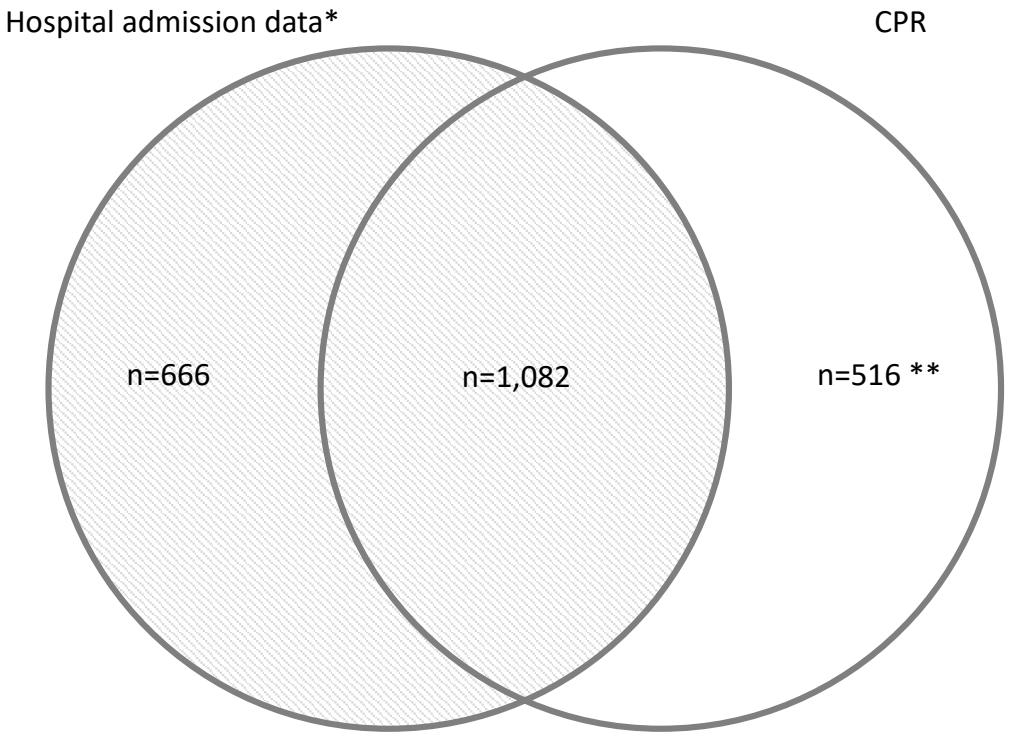
Sociodemographic characteristic	CPR (n=1,598) n (%)	APDC* (n=1,748) n (%)	SMD#
Sex			0.034
Male	914 (57.2)	1003 (57.4)	
Female	684 (42.8)	744 (42.6)	
Birth Years			0.115
2001-2005	807 (50.5)	983 (56.2)	
2006-2010	791 (49.5)	765 (43.8)	
Index of Relative Socioeconomic Disadvantage			0.101
1 (most disadvantaged)	290 (18.1)	323 (18.5)	
2	288 (18)	288 (16.5)	
3	315 (19.7)	304 (17.4)	
4	327 (20.5)	290 (16.6)	
5 (least disadvantaged)	290 (18.1)	239 (13.7)	
Unknown	88 (5.5)	304 (17.4)	
Remoteness			0.182
Major city	876 (54.8)	1136 (65)	
Regional	625 (39.1)	526 (30.1)	
Remote	10 (0.6)	13 (0.7)	
Unknown	87 (5.4)	73 (4.2)	

* Cerebral palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis
# Differences in data sources was considered where SMD >0.1
APDC Admitted Patient Data Collection; CPR Cerebral Palsy Register; SMD Standardised Mean Difference

Data linkage showed n=1,082 children and young people to be identified in both NSW/ACT CPR and APDC-G80 cohorts (67.7% of children in CPR, 61.9% of children in APDC-G80) (**Figure 4.1**). Comparison of clinical characteristics of children identified in the NSW/ACT CPR or APDC-G80 cohort using variables available through data linkage is shown in **Table 4.4**. When compared to children identified in the NSW/ACT CPR, a higher proportion of children and young people in the APDC-G80 cohort had comorbidities including epilepsy (31.2% v 26.3%), gastrostomy (21.3% v 13.6%), intellectual disability

(51.4% v 49.0%) and autism (14.9% v 11.0%) (SMD >0.1). Where clinical characteristics were available in multiple data sources, there was at least moderate agreement between sources (**Table 4.5**).

Figure 4.1. Overlap of children with a diagnosis of cerebral palsy ascertained from either a cerebral palsy register or hospital admission data.



* Cerebral palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis
** Includes n=44 cases in CPR where no admission during study period, and n=472 cases in CPR where cases had one or more admissions but no admissions with G80 diagnosis
CPR NSW / ACT Cerebral Palsy Register

Table 4.4. Comparison of clinical characteristics of children and young people ascertained using data linkage to various data sources based on identification of cerebral palsy using either CP Register or hospital admission data (APDC).

Data source used to identify clinical characteristic	Clinical characteristic	Data source used to identify cerebral palsy		SMD
		CPR n (%)	APDC* n (%)	
CPR		1598 (100)	1082 (61.9)	
	Epilepsy			0.128
	Yes	420 (26.3)	338 (31.2)	
	No or resolved	865 (54.1)	564 (52.1)	
	Unknown	313 (19.6)	180 (16.6)	
	Intellectual Disability			0.048
	Yes	783 (49)	556 (51.4)	
	No	610 (38.2)	395 (36.5)	
	Unknown	205 (12.8)	131 (12.1)	
PDC		1331 (83.3)	1357 (77.6)	
	Gestation at birth			0.133
	Preterm	488 (36.7)	448 (33)	
	Term	842 (63.3)	908 (66.9)	
APDC		1554 (97.2)	1748 (100)	
	Gastrostomy use			0.152
	Yes	211 (13.6)	373 (21.3)	
	No	1343 (86.4)	1375 (78.7)	
FACS		1413 (88.4)	1454 (83.2)	
	Autism			0.178
	Yes	156 (11)	217 (14.9)	
	No	1257 (89)	1237 (85.1)	
BDM				
	Death			0.228
	Yes	47 (2.9)	142 (8.1)	
	No	1551 (97.1)	1606 (91.9)	

*Cerebral Palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis

[#] Differences in data sources was considered where SMD >0.1

APDC Admitted Patient Data Collection; CPR New South Wales / Australian Capital Territory Cerebral Palsy Register; FACS Family and Community Services dataset; PDC Perinatal Data Collection**;** BDM Registry of Births, Deaths and Marriages; SMD Standard Mean Difference

Table 4.5. Agreement between clinical variables ascertained from either a cerebral palsy register (CPR) or hospital admission data (APDC) in children with cerebral palsy

Clinical variable	Source dataset 1	Source dataset 2	Kappa (95% CI)
Epilepsy	CPR	APDC	0.70 (0.66, 0.75)
Prematurity	CPR	PDC	0.93 (0.91, 0.96)
Intellectual disability	CPR	FACS	0.44 (0.39, 0.50)

APDC Admitted Patient Data Collection; CI confidence interval; CPR New South Wales / Australian Capital Territory Cerebral Palsy Register; FACS Family and Community Services dataset; PDC Perinatal Data Collection

A higher proportion of children in the APDC-G80 cohort also died during the study (8.1% v 2.9% in CPR) (SMD >0.1), and these deaths occurred at a median earlier age 6.3 years (interquartile range (IQR) 3.4-9.7 years), compared with 8.4 years (IQR 5.1-11.1 years) in the NSW/ACT CPR. The top three underlying causes of death was similar between the groups (diseases of the nervous system, diseases of the respiratory system, congenital malformations).

Compared with children in the NSW/ACT CPR cohort but not in the APDC-G80 cohort, those identified in both cohorts were more likely to have spastic bilateral or dyskinetic motor types, be GMFCS IV-V and/or MACS IV-V and were more likely to have epilepsy and/or intellectual disability (**Supplementary Table 9.4**).

4.5 Discussion

4.5.1 Principal findings

Hospital admission data and a common case definition (one or more admissions with G80 code) had a sensitivity of 70% and a PPV of 62% for identifying CP in children compared with a gold standard of a population CP register (NSW/ACT CPR). Sensitivity varied from 0.40-0.74 and PPV varied from 0.47-0.73 when using other case definitions with hospital admission data. When compared with children identified in the NSW/ACT CPR, a higher proportion of children with CP identified in hospital admission data had comorbidities, lived in major cities, and had died during the study period.

4.5.2 Strengths of the study

Strengths of this study include the use of data linkage of multiple datasets to assess differences between CP register and hospital admission data cohorts, which supported a more comprehensive description of the populations. Our results can be used by researchers and research end-users alike when considering bias influencing the interpretation of epidemiological CP research.

4.5.3 Limitations of the data

The main limitation of this study is the availability of diagnosis (ICD10-AM) codes only in the hospital admission data (APDC), and not in other data sources (e.g. outpatient, insurance claim) as in some other studies.(8, 10, 12-14) However, as almost all (97.3%) participants had at least one hospital admission, this is not likely to have influenced the results. The small number of children who have opted out of the NSW/ACT CPR may also have influenced the sensitivity and PPV rates that we have reported, although likely not substantially. Potential errors in coding that often exist in hospital admission data should also be considered; we have attempted to minimise these by including data from multiple admissions where available, and linkage with other data sources. Our study was also limited to children, with the oldest participant being 19 years old. It is therefore unclear whether our findings would be replicated in adults. Future research focused on adults with CP would be helpful to explore this further.

4.5.4 Interpretation

Understanding the differences between cohorts from administrative health data such as hospital admission data and a CP register is crucial for researchers to consider when planning studies, and for research end-users when interpreting study results. Our findings suggest that studies with populations derived from hospital admission data will include a population with more comorbidities and greater severity. These factors are known to be associated with greater health service utilisation(26) and adverse health

outcomes(27, 28) and results based on such populations are likely to overstate these (negative) outcomes. We also noted that children with CP identified from hospital admission data were more likely to be older than those from a CP register. This may be due to these children having a longer-lived period to experience a hospital admission. Researchers should ensure a long enough period to capture health care episodes (such as hospital admissions) when using health administrative data such as hospital admission data to ascertain their population.

Our findings also highlight that many (27-53%) CP cases defined using administrative health data are not found in a population-based CP register. Some of these cases may represent families who have chosen to not have their details included in a CP register (opted out) but are still counted for prevalence estimates (we estimate n=120 (7.5%) cases in our study). Some may represent cases of CP missed by a CP register, with a recent Norwegian study reporting up to 60% of cases of CP ascertained in administrative health data but not in a CP population registry were correct.(18) Other cases may represent known complexities in diagnosis of CP, including definitions based on age at injury (post-neonatal causes), children who would 'lose' a CP diagnosis following clinical review, when another, progressive condition is identified (false positives)(17) or those described as 'at risk of CP' who have no discernible functional impairment at the age of five years when a diagnosis of CP might be confirmed, and overlap with other neurodevelopmental disorders.(29, 30)

There are limited studies that have compared CP population registers and administrative health data. Comparison of the APDC with the CPR in our study was similar to that found in a recent study based on inpatient and outpatient visits in Quebec, when using a similar case definition.(12) A study that used a broader case definitions (e.g. including G81, G82 or G83 ICD-10AM codes) is likely to have included a larger proportion of individuals who are not in CP population registers.(8)

In recent years there has been a substantial increase in research about adults with CP,(31) including several epidemiological studies that have used administrative health datasets to explore a range of outcomes.(13, 14) Many (younger) CP registers may not have sufficient data on adults with CP at the present time, and in this group administrative health data may be particularly useful. Researchers should be aware of impact of case definitions on sensitivity and PPV when using administrative data to ascertain CP and choose case definitions that best meet the needs of the research question.

Where available, the use of CP registers to define a research population offer substantial advantages over using administrative health data, and data linkage can further enhance their value for research. CP registers often include condition-specific clinical data including motor type, markers of functional severity (e.g., GMFCS), and comorbidities such as developmental disabilities which are often not captured or incompletely captured in more general administrative health data. Data linkage of CP registers with

other data sets can enable a comprehensive picture of health and education outcomes and service utilisation. Data linkage research of large datasets may enable the identification of patterns of treatment outcomes, supporting health care providers to develop more personalised treatment programs such as medication or therapy interventions.

4.5.5 Conclusions

In summary, this study has shown that populations of children with CP derived from CP registers and hospital admission data are likely to differ in severity and associated comorbidities. The chosen method for population ascertainment should be considered when interpreting the results of epidemiological research.

4.6 Chapter Synopsis

In **Chapter 4**, clinically meaningful differences were described between populations of children with CP either in a CP population register (NSW/ACT CP Register) or in hospital admission data. Children identified through a common case definition in hospital admission data were older, with a higher proportion of comorbidities and were more likely to die during the study period. These findings are important for end users when interpreting the results of research using populations derived from health administrative

data. The study was published in *Paediatric and Perinatal Epidemiology*. **Chapters 5**, **6** and **7** will focus on different dimensions of outpatient access for children including non-attendance (**Chapter 5**), telemedicine (**Chapter 6**), and continuity of care (**Chapter 7**).

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Chapter 5 Non-attendance at outpatient clinic appointments by children with cerebral palsy

5.1 Published Manuscript

This chapter has been reformatted for publication as part of this thesis.

The citation for the published manuscript is:

> Paget SP, McIntyre S, Goldsmith S, et al. Non-attendance at outpatient clinic appointments by children with cerebral palsy. Dev Med Child Neurol. 2022;64(9):1106-1113. doi:10.1111/dmcn.15197

A copy of the published article as well as a statement of the specific contributions of the co-authors can be found in **Chapter 10 - Appendix B**.

The thesis concept map for Chapters 5, 6 and 7 is shown in **Figure 5.1**.

Figure 5.1. Thesis Concept Map for Chapters 5, 6 and 7.

	Knowledge Gap	Objective	Chapter	Publication	Key Findings
Introduction		Background and literature review	Chapter 1		
	What is already known about health service access in CP?	(I) To systematically review the existing literature regarding determinants of hospital-based health service access among people with CP.	Chapter 2	Determinants of Hospital-Based Health Service Utilisation in CP: a Systematic Review. *doi:10.1016/j.apmr.2021.12.003*	• Hospital health service utilisation associated with age, severity and comorbidities.
Study Methods		Introduce the NSW/ACT CP Register, administrative data sets and data linkage.	Chapter 3		
	How do study populations differ between a CP population register and hospital admission data?	(II) To compare the sociodemographic and clinical characteristics of children with CP identified from a CP population register and hospital admission data.	Chapter 4	A comparison of cohorts of children with CP from a population register and hospital admission data. *doi:10.1111/ppe.13024*	• Hospital admission data has sensitivity of 0.7 for a diagnosis of CP. • Children with CP from hospital admission data are older, live in major cities, more comorbidities and early death.
Dimensions of outpatient health service access and CP	What factors influence non-attendance at specialty outpatient clinics?	(III) To explore the factors associated with non-attendance by children with CP at specialty outpatient clinics.	**Chapter 5**	Non-attendance at outpatient clinic appointments by children with CP. *doi:10.1111/dmcn.15197*	• Non-attendance associated with older age, socioeconomic disadvantage and previous non-attendance.
	How does telemedicine influence equity of access to outpatient care?	(IV) To determine how telemedicine supports equity of access to specialist outpatient care for children with CP.	**Chapter 6**	Telemedicine for children with CP before, during and after the COVID-19 pandemic. *Under review.*	• Telemedicine use lower in children living in regional and remote areas.
	How have telemedicine rates have changed since the COVID-19 pandemic?	(V) To determine the impact of the COVID-19 pandemic on telemedicine use in children with CP.			• Telemedicine use increased substantially during COVID-19 pandemic and since has declined to near baseline.
	What factors influence access to outpatient services?	(VI) To determine how clinical and sociodemographic determinants influence hospital outpatient service access in CP.	**Chapter 7**	Outpatient encounters, continuity of care and unplanned hospital care for children and young people with CP. *doi:10.1111/dmcn.15800*	• Outpatient service use associated with metropolitan areas, GMFCS IV-V, epilepsy, intellectual disability.
	Are CP outpatient services accessible for Aboriginal and/or Torres Strait Islander children?	(VII) To determine hospital outpatient service utilisation for Aboriginal and/or Torres Strait Islander children and young people with CP.			• Rates of outpatient service use for Aboriginal and/or Torres Strait Islander children are similar to non-indigenous population.
	How does outpatient continuity of care impact unplanned hospital care?	(VIII) To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.			• Decreased continuity of care is associated with increased unplanned hospital care.
Summary and Conclusions		Summary of key findings and implications for clinical practice, policy and research.	Chapter 8		

5.2 Introduction

Cerebral Palsy (CP) is a neurodevelopmental condition characterised by a permanent disorder of movement and posture and attributed to non-progressive disturbances in early brain development.(1) CP is the most common cause of physical disability in childhood, with a birth prevalence of approximately 2.0 per 1000 live births in most high-income countries (2) although recent birth years in Australia are showing a decline.(3) For many children with CP, the motor disorder is accompanied by neurological disorders (e.g., epilepsy), diseases of other body systems (e.g., respiratory, digestive system)(4) and musculoskeletal deformities(5, 6) (e.g., scoliosis, hip displacement) that further complicate their health.

The long-term management of CP and its associated health conditions and complications is a focus for health services,(7) with most management occurring in outpatient settings. As children with CP often have complex health needs, specialty medical and surgical outpatient services are generally centralised in children's hospitals and frequently involve multidisciplinary teams including medical, nursing and allied health professionals.

Non-attendance at scheduled outpatient appointments is recognised as a major issue across the health care system and health conditions. At a patient level, non-attendance may represent a missed opportunity for the early diagnosis of a health-related problem, or the initiation of an intervention to improve health or wellbeing, and in children with neurological conditions, may result in increased unplanned health care use such as emergency department presentations.(8) At a health service level, non-attendance is recognised to increase health care costs, decrease services' effective capacity, and add to waiting times for consultations and procedures.(9)

In this context, understanding factors associated with non-attendance at outpatient appointments is important to enable the identification of strategies to improve attendance and health outcomes for children with CP.(10) A systematic review of non-attendance across patient groups identified multiple factors are likely to be relevant, including factors related to the individual (younger age, lower socioeconomic status, history of previous non-attendance), and those related to the clinic (e.g., specialty type) and service.(9) Given the complexity and diversity of CP, factors such as severity of CP and comorbidities may also be important. We aimed to explore the factors associated with non-attendance by children with CP at specialty outpatient clinics located at two locations across a tertiary children's hospitals network.

5.3 Methods

5.3.1 Study population and data sources

We conducted a retrospective data linkage study of children with CP, born 2005-2017, who attended outpatient clinics at two children's hospitals in New South Wales (NSW), that provide services for children in NSW and the Australian Capital Territory (ACT). Children with CP were identified from the NSW/ACT CP Register (n=1764), a population-based database with multiple ascertainment strategies. The Register contains details of individuals with CP who were born or live in NSW or ACT, including demographic and clinical (motor type, severity of CP, presence of comorbidities) information. For each child, corresponding information on outpatient appointments scheduled at either of two tertiary paediatric hospitals in metropolitan Sydney, NSW Australia: Sydney Children's Hospital, Randwick and the Children's Hospital at Westmead (as part of Sydney Children's Hospitals Network (SCHN)) between 1st January 2012 and 31st December 2019 was ascertained. This timeframe was chosen as data before 2012 were incomplete due to changes in data collection processes. Outpatient data were obtained from the SCHN Non-Admitted Patient (NAP) administrative data collection. The SCHN NAP is based on two data sources: data documenting scheduled outpatient appointments and patient-level clinician activity including demographic information, clinical specialty type, location, attendance/non-attendance, and clinician discipline. The accuracy of the NAP data collection is ensured as it is a statutory data collection with the NSW Ministry of Health mandating the collection and reporting of patient level non-admitted activity for

all clinical and/or therapeutic services provided or contracted by NSW Health. Outpatient clinics at both hospitals are provided under a government universally funded system (either state-funded or Medicare) without fee to the patient, during office hours (typically 8am to 5pm), and on weekdays (Monday to Friday).

5.3.2 Study outcomes

The main study outcome was frequency of scheduled outpatient appointments categorised as attended or not attended. The data available in the SCHN NAP did not discriminate between appointments rescheduled or cancelled by the hospital for administrative reasons (e.g., staff being unavailable) and those rescheduled by families. Scheduled outpatient appointments were categorised based on clinical specialty listed in **Supplementary Table 9.5** and health care professionals seen were categorised by discipline (medical/dental, nursing, allied health, other). To adjust for varied scheduling practices (e.g., some specialties scheduled multiple appointments with health care professionals of different disciplines on the same day), scheduled outpatient appointments were converted to outpatient days. At each (attended) outpatient day a child could be reviewed by multiple (different) clinical specialties and seen by multiple health care professionals of different disciplines. A flow diagram presenting an overview of the study processes including study exclusions is shown in **Supplementary Figure 9.3**.

5.3.3 Patient socio-demographic, clinical and process of care factors

Patient socio-demographic and clinical factors were collected from the CP Register and SCHN NAP, and included demographic information on age at appointment, sex, preferred language, and country of birth. Postcode of residence was used to estimate socioeconomic disadvantage and geographical remoteness. Socioeconomic disadvantage was measured with reference to the general population, using the Index of Relative Socioeconomic Disadvantage (IRSD) and categorising into quintiles (quintile 1 being the most disadvantaged, and quintile 5 being the least disadvantaged).(11) Geographical remoteness was defined using Australian Statistical Geography Standard, which categorises populated localities as major cities, inner/outer regional and rural/remote areas) based on ease of access to services via road network.(12) Clinical variables included Gross Motor Function Classification System (GMFCS) classification (dichotomised into levels I-III (ambulant) and IV-V (non-ambulant)),(13) predominant motor type (grouped into spastic, dyskinetic, and other (ataxia, hypotonia, those identified as 'early and at risk' of CP)), and the presence of comorbidities of epilepsy and intellectual disability (dichotomised as 'yes' or 'no').

Process of care factors were identified using NAP data. Recent multidisciplinary team (MDT) care was defined as review by two or more health care professionals from different disciplines at the previous outpatient day (visit). Recent experience of care coordination was defined as review by two or more different clinical specialties at the previous

outpatient day. Recent non-attendance was defined as non-attendance at the previous outpatient day. Appointments that were rescheduled or cancelled were also identified. Recent rescheduled or cancelled appointments were defined as one or more rescheduled / cancelled appointment in the previous six months. Where there was no previous recorded appointment, (e.g., at the first scheduled outpatient day during the study period) these process of care factors were classified as 'no'.

5.3.4 Statistical analysis

Children who were scheduled to attend clinic appointments at either of the two hospitals on at least one occasion (n=1,395) were described in terms of their demographic and clinical features. Proportions, counts, and rates of scheduled appointments by specialty type were compared. Characteristics of children reviewed by major specialties, and proportions of scheduled outpatient appointments by age group were compared using Chi-squared tests. Associations between patient factors and non-attendance were assessed using univariable and multivariable logistic regression including date of appointment, child socio-demographic, and clinical factors and process of care measures. Multivariable analyses were conducted using generalised estimating equations and exchangeable correlation structure to account for repeated outpatient attendances by the same child. Analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC, USA). The study was approved by the SCHN Human Research Ethics Committee (2019/ETH11829).

5.4 Results

We identified 1,395 children from the NSW/ACT CP Register who had one or more outpatient appointments scheduled during the study period (**Table 5.1**). 59.6% (n=831) of children were male; most (96.1%, n=1,340) lived either in major cities or inner regional areas.

Table 5.1. Characteristics of children with cerebral palsy with scheduled outpatient appointments, 2012-2019.

Demographic / Clinical Factor	N= 1,395 n (%)
Gender	
Male	831 (59.6)
Female	564 (40.4)
Country of Birth	
Australia	1298 (93.7)
Overseas	88 (6.3)
Preferred Language	
English	1214 (91)
Other	120 (9)
Remoteness	
Major cities of Australia	941 (67.7)
Inner Regional Australia	399 (28.7)
Outer Regional Australia	42 (3)
Remote Australia	7 (0.5)
State / territory of residence	
NSW	1323 (95.9)
ACT	57 (4.1)
IRSD Quintile	
1 (most disadvantaged)	274 (19.7)
2	210 (15.1)
3	275 (19.8)
4	288 (20.7)
5 (least disadvantaged)	342 (24.6)
GMFCS	
I-III	998 (74.5)
IV-V	342 (25.5)
Predominant motor type	
Spastic	1010 (73.6)
Dyskinetic	192 (14.0)
Other	171 (12.5)
Intellectual Disability	
Yes	645 (46.2)
No	515 (36.9)

Not reported	235 (16.8)
Epilepsy	
Yes	394 (28.2)
None or resolved	796 (57.1)
Not reported	205 (14.7)

ACT Australian Capital Territory; GMFCS Gross Motor Function Classification System; IRSD Index of Relative Disadvantage; NSW New South Wales

There was a total of 50,121 scheduled outpatient days during 2012-2019; each child had a median of 4.8 (interquartile range (IQR) 2.0-7.9) scheduled appointments per year. There was variation in the frequency and involvement of different specialties (Table 5.2). Most children were reviewed one or more times by rehabilitation medicine (82.2%), allied health (78.9%) and neurology/neurosurgery (55.6%) clinics. These clinics were also the most frequently attended (**Table 5.2**). There were differences between the groups of children reviewed by different specialties (**Supplementary Table 9.6**). Compared with children without each respective comorbidity, children with epilepsy (odds ratio (OR) 6.28; 95% confidence interval (95%CI) 4.68-8.44) and intellectual disability (OR 3.06; 95%CI 2.41-3.90) were substantially more likely to be seen by neurology / neurosurgery; children with non-ambulant CP were more likely to be reviewed by orthopaedics (OR 3.69; 95%CI 2.84-4.79) (**Supplementary Table 9.6**). There were also differences in specialty scheduled outpatient days between age groups **(Supplementary Table 9.7)**. The 0-4 year age group attended 54% of neurology outpatient days (compared with 40% of total outpatient days) and the 10-14 year age group attended 11% of neurology outpatient days (compared with 15% of total outpatient days). In contrast, the 0-4 year

age group attended 18% of orthopaedic outpatient days and the 10-14 year age group attended 30% of orthopaedic outpatient days.

Table 5.2. Number and proportion of children with cerebral palsy attending scheduled outpatient appointments and non-attendance by specialty group, 2012-2019

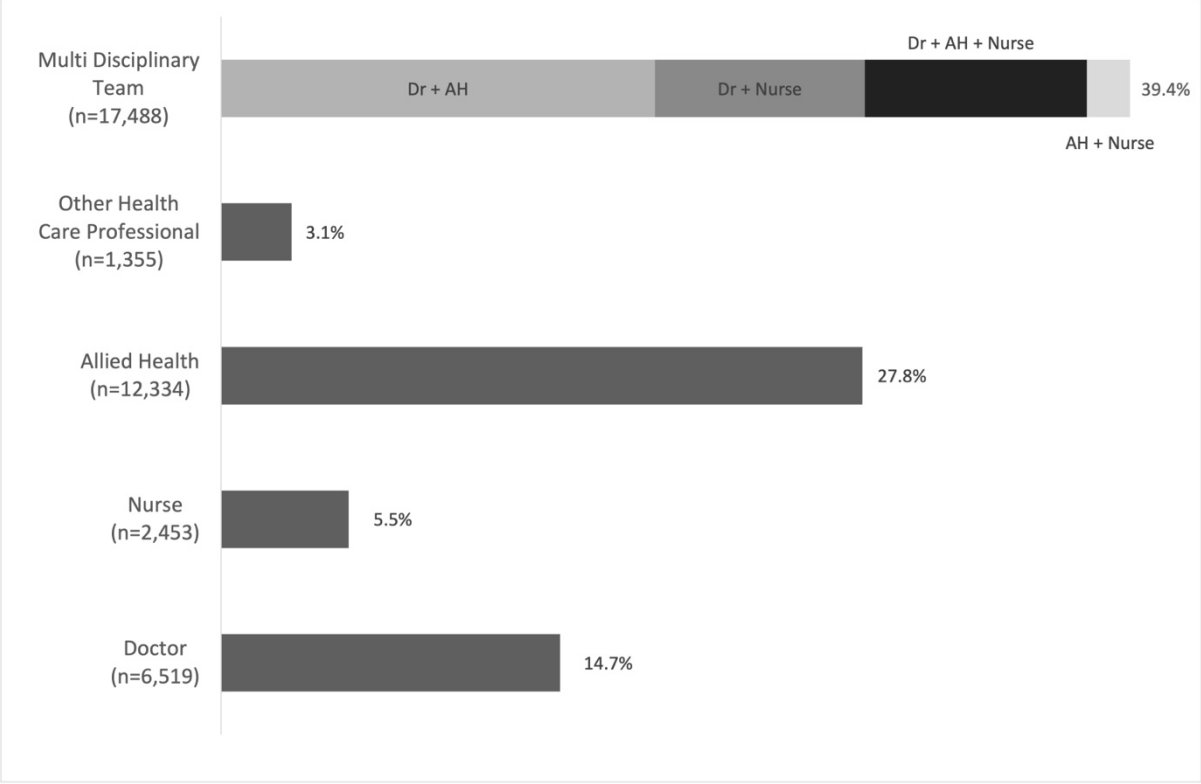
Specialty Group	Children attending outpatient clinics n (%)	Frequency of outpatient days n (%)	Number of scheduled appointments/ year mean (SD)	Scheduled appointments not attended n (%)
Allied Health	1,100 (78.9)	19,008 (37.9)	2.2 (2.7)	2,187 (11.5)
General Medicine	297 (21.3)	1,863 (3.7)	0.2 (0.9)	416 (22.3)
Rehabilitation Medicine	1,147 (82.2)	14,918 (29.8)	1.8 (2.1)	1,651 (11.1)
Neurology / Neurosurgery	776 (55.6)	4,982 (9.9)	0.7 (1.6)	469 (9.4)
Other Medical Specialty	776 (55.6)	6,172 (12.3)	0.8 (2)	669 (10.8)
General Surgery	305 (21.9)	1,030 (2.1)	0.1 (0.5)	127 (12.3)
Orthopaedics	604 (43.3)	4,433 (8.8)	0.5 (0.9)	655 (14.8)
Other Surgical Specialty	681 (48.8)	4,559 (9.1)	0.6 (1)	604 (13.2)
Medical Imaging	625 (44.8)	4,620 (9.2)	0.6 (1.2)	214 (4.6)
Total	1,395 (100)	50,121 (100)	6.1 (6.1)	5773 (11.5)

Numbers and percentages do not sum to totals as children may attend multiple specialty clinics

Most (88.5%, n=44,348) scheduled outpatient days were attended; with children seen by a single provider in about half (51.1%) of all attended outpatient days (**Figure 5.2**). MDT

care was provided in one-third (39.4%) of attended outpatient days and most involved a doctor and allied health professional (47.7%), doctor and nurse (23.1%), or doctor, nurse and allied health professional (24.4%) **(Figure 5.2)**. Care coordination of multiple specialty appointments occurred in 19.9% (n=8,813) of all attended outpatient days.

Figure 5.2. Proportion of outpatient days attended by children with cerebral palsy (by health care professional discipline).

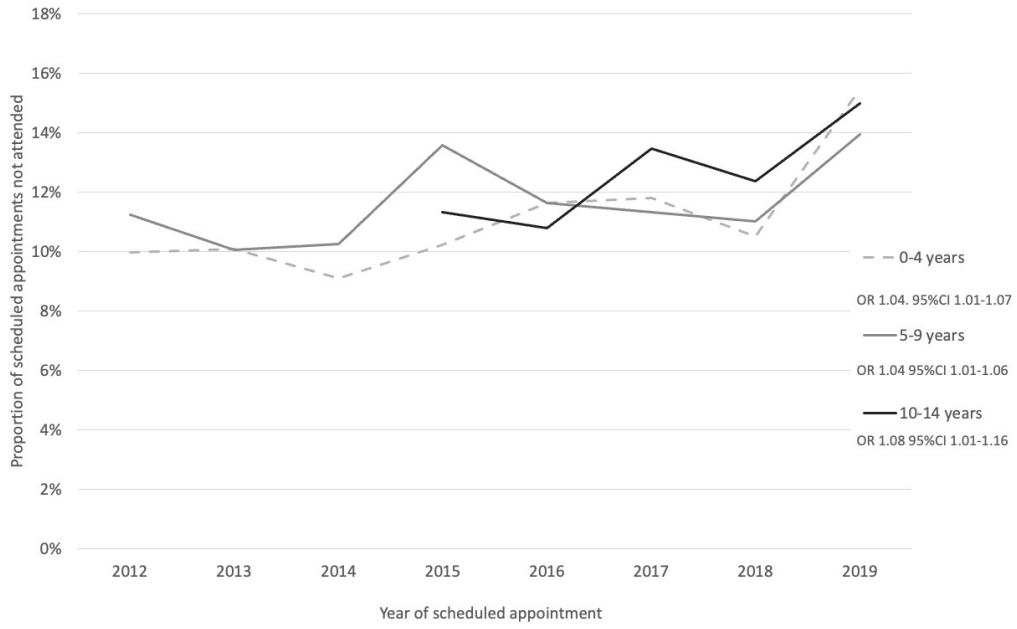


*Health care professional involved unknown in 4199 (9.5%) outpatient days, all groups are mutually exclusive

AH Allied Health; Dr Doctor

A total of 5,773 (11.5%) of scheduled outpatient days were not attended (**Table 5.2**). The rate of non-attendance increased on average by 5% per year (odds ratio (OR) 1.05; 95% confidence interval (CI) 1.04-1.07) from 11.5% in 2012 to 14.2% in 2019. The rate of increase was similar among 0-4 year (OR 1.04; 95%CI 1.01-10.07) and 5-9 year (OR 1.04; 95%CI 1.01-1.06), and 10-14 year age groups (OR 1.08; 95%CI 1.01-1.16) (**Figure 5.3**).

Figure 5.3. Proportion of scheduled outpatient clinic appointments not attended by year and age group in children with cerebral palsy at a children's hospitals network.



OR Odds Ratio

The association between socio-demographic, clinical, and process of care factors and non-attendance is shown in **Table 5.3**. After adjusting for all factors, increased likelihood of non-attendance was associated with older aged children 5-9 years (adjusted odds ratio (aOR) 1.11; 95%CI 1.02-1.22) and 10-14 years (aOR 1.17; 95%CI 1.03-1.34) and greater socioeconomic disadvantage (IRSD quintile 1: aOR 1.29 95%CI 1.11-1.50 and IRSD quintile 2: aOR 1.50 95%CI 1.29-1.76) (**Table 5.3**). There was no statistical evidence of an association between clinical variables such as GMFCS and predominant motor type and likelihood of non-attendance. Recent experience of multidisciplinary team or coordinated care was not associated with likelihood of non-attendance. However, children with recent rescheduled or cancelled appointment (aOR 1.08; 95%CI 1.01- 1.16) or previous non-attendance (aOR 1.38; 95%CI 1.23- 1.53) had increased odds of non-attendance.

Table 5.3. Association between socio-demographic, clinical and process of care factors with non-attendance at outpatient clinics for children with cerebral palsy.

Socio-demographic, clinical and process of care factors	Univariable OR (95% CI)	Multivariable OR (95% CI)
Socio-demographic factors		
Year of appointment	1.05 (1.04- 1.07)	1.04 (1.02- 1.06)
Sex		
Male	1.04 (0.94- 1.15)	1.01 (0.92- 1.12)
Female	Reference	Reference
Age		
0-4 years	Reference	Reference
5-9 years	1.22 (1.12- 1.34)	1.12 (1.03- 1.23)
10-14 years	1.45 (1.3- 1.63)	1.19 (1.04- 1.35)
Country of Birth		
Australia	Reference	Reference
Overseas	1.11 (0.92- 1.34)	1.06 (0.87- 1.27)
Preferred Language		
English	Reference	Reference
Other	1.08 (0.92- 1.26)	0.98 (0.83- 1.16)
IRSD Quintile		
1 (most disadvantaged)	1.32 (1.13- 1.53)	1.30 (1.12- 1.52)
2	1.50 (1.28- 1.76)	1.52 (1.30- 1.78)
3	1.20 (1.04- 1.40)	1.20 (1.03- 1.39)
4	1.12 (0.96- 1.30)	1.13 (0.97- 1.30)
5 (least disadvantaged)	Reference	Reference
Remoteness		
Major Cities of Australia	Reference	Reference
Regional / Remote	1.04 (0.92- 1.16)	0.95 (0.85- 1.05)
Clinical factors		
GMFCS		
I-III	Reference	Reference
IV-V	1.11 (1.00- 1.24)	1.08 (0.95- 1.23)
Predominant motor type		
Spastic	Reference	Reference
Dystonic	0.94 (0.83- 1.08)	0.92 (0.79- 1.06)
Other	0.96 (0.82- 1.12)	0.97 (0.82- 1.14)

Intellectual Disability		
Yes	1.12 (1.01- 1.23)	1.09 (0.97- 1.22)
No	Reference	Reference
Epilepsy		
Yes	0.99 (0.89- 1.10)	0.92 (0.82- 1.04)
No	Reference	Reference
Process of Care Factors		
Last appointment with MDT Care		
Yes	1.05 (0.99- 1.12)	1.01 (0.94- 1.07)
No	Reference	Reference
Last appointment with Care Coordination		
Yes	1.05 (0.97- 1.14)	1.03 (0.95- 1.12)
No	Reference	Reference
Last appointment not attended		
Yes	1.40 (1.26- 1.56)	1.32 (1.17- 1.48)
No	Reference	Reference
Recent cancelled or rescheduled appointment		
Yes	1.14 (1.06- 1.22)	1.08 (1.01- 1.16)
No	Reference	Reference

GMFCS Gross Motor Function Classification System; IRSD Index of Relative Socioeconomic Disadvantage; MDT Multidisciplinary team care

5.5 Discussion

Non-attendance at outpatient clinics for children with CP is a little-researched area. We found non-attendance to be associated with four factors: increasing age, socioeconomic disadvantage, previous non-attendance at an outpatient clinic and recent cancellation or rescheduling of an appointment. Non-attendance was not associated with area of residence, CP severity nor the presence of major comorbidities. Non-attendance was

also not associated with recent multidisciplinary team or coordinated care. Rates of non-attendance increased during the study period.

Outpatient clinics are the dominant model through which the health system provides support for the management of chronic health conditions. Non-attendance at outpatient clinics can therefore have important consequences for children with CP. Not attending an outpatient clinic appointment means a child misses an opportunity to receive timely (and evidence-based) health interventions and/or engage in health surveillance and education. This may result in them using unplanned heath care (e.g., emergency departments) to support their needs,(8) and contribute over time to worse health outcomes. Our results suggest that children at greater socioeconomic disadvantage, who are already known to have higher rates of CP severity, intellectual disability, and comorbidities,(14) are also inequitably exposed to these risk. It is encouraging that overseas birth and non-English speaking backgrounds, or those from regional or remote areas were not associated with non-attendance.

That non-attendance increases with age also requires further investigation. While this may represent changing priorities as children grow older, greater need for young person engagement or reduced perceived need, some health conditions associated with CP are known (for the most part) to only become apparent with increasing age. Examples of this include scoliosis,(5) and cognitive (e.g. attention deficit hyperactivity disorder), affective

and anxiety disorders, which are also known to be more prevalent in children and adolescents with CP than other children.(15) Our results support this finding, for example the development of musculoskeletal problems indicated by increased use of orthopaedic services in older age groups. Care fragmentation among multiple specialties as children grow older and new priorities arise may also result in children missing important aspects of care which are not typically addressed by all specialties. Awareness of this issue and ensuring services are adapted to be sensitive to changing needs and age-appropriate is important.

Our findings are largely consistent with the research in non-attendance at outpatient clinics in children (with CP and other health conditions). The rate of non-attendance that we report is similar to that reported in a recent study in children with neurological conditions,(8) although the reported rate of non-attendance can vary substantially depending on setting. Studies in paediatric settings have suggested that factors relating to both individuals (e.g., sociodemographic factors, ethnicity, insurance status) and systems (e.g., waiting times for appointments, administrative error) are associated with non-attendance.(16, 17) Studies of adults in outpatient (18) and primary care settings (19) have also identified social deprivation and age to be associated with non-attendance (with younger adults more likely to not attend than older adults), suggesting that our results here may reflect broader trends and may be applicable to other childhood patient groups. The reasons that families do not attend outpatient clinic appointments has also been the subject of recent qualitative studies.(20, 21) Common reasons reported

included travel difficulties, competing priorities and administrative issues (e.g., not receiving an appointment, difficulties in changing an appointment), that highlight the complexity that families manage to balance their child's health and other priorities.(22) These perspectives can help us reconsider non-attendance as a weakness in the model of care provided through outpatient clinics, in that they rely on face-to-face contact between a patient and health care professional at a particular point in time. There is a need for increased acknowledgement that health care is not 'one size fits all' and personalising health care delivery should sit alongside the agenda to personalise therapeutics. Strategies such as mHealth (e.g., SMS reminders (23)) and telemedicine may help support this agenda; increased use of telemedicine during the COVID-19 pandemic has suggested that this is an acceptable alternative for many face-to-face consultations.(24) Our results also underpin the need for improved coordination of care, and integration of speciality care with a child's local health care service and primary care team to ensure all opportunities to optimise health and development are pursued. The increasing rate of non-attendance that we identified makes these requirements time sensitive.

Strengths of our study include its size and the use of data from a CP register, which improved precision of our study population and availability of clinical and socio-demographic descriptors, and the use of routinely collected administrative data which is mandated by the NSW state government, which ensured accurate estimation of attendance rates and service events. However, our methodology was not designed to

explore the reasons that families did not attend, nor the reasons for the increase in non-attendance over time. Other limitations of our methodology include missing pertinent factors in our available data, including residency status. Our data also did not allow us to identify appointments cancelled by families prior to their appointment (distinct from those cancelled by the hospital). As others have identified (25) this is another clinically important group, as they too represent a missed opportunity for health care.

Our study shows an association between non-attendance at outpatient appointments and socioeconomic disadvantage, increasing age, recent non-attendance and cancelled or rescheduled appointments. These factors are readily identified and should be targeted when considering strategies to support families who may be experiencing difficulties with health care access. For example, clinicians can follow-up with families who have missed appointments and consider alternative ways of supporting children's health where possible. Future studies to investigate barriers and facilitators for attendance to outpatient clinics, and interventions to improve health care accessibility are warranted. This will enable design/ implementation of appropriate measures for uptake and access to care and services.

5.6 Chapter Synopsis

In **Chapter 5**, socioeconomic disadvantage, increased age, previous non-attendance and recent cancelled or rescheduled appointments were found to be associated with non-attendance at specialty children's hospital outpatient clinics. The study was published in *Development Medicine and Child Neurology*. **Chapter 6** will focus on access to telemedicine for children with CP.

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Chapter 6 Telemedicine for children with cerebral palsy before, during and after the COVID-19 pandemic

6.1 Published Manuscript

This chapter has been reformatted for publication as part of this thesis. It is currently in peer review with Journal of Child Neurology.

The citation for the manuscript is:

> Paget SP, McIntyre S, von Huben A, Stewart K, Williams T, Maly E, Ford K, Woolfenden S, Nassar N. Telemedicine for children with cerebral palsy before, during and after the COVID-19 pandemic: a cohort study. J Child Neurol. (Under review).

A statement of the specific contributions of the co-authors can be found in **Chapter 10 - Appendix B**.

6.2 Introduction

Telemedicine describes the delivery of health care services, by healthcare professionals using information and communications technologies for the exchange of information.(1) This is particularly pertinent when distance is a critical factor. Telemedicine includes outpatient encounters where consultation occurs through video or telephone, but the term is often used to include broader approaches including the sharing of medical images and remote monitoring of vital signs (e.g., heart rate, blood pressure, temperature).(2) Telemedicine is an important component of Australian digital health strategy,(3) and may improve equity of access to some health care services,(4, 5) particularly for patients living in regional and remote areas.(6, 7)

Cerebral palsy (CP) is a common childhood chronic health condition, and the most common cause of physical disability with a birth prevalence of 1.5 per 1000 live births in Australia.(8) Children with CP are frequent users of health care:(9) one quarter use outpatient services more than once monthly, and across multiple specialties,(10) and one third live outside metropolitan areas.(8) Complexities of travel and managing time away from work and other caring roles may make in-person attendance for health care consultations challenging. Telemedicine, therefore, may have an important role in improving access to health care and health outcomes for these children.

The COVID-19 pandemic was associated with a rapid and substantial increase in telemedicine as public health restrictions and the direct impacts of the virus limited access to in-person health care,(11) including for children with CP.(12) Studies conducted during this period, particularly among children with CP and other neurodevelopmental disorders, described increased telemedicine use for medical consultations (12) and explored the potential for telemedicine, for example, to provide rehabilitation programs to improve gross motor function (13) or manage common comorbidities such as epilepsy.(14) One study suggested that telemedicine did not suit all aspects of care for children with neurodevelopmental disabilities.(15) Other studies highlighted inequities in telemedicine access (14) that are also seen more broadly in society,(16) such as availability of information technologies (e.g., smartphones, broadband internet) and usability of digital platforms for those with less experience of technology.(17) Less is known about whether patterns of telemedicine established during the pandemic have been maintained since public health restrictions have ceased. We aimed to determine to what extent telemedicine supports equity of access to specialist outpatient care for children with CP experiencing neighbourhood socioeconomic disadvantage and/or in regional and remote areas. We also aimed to examine patterns of telemedicine for children with CP in time periods before, during and since the COVID-19 pandemic.

6.3 Methods

We conducted a retrospective data linkage study using administrative health data. Ethical approval was granted by Sydney Children's Hospitals Network Human Research Ethics Committee (2019/ETH11829). We identified children with CP, born from 2005 to 2017, from the New South Wales (NSW) / Australian Capital Territory (ACT) CP Register (n=1,764). The NSW/ACT CP Register is a population-level database with multiple ascertainment strategies.

We identified outpatient encounters for these children at two tertiary paediatric hospitals in metropolitan Sydney, NSW Australia (Sydney Children's Hospital, Randwick and the Children's Hospital at Westmead). Combined, these two hospitals provide the majority of speciality paediatric services for children living in NSW. Data were obtained from the hospitals' Non-Admitted Patient (NAP) data collection, which collects patient-level activity for all clinical and/or therapeutic services provided in non-admitted settings at the hospitals.

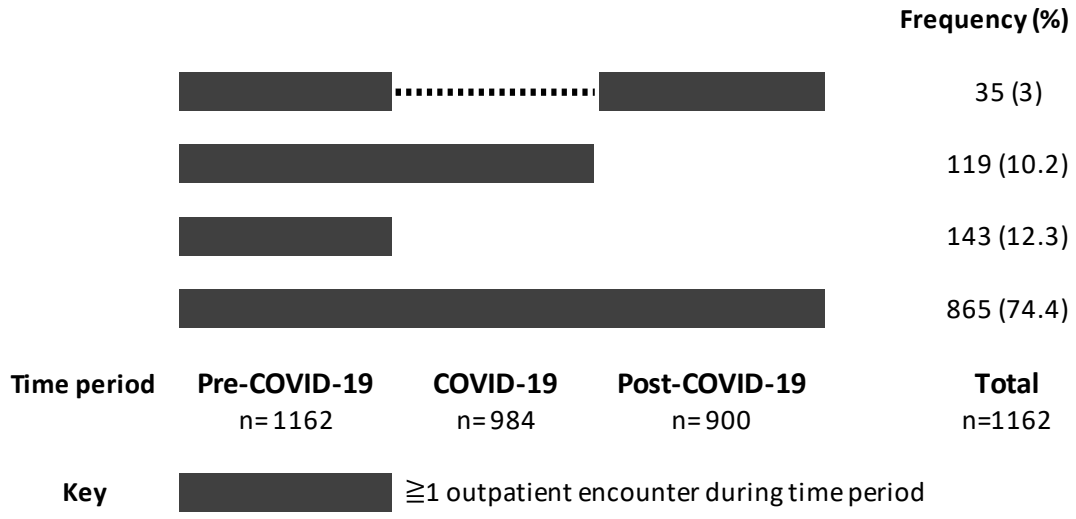
Data available from the NSW/ACT CP Register included age, sex, clinical information (Gross Motor Function Classification System (GMFCS), epilepsy, intellectual disability). Postcode of residence was determined from NAP data, or where not available, from the

NSW/ACT CP Register. Neighbourhood socioeconomic disadvantage was determined by assigning each individual residential postcode to its corresponding Australian Bureau of Statistics Index of Relative Socioeconomic Disadvantage (IRSD) score, and then categorized into quintiles (quintile 1 being the most disadvantaged and quintile 5 being the least disadvantaged).(18) Geographic remoteness was defined using the Australian Statistical Geography Standard, which categorize each residential postcode as a major city, inner/outer regional, and remote/very remote area) based on ease of access to services via road network.(19)

Data from the NAP data collection were available from 1st January 2018 to 31st May 2023. We defined three time periods: a 'pre-COVID-19' time period (1st January 2018 to 29th Feb 2020 (26 months)), a 'COVID-19' time period (1st March 2020 to 31st December 2021 (22 months)) and a 'post-COVID-19' period (1st January 2022 to 31st May 2023 (17 months)). The 'COVID-19' time period corresponded to the commencement of COVID-19 related restrictions in Australia (March 2020) to the easing of restrictions (e.g., opening of international borders) in December 2021, and included periods of stringent public health measures in NSW during March-July 2020 and July-October 2021. We categorised modality of outpatient encounters as in-person or telemedicine (including telephone and audiovisual encounters). We excluded encounters with no direct patient contact (e.g., case conferences, case planning, email). We identified the four most frequent clinical speciality types (Tier 2 class) based on frequency of (either in-person or telemedicine) encounters during the study period.

In order to improve understanding of how telemedicine use changed during the study, we only included children in analyses where they had one or more outpatient encounters in the 'pre-COVID-19' time period (**Figure 6.1**). We undertook descriptive analyses of the children and outpatient encounters. We determined the representativeness of our study population by comparison to children in the NSW/ACT CP Register using contingency tables, t-tests for continuous variables (age), chi-squared tests for categorical variables and Fisher exact test where expected frequencies were <5.

Figure 6.1. Frequency of children with cerebral palsy having one or more outpatient encounters at two children's hospitals in time periods during the COVID-19 pandemic.



We performed descriptive analyses of total outpatient encounters and telemedicine encounters, including for the four most frequent clinical specialty types. We determined average monthly rates of outpatient encounters (total, telemedicine) per 100 persons reported as medians and interquartile ranges (IQR), comparing rates between demographic and clinical descriptor groups (e.g., sex, socioeconomic disadvantage, geographical remoteness, GMFCS, epilepsy, intellectual disability).

To determine changes in telemedicine over time we determined monthly rates of total outpatient encounters and telemedicine encounters (median encounters per 100 persons per month) and the proportion of total outpatient encounters by telemedicine. We compared median rates across the three time periods using the Kruskall Wallis test. We compared changes in rates across time periods by age groups. Initial analyses suggested significant variation in rates of total outpatient encounters by calendar month (lower in January and December, corresponding to a 'low activity' period including Christmas and summer holidays in New South Wales) (**Supplementary Figure 9.4**). We therefore performed sensitivity analyses calculating rates of total outpatient encounters and telemedicine encounters for each time period removing encounters in January or December. To further examine changes in telemedicine over time we analysed the monthly proportion of total outpatient encounters provided by telemedicine using joinpoint regression. Joinpoint is a trend analysis software which describes changes in data by connecting several different line segments on a log scale at "joinpoints". Analysis starts with a minimum number of joinpoints (i.e., zero join points), and adds joinpoints to

the model where they improve model fit, using Monte Carlo permutation to test significance.(20) The joinpoint regression also calculates monthly percent change and 95% confidence intervals.

We performed stratified analyses of rates of total outpatient encounters, telemedicine encounters and proportion of telemedicine between groups of children based on socioeconomic disadvantage (IRSD 1 v IRSD 2-5) and geographical remoteness (major cities v regional and remote) using the Kruskall Wallis test. Analyses were performed in SAS 9.4 (SAS Institute, Cary, NC, USA) and Joinpoint Regression Program, V5.0.2 (National Cancer Institute, USA).

This study was conducted in partnership with two research partners (EM, KF) with lived experience from CP Quest https://cerebralpalsy.org.au/get-involved-research/cp-quest/. Discussions with both partners guided the prioritisation of research questions and supported contextual understanding of the data and initial results. These insights were used to inform the discussion of this paper.

6.4 Results

We identified 1,162 children with cerebral palsy (59.7% male, mean age at study commencement 7.3 years, standard deviation 3.4 years) who had one or more outpatient encounters in the pre-COVID-19 and in the COVID-19 and/or post-COVID-19 time periods (**Table 6.1**). Almost three-quarters of children (74.4%) had outpatient encounters in all three time periods (**Figure 6.1**), and over 85% (n=993) had one or more telemedicine encounter. Compared with children identified in the NSW/ACT CP Register without an outpatient encounter, children with at least one encounter were younger, more likely to live in major cities and neighbourhoods with less socioeconomic disadvantage and more likely to have severe CP (GMFCS levels IV-V) and/or comorbidities**.**

A total of 48,896 encounters were recorded between January 2018 and May 2023, one-quarter (n=11,929, 24.4%) involved telemedicine. In the pre-COVID-19 time period only 1.4% (n=51) telemedicine encounters used an audiovisual medium (the remainder using telephone). The proportion of audiovisual telemedicine encounters increased during the COVID-19 period (29.1%, n=1,590) and was sustained post-COVID-19 (19%, n=525). Rates of total encounters and telemedicine encounters were higher in children in IRSD quintile 1 (most disadvantaged), in children living in major cities, and in children GMFCS levels IV-V (compared with those GMFCS levels I-III), and in those with epilepsy and/or intellectual disability (**Table 6.1**).

Table 6.1. Demographic and clinical details of children with cerebral palsy attending outpatient encounters at two children's hospitals, rates of total outpatient encounters and telemedicine encounters.

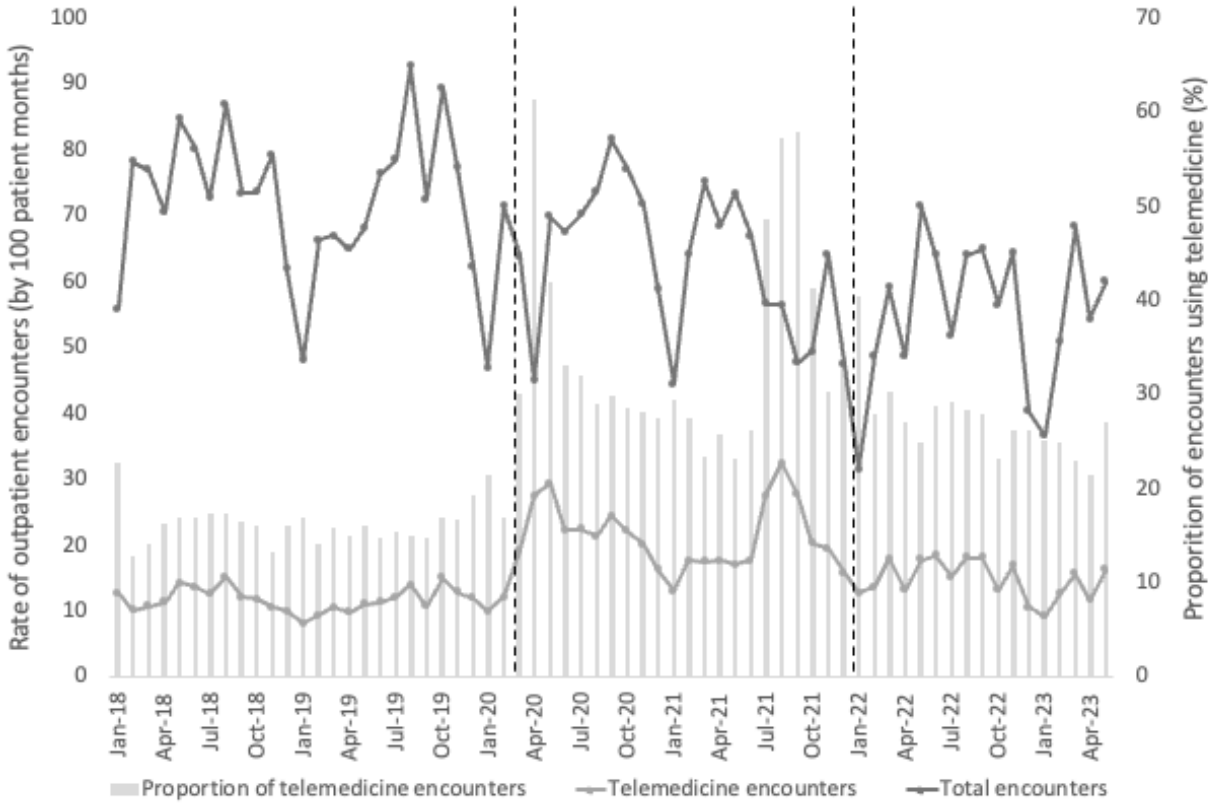
Demographic / clinical factor	Total children attending outpatient services n (%)	Total encounters per 100 persons Median (IQR)	P-value	Telemedicine encounters per 100 persons Median (IQR)	P-value
Total	1162 (100)	66.4 (56.5-73.5)		14.3 (11.9-18.2)	
Sex			0.71		0.83
Male	694 (59.7)	67.3 (57.5-73.1)		14 (11.5-18.9)	
Female	468 (40.3)	65.2 (53.2-76.7)			
Socioeconomic disadvantage (quintiles)			<0.001		<0.001
1 (most disadvantaged)	208 (18.2)	77.9 (65.4-89.9)		16.3 (13.5-21.2)	
2	157 (13.7)	49 (35.7-55.4)		13.4 (9.6-15.9)	
3	248 (21.7)	55.2 (48-73)		13.7 (10.9-16.9)	
4	237 (20.7)	65.4 (56.5-77.6)		15.2 (10.5-19.8)	
5 (least disadvantaged)	294 (25.7)	68 (57.1-77.9)		14.6 (11.9-20.4)	
Geographical remoteness			<0.001		<0.001
Major city	810 (70.7)	73.2 (59.9-80.6)		15.1 (12.6-19.4)	
Inner regional	302 (26.4)	49.3 (42.4-60.6)		13.2 (9.6-15.6)	
Outer regional / remote	34 (3.0)	41.2 (26.5-61.8)		11.8 (8.8-17.6)	

GMFCS			<0.001		<0.001
I-III	816 (70.2)	47.3 (39.5-53.3)		9.7 (8-12.4)	
IV-V	303 (26.1)	116.5 (102.9-127.3)		27.1 (22.4-35)	
Unknown	43 (3.7)	55.8 (39.5-72.1)		11.6 (9.3-18.6)	
Epilepsy			<0.001		<0.001
Yes	338 (29.1)	85.8 (68-93.3)		19.5 (16.2-25.1)	
None or resolved	665 (57.2)	51.7 (43-58.6)		10.7 (8.4-13.4)	
Unknown	159 (13.7)	55.8 (39.5-72.1)		11.6 (9.3-18.6)	
Intellectual disability			<0.001		<0.001
Yes	550 (47.3)	85.8 (68-93.3)		19.5 (16.2-25.1)	
None	432 (37.2)	42.4 (35.2-49.1)		8.1 (6.7-10.9)	
Unknown	180 (15.5)	60 (47.2-70)		12.8 (10-18.9)	

GMFCS Gross Motor Function Classification System; IQR Interquartile range

Monthly rates and proportion of total and telemedicine encounters changed over time (**Figure 6.2**). The median rate of total encounters decreased in consecutive time periods from 76.1 per 100 persons per month (IQR 66.4-78.6) (pre-COVID-19); to 65.6 (IQR 56.5-71.7) (COVID-19); and 56.5 (IQR 48.8-64.1 post-COVID-19), p< 0.001)). This decline was also noted across consecutive years when adjusting for age group (**Supplementary Table 9.8**). The median rate of telemedicine encounters increased between the pre-COVID-19 (rate 11.7 per 100 persons per month (IQR 10.6-12.7)) and COVID-19 time periods (rate 20.2 per 100 persons per month (IQR 17.6-24.4)) and decreased in the post-COVID-19 time period (rate 15.2 per 100 persons per month (IQR 12.7-17.8)) (p <0.001). Sensitivity analyses (excluding December and January) showed similar relationships between time periods. Differences in patterns of telemedicine rates and proportions were identified between major specialty types (**Table 6.2**). Some specialties (Rehabilitation and Neurology) provided a higher proportion of outpatient encounters using telemedicine across time periods (e.g. 29.9% and 39.6% in the pre-COVID-19 period, respectively), whereas others (Allied Health, Orthopaedics) provided a lower proportion (e.g., 4.7% and 3.4% in the pre-COVID-19 period, respectively). The proportion of encounters provided using telemedicine increased in all specialty groups during the COVID-19 period and declined post-COVID-19. Post-COVID-19, higher telemedicine use was sustained in Rehabilitation (40.0%) and Neurology (61.8%) specialties.

Figure 6.2. Rates of total and telemedicine outpatient encounters and proportion of telemedicine encounters for children with cerebral palsy at two specialist children's hospitals.



Dashed lines indicate change between COVID-19 time periods

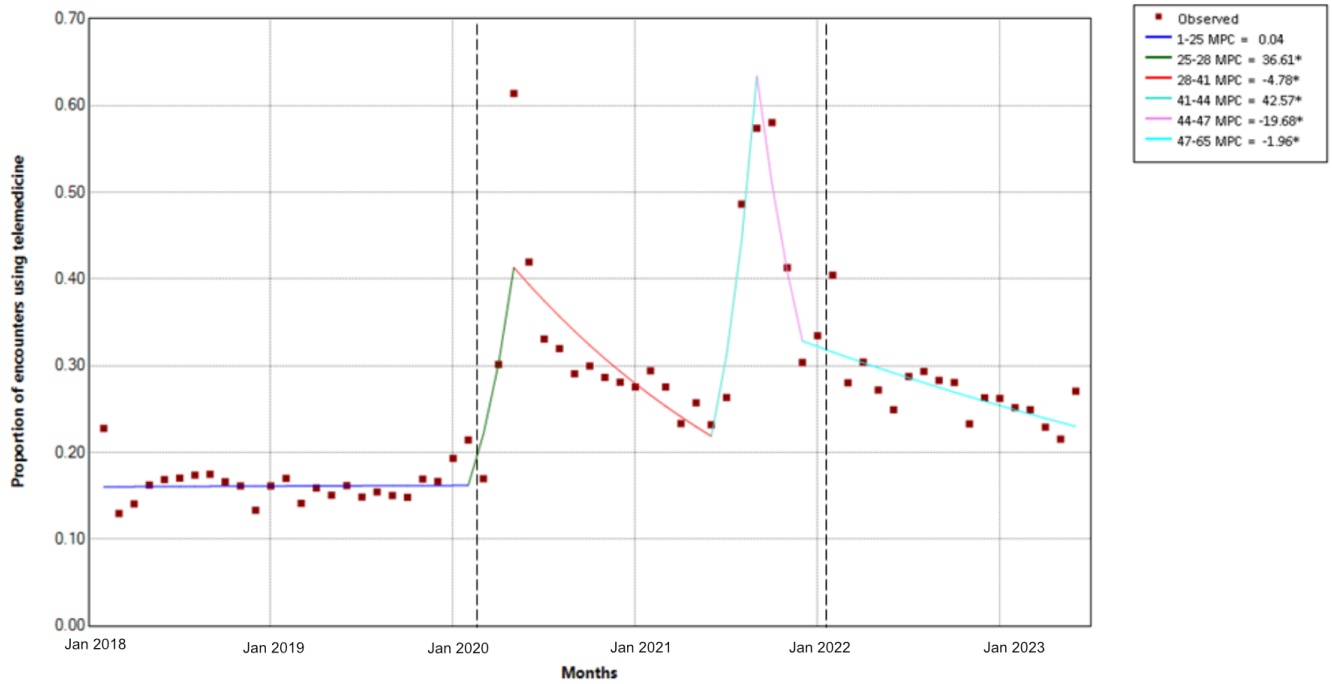
Table 6.2. Proportion of outpatient encounters provided using telemedicine for children with cerebral palsy by major specialty types during the COVID-19 pandemic.

	Rehabilitation (n=15 813)		Allied Health (n=150105)		Neurology (n= 3 120)		Orthopaedics (n= 4 862)		Other (n= 9 923)	
	Telemedicine	In-person	Telemedicine	In-person	Telemedicine	In-person	Telemedicine	In-person	Telemedicine	In-person
Pre-COVID-19 n (%)	1938 (29.9)	4551 (70.1)	323 (4.7)	6601 (95.3)	559 (39.6)	854 (60.4)	61 (3.4)	1738 (96.6)	666 (12.8)	4519 (87.2)
COVID-19 n (%)	2768 (48.8)	2905 (51.2)	320 (6.5)	4578 (93.5)	824 (74.6)	280 (25.4)	242 (14.5)	1428 (85.5)	1303 (45.5)	1562 (54.5)
Post-COVID-19 n (%)	1459 (40)	2192 (60)	209 (6.4)	3074 (93.6)	418 (61.8)	258 (38.2)	103 (7.4)	1290 (92.6)	736 (39.3)	1137 (60.7)

There were notable peaks in telemedicine use during the COVID-19 pandemic in March 2020 (61.4% total encounters) and August and September 2021 (57.4%, 58.0% total encounters) (**Figure 6.2**). Joinpoint analysis of the proportion of monthly outpatient encounters provided by telemedicine is shown in **Figure 6.3**. In the pre-COVID-19 period the proportion of telemedicine was stable (16%). There were two periods of rapid increase in telemedicine starting in February 2020 (monthly percent change 36.7%) and May to August 2021 (monthly percent change 24.9%) separated by a 13-month period where telemedicine declined by 4.6% per month. After August 2021, the proportion of encounters provided using telemedicine rapidly decreased by 19.7% per month for three months and after November 2021 continued to decline more slowly by 2% per month.

Children living in neighbourhoods of IRSD quintile 1 (most disadvantaged) had a higher median rate of outpatient encounters than children living in neighbourhoods of IRSD quintiles 2-5 in all time periods (**Figure 6.4a**). Children living in IRSD quintile 1 had a slightly higher median rate of telemedicine encounters in the pre-COVID-19 period, but there was no difference in rates by IRSD in subsequent time periods (**Figure 6.4b**). There was no evidence of a difference in the proportion of telemedicine encounters related to neighbourhood socioeconomic disadvantage across time periods.

Figure 6.3. Joinpoint regression of proportion of outpatient encounters using telemedicine for children with cerebral palsy (January 2018 and May 2023).

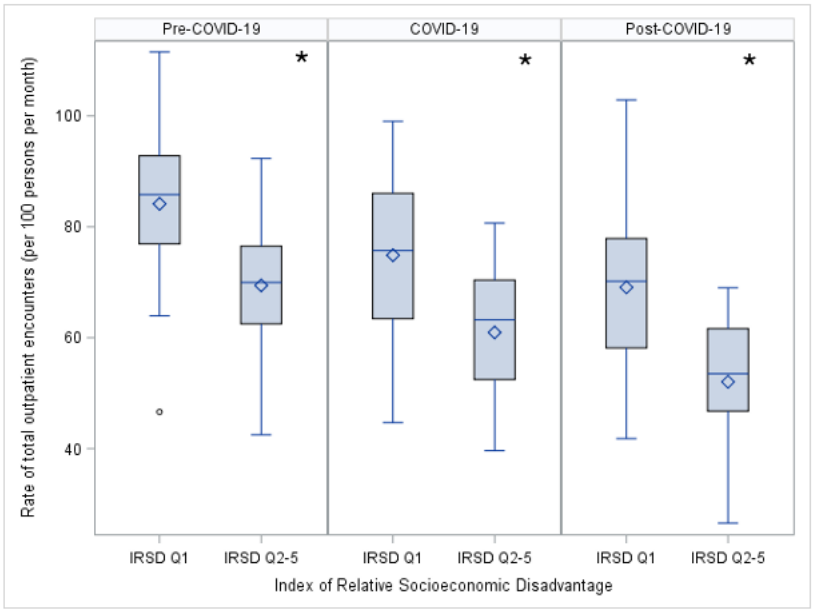


* indicates that the Monthly Percent Change (MPC) is significantly different from zero at the alpha= 0.05

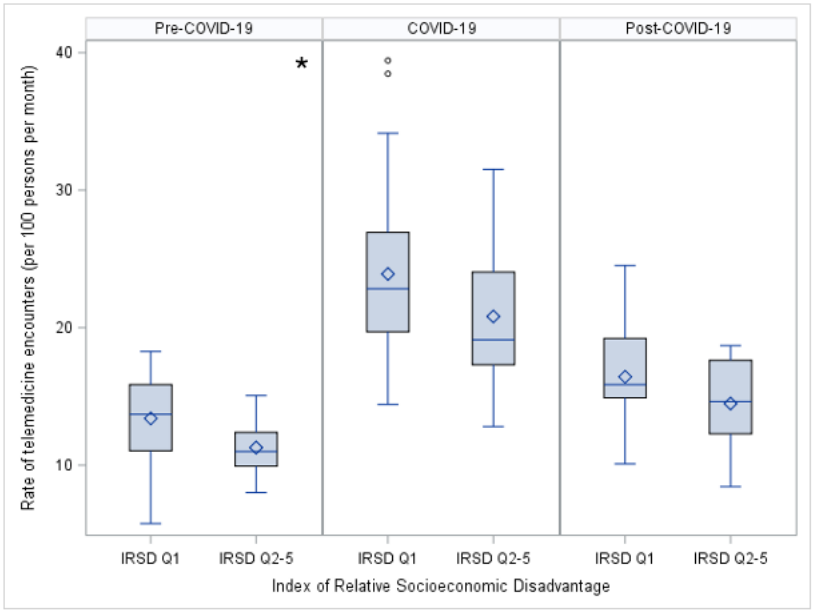
Dashed lines indicate change between COVID-19 time periods

Figure 6.4. Comparison of (a) rate of outpatient encounters and (b) rate of telemedicine encounters for children with cerebral palsy living in most socioeconomically disadvantaged areas (IRSD quintile 1) and children with cerebral palsy living in less disadvantaged areas (IRSD quintiles 2-5).

(a)



(b)



* indicates statistical evidence of difference ($p<0.05$)

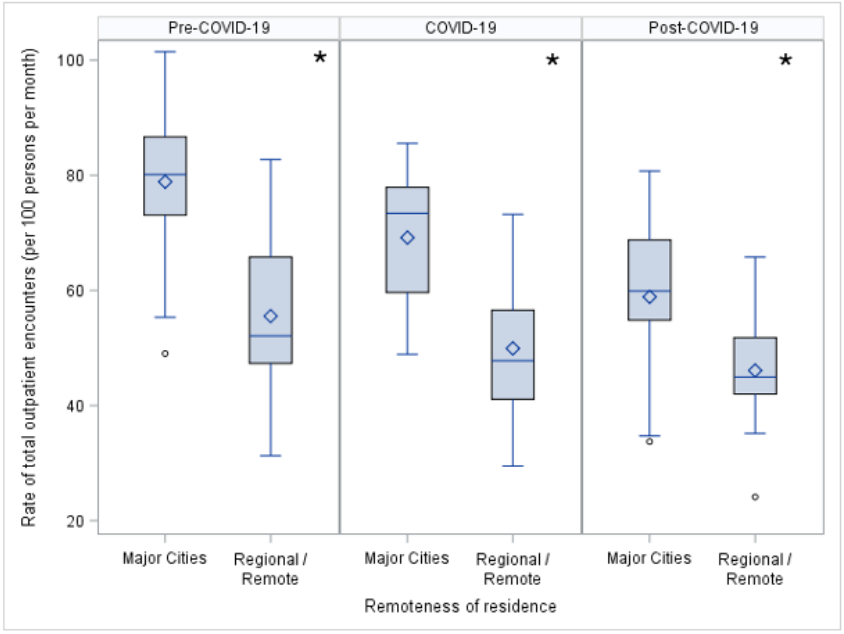
Children with CP living in regional and remote areas had lower median rates of total outpatient encounters than those living in major cities during all time periods (**Figure 6.5a**). Children living in regional and remote areas also had lower median rates of telemedicine encounters in pre-COVID-19 and COVID-19 periods, but there was no difference post-COVID-19 between groups (**Figure 6.5b**). There was no statistical evidence of a difference in the proportion of telemedicine encounters related to geographical remoteness across time periods.

6.5 Discussion

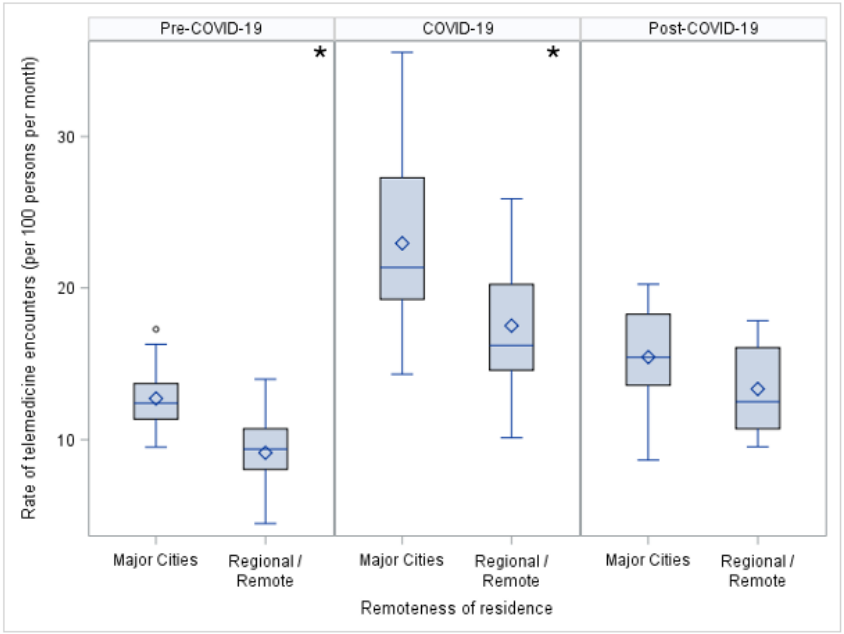
The COVID-19 pandemic and associated public health restrictions necessitated a profound change in health service provision including a substantial increase in telemedicine, particularly in high income countries.(21) Our results illustrate these changes for children with CP and suggest, following peaks of telemedicine use during the most severe COVID-19 restrictions in NSW, that the use of telemedicine for specialist and allied health outpatient telemedicine consultations for children with CP have continued to decline towards pre-pandemic levels.

Figure 6.5. Comparison of (a) rate of outpatient encounters and (b) rate of telemedicine encounters for children with cerebral palsy living in major cities and children with cerebral palsy living in regional and remote areas.

(a)



(b)



* indicates statistical evidence of difference ($p<0.05$)

We found inequity of access to these services for children living in regional and remote areas, with lower rates of total and telemedicine outpatient encounters for these children than those living in major cities before and during COVID-19. This inequity persisted for total outpatient encounters post-COVID-19, but the difference narrowed as the rates of telemedicine encounters declined much faster for children living in major cities. We found no evidence of inequity of access to these services with children experiencing neighbourhood socioeconomic disadvantage. We also found a decline in the frequency of total outpatient encounters for children with CP over time, which persisted even when adjusting for the age of the child.

The ongoing decline in telemedicine we have observed is an interesting finding given the political investment in sustaining telemedicine after the pandemic.(3) Some of this decline may be due to patient and clinician preference for in-person consultations,(22, 23) including for families of children with neurodevelopmental disorders,(15) reported during the pandemic. Studies have also described challenges that accompanied pandemic telemedicine experiences, including difficulties with managing the technology, difficulties for families to implement and keep children engaged in therapy programs (23, 24) and clinician-reported challenges of physical assessment, and establishing and maintaining rapport,(12, 25) perhaps influenced by (at the time) less familiarity with service delivery model, the requirement for different skills, and the known learning curve.(26) Similar to another study,(14) our results suggest that telemedicine may be harder to incorporate in some types of consultations such as those with strong

reliance on physical examination (e.g., allied health, orthopaedics), or where an intervention is required (e.g., botulinum toxin injections, serial casting). There is less reporting of more positive experiences of telemedicine in children with CP, and our research partners also identified advantages of telemedicine for families, including reducing the burden of in-person attendances and time away from other responsibilities (e.g., employment, caring roles), and less likelihood of cancellation related to intercurrent illnesses.

An often-reported benefit of telemedicine is improved patient access to health care by reducing travel.(27, 28) The inequities in telemedicine outpatient encounters for children living in regional and remote areas in our study pre and during COVID-19 is therefore a key service delivery concern. Potential contributing factors may include known limitations of digital infrastructure in rural areas, and potential differences in digital literacy and access to internet devices.(29) Some of these children may also access services local to them and/or outside of Sydney, although availability of specialist paediatric services outside metropolitan areas in Australia is typically limited. The improvement in equity of telemedicine for children and families from regional and remote areas noted since the pandemic is reassuring and may indicate that these families are now more routinely offered choice of telemedicine.

That we found no evidence of differences in telemedicine use related to socioeconomic disadvantage is also reassuring. Inequity in access to telemedicine related to socioeconomic disadvantage has been found in similar patient groups.(14) It may be that more sensitive measures of socioeconomic disadvantage are required to identify where digital poverty impacts telemedicine use, or that telephone encounters are less impacted.

The decline in total outpatient encounters for these children over time requires further investigation. Of primary consideration is whether these children have unmet medical needs, and whether these have been impacted by less frequent outpatient visits. Alternatively, the reduction may reflect a perceived decreased need for frequent specialist visits and/or better integration with local services. At a service level this reduction may be impacted by known workforce issues in the health system (e.g., staff retention) during the time studied (particularly after the pandemic).(30)

Strengths of this study include use of a CP register to identify the study population and the timeframe enabling a description of telemedicine use before, during and after the COVID-19 pandemic. Limitations include the restriction to two metropolitan specialist children's hospitals. NSW has a third paediatric hospital (John Hunter Children's Hospital), that provides a range of specialist outpatient services and may account for some children seen in the NSW/ACT CP Register who are not seen in our cohort. We have

also not collected information on health care provided elsewhere within the public health system, or the primary care and private health systems. Whether the changes we have observed will generalise to other settings is also uncertain.

Further research is needed to better delineate the role(s) of telemedicine for children with CP and other neurodevelopmental disorders into the future. First amongst this is re-evaluating the telemedicine preferences of families after the pandemic, as much of the recent research was performed during the extraordinary times during the pandemic.(12) Additionally, further work is required to identify effective digital health technologies that can support the identified challenges of telemedicine (e.g., physical examination, developing and maintaining rapport). Studies among children with other chronic health conditions have identified that technology (e.g. SMS technology, apps), can promote positive health outcomes when specifically targeted at a component of care.(31, 32) Digital health technology is an acceptable medium for health care for adolescents and young adults,(33) but must be designed acknowledging concerns around privacy, risks of labelling and identity and accessibility,(34) and consider equity of data and device access from the outset.(35) Children and young people living in regional and remote areas must be prioritised in this work.

Telemedicine has been identified as an important part of digital health strategy, but our results suggest that further support will be required to sustain rates of use higher than

that seen pre-pandemic. This support includes reducing barriers to access including broadband internet, developing best practices for telemedicine, and identifying components of care in children and young people with CP that can be addressed through digital health technologies outside of in-person outpatient attendances (e.g., coordination of investigations). Combined, this work can help deliver on the promise of telemedicine, including to improve access to health care and health outcomes for children with CP.

6.6 Chapter Synopsis

In **Chapter 6**, telemedicine for outpatient services at specialist children's hospitals was found to be declining in the period after COVID-19 pandemic public health restrictions and approaching levels seen pre-pandemic. Compared with children with CP living in major cities, telemedicine access was reduced in children with CP living in regional and remote areas. This study is submitted for peer review in Journal of Child Neurology. **Chapter 7** will focus on outpatient services, including for Aboriginal and/or Torres Strait Islander children with CP, continuity of care and unplanned hospital care.

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Chapter 7 Outpatient encounters, continuity of care and unplanned hospital care for children and young people with cerebral palsy in New South Wales, Australia.

7.1 Published Manuscript

This chapter has been adapted and reformatted for publication as part of this thesis.

The citation for the published manuscript is:

> Paget SP, Mcintyre S, Schneuer FJ, et al. Outpatient encounters, continuity of care, and unplanned hospital care for children and young people with cerebral palsy. Dev Med Child Neurol. 2024;66(6):733-743. doi:10.1111/dmcn.15800

A copy of the published article as well as a statement of the specific contributions of the co-authors can be found in **Chapter 10 - Appendix B**.

7.2 Introduction

Cerebral palsy (CP) is the most common cause of physical disability in childhood and is defined by disorder(s) of movement and posture that arise from a disturbance of early brain development.(1) Comorbidities, such as epilepsy, respiratory and gastrointestinal diseases, are common in CP (2) and a frequent cause of (planned and/or unplanned) hospital admissions and emergency department (ED) presentations.(3, 4) Children and young people with CP require hospital services substantially more than the general population,(5, 6) with Gross Motor Function Classification System (GMFCS) levels IV-V, epilepsy, gastrostomy use, and intellectual disability associated with increased health service utilisation.(7)

Outpatient services have an important role in the management of CP, providing early detection and preventive measures for health problems, therapy, medical interventions, and assessment for and planning of surgery.(8) Good outpatient management of CP and associated comorbidities may reduce adverse health outcomes. Missing outpatient appointments has been associated with unplanned hospital admissions and ED presentations (unplanned hospital care) in children with CP and related neurological conditions.(9)

Access to health services is a key measure of health system performance. Health service access may be influenced by sociodemographic factors such as private health insurance, distance to major metropolitan hospitals, socioeconomic disadvantage, and parental employment status.(7, 10) Continuity of care is a key measure of the quality of outpatient services.(11, 12) Improved continuity of care has been associated with decreased hospital admission duration in children with CP,(13) and decreased hospital admissions and emergency department presentations in other childhood populations.(14-16)

Aboriginal and/or Torres Strait Islander peoples comprise 3.8% of the total Australian population.(17) Aboriginal and/or Torres Strait Islander Australians experience substantial health inequities, caused in part by the continuing effects of colonisation and barriers to culturally appropriate and accessible health care. Addressing these health inequities is recognised as a major health care priority.(18) There is a lack of research exploring equity of access to health services for Aboriginal and/or Torres Strait Islander people living with CP.

Little is known about the relationships between outpatient encounters, quality of care and unplanned hospital care utilisation in people with CP. We aimed to (i) explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP and (ii) describe patterns of

outpatient encounters for children and young people with CP and identify if there are variations related to sociodemographic and clinical factors, and for Aboriginal and/or Torres Strait Islander children and young people.

7.3 Methods

7.3.1 Population

We conducted a population-based record linkage study of children and young people with CP using multiple administrative health data sets. The study population was derived from the New South Wales (NSW)/Australian Capital Territory (ACT) Cerebral Palsy Register. We included all children with confirmed CP on the Register at the time of linkage (January 20, 2019), born between 1994-2018 and who were alive at the beginning of the study period (1st July 2015 to 30th June 2020).

7.3.2 Data sources

The NSW/ACT CP Register is a population-based database, with multiple ascertainment strategies, of individuals with CP who were born or live in NSW or ACT. Health data sets included the NSW Non-Admitted Patient Data Collection (NAP) which contains data from all non-admitted (outpatient) encounters with clinical and/or therapeutic content provided by NSW Health facilities; NSW Emergency Department Data Collection (EDDC)

which contains data from presentations to NSW public hospital emergency departments; and NSW Admitted Patient Data Collection (APDC) which contains data from all admissions to NSW hospitals (public and private). Deaths of individuals were identified through the NSW Registry of Births, Deaths and Marriages (RBDM) Death Registrations. Datasets were probabilistically linked by the New South Wales (NSW) Centre for Health Record Linkage.

7.3.3 Clinical and sociodemographic variables

Clinical CP variables derived from the NSW/ACT CP Register included predominant motor type, topography, GMFCS (categorised into GMFCS levels I-III and GMFCS levels IV-V) and the presence of epilepsy and/or intellectual disability. Sociodemographic variables of children included sex and maternal country of birth (dichotomised as born in Australia or overseas). Postcode of residence was used to determine geographical remoteness and socioeconomic disadvantage. Socioeconomic disadvantage was derived from the Index of Relative Socioeconomic Disadvantage (IRSD) and grouped into quintiles (quintile 1 most disadvantaged, and quintile 5 least disadvantaged).(19) Geographical remoteness (metropolitan; regional/remote areas) was defined using Australian Statistical Geography Standard.(20)

7.3.4 Aboriginal and/or Torres Strait Islander status

Children and young people who identify as Aboriginal and/or Torres Strait Islander were identified by applying the "Enhanced Reporting of Aboriginality" algorithm (21) to "self-reported Aboriginality" as captured in the APDC and EDDC. It is important to acknowledge that being of Aboriginal and/or Torres Strait Islander background is not considered itself as a risk factor; rather our focus is the recognised systemic health inequities that Aboriginal and Torres Strait Islander people face.

7.3.5 Study measures

To evaluate outpatient service utilisation, outpatient clinic encounters between 1st July 2015 and 30th June 2020 (five years) were identified for each child and young person. Each child was dichotomised as having any outpatient encounters or not. The frequency of outpatient encounters during the study period was not normally distributed, with substantial positive skew. The total number of outpatient encounters for each child was categorised into tertiles (no use, tertile 1 ('low use'), tertile 2 ('medium use'), tertile 3 ('high use')). Continuity of care was defined using the Usual Provider of Care Index (UPCI). (22) We designated each child a usual provider of care by identifying the specialty service (acknowledging location and specialty type (e.g., rehabilitation, general medicine, neurology)) with whom they had the highest frequency of outpatient encounters during the study period. The UPCI was then defined as the proportion of total outpatient encounters conducted with the usual provider of care. We decided to calculate UPCI only

for those children who had five or more outpatient encounters during the study period; those with less than five (including none) encounters (n=813) were assigned a UPCI score of zero. Multidisciplinary clinics were categorised to the primary service specialty. We categorised continuity of care at an individual level into three categories, a-priori and consistent with recent studies:(15, 23) low continuity of care (UPCI <0.4), medium continuity of care (0.4≤ UPCI <0.7) and high continuity of care (UPCI ≥0.7). Outpatient encounters were also evaluated by hospital type (paediatric/metropolitan/regional), mode of delivery (in-person/virtual care (including telephone), professional grouping (medical/allied health and nursing/procedural/diagnostic) and specialty groups.

Unplanned hospital care was defined as: (i) any presentation to an emergency department; and (ii) urgent hospital admission due to an emergency presentation. The frequency of emergency department presentations was highly skewed and was therefore categorised into clinically and numerically meaningful groups defining none, one, two-three, and four or more presentations. Principal diagnosis for urgent hospital admissions was also identified based on the International Classification of Diseases, Revision 10, Australian Modification (ICD10-AM) and categorised according to disease chapters. Where the principal diagnosis was classified as either cerebral palsy (ICD10-AM G80.0-G80.9) or 'Symptoms, signs and abnormal clinical and laboratory findings' ICD10-AM R00-R99), the next diagnosis code was used.

7.3.6 Statistical analysis

Initial analysis of the frequency of outpatient encounters, ED presentations and urgent hospital admissions highlighted these data to be skewed and they were therefore described using non-parametric measures (medians, interquartile ranges (IQR)). Clinical and sociodemographic variables were compared for those who had and had not accessed each type of health service using contingency tables and chi-squared tests where a p-value <0.05 was considered statistically significant. Rates of outpatient encounters, ED presentations and urgent hospital admissions were calculated as time-at-risk and reported as per 100 person years. Correlation between continuous variables (e.g., UPCI and frequency of outpatient encounters) was examined using Pearson's correlation coefficient.

The association between outpatient encounters and continuity of care with ED presentations and urgent hospital admissions was evaluated using multivariable multinomial (ED presentations) and binomial (urgent hospital admissions) logistic regression models adjusted for clinical and sociodemographic variables. For the logistic regression models, only children who completed the whole study period (five years) (n=3,058) were included, and where appropriate variables were dichotomised (present / not present or unknown). Associations between outpatient encounter or UPCI tertiles and unplanned hospital care was described as odds ratios with 95% confidence intervals. Stratified analysis by sociodemographic factors and clinical severity, and

Aboriginal and/or Torres Strait Islander status was conducted to explore any potential differences in the association by specific sub-group. Analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC, USA). The study was approved by the NSW Population and Health Services Research Ethics Committee (2019/ETH11532), including a waiver of consent for participants, and the Aboriginal Health and Medical Research Council of NSW Human Research Ethics Committee (1861/21).

7.4 Results

Of 3,267 children and young people with CP, we identified 2738 (83.8%) who attended at least one outpatient encounter over the five-year study period (**Table 7.1**). The youngest child was in their first month of life at their first encounter and the oldest person was 26 years at their last encounter. A higher proportion of those with at least one outpatient encounter lived in metropolitan areas (58.6% vs. 52.3%), had predominant dyskinetic motor type (11.6% vs. 5.7%) and/or epilepsy (27.1% vs. 18.3%) (**Table 7.1**).

Table 7.1. Outpatient encounters and continuity of care for children and young people with cerebral palsy

Demographic, Clinical Factor and Aboriginal status	Total	Outpatient Encounters			Level of Continuity of Care (UPCI)			
		Persons with 1+ encounter N (%)	Persons with no encounters N (%)	P	Low (UPCI < 0.4) N (%)	Medium (UPCI 0.4-0.7) N (%)	High (UPCI > 0.7) N (%)	P
Total	3267 (100)	2738 (83.8)	529 (16.2)		1625 (49.7)	942 (28.8)	700 (21.4)	
Sex				0.056				0.9
Male	1903 (58.2)	1576 (57.6)	327 (61.8)		952 (58.6)	543 (57.6)	408 (58.3)	
Female	1364 (41.8)	1162 (42.4)	202 (38.2)		673 (41.4)	399 (42.4)	292 (41.7)	
Maternal Country of Birth				0.896				<0.001
Australia	2215 (73.6)	1860 (73.5)	355 (73.8)		1151 (76.7)	607 (69.1)	457 (72.2)	
Overseas	796 (26.4)	670 (26.5)	126 (26.2)		349 (23.3)	271 (30.9)	176 (27.8)	
Aboriginality				0.016				0.22
Aboriginal	262 (8)	234 (8.5)	28 (5.3)		126 (7.8)	87 (9.2)	49 (7)	
Not Aboriginal	3005 (92)	2504 (91.5)	501 (94.7)		1499 (92.2)	855 (90.8)	651 (93)	
Remoteness				0.013				<0.001
Metropolitan Areas	1851 (57.6)	1597 (58.6)	254 (52.3)		764 (50.5)	803 (51)	598 (63.5)	
Regional / Remote Areas	1361 (42.4)	1128 (41.4)	233 (47.9)		750 (49.5)	771 (49)	343 (36.5)	
IRSD Quintile				0.326				<0.001
1 (most disadvantaged)	591 (18.4)	518 (19)	73 (15)		286 (18.2)	188 (20)	117 (16.8)	
2	623 (19.4)	526 (19.3)	97 (20)		325 (20.6)	174 (18.5)	124 (17.8)	
3	705 (22)	591 (21.7)	114 (23.5)		360 (22.9)	217 (23.1)	128 (18.4)	
4	669 (20.8)	563 (20.7)	106 (21.8)		333 (21.2)	185 (19.7)	151 (21.7)	

5 (least disadvantaged)	623 (19.4)	527 (19.3)	96 (19.8)		269 (17.1)	177 (18.8)	177 (25.4)	
GMFCS				<0.001				<0.001
I-III	2141 (65.5)	1758 (64.2)	383 (72.4)		1037 (63.8)	563 (59.8)	541 (77.3)	
IV-V	768 (23.5)	679 (24.8)	89 (16.8)		386 (23.8)	275 (29.2)	107 (15.3)	
Unknown	358 (11)	301 (11)	57 (10.8)		202 (12.4)	104 (11)	52 (7.4)	
Motor type				<0.001				<0.001
Spastic Unilateral	1070 (35.3)	860 (26.4)	210 (42.5)		508 (34.3)	259 (29.4)	303 (45.5)	
Spastic Bilateral	1381 (45.6)	1157 (35.5)	224 (45.3)		694 (46.8)	420 (47.7)	267 (40.1)	
Dyskinetic	322 (10.6)	294 (9.0)	28 (5.7)		134 (9)	124 (14.1)	64 (9.6)	
Ataxic	128 (4.2)	103 (3.2)	25 (5.1)		78 (5.3)	31 (3.5)	19 (2.9)	
Other	129 (4.3)	122 (3.7)	7 (1.4)		69 (4.7)	47 (5.3)	13 (2.0)	
Intellectual Disability				<0.001				<0.001
Yes	1551 (47.5)	1321 (48.2)	230 (43.5)		777 (47.8)	505 (53.6)	269 (38.4)	
No	1222 (37.4)	998 (36.4)	224 (42.3)		580 (35.7)	298 (31.6)	344 (49.1)	
Unknown	494 (15.1)	419 (15.3)	75 (14.2)		268 (16.5)	139 (14.8)	87 (12.4)	
Epilepsy				<0.001				<0.001
Yes	840 (25.7)	743 (27.1)	97 (18.3)		408 (25.1)	299 (31.7)	133 (19)	
None or resolved	1728 (52.9)	1405 (51.3)	323 (61.1)		843 (51.9)	452 (48)	433 (61.9)	
Unknown	699 (21.4)	590 (21.5)	109 (20.6)		374 (23)	191 (20.3)	134 (19.1)	

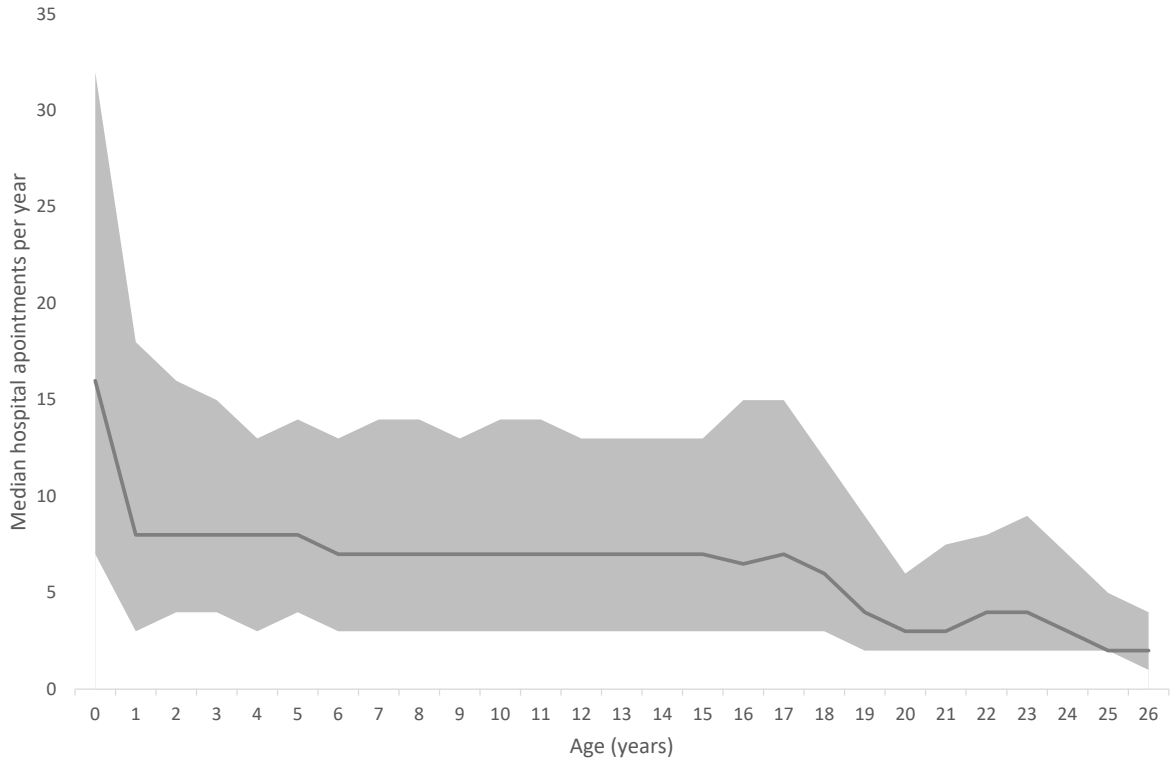
GMFCS Gross Motor Function Classification System; IRSD Index of Relative Socioeconomic Disadvantage; UPCI Usual Provider of Care Index

P values relate to differences between health service use groups by demographic, clinical factors or Aboriginal status; Motor type 'Other' group combines Hypotonia and Early at risk

Totals may not sum where missing data < 10%

There was a total of 123,463 outpatient encounters during a total of 13,376 person-years, with a median rate of 620 per 100 person years (IQR 240-1200). The median number of encounters per year for each individual was stable through childhood (median=6) and declined into adulthood (median=3) (**Figure 7.1**).

Figure 7.1. Median rate of hospital outpatient encounters by age for children and young people with cerebral palsy.



* Shaded area represents interquartile range

For children aged 0-18 years, 65.5% of outpatient encounters occurred at children's hospitals and 18% of outpatient encounters occurred using virtual care (**Table 7.2**). Half

of outpatient encounters (48.1%) were coordinated with at least one other encounter that day (28.3% two, 12.0% three, and 7.8% four or more encounters). Almost all encounter types were either medical (49%) or allied health and nursing (48%) (**Table 7.2**). Rehabilitation (35%), neurology (14.5%) and general medicine (14%) were the most common medical specialty types.

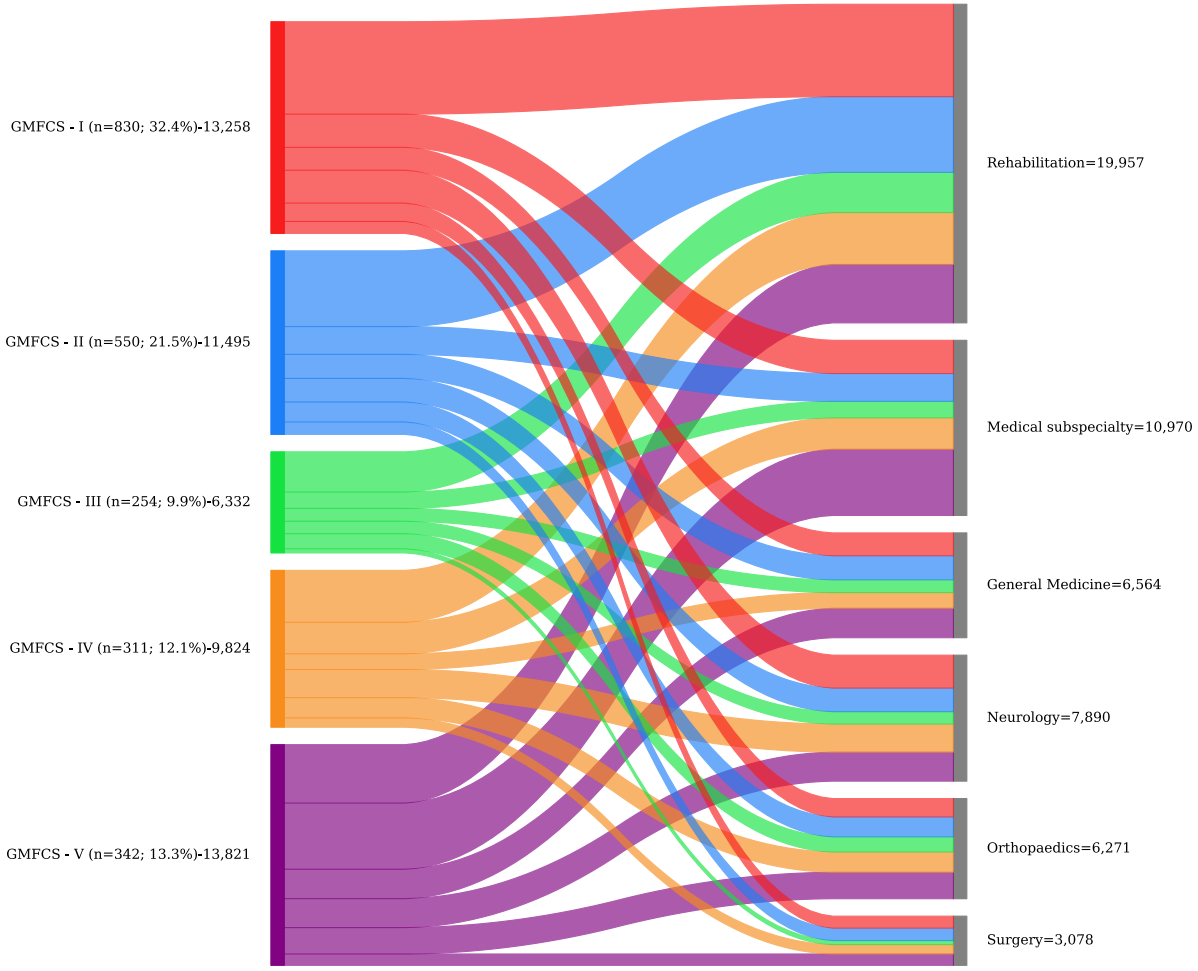
Table 7.2. Comparison of frequency and proportions of outpatient encounters by age group for children and young people with cerebral palsy in 2015-2020.

Age Group	Total (N)	Facility Location N (%)			Contact Mode N (%)		Outpatient Encounter type N (%)		
		Paediatric	Metro	Regional / Rural	In Person	Virtual Care	Medical	Allied Health and Nursing	Diagnostic and Procedural
0-4 years	27340	15933 (58.3)	6930 (25.3)	4477 (16.4)	22639 (82.8)	4701 (17.2)	12685 (46.4)	14203 (51.9)	161 (0.6)
5-9 years	36195	24687 (68.2)	4116 (11.4)	7392 (20.4)	29884 (82.6)	6311 (17.4)	19076 (52.7)	16385 (45.3)	181 (0.5)
10-14 years	33057	22746 (68.8)	4582 (13.9)	5729 (17.3)	27403 (82.9)	5654 (17.1)	15592 (47.2)	16798 (50.8)	110 (0.3)
15-19 years	22062	12548 (56.9)	4645 (21.1)	4869 (22.1)	17108 (77.5)	4954 (22.5)	11356 (51.5)	9984 (45.3)	146 (0.7)
20+ years	4809	121 (2.5)	3194 (66.4)	1494 (31.1)	3841 (79.9)	968 (20.1)	2244 (46.7)	2050 (42.6)	95 (2)
Total	**123463**	**76035 (61.6)**	**23467 (19)**	**23961 (19.4)**	**100875 (81.7)**	**22588 (18.3)**	**60953 (49.4)**	**59420 (48.1)**	**693 (0.6)**

Totals may not add up to totals due to missing data

Children and young people were reviewed by a median of 7 (IQR 2-16) different specialty services during the five-year period. Although representing one-quarter of the population, those with GMFCS IV-V had disproportionally high outpatient services use across different specialty types (**Figure 7.2**). Similar relationships were seen with major comorbidities (**Supplementary Figure 9.5**). Two-thirds of children (65.7%) had an identified usual provider of care (UPC); most of these were based at a children's hospital (88.2%). Common UPC specialty types were rehabilitation (47.9%), general medicine (14.6%) and neurology (15.5%). The median UPCI for children was 0.56 (IQR 0.40-0.78), indicating almost 60% of total outpatient appointments were conducted with their UPC (**Supplementary Figure 9.6**). Children receiving high levels of continuity of care (UPCI >0.7) were over-represented in those with a mother born overseas, living in metropolitan areas, from less socioeconomic disadvantaged areas, spastic unilateral motor type and neither epilepsy nor intellectual disability (**Table 7.1**). Outpatient utilisation (total number of outpatient encounters) and UPCI were only weakly correlated (Pearson's correlation coefficient 0.12).

Figure 7.2. Major outpatient speciality use in children with cerebral palsy grouped by Gross Motor Function Classification System.



Two-thirds (65.9%) of children and young people had at least one ED presentation (median 3, IQR 1-6)) and 33.4% had at least one urgent hospital admission (median 2, IQR 1-4) (**Supplementary Table 9.9**). For children aged 0-18 years, 42.4% of urgent admissions occurred at children's hospitals. The most common principal diagnoses at admission were diseases of the respiratory system (30.1%) and diseases of the nervous

system (17.8%), although in early adulthood injuries, diseases of the digestive system and mental health disorders became more common (**Supplementary Table 9.10**).

7.4.1 Outpatient utilisation, continuity of care and unplanned hospital care

Multinomial and binomial analysis of factors associated with ED presentations and urgent hospital admissions are shown in **Table 7.3**. Children and young people with high outpatient utilisation had substantially increased odds of having 4+ ED presentations (aOR 5.00; 95%CI 3.62-6.92) and an urgent hospital admission (aOR 3.38 95%CI 2.63-4.35). Compared with high continuity of care, low and medium continuity of care were also associated with increased odds of having 4+ ED presentations (low: aOR 2.34; 95%CI 1.71-3.19; medium: aOR 1.63; 95%CI 1.21-2.20) and one or more urgent hospital admissions (low: aOR 2.02; 95%CI 1.57-2.61; medium: aOR 1.88; 95%CI 1.48-2.39). Stratified analysis using major sociodemographic and clinical factors and for Aboriginal and/or Torres Strait Islander people is shown in **Figure 7.3** and **Supplementary Figure 9.7**. These analyses suggested that associations between UPCI and unplanned hospital care persisted when limited to children with CP and markers of less severity (e.g., GMFCS levels I-III, no epilepsy and/or no intellectual disability).

Table 7.3. Univariable, multivariable and multinomial logistic regression of Outpatient factors associated with Emergency Department presentations and urgent hospital admissions in children and young people with cerebral palsy.

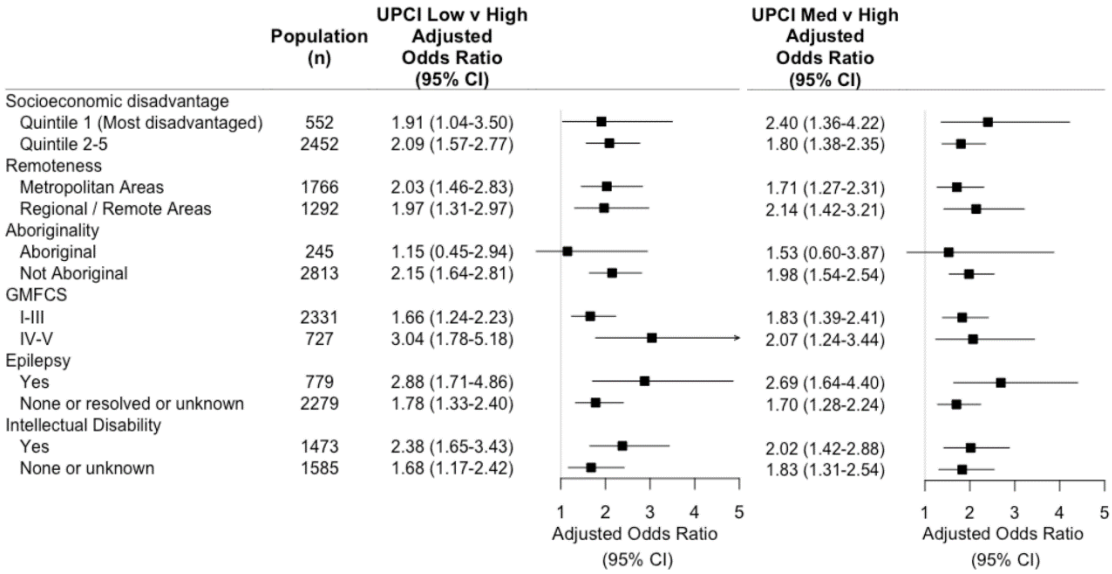
Outpatient Factors	ED Presentations*				Urgent Hospital Admissions*	
	Univariable OR (95%CI)	Multivariable 1 Presentation OR (95%CI)	Multivariable 2-3 Presentations OR (95%CI)	Multivariable 4+ Presentations OR (95%CI)	Univariable OR (95%CI)	Multivariable OR (95%CI)
Outpatient Use						
No use	0.24 (0.19 - 0.30)	0.37 (0.27 - 0.52)	0.21 (0.14 - 0.3)	0.15 (0.09 - 0.23)	0.22 (0.15 - 0.33)	0.22 (0.15 - 0.34)
Low (1st tertile)	REF	REF	REF	REF	REF	REF
Medium (2nd tertile)	1.40 (1.14 - 1.72)	1.09 (0.81 - 1.47)	1.47 (1.10 - 1.97)	2.40 (1.76 - 3.27)	1.78 (1.43 - 2.21)	1.90 (1.48 - 2.43)
High (3rd tertile)	2.34 (1.88 - 2.93)	1.23 (0.88 - 1.72)	1.88 (1.36 - 2.58)	5.00 (3.62 - 6.92)	4.09 (3.31 - 5.07)	3.38 (2.63 - 4.35)
Usual Provider of Care Index						
Low	0.72 (0.60 - 0.87)	1.11 (0.81 - 1.52)	1.38 (1.02 - 1.87)	2.34 (1.71 - 3.19)	1.17 (0.95 - 1.44)	2.02 (1.57 - 2.61)
Medium	1.68 (1.35 - 2.09)	1.35 (1.00 - 1.81)	1.3 (0.97 - 1.73)	1.63 (1.21 - 2.20)	2.48 (1.98 - 3.09)	1.88 (1.48 - 2.39)
High	REF	REF	REF	REF	REF	REF

ED Emergency Department, OR odds ratio, 95%CI 95% confidence interval, REF reference group

Multivariable models adjusted for demographic variables (sex, age, country of birth, index of relative socioeconomic disadvantage, residential location), Aboriginal and/or Torres Strait Islander background and clinical variables (Gross Motor Function Classification System, motor type, intellectual disability, epilepsy

* Odds ratios presented compared with no ED presentations and no urgent hospital admissions respectively

Figure 7.3. Association between continuity of care and urgent hospital admissions stratified by major sociodemographic and clinical factors, and Aboriginal status.



95% CI 95% Confidence Interval; GMFCS Gross Motor Function Classification System; UPCI Usual Provider of Care Index

7.4.2 Aboriginal and/or Torres Strait Islander children with CP

Most (89%; n=234/262) Aboriginal and/or Torres Strait Islander children and young people with CP attended at least one outpatient encounter during the five years (rate 770 per 100 person years (IQR 300-1580)). During the same timeframe, 79.7% (n=209) of Aboriginal and/or Torres Strait Islander children had at least one ED presentation and 44.3% (n=116) at least one urgent hospital admission. Almost half (47.3%, n=124) of Aboriginal and/or Torres Strait Islander children had 4+ ED presentations and they were more likely to live

in regional or remote areas (76.4%) than those with <4 ED presentations (64.4%) (p=0.036). Stratified analysis showed no statistical evidence of a relationship between UPCI and urgent admissions (OR (low UPCI v high UPCI) 1.15; 95% CI 0.45-2.94) or 4+ ED presentations (OR low UPCI v high UPCI) 1.95; 95% CI 0.62-6.13).

7.5 Discussion

This study is one of the first to examine outpatient service utilisation in children and young people with CP at a population level, and the first epidemiological study of outpatient service utilisation in Aboriginal and/or Torres Strait Islander children and young people with CP. Our results highlight that over four in five children with CP access outpatient services, with many using services frequently. Continuity of care was inequitably distributed, with a higher proportion of high continuity of care (high UPCI) for children living in metropolitan areas and in least socioeconomically disadvantaged areas. Less access to continuity of care was strongly associated with increased unplanned hospital care utilisation. These associations persisted when analyses were restricted to those with less severe CP.

Our findings build on other studies that show the frequency and complexity with which children and young people with CP access the health system (24) and the associated carer burden.(25) Our findings emphasise the need for better continuity of care and care coordination, particularly in those with complex health needs. Our results suggest that

continuity of care is less accessible for those living in the most socioeconomically disadvantaged and in regional and remote areas, reflecting the known challenges of recruiting and retaining health service staff in these areas.(26) In other CP populations, access to care coordination has also shown a socioeconomic gradient, with unmet need for care coordination increasing with decreasing household income.(27)

Recent Australian epidemiological research has highlighted that while one in five Aboriginal and/or Torres Strait Islander children with CP live in socioeconomically disadvantaged remote areas, most live in more populated areas.(28) While our study suggests that broadly access to outpatient services for Aboriginal and/or Torres Strait Islander children with CP is similar to that of the whole population, it is essential that a focus on equity of access is maintained, prioritising the needs of those identified as having barriers to access to continuity of care and care coordination, and that care is provided in culturally appropriate ways.(29, 30)

Continuity of care and interrelated concepts such as care coordination acknowledge the importance of the health care system working as a cohesive whole (integrated care). Continuity of care highlights the importance of stable relationships between patient and provider(s) and between providers within and between settings, and sharing of information and knowledge about patients, consistent with patients' needs and preferences.(23, 31) Continuity of care is the more frequently used concept in research, likely as it is easier to measure.(32) Improved continuity of care has been associated with

reductions in ED presentations (14, 33) and hospitalisations (14, 23, 33) overall and for children with CP.(13) Coordinated care programs that provide standardised systems of musculoskeletal evaluation for children with CP are available in some countries (34, 35) have been credited with decreasing incidence of musculoskeletal deformities.

Recent government reports have highlighted the need to support navigation of the health care system for families with children with complex needs and offered suggestions for improvement.(36) The COVID-19 pandemic has accelerated the uptake of telemedicine, including for children with neurodevelopmental disabilities, and offers the potential to decrease the burden and costs associated with frequent outpatient visits for both families and the health system. There is an opportunity to further develop telemedicine consultations (37) to augment in-person health care and reduce travel and time lost from work, school and family life. Similarly, better care coordination between speciality teams and hospitals, including development of integrated electronic health records, (38) use of care navigators, and outreach clinics can improve consumer experience of health care quality,(39) reduce travel,(40) and potentially reduce hospital admissions (41) and improve continuity of care.

During the last ten years there has been an increased focus on CP as a lifelong health condition. Our findings highlight that outpatient service use declines during transition to adult services. Further work is needed in this area to understand the reasons for this decline and its impact. Our results also highlight that while diseases of the respiratory

system continue to be a major cause of hospitalisation in CP into adulthood,(42) early adulthood is associated with emergence of other health problems (e.g., mental health disorders) (43) that will likely require a change of focus for clinicians to ensure efforts for early detection / intervention during adolescence are successful.

Strengths of our study include the use of a large population-based data set. Using data linkage of a CP register with administrative health data enables ascertainment of CP severity and comorbidities, ensures complete follow-up and minimises selection bias observed when using administrative health data alone.(44) The broad health care setting including specialist children's hospitals and general hospitals also supports a more complete picture of outpatient health service utilisation. Our study also has some limitations. In Australia, primary health care and some specialist care is also available in private settings and these data were not available. However, previous studies suggest that 80% of children have seen a general practitioner and just under 40% of children have seen a private paediatric medical specialist in the preceding year.(45) Access to National Disability Insurance Scheme-funded services may also influence health service utilisation, and this should be a focus of future work. There was also 16% of children without any record of outpatient encounters. This may be due to those living near state borders who may have travelled interstate for their health care, or those who may be only accessing private services. There are known relationships between measures of severity (e.g., GMFCS, epilepsy, intellectual disability) and health service utilisation. Associations between these and UPCI may have impacted our regression models, although our stratified analysis supports an association in less severe CP. There are also limitations in

the use of continuity of care and application of the UPCI in the context of children with CP with multiple and complex health conditions where best care may be better achieved by seeing multiple providers of different specialties.(32) Future research should focus on the quality of care children and young people with CP receive and identifying best ways of measuring this quality.

In conclusion, our study highlights the importance of coordination of outpatient services for children and young people with CP. Establishing better continuity of care, preferentially prioritising this for families residing in rural/remote and from most disadvantaged areas, may improve quality of care and decrease unplanned hospital care utilisation for these groups. Achieving this will require a whole of system approach.

7.6 Chapter Synopsis

In **Chapter 7**, better continuity of care at outpatient services was found to be associated with decreased unplanned hospital care. Continuity of care was inequitably distributed, and less available outside of major cities and in more socioeconomically disadvantaged areas. The study was published in *Development Medicine and Child Neurology*. This chapter concludes the component studies of this thesis. **Chapter 8** will provide a summary of the thesis findings and discuss clinical, policy and research implications.

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Chapter 8 Summary and Conclusions

8.1 Introduction

This thesis aimed to examine access, and equity of access to hospital-based outpatient services that are intended to support children and young people with CP. This final chapter of the thesis will summarise the key findings and highlight implications for clinical practice, policy and research. It will also outline the strengths and limitations of this doctoral program of research. The concept map for this thesis is shown in **Figure 8.1**.

Figure 8.1. Thesis Concept Map for Chapter 8

	Knowledge Gap		Objective	Chapter	Publication	Key Findings
Introduction			Background and literature review	Chapter 1		
	What is already known about health service access in CP?	I	To systematically review the existing literature regarding determinants of hospital-based health service access among people with CP.	Chapter 2	Determinants of Hospital-Based Health Service Utilisation in CP: a Systematic Review. *doi:10.1016/j.apmr.2021.12.003*	• Hospital health service utilisation associated with age, severity and comorbidities.
Study Methods			Introduce the NSW/ACT CP Register, administrative data sets and data linkage.	Chapter 3		
	How do study populations differ between a CP population register and hospital admission data?	II	To compare the sociodemographic and clinical characteristics of children with CP identified from a CP population register and hospital admission data.	Chapter 4	A comparison of cohorts of children with CP from a population register and hospital admission data. *doi:10.1111/ppe.13024*	• Hospital admission data has sensitivity of 0.7 for a diagnosis of CP. • Children with CP from hospital admission data are older, live in major cities, more comorbidities and early death.
Dimensions of outpatient health service access and CP	What factors influence non-attendance at specialty outpatient clinics?	III	To explore the factors associated with non-attendance by children with CP at specialty outpatient clinics.	Chapter 5	Non-attendance at outpatient clinic appointments by children with CP. *doi:10.1111/dmcn.15197*	• Non-attendance associated with older age, socioeconomic disadvantage and previous non-attendance.
	How does telemedicine influence equity of access to outpatient care?	IV	To determine how telemedicine supports equity of access to specialist outpatient care for children with CP.	Chapter 6	Telemedicine for children with CP before, during and after the COVID-19 pandemic. *Under review.*	• Telemedicine use lower in children living in regional and remote areas.
	How have telemedicine rates have changed since the COVID-19 pandemic?	V	To determine the impact of the COVID-19 pandemic on telemedicine use in children with CP.			• Telemedicine use increased substantially during COVID-19 pandemic and since has declined to near baseline.
	What factors influence access to outpatient services?	VI	To determine how clinical and sociodemographic determinants influence hospital outpatient service access in CP.	Chapter 7	Outpatient encounters, continuity of care and unplanned hospital care for children and young people with CP. *doi:10.1111/dmcn.15800*	• Outpatient service use associated with metropolitan areas, GMFCS IV-V, epilepsy, intellectual disability.
	Are CP outpatient services accessible for Aboriginal and/or Torres Strait Islander children?	VII	To determine hospital outpatient service utilisation for Aboriginal and/or Torres Strait Islander children and young people with CP.			• Rates of outpatient service use for Aboriginal and/or Torres Strait Islander children are similar to non-indigenous population.
	How does outpatient continuity of care impact unplanned hospital care?	VIII	To explore the relationship between outpatient service utilisation, continuity of care, and unplanned hospital care for children and young people with CP.			• Decreased continuity of care is associated with increased unplanned hospital care.
Summary and Conclusions			Summary of key findings and implications for clinical practice, policy and research.	**Chapter 8**		

8.2 Key findings

8.2.1 Determinants of hospital health service access.

The first objective of this thesis was to identify determinants of hospital health service access. Systematic review methodology was used to synthesise the available literature. Key findings were the increased health service utilisation across inpatient, emergency department and outpatient settings related to individual need determinants including GMFCS levels IV-V, epilepsy, gastrostomy and intellectual and additional developmental disabilities (e.g., autism, attention deficit hyperactivity disorder). These findings were not surprising. Management of a comorbidity of epilepsy and/or gastrostomy represent common primary reasons for hospital health service use, both in outpatient, emergency department and inpatient settings.(1-4) GMFCS levels IV-V and increased health service use is more complex, and relates both to the known associations with musculoskeletal complications of CP (e.g., hip subluxation, scoliosis),(5, 6) and the association of GMFCS levels IV-V and comorbidities such as epilepsy, respiratory diseases and gastrointestinal diseases.(7-9) Intellectual disability and other developmental disabilities are also associated with increased prevalence of other health conditions.(10) In addition, children with intellectual and developmental disabilities often experience additional challenges in accessing safe and high-quality healthcare.(11)

The systematic review identified very few studies exploring individual enabling factors, such as adverse social determinants of health, or contextual determinants, such as continuity of care. One Australian survey study, using self-report of health service access

and individual measures of socioeconomic position, reported full-time employment, private health insurance and regional residential location to be associated with increased use of private paediatric medical specialists.(12) One Taiwanese study showed an association between continuity of care and reduced inpatient admission rates, but this finding had not been replicated elsewhere.(13)

These knowledge gaps were a strong influence on the direction of the subsequent studies in this thesis, which included data linkage studies that explored how socioeconomic disadvantage and residential remoteness influence different aspects of equity of access to hospital-based outpatient services and associations between continuity of care and unplanned hospital care.

8.2.1.1 Implications for clinical practice, policy and research

The systematic review highlighted the high rates with which children and adults living with CP access hospital health services, and the complexity of that health care utilisation related to motor type, gross motor function and comorbidities. The findings emphasised the ongoing importance of ensuring equity of access to health care for people with CP: ensuring spatial availability of health services, staff that are knowledgeable about CP and ensuring health services are responsive to individual (child/family) needs (e.g., physical access, communication preferences, intellectual functioning and language and cultural background).

Getting this right, particularly for outpatient services that focus on preventive and early management offers the best opportunity to promote health and wellbeing for people with CP. The review also described how the hospital health care needs of people with CP change with time and age and highlighted the opportunity for health services to develop a lifespan approach to CP management. Soon after this review was published, another systematic review focused on health service use among adults with CP was published with consistent results.(14) The time of transition from paediatric to adult services presents a key opportunity to promote long-term health and engagement with the health system.(15) Improving health care transition for young people with CP should be a priority.

The review highlighted the multiple ways in which health care access can be measured; meta-analysis was limited by the lack of standardisation. It is recommended that future health service utilisation research in this area adopts a standard set of agreed measurements. Proportions (proportion of people having accessed a particular health service over a defined time period), and rates of use (uses per person year) are dominant measures, easily understood and should be considered. Adopting standardisation of measures will enable more direct comparisons between types of health service and regions that will help with benchmarking and identifying local priorities for improved access.

8.2.2 Identifying children with CP using different data sources

The second objective of this thesis was driven by a review of the research literature and identifying studies that defined study populations using either CP population registers or administrative health data and International Classification of Diseases (ICD-9, ICD-10) codes for CP. Key findings included that, compared with children in the NSW/ACT CP Register, children identified as having CP from hospital admission data were older, more likely to live in major cities and to have higher proportions of gastrostomy use, epilepsy, intellectual disability and autism. A higher proportion of children identified from hospital admission data also died during the study period. Case definition impacted both the sensitivity of hospital admission data for diagnosis of CP (range 0.40–0.74) and the positive predictive value (0.47–0.73). The study also highlighted that many (27-53%) CP cases defined using administrative health data are not found in a population-based CP register. Some may represent cases of CP missed by a CP register - a recent Norwegian study reported up to 60% of cases of CP ascertained in administrative health data but not in a CP population registry were correct.(16) Other cases may represent known complexities in diagnosis of CP, including overlap with other neurodevelopmental disorders, age at injury (post-neonatal causes), and children who 'lose' a CP diagnosis either, when another, progressive condition is identified (false positives)(17) or when those described as 'at risk of CP' who have no discernible functional impairment when rechecked at the age of 5 years.(18, 19)

8.2.2.1 Implications for clinical practice, policy and research

These results highlight the strength of using, where available, a CP register in data-linkage, epidemiological and health services research, to provide a more accurate representation of the whole population. This is particularly important where the focus of research is equity of access to health services across the population. It is of the highest importance that CP registers include (as close to as possible) the total population, and that multiple strategies are used to optimise ascertainment. Our findings support ongoing collaboration of the NSW/ACT CP Register with paediatric rehabilitation services in NSW Health (SCHN, Hunter New England Local Health District) to review Electronic Medical Records (EMR) to improve ascertainment, including review of children attending acquired brain injury clinics.(20)

CP registers are not available in all jurisdictions and may not be available for data linkage research. In these cases, researchers should be mindful that populations derived from hospital admission data will include a population with more comorbidities and should consider the case definition used. As comorbidities are known to be associated with greater health service utilisation and adverse health outcomes, studies based on such populations are likely to overstate these (negative) outcomes.(21, 22)

8.2.3 Equity of access to hospital outpatient care

Objectives iii, iv, vi, vii and viii of the thesis aimed to explore equity of access to various aspects of hospital outpatient care: for hospital outpatients across NSW (objective vi), for hospital outpatients for Aboriginal and/or Torres Strait Islander children and young people with CP (objective vii), for non-attendance at clinics (objective iii) and telemedicine (objective iv). Data linkage research methods were used to achieve these objectives.

Key findings included: a higher proportion of children living in regional/remote areas had no outpatient encounters; access to outpatients for Aboriginal and/or Torres Strait Islander children with CP was similar to the non-indigenous population; Aboriginal and/or Torres Strait Islander children with CP were frequent users of emergency department services, particularly those living in regional/remote areas; socioeconomically disadvantaged children were more likely to not attend outpatient clinics appointments; and children in regional/rural areas had less access to telemedicine. These findings helped address the literature gap identified in the systematic review (objective i) and highlighted how differences in the social determinants of health (e.g., socioeconomic disadvantage, residential remoteness), and being Aboriginal and/or Torres Strait Islander influence hospital outpatient service access for children with CP in Australia. It is notable that these findings occurred in the context of publicly funded health system, and they have important implications for the current health system, and for health system planning.

8.2.3.1 Implications for clinical practice, policy and research

Health care professionals and clinical services should consider social and cultural circumstances as well as clinical factors when considering access to health care for children with CP and their families. The findings from these studies suggest that children with CP living in regional/remote areas should be prioritised to support referral pathways to specialist outpatient care and define the future role of telemedicine to support care over distance. Additionally, action is required to understand the reasons for increased non-attendance amongst children living with socioeconomic disadvantage, and design interventions that aim to reduce the impact of a missed appointment on health care outcomes for the child. This should include ensuring a strengths-based approach that ensures families experiencing barriers are not judged or labelled (e.g. 'non-attenders', 'non-compliant') and are not discharged from services whilst having more needs for support.(23)

These studies findings emphasise that health care services are not 'one size fits all'. Families are varied and complex, and their health care preferences can differ, both between families and over time.(24) It is also important that whilst variations between socioeconomic, geographical and cultural groups are acknowledged, that the substantial intersectionality between environmental factors (e.g., socioeconomic conditions, geography) and personal factors (e.g., age, sex, ethnicity) is also recognised.(25) In the face of this complexity, there is a (simple) solution - health care professionals must embed the principles of family-centred care:(26) recognise that each

family is unique; the family is the constant in the child's life; and that parents/carers are the experts on the child's abilities and needs.

Services should consider how they can better understand and accommodate personal circumstances and preferences, and help support to coordinate, and provide holistic and personalised care.(27) At a health service level, agencies must partner with people with lived experience when designing new services and service development (co-design). (28) Including the perspectives of priority populations, such as indigenous populations is essential to understand the needs and preferences of these peoples, and improve cultural safety.(28-30) It is important that as health services develop and integrate new technologies and models of care that the focus remains on equity of access rather than the new technologies themselves.

There is also an opportunity to make health care more holistic, through screening for differences in the social determinants of health (experienced as unmet social needs) and by direct action on these unmet social needs. Multiple screening tools now exist for screening of unmet social needs in clinical settings,(31) and intervention pathways (e.g., community resources, a navigator, clinician training) have been shown to reduce unmet social needs.(32) Current research is exploring the feasibility of an adapted screening tool and care navigator in CP rehabilitation clinics.(33)

The research presented in this thesis has measured equity of access in terms of *Realised Access* (i.e., utilisation). There is a need for research that includes other measures of access to health care. One approach is to measure "unmet health care needs" i.e., a need for health care that remains because appropriate health care was not received. Unmet health care needs can be assessed in two ways: (i) through the development of clinical guidelines that define appropriate care (such as those developed for CP hip surveillance),(34) and examining equity of access compared to this benchmark; and (ii) measuring unmet health care needs as experienced by people with CP and their parents/carers.(35) Questions about unmet health care needs have been integrated as part of some government surveys (e.g. National Survey of Children with Special Health Care Needs (US)),(36, 37) and adapted surveys have been used in adults with CP,(38) and at the time of transition.(39) Research examining unmet health care needs of children and young people with CP in Australia would help identify inequities and indicate priorities (and regional priorities) in service development.

8.2.4 Telemedicine use in specialty children's hospitals outpatients

The fifth objective of this thesis was influenced by my experiences as a health professional during the COVID-19 pandemic and witnessing the extensive changes to health care provision during this time, including widespread uptake of telemedicine. Rates and proportions of telemedicine use were measured prior to, during and after COVID-19 related public health restrictions. The key finding was that following peaks of telemedicine use during the most severe COVID-19 restrictions in NSW, the use of

telemedicine for specialist medical and allied health outpatient consultations for children with CP declined towards pre-pandemic levels.

Whilst some decline in telemedicine use was anticipated, the extent of the decline observed was surprising given the expansion of telemedicine services during COVID-19 for children with disabilities(40) and the political investment in sustaining telemedicine after the pandemic.(41) The reasons for the decline are likely to include known patient and clinician preference for in-person consultations,(42-44) and challenges reported including managing technology, child engagement, physical examination and maintaining rapport.(40, 44-46) Similar to another study,(47) the results suggested that telemedicine may be harder to incorporate in consultations with a strong reliance on physical examination (e.g., allied health, orthopaedics).

8.2.4.1 Implications for clinical practice, policy and research

These results suggest that further work is required to embed telemedicine approaches into routine care for health services for children with CP. There is a need for research that re-evaluates the telemedicine preferences of families after the pandemic to help guide this service delivery, including co-design of telemedicine's role in developing models of care. Similarly, there is a need for research to determine the effectiveness, safety, acceptability and cost effectiveness of digital health technologies in CP, as there is in other areas of health care.(48) Additionally, there are opportunities to integrate more digital technologies into CP health care, for example, studies among children with other

chronic health conditions have identified that technology (e.g., SMS technology, apps) can promote positive health outcomes when specifically targeted at a component of care.(49, 50) There is an emerging evidence base for inclusion of wearable sensors (including wearable electromyography (EMG),(51) accelerometers and gyroscopes(52)) in assessment of CP. One potential advantage of these systems is the assessment can occur remotely, and in a child's natural environment.(53) There is a need to explore whether wearable sensors can help bridge the difficulties of physical examination in telemedicine to help improve access for children with CP who live in regional and remote areas.

8.2.5 Continuity of care and unplanned hospital care

The eighth and last objective of this thesis explored the association between continuity of care and unplanned hospital care. Key findings were that (i) continuity of care was inequitably distributed, with high continuity of care more common in children living in major cities and least socioeconomically disadvantaged areas; and (ii) in adjusted analyses, reduced continuity of care was associated with increased use of unplanned hospital services. While measures of continuity of care have been most commonly used in primary care settings,(54-56) and would benefit from further validation in specialist CP outpatient care,(57) this was an important finding. The result was also consistent with a Taiwanese study in which lower continuity of care in children with CP was associated with increased hospital admissions (measured in inpatient days).(13) The results also highlighted the complexity of health system interactions experienced by children with CP

and their families. Similar to another study, (58) children with CP were reviewed by median of seven different specialties (and 25% children more than 16) and a median of six appointments per year with 25% children having more than 12 encounters per year.

These findings highlight the need for the health system to work as an integrated and cohesive whole to improve health care outcomes and experience for children with CP and the families. Improving continuity of care, 'the extent to which a series of discrete health care events is experienced by people as coherent and interconnected over time and consistent with their health needs and preferences'(59) and related concepts such as care coordination, 'a proactive approach to bringing together care professionals and providers to meet the needs of service users, to ensure that they receive integrated, person-focused care across various settings'(59) are important ways to achieve this aim.

8.2.5.1 Implications for clinical practice, policy and research

Health care professionals and clinical services can improve patient outcomes and health care experiences through supporting continuity of care and care coordination. While relational aspects of continuity of care (the ongoing therapeutic relationship between provider and patient) are of high importance, teams, rather than individuals, are important functional units in health care for children and young people with complex disabilities. Continuity of care can also be supported by initiatives that support informational continuity (using knowledge of past events and personal history to influence current management) and/or management continuity (consistency in

management of a health condition across providers). (60) Teams have opportunities to work together to better understand each other's roles and processes, to develop guidelines that improve consistency in management, and share responsibility for supporting best patient outcomes. The electronic medical record (EMR) is well placed to improve the sharing and updating of relevant clinical information that supports personalised care and should be harnessed to meet this need. While current EMR systems tend to be limited to one organisation, the planned development of the Single Digital Patient Record (SDPR) in NSW combined with the statewide integration of EMR is an opportunity for health services to work across current boundaries to achieve this goal.(61)

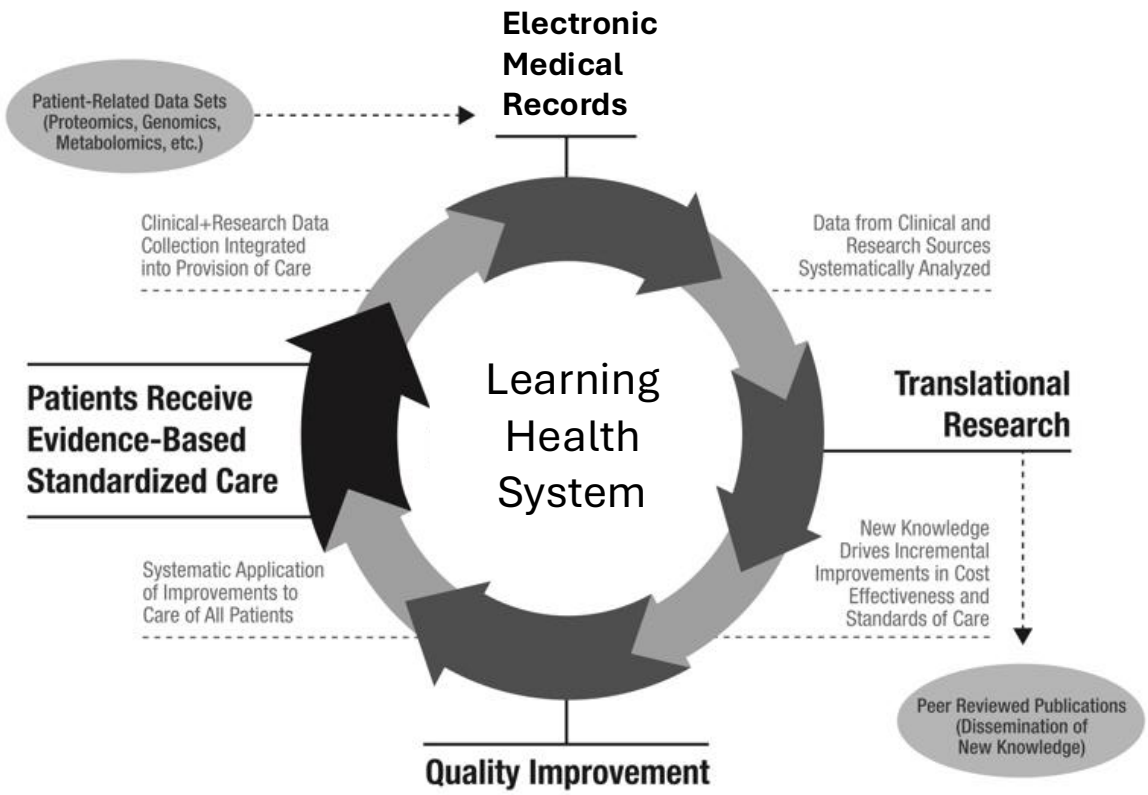
At an organisational level, formalised partnerships between services in different organisations, such as those that support the better coordination of paediatric rehabilitation services in NSW,(62) can support a population perspective, and ensure equitable access to specialised services (e.g., selective dorsal rhizotomy, intrathecal baclofen). Developing integrated care models that include clinicians in primary care, secondary care and specialist settings can improve access to health care.(63) Examples of this include care coordination services, involving a Care Coordinator role for children with chronic and/or complex health conditions. Introduction of care coordination services in metropolitan NSW has been associated with reduced emergency department attendances, health care costs and family travel,(64) and there are now studies to explore the efficacy of similar models in regional/rural settings.(65) Research is needed that examines families' experience of integration of their child's health care in the context of

CP. Likewise, research is needed that develops interventions to improve care coordination and/or continuity of care for children with CP and explores the impact on health care access and health outcomes.

8.2.6 Thesis as a whole

Data is key to advances in health care. Population-level data are particularly important in evaluating access to the health system and the safety and quality of health services provided. The program of research presented in this thesis provides strong representation of the value of research using health administrative data, particularly using data linkage techniques and with a population-level CP register that enables accurate identification and description of a population in clinically meaningful ways. The integration of EMR in health care has promoted the development of 'Learning Health Systems' (LHS), EMR systems in which routinely-collected clinical and service-level data are collected and used to generate new knowledge which can drive improvements in health care.(66) (**Figure 8.2**).

Figure 8.2. An example model of processes involved in a Learning Health System



Adapted from Lowes *et al* (67)

LHS have been established in some Australian states (e.g., Queensland, Western Australia), through partnerships between government agencies, service providers and research organisations. These LHS visualise area-level data using dashboards that profile child health, education and well-being, in order to inform priorities in service development.(68, 69) However, challenges have been identified for similar projects in NSW including inconsistencies in current data structures in EMR, differing requirements of end users, and barriers to using available data (e.g., data skills, time).(70)

In other settings, LHS have been shown to provide clinical and research benefits for patients, clinicians, health systems and researchers, by improving opportunities to track symptoms and health issues, measure compliance with evidence-based guidelines and to support comparative effectiveness trials.(66) Establishing LHSs for CP has been suggested as a way to better understand which treatments work best for which children.(71) A LHS established specifically for treatment of children with CP at a single children's hospital in Ohio, US has showed early evidence of benefits, with reductions in inpatient admissions, emergency department attendances and urgent care encounters.(67)

8.2.6.1 Implications for clinical practice, policy and research

Establishing LHS for CP in the NSW health system would provide numerous opportunities to improve clinical care and for research. In order to capitalise on this opportunity, foundational work should be prioritised to form consensus on essential data fields, minimising variations in assessment, and engaging with children with CP and their families to understand their priorities. To maximise research potential, it is important that LHS for CP are developed and supported by inter-agency research teams, including custodians of the NSW/ACT CP Register. Research has a key role in developing consensus around what optimal care looks like, both in terms of health service access and outcomes.

8.3 Strengths and Limitations

Strengths and limitations of the individual component studies detailed in **Chapters 4-7** have been discussed in their relevant chapters. Overarching strengths and limitations of the thesis will be outlined here.

8.3.1 Strengths

A particular strength of this thesis has been the diversity and expertise of the research team that supported the component studies. While the research questions initiated from my own clinical experience and the research expertise of my supervisors (Professor Natasha Nassar, Adjunct Professor Sarah McIntyre), the research questions, (and the thesis as a whole) benefited enormously from the expertise and guidance of other clinicians (Mary-Clare Waugh, Maria Kyriagis, Kirsty Stewart, Tracey Williams, Heather Burnett), researchers (Katarina Ostojic, Shona Goldsmith, Samantha Lain, Francisco Schneuer, Amy von Huben, Sue Woolfenden, Tan Martin), policy makers (Louise Sellars, Sophie Price) and consumers (Emma Maly, Katrina Ford).

The thesis was also strengthened by consultation with the Cerebral Palsy Alliance CP Quest (community and research group), the Australian Cerebral Palsy Register Community, Aboriginal and Torres Strait Islander Reference Group, and the Aboriginal Health and Medical Research Council of NSW; these conversations have sharpened

research questions, and supported the strengths-based approach to reporting results for Aboriginal and/or Torres Strait Islander children and young people with CP.

The aim and objectives of this thesis were best met through epidemiological research methods, and the component studies have strengths in their use of a CP population register, and data linkage with administrative health datasets. The use of the NSW/ACT CP Register to define study populations in the component studies means that these populations are truly representative of the CP population, with substantially reduced selection bias (e.g., studies that are reliant on a 'health service using' population) and improved certainty about the diagnosis. The use of the NSW/ACT CP Register also enabled broad clinical description of the study populations, subgrouping in clinically meaningful ways including motor disorder type, GMFCS level and comorbidities. Using the NSW/ACT CP Register in data linkage research highlights the substantial importance of these resources and the findings from this thesis will encourage others to seek their use in the future.

Data linkage of the NSW/ACT CP Register with hospital outpatient data has enabled this thesis to fill important gaps in the literature and build on previous Australian work that used data linkage techniques to improve understanding of ED and hospital admissions in children with CP.(72) Data linkage with large administrative datasets (i.e., SCHN (**Chapters 5,6**) outpatient data and NSW Health (**Chapters 4,7**)) enabled analysis of health administrative data at a population scale and scope that would not be feasible

through other research methodologies. Use of administrative health data also reduces the recall bias present in patient surveys of health utilisation, and linkage minimises the loss to follow up inherent in cohort studies.

8.3.2 Limitations

The limitations in the studies in this thesis primarily relate to the limitations of the datasets and the research methodologies used. Firstly, observational study techniques, including multivariable logistic regression, are not structured to explore causative relationships, and other research methodologies will be required to support whether interventions can change the outcomes reported in this thesis. Secondly, while multivariable regression analyses have controlled for confounding variables, administrative health data and the NSW/ACT CP Register do not capture every pertinent clinical variable (e.g., some common comorbidities such as chronic pain), nor information about lifestyle and treatments. Thus, the studies have a potential for unmeasured confounding.

Additionally, the nature of the datasets means that while we have been able to describe health service utilisation (*Realised Access*) at a population level, we have not been able to determine why individual health service encounters occurred, what health issues were discussed and managed and whether the encounter influenced health outcomes. As described above, further research exploring how health needs are met (or unmet) would help explore these relationships further. As detailed, we have largely used area level

measures for socioeconomic disadvantage, and using more individualised measures (e.g., educational attainment, parental occupation, income) may have provided additional useful information.

Finally, the regional nature of the work based in New South Wales, Australia, and its restriction to hospital care may impact the generalisability of some of the findings. Research that explores equity of access to other important aspects of the Australian health system, including the primary health care system and private health care, and the National Disability Insurance Scheme is a priority.

8.4 Conclusion

This thesis aimed to examine access, and equity of access to hospital-based outpatient services that are intended to support children and young people with CP. This final chapter has summarised the key findings, strengths and limitation of the research program and identified implications for future clinical practice, policy and research. Main conclusions include that access to hospital-based outpatient services for children with CP is influenced by differences in the social determinants of health including socioeconomic disadvantage and residential remoteness; and that continuity of care can influence unplanned health care use. These findings provide a strong rationale for strengthening the health system for children with CP and identifies key areas of outpatient care such as non-attendance, telemedicine where the focus of attention

should be prioritised. Further work is planned in this area, with the hope of improving access to health care for children and young people with CP.

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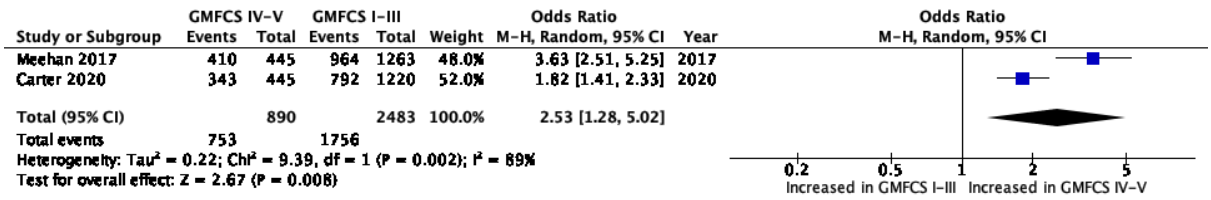
Chapter 9 Appendix A – Supplementary Information

This chapter contains supplementary tables and figures referenced in **Chapters 2,4-7**.

9.1 Chapter 2 – Supplementary Information

Supplementary Figure 9.1. Forest plot of Gross Motor Function Classification System and inpatient admissions.



Supplementary Table 9.1. MEDLINE Search Strategy.

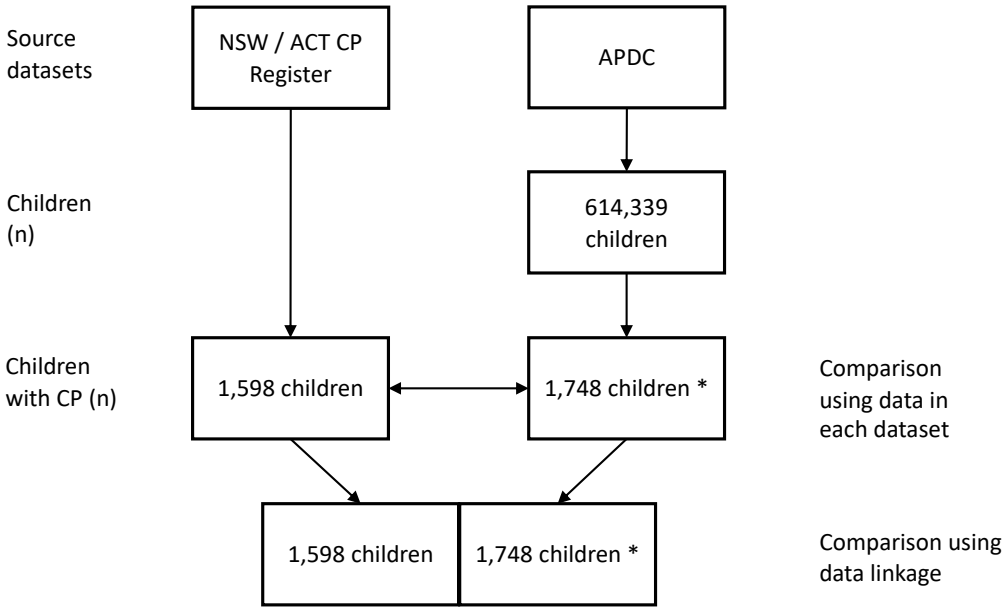
Search Term	Results
1. exp Cerebral Palsy/	20275
2. cerebral palsy.tw	21516
3. 1 or 2	26842
4. exp health services/sn	201371
5. exp emergency medical services/	137120
6. exp ambulatory care facilities/	54325
7. exp hospital departments/	180659
8. exp hospitals, pediatric/	13081
9. exp health services accessibility/	108785
10. (use* or utili* or attend* or present* or admi*) adj2 (department* or health care* or service* or hospital*)).tw	146253
11. 4 or 5 of 6 or 7 or 8 or 9 or 10	640611
12. 3 and 11	551

Supplementary Table 9.2. Risk of bias and quality assessment.

Source	Question	Domain Addressed
Cohort studies		
Newcastle-Ottawa Scale – Cohort (Wells 2000)	Representativeness of the exposed cohort	Selection
	Selection of the non-exposed cohort	Selection
	Ascertainment of exposure	Exposure
	Comparability of cohorts on the basis of design or analysis	Selection
	Assessment of outcome	Outcome
	Was follow-up long enough for outcomes to occur	Loss to follow up
	Adequacy of follow up of cohorts	Loss to follow up
AHRQ - Viswanathan 2013	Are any important primary outcomes missing from results?	Selective reporting
Downs & Black 1998	Were the main outcome measures used accurate (valid and reliable)?	Outcome
	Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?	Confounding
	Were the statistical tests used to assess the main outcomes appropriate	Analysis
Cross-sectional studies		
Newcastle-Ottawa Scale – Cohort (Herzog 2013)	Representativeness of the sample	Selection
	Sample size	N/A
	Non-respondents	Selection
	Ascertainment of the exposure	Exposure
	The subjects in different outcome groups are comparable, based on the study design or analysis. Confounding factors are controlled	Confounding
	Assessment of the outcome	Outcome
	Statistical test	Analysis
AHRQ - Viswanathan 2013	Are any important primary outcomes missing from results?	Selective reporting

9.2 Chapter 4 – Supplementary Information

Supplementary Figure 9.2. Study flow chart showing comparison of datasets used to identify children/young people with cerebral palsy.



Supplementary Table 9.3. Examples of research studies using different algorithms based on International Classification of Diseases codes in health administrative datasets.

Citation	First Author	Administrative health dataset type(s)	Case definition
[1]	Oskoui, M *et al*	Inpatient Outpatient	1 or more G80 code, 2 years or older
[2]	Carter, B *et al*	Primary care Inpatient Outpatient Mortality	1 or more G80, G81, G82, G83 codes before 25 years
[3, 4]	Gill, J *et al*; Morgan, P *et al*	Inpatient Emergency Department *	1 or more G80 codes
[5]	Whitney, D et al **	Inpatient Outpatient Pharmacy Laboratory	1 or more G80 codes
[6]	Peterson, MD *et al* **	Inpatient Outpatient Pharmacy Laboratory	1 or more G80 codes
[7]	Li, Q *et al*	Inpatient Insurance claims	1 or more 343.X codes (inpatient), 2 or more 343.x codes (insurance) ***
[8]	Toyokawa, S *et al*	Insurance claims	1 or more G80 codes

* CP diagnosis identified through linkage to inpatient health administrative dataset

** study only including adults

*** ICD-9

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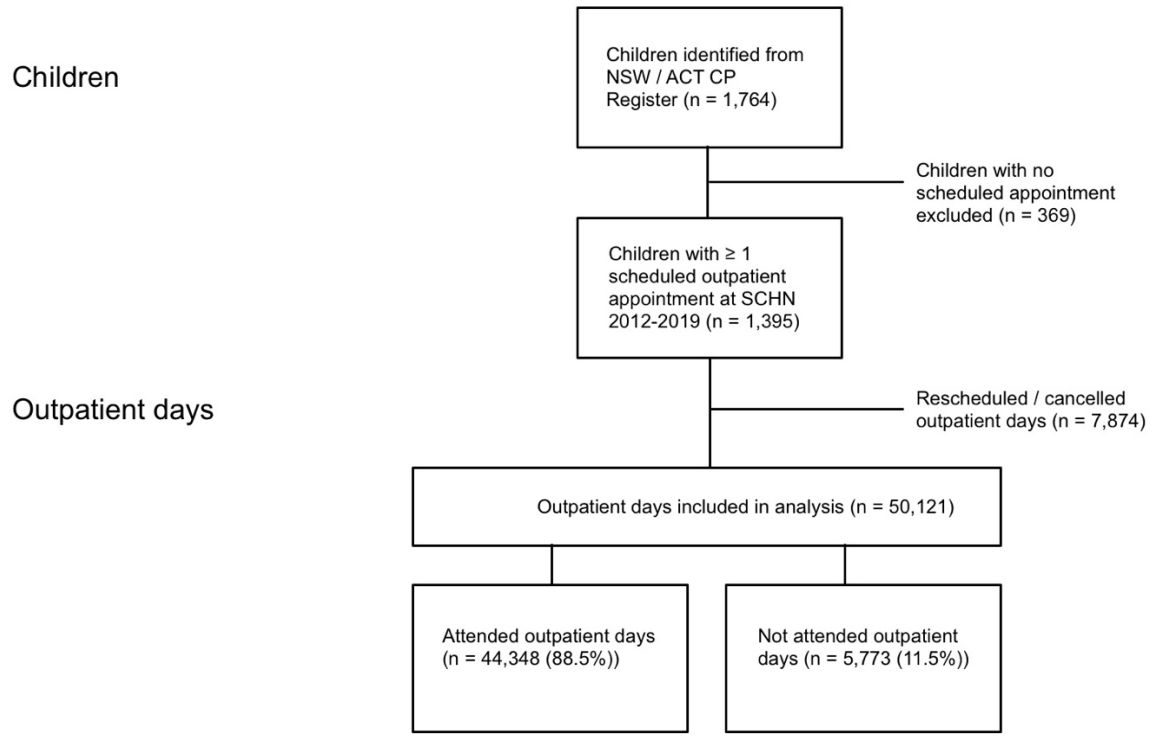
Supplementary Table 9.4. Comparison of motor disorder types, functional classifications, and common comorbidities for children identified in either the CP Registry alone and CP Registry and Hospital admission data (APDC)*.

Clinical characteristics	In CPR, not APDC* (n=472)	In CPR and APDC* (n=1082)	SMD
Motor Type (CPR) n (%)			0.477
Spastic Unilateral	198 (41.9)	329 (30.4)	
Spastic Bilateral	169 (35.8)	547 (50.6)	
Dyskinetic	28 (5.9)	131 (12.1)	
Ataxia	39 (8.3)	34 (3.1)	
Hypotonia	13 (2.8)	21 (1.9)	
Early at risk CP	0 (0)	1 (0.1)	
Unknown	25 (5.3)	19 (1.8)	
GMFCS (CPR) n (%)			0.793
I-III	393 (83.3)	683 (63.1)	
IV-V	41 (8.7)	360 (33.3)	
Unknown	38 (8.1)	39 (3.6)	
MACS (CPR) n (%)			0.679
I-III	335 (71)	619 (57.2)	
IV-V	19 (4)	257 (23.8)	
Unknown	118 (25)	206 (19)	
Epilepsy (CPR) n (%)			0.389
Yes	75 (15.9)	338 (31.2)	
None or resolved	276 (58.5)	564 (52.1)	
Unknown	121 (25.6)	180 (16.6)	
Intellectual Disability (CPR) n (%)			0.122
Yes	214 (47.5)	556 (53.7)	
No	192 (42.6)	395 (38.2)	
Unknown	66 (14.6)	131 (12.7)	

* Based on the ascertaining CP using the APDC (one or more admissions with G80 code); SMD: Standardised mean difference

9.3 Chapter 5 – Supplementary Information

Supplementary Figure 9.3. Flow diagram of inclusions and exclusions in study.



Supplementary Table 9.5. List of medical and surgical specialties.

Specialty Group	Specialties	
Allied Health	Physiotherapy	Social Work
	Occupational Therapy	Psychology
	Speech Pathology	Child life therapy
	Dietetics	Orthotics
General Medicine	General Medicine	
Rehabilitation	Rehabilitation Medicine	
Neurology / Neurosurgery	Neurology	Neurosurgery
Other Medical Specialties	Adolescent Medicine	Neonatology
	Allergy & Immunology	Oncology
	Audiology	Pain
	Cardiology / Cardiothoracic	Palliative Care
	Developmental Medicine	Psychological Medicine
	Endocrinology	Renal Medicine
	Gastroenterology	Respiratory and Sleep Medicine
	Haematology	Rheumatology
	Infectious Diseases	Sports Medicine
	Metabolic medicine	Weight Management
General Surgery	General Surgery	
Orthopaedics	Orthopaedics	
Other Surgical Specialties	Anaesthetics	Ophthalmology
	Craniofacial	Gynaecology
	Dental	Plastics
	Ears, Nose, Throat	Urology
	Cleft lip / palate	Burns

Supplementary Table 9.6. Proportions of children and likelihood of scheduling for major specialty outpatient clinics in children with cerebral palsy, 2012-2019.

Demographic and clinical factors	Scheduled appointments	Rehabilitation Medicine appointments		Allied Health appointments		Neurology appointments		Orthopaedic appointments	
	n (%)	n (%)	OR (95%CI)	n (%)	OR (95%CI)	n (%)	OR (95%CI)	n (%)	OR (95%CI)
Total	1395 (100)	1147 (100)		1100 (100)		776 (100)		604 (100)	
Sex									
Male	831 (59.6)	674 (58.8)	0.83 (0.62-1.1)	661 (60.1)	0.83 (0.62-1.1)	445 (57.3)	0.81 (0.65-1.01)	338 (56)	0.77 (0.62-0.95)
Female	564 (40.4)	473 (41.2)	REF	439 (39.9)	REF	331 (42.7)	REF	266 (44)	REF
Country of Birth									
Australia	1298 (93)	1058 (92.2)	REF	1027 (93.4)	REF	727 (93.7)	REF	561 (92.9)	REF
Overseas	88 (6.3)	83 (7.2)	3.77 (1.51-9.39)	71 (6.5)	3.77 (1.51-9.39)	46 (5.9)	0.86 (0.56-1.33)	42 (7)	1.2 (0.78-1.85)
Preferred Language									
English	1214 (87)	1005 (87.6)	REF	964 (87.6)	REF	672 (86.6)	REF	538 (89.1)	REF
Other	120 (8.6)	97 (8.5)	0.88 (0.54-1.42)	94 (8.5)	0.88 (0.54-1.42)	77 (9.9)	1.44 (0.98-2.13)	61 (10.1)	1.3 (0.89-1.89)
Remoteness									
Major cities	941 (67.5)	784 (68.4)	REF	748 (68)	REF	539 (69.5)	REF	539 (89.2)	REF
Regional / Remote	448 (32.1)	357 (31.1)	0.79 (0.59-1.05)	346 (31.5)	0.79 (0.59-1.05)	233 (30)	0.81 (0.64-1.01)	233 (38.6)	0.9 (0.71-1.13)
IRD Quintile									
1 (most disadvantaged)	274 (19.6)	218 (19)	0.57 (0.37-0.89)	212 (19.3)	0.57 (0.37-0.89)	164 (21.1)	1.22 (0.88-1.69)	124 (20.5)	1.05 (0.76-1.44)
2	210 (15.1)	165 (14.4)	0.54 (0.34-0.86)	154 (14)	0.54 (0.34-0.86)	106 (13.7)	0.83 (0.59-1.18)	89 (14.7)	0.93 (0.66-1.32)
3	275 (19.7)	216 (18.8)	0.54 (0.35-0.83)	205 (18.6)	0.54 (0.35-0.83)	146 (18.8)	0.93 (0.67-1.27)	105 (17.4)	0.78 (0.57-1.08)
4	288 (20.6)	244 (21.3)	0.82 (0.52-1.29)	236 (21.5)	0.82 (0.52-1.29)	168 (21.6)	1.15 (0.84-1.57)	130 (21.5)	1.04 (0.76-1.43)
5 (least disadvantaged)	342 (24.5)	298 (26)	REF	287 (26.1)	REF	188 (24.2)	REF	151 (25)	REF

GMFCS									
I-III	998 (71.5)	819 (71.4)	REF	756 (68.7)	REF	492 (63.4)	REF	360 (59.6)	REF
IV-V	342 (24.5)	309 (26.9)	2.05 (1.38-3.03)	312 (28.4)	2.05 (1.38-3.03)	248 (32)	2.71 (2.08-3.55)	231 (38.2)	3.69 (2.84-4.79)
Predominant motor type									
Spastic	1010 (72.4)	865 (75.4)	REF	815 (74.1)	REF	502 (64.7)	REF	457 (75.7)	REF
Dyskinetic	192 (13.8)	168 (14.6)	1.17 (0.74-1.86)	164 (14.9)	1.17 (0.74-1.86)	135 (17.4)	2.4 (1.72-3.34)	86 (14.2)	0.98 (0.72-1.34)
Other	171 (12.3)	110 (9.6)	0.3 (0.21-0.43)	109 (9.9)	0.3 (0.21-0.43)	123 (15.9)	2.59 (1.82-3.7)	56 (9.3)	0.59 (0.42-0.83)
Intellectual Disability									
Yes	645 (46.2)	526 (45.9)	0.66 (0.48-0.91)	527 (37.8)	0.66 (0.48-0.91)	433 (67.8)	3.06 (2.41-3.9)	331 (62.7)	1.7 (1.34-2.15)
No	515 (36.9)	448 (39.1)	REF	404 (29)	REF	206 (32.2)	REF	197 (37.3)	REF
Not reported	235 (16.8)	235 (0)		235 (0)		(0)		(0)	
Epilepsy									
Yes	394 (28.2)	323 (32.3)	0.79 (0.57-1.09)	326 (34.1)	0.79 (0.57-1.09)	325 (48.8)	6.28 (4.68-8.44)	208 (39.6)	1.69 (1.32-2.16)
None or resolved	796 (57.1)	678 (67.7)	REF	631 (65.9)	REF	341 (51.2)	REF	317 (60.4)	REF
Not reported	205 (14.7)	205 (0)		(0)		(0)		(0)	

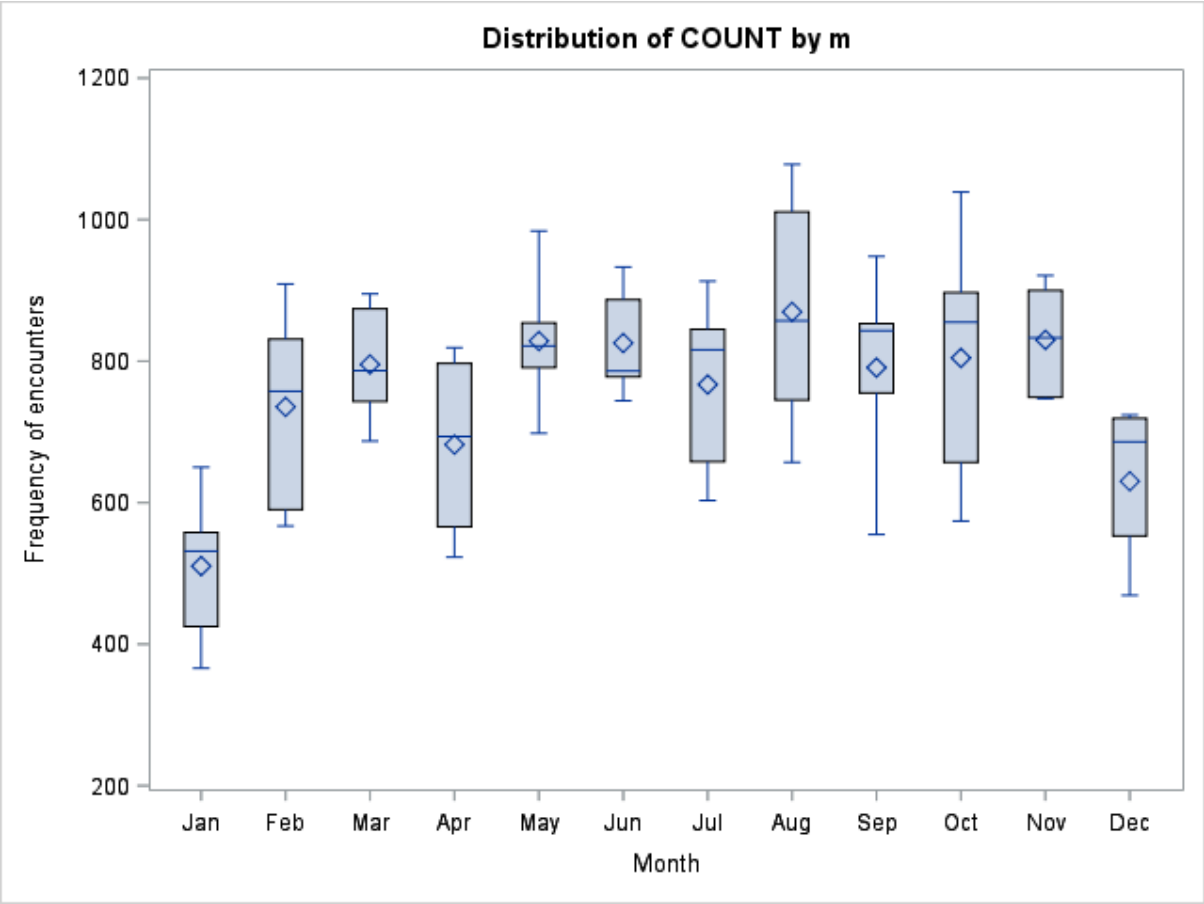
GMFCS Gross Motor Function Classification System; IRD Index of Relative Socioeconomic Disadvantage; OR Odds Ratio

Supplementary Table 9.7. Rates and proportions of scheduled outpatient days by age group and major specialty outpatient clinics in children with cerebral palsy, 2012-2019.

Specialty Group	Total outpatient days (n)	Total outpatient days by age group n (%)			P
		0 to 4 years	5 to 9 years	10 to 14 years	
Total outpatient days	50121	20204 (40)	22242 (44)	7675 (15)	<0.0001
Specialty Type					
Allied Health	19008	7104 (37)	9023 (47)	2881 (15)	<0.0001
General Medicine	1863	813 (44)	822 (44)	228 (12)	0.0002
Rehabilitation Medicine	14918	4874 (33)	7397 (50)	2647 (18)	<0.0001
Neurology	4982	2707 (54)	1741 (35)	534 (11)	<0.0001
General Surgery	1030	552 (54)	364 (35)	114 (11)	<0.0001
Orthopaedics	4433	804 (18)	2306 (52)	1323 (30)	<0.0001

9.4 Chapter 6 – Supplementary Information

Supplementary Figure 9.4. Average frequency of total outpatient encounters by calendar month in children with cerebral palsy.



Difference between months (p=0.0024 (Kruskall-Wallis)).

Supplementary Table 9.8. Annual median outpatient encounters for children with cerebral palsy at two children's hospitals by age group.

Age group	Annual Median Encounters Median (IQR)				
	2018	**2019**	**2020**	**2021**	**2022**
0 to 4 y	6 (2-11), (n=228)	6 (2-12), (n=150)	5 (1-10), (n=82)	NA	NA
5 to 9 y	5 (2-10), (n=516)	5 (2-9), (n=503)	4 (1-9), (n=460)	3 (1-9), (n=394)	3 (0-7), (n=323)
10 to 14 y	5 (2-11), (n=418)	5 (2-11), (n=509)	5 (1-9), (n=530)	3 (1-8), (n=532)	3 (1-9), (n=534)
15+ y	NA	NA	4 (1-8), (n=90)	3.5 (1-8), (n=198)	3 (0-8), (n=305)

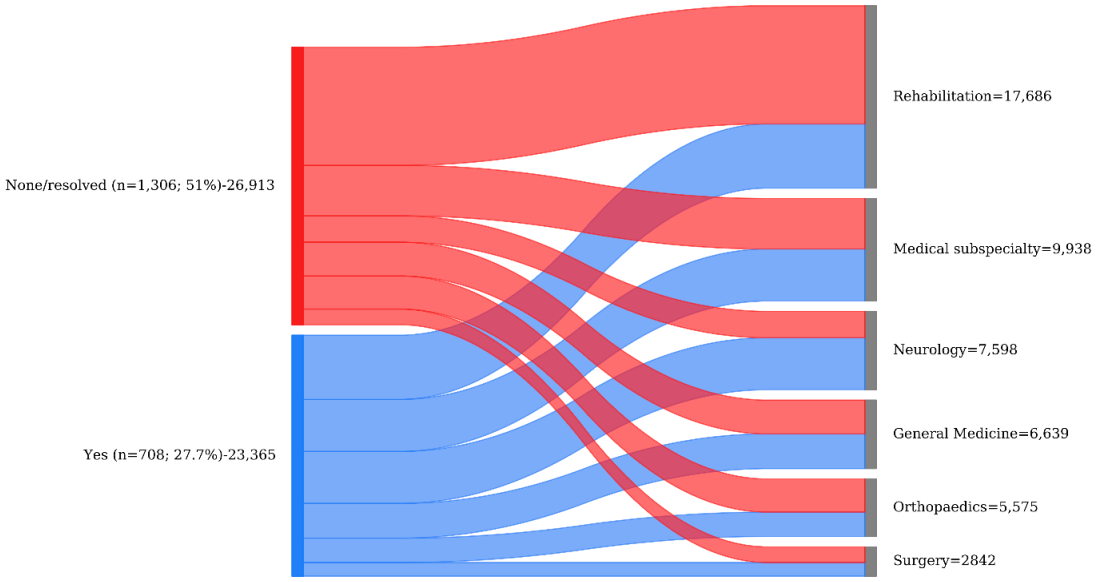
IQR Interquartile range; NA Not Applicable (i.e., no children of this age range at this time point)

NB Only including complete years (2023 not included), n= refers to number of children of this age group at this time point

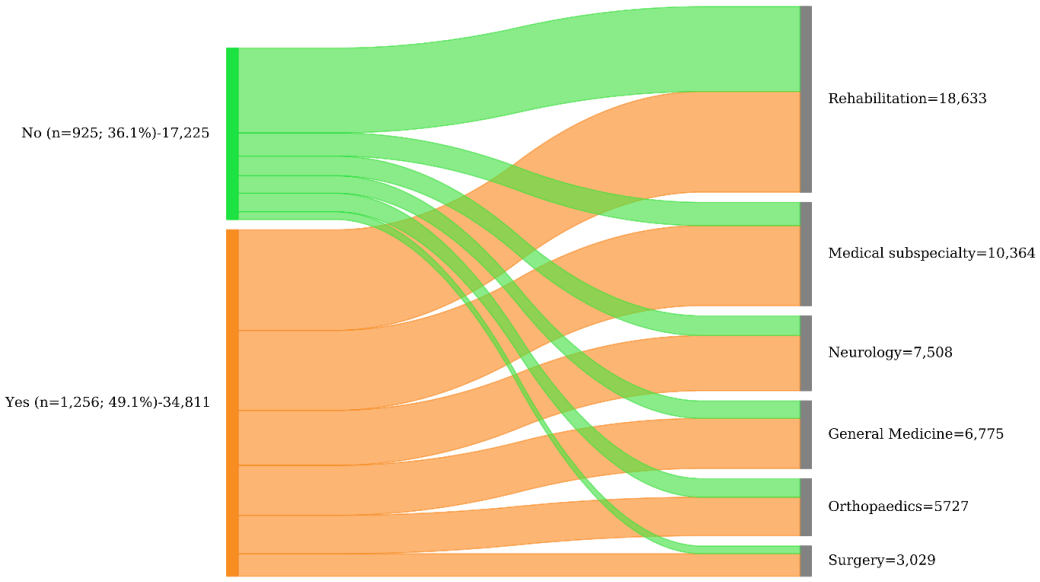
9.5 Chapter 7 – Supplementary Information

Supplementary Figure 9.5. Major outpatient speciality use in children with cerebral palsy.

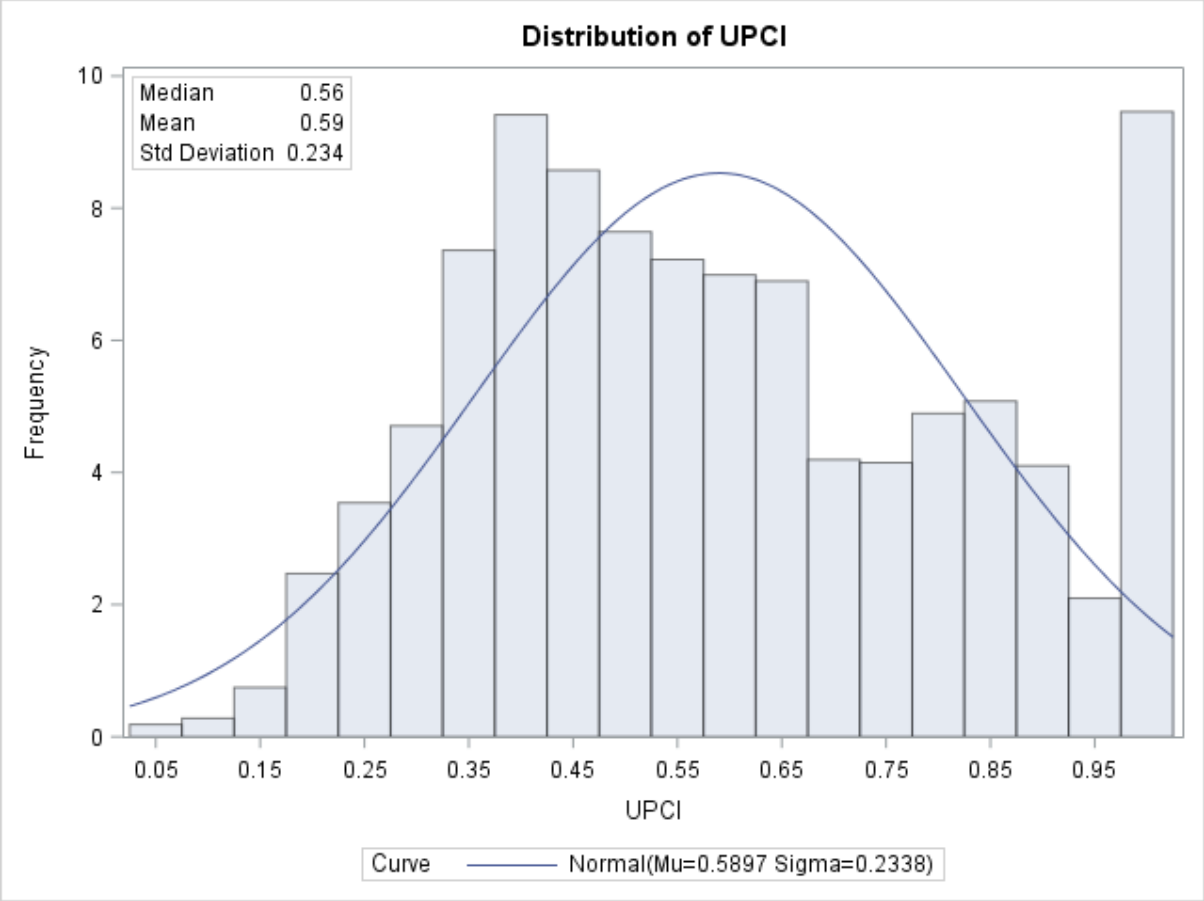
(a) Epilepsy



(b) Intellectual Disability



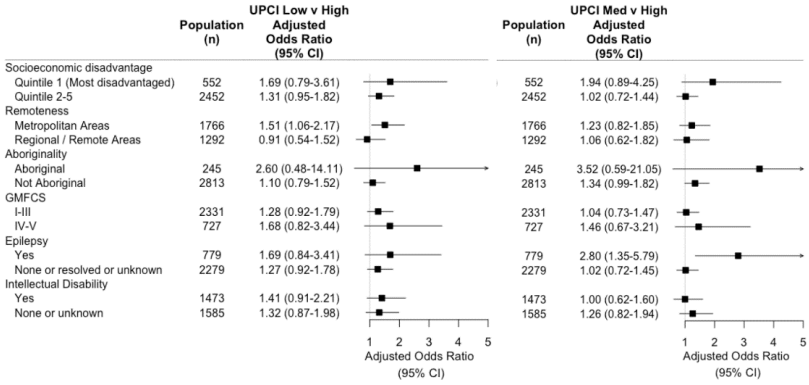
Supplementary Figure 9.6. Frequency of Usual Provider of Care Index for children and young people with cerebral palsy.



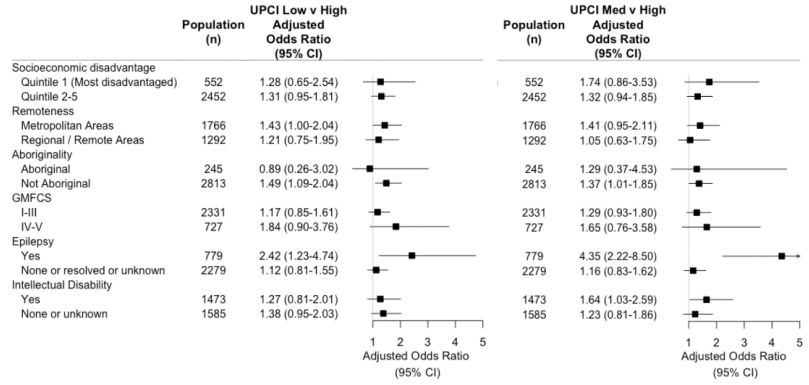
* figure includes only those with an identified usual provider of care

Supplementary Figure 9.7. Association between continuity of care and Emergency Department Presentations stratified by major sociodemographic and clinical factors, and Aboriginal status.

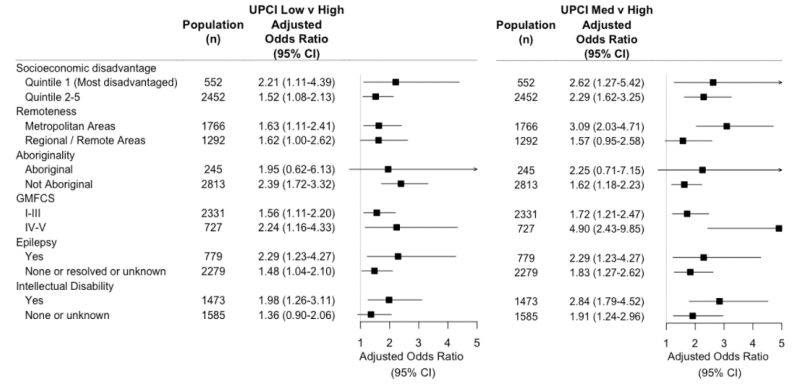
(a)



(b)



(c)



(a) 1 ED presentation
(b) 2-3 ED presentations
(c) 4+ ED presentations

ED Emergency Department; GMFCS Gross Motor Function Classification System; UPCI Usual Provider of Care Index

Supplementary Table 9.9. Comparison of proportions of emergency department attendances and urgent hospital admissions for children and young people with cerebral palsy in 2015-2020.

Demographic, Clinical Factors and Aboriginal status	Total N (%)	ED Presentations				Urgent Hospital Admissions	
		1 Presentation N (%)	2 or 3 Presentations N (%)	4+ Presentations N (%)	No Presentations N (%)	1+ Admissions N (%)	No Admissions N (%)
Total	3267 (100)	587 (18)	684 (20.9)	883 (27)	1113 (34.1)	1091 (33.4)	2176 (66.6)
Sex							
Male	1903 (58.2)	324 (55.2)	405 (60.1)	535 (60.6)	639 (57.4)	647 (59.3)	1256 (57.7)
Female	1364 (41.8)	263 (44.8)	279 (41.4)	348 (39.4)	474 (42.6)	444 (40.7)	920 (42.3)
Maternal Country of Birth							
Australia	2215 (73.6)	382 (71.4)	474 (74.5)	643 (77.7)	716 (70.8)	733 (71.7)	1482 (74.5)
Overseas	796 (26.4)	153 (28.6)	162 (25.5)	185 (22.3)	296 (29.2)	289 (28.3)	507 (25.5)
Aboriginality							
Aboriginal	262 (8)	29 (4.9)	56 (8.2)	124 (14)	53 (4.8)	116 (10.6)	146 (6.7)
Not aboriginal	3005 (92)	558 (95.1)	628 (91.8)	759 (86)	1060 (95.2)	975 (89.4)	2030 (93.3)
Remoteness							
Metropolitan Areas	1851 (57.6)	356 (61.5)	367 (54.1)	442 (50.3)	686 (63.8)	635 (58.4)	1216 (57.2)
Regional / Remote Areas	1361 (42.4)	223 (38.5)	311 (45.9)	437 (49.7)	390 (36.3)	452 (41.6)	909 (42.8)
IRSD Quintile							
1 (most disadvantaged)	591 (18.4)	106 (18.3)	132 (19.5)	191 (21.7)	162 (15.1)	213 (19.6)	378 (17.8)
2	623 (19.4)	111 (19.2)	129 (19)	193 (22)	190 (17.7)	212 (19.5)	411 (19.3)
3	705 (21.9)	118 (20.4)	158 (23.3)	207 (23.5)	222 (20.7)	242 (22.3)	463 (21.8)

4	669 (20.8)	114 (19.7)	150 (22.1)	165 (18.8)	240 (22.3)	232 (21.3)	437 (20.6)
5 (least disadvantaged)	623 (19.4)	130 (22.5)	109 (16.1)	123 (14)	261 (24.3)	188 (17.3)	435 (20.5)
GMFCS							
I-III	2141 (65.5)	414 (70.5)	467 (69.3)	437 (49.5)	823 (73.9)	517 (47.4)	1624 (74.6)
IV-V	768 (23.5)	115 (19.6)	131 (19.4)	317 (35.9)	205 (18.4)	414 (37.9)	354 (16.3)
Unknown	358 (11)	58 (9.9)	86 (12.8)	129 (14.6)	85 (7.6)	160 (14.7)	198 (9.1)
Motor type							
Spastic Unilateral	1070 (35.3)	208 (37.7)	236 (34.6)	211 (24)	415 (39.3)	252 (25.6)	818 (40)
Spastic Bilateral	1381 (45.6)	238 (43.2)	283 (41.4)	376 (42.7)	484 (45.9)	473 (48)	908 (44.4)
Dyskinetic	322 (10.6)	59 (10.7)	64 (9.4)	118 (13.4)	81 (7.7)	152 (15.4)	170 (8.3)
Ataxic	128 (4.2)	21 (3.8)	23 (3.4)	31 (3.5)	52 (4.9)	33 (3.4)	95 (4.6)
Hypotonic	67 (2.2)	12 (2.2)	13 (1.9)	26 (3)	16 (1.5)	36 (3.7)	31 (1.5)
Early at risk	62 (2)	13 (2.4)	64 (9.4)	118 (13.4)	7 (0.7)	39 (4)	23 (1.1)
Intellectual Disability							
Yes	1551 (47.5)	277 (47.2)	302 (44.8)	506 (57.3)	466 (41.9)	635 (58.2)	916 (42.1)
No	1222 (37.4)	226 (38.5)	279 (41.4)	216 (24.5)	501 (45)	273 (25)	949 (43.6)
Unknown	494 (15.1)	84 (14.3)	103 (15.3)	161 (18.2)	146 (13.1)	183 (16.8)	311 (14.3)
Epilepsy							
Yes	840 (25.7)	111 (18.9)	155 (23)	354 (40.1)	220 (19.8)	428 (39.2)	412 (18.9)
None or resolved	1728 (52.9)	343 (58.4)	370 (54.9)	337 (38.2)	678 (60.9)	429 (39.3)	1299 (59.7)
Unknown	699 (21.4)	133 (22.7)	159 (23.6)	192 (21.7)	215 (19.3)	234 (21.4)	465 (21.4)

ED Emergency Department; GMFCS Gross Motor Function Classification System; IRSD Index of Relative Socioeconomic Disadvantage

Supplementary Table 9.10. Principal diagnosis for urgent hospital admissions for children and young people with cerebral palsy.

Age group	Infectious and parasitic diseases	Endocrine and metabolic diseases	Diseases of the Nervous System	Diseases of the respiratory system	Diseases of the digestive system	Mental and behavioural disorders	Injury	Other	Total
0-4 years	141 (14.6)	26 (2.7)	176 (18.3)	357 (37.1)	29 (3)	4 (0.4)	60 (6.2)	170 (17.7)	963
5-9 years	98 (12.3)	20 (2.5)	183 (22.9)	213 (26.7)	31 (3.9)	7 (0.9)	84 (10.5)	82 (10.3)	799
10-14years	43 (7.7)	15 (2.7)	93 (16.6)	159 (28.4)	61 (10.9)	8 (1.4)	58 (10.4)	61 (10.9)	560
15-19 years	33 (6)	31 (5.6)	68 (12.4)	155 (28.2)	62 (11.3)	24 (4.4)	81 (14.8)	63 (11.5)	549
20+ years	16 (5.1)	6 (1.9)	47 (15)	75 (23.9)	43 (13.7)	32 (10.2)	46 (14.6)	31 (9.9)	314
Total	331 (10.4)	98 (3.1)	567 (17.8)	959 (30.1)	226 (7.1)	75 (2.4)	329 (10.3)	170 (17.7)	3185

* 30 cases no principal diagnosis assigned

Principal diagnosis classified using International Classification of Diseases, Tenth Revision, Australian Modification (ICD-10-AM)

Chapter 10 Appendix B – Statements of Contribution and Publications

This chapter contains statements of contribution and published manuscripts for **Chapters 2, 4-7**.

10.1 Chapter 2

10.1.1 Statement of Contribution

Chapter 2 was published as a manuscript in *Archives of Physical Medicine and Rehabilitation* as:

> Paget S, Ostojic K, Goldsmith S, Nassar N, McIntyre S. Determinants of Hospital-Based Health Service Utilization in Cerebral Palsy: a Systematic Review. Arch Phys Med Rehabil. 2022;103(8):1628-1637. doi:10.1016/j.apmr.2021.12.003

The co-authors made the following contributions to the manuscript:

- Simon Paget conceived and designed the study, screened titles and abstracts, screened full text articles, performed all statistical analyses, lead data

interpretation, drafted the initial manuscript and lead subsequent revisions of the manuscript.

- Katarina Ostojic screened full text articles, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Shona Goldsmith screened full text articles, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Natasha Nassar contributed to the conception and design of the study, provided supervision, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Sarah McIntyre contributed to the conception and design of the study, provided supervision, screened full text articles, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

10.1.2 Acknowledgements

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Archives of Physical Medicine and Rehabilitation 2022;103: 1628–37

REVIEW ARTICLE (META-ANALYSIS)

Determinants of Hospital-Based Health Service Utilization in Cerebral Palsy: a Systematic Review

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Abstract

Objective: To systematically review and synthesize evidence of determinants associated with hospital-based health service utilization among individuals with cerebral palsy (CP).

Data Sources: Electronic databases MEDLINE, Embase, APA Psycinfo were searched from January 2000 to April 2020.

Study Selection: Observational studies were included that described people with CP, reported quantitative measures of hospital-based health service utilization (inpatient, outpatient, emergency department), and based in high-income countries. We excluded studies that included only subsets of people with CP, or those that only reported therapy service utilization.

Data Extraction: After initial screen, 2 reviewers reviewed full texts for inclusion and performed data extraction and risk of bias assessment using the Newcastle-Ottawa scale. Determinants of health service utilization were identified and categorized using the Andersen behavioral model.

Data Synthesis: Seventeen studies met inclusion criteria. Study quality was high. Twenty-six determinants were reported across 8 Andersen model characteristics. Individual predisposing factors such as sex showed no difference in health service utilization; inpatient admissions decreased with increasing age during childhood and was lower in adults. Increased health service utilization was associated with "individual need" including severe gross motor disability, epilepsy, developmental/ intellectual disability and gastrostomy-use across inpatient, outpatient and emergency department settings. There was little information reported on socio-demographic and health system contextual determinants.

Conclusions: CP health service utilization is associated with age, severity and comorbidities. Improved understanding of determinants of health service utilization can support health service access for people with CP.

Archives of Physical Medicine and Rehabilitation 2022;103:1628–37

Cerebral palsy (CP) is the most common cause of physical disability in childhood, with a current estimated worldwide incidence of 2 per 1000 live births.[1] In addition to the motor disorder that defines the condition, almost all children with CP have comorbidities (eg, epilepsy, intellectual disability) and/or complications (eg, hip subluxation, scoliosis),[2] and in adulthood, people with CP experience higher rates of many non-communicable diseases compared with adults without CP.[3]

Surveillance for and treatment of these health problems is the primary role of health services. Ensuring that people with CP have access to "the right [health] service at the right time in the right place"[4(p866)] can support improved health outcomes, decreased costs for the health system,[5] and reduced unplanned health care use.[6]

Studies exploring other health conditions (eg, HIV infection,[7] common mental disorders[8]) suggest that health service utilization is influenced not only by need. The Andersen behavioral model is a widely used framework that represents health service access and utilization as an interplay between individual and contextual (health organization, provider, and community) characteristics and the predisposing, enabling and need determinants that underpin these characteristics (fig 1).[9,10] Examples of individual factors

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Disclosures: none.

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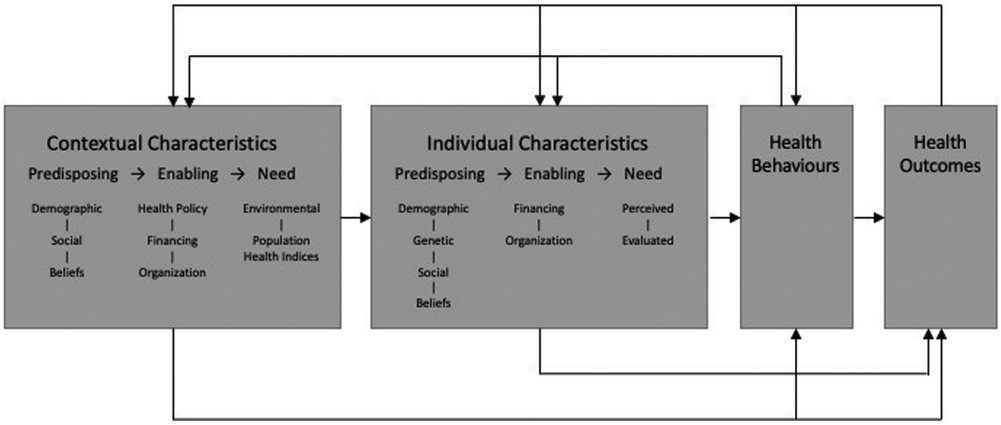

Fig 1 The Andersen behavioral model of health service access and use.[10]

incorporated in the model include age and sex (individual predisposing factors); personal finances and transport (individual enabling factors); and perceived health status, symptom severity, and duration (individual need factors).

Achieving a better understanding of the factors that influence health service utilization for people with CP is an important first step to improve equity of access. Use of the Andersen model can highlight where utilization is driven by determinants other than need. The aim of this systematic review was to identify evidence of determinants associated with hospital-based health service use among people with CP using the Andersen model as a framework.

Methods

The systematic review was conducted following Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and protocol was registered with the International Prospective Register of Systematic Reviews (PROSPERO) on April 28, 2020 (registration number CRD42020176271).

Search strategy

We searched MEDLINE (January 2000-March 25, 2020), Embase (January 2000-March 25, 2020) and APA Psycinfo (January 2000 through the fourth week of March 2020) through the Ovid platform (date of search was March 3, 2020; updated April 20, 2020). The search strategies are shown in supplemental table S1 (available online only at http://www.archives-pmr.org/), with the search adjusted as appropriate for each database. The references of included studies were reviewed for additional inclusions.

List of abbreviations:

CP	**cerebral palsy**
GMFCS	**Gross Motor Function Classification System**
IRR	**incident rate ratio**
OR	**odds ratio**

Eligibility criteria

The inclusion criteria for studies were those that: (1) identified people with CP (of any age), including studies where data were available and attributed distinctly to people with CP among other health conditions or developmental disabilities; (2) reported, in observational studies, quantitative measures of formal utilization of medical and/or multidisciplinary hospital-based health services (hospital inpatient, emergency department, and outpatient services) (including those in private/clinic settings); (3) reported determinants that show a relationship with rates and/or types of hospital-based health service utilization; and (4) were conducted in high-income countries based on the World Bank classification of national economies.[11]

We excluded studies: (1) that were limited to a subset of people with CP not representative of the whole population (eg, studies limited to only those with severe intellectual disability, studies limited to those attending a specific medical subspecialty); and (2) that only reported therapy service utilization (hospital-based or community). We excluded therapy services after piloting the study selection process, which identified a wide range of settings for therapy services (hospital, community) in different countries that we felt would make any appropriate comparison and synthesis challenging. We also felt this approach would be more consistent with exclusion of studies reporting only on specific medical subspecialties. Further minor protocol amendments are documented in PROSPERO.

Study selection

All references selected were imported into Endnote (Endnote X9; Thomson Reuters) and duplicates removed. One author (S.P.) independently screened the titles and abstracts of the selected references. Titles and/or abstracts not published in English were excluded. Full texts were retrieved for all articles included after title/abstract screen. Studies that met title and abstract inclusion criteria with English-language title and abstract, but non-English full-text articles, were translated by native speakers of the language. Full texts were reviewed by 2 authors for inclusion (S.P. in all cases with K.O., S.G., or S.M.) and reasons for exclusion were

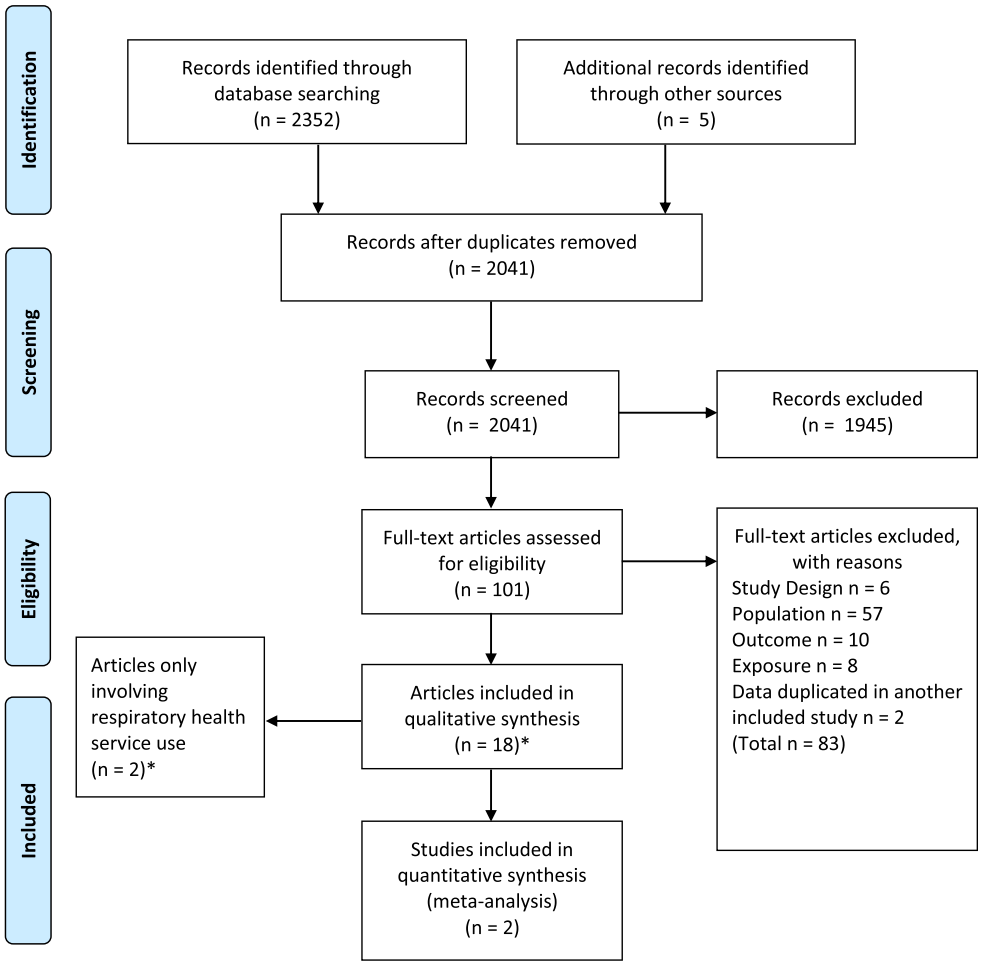

Fig 2 PRISMA flow diagram.

recorded (fig 2). Where disagreements arose, these were resolved through discussion between reviewers.

Risk of bias assessment

We evaluated the quality and risk of bias of included cohort studies using the Newcastle-Ottawa Scale[12] and a modified Newcastle-Ottawa Scale for cross-sectional studies.[13] We supplemented questions from 2 other risk of bias tools[14,15] to meet the domains identified to be important in the methodological design of observational studies (supplemental table 2, available online only at http://www.archives-pmr.org/).[16] Consistent with advice,[16,17] summary scores were not calculated; rather, risk of bias assessment for each study reviewed was presented as a matrix with compliance to criteria noted (table 1). For each study, risk of bias was independently assessed by 2 authors (S.P. in all cases with K.O., S.G., or S.M.), with any differences in scores resolved by discussion.

Data extraction

The data extracted from the included studies were study design, country of origin, sample size, and age range. Determinants of health service utilization reported in included studies were classified consistent with the Andersen behavioral model into individual predisposing, enabling, need, and contextual factors by 2 authors (S.P. in all cases with K.O., S.G., or S.M.).[9] Outcomes were defined as rates and/or proportions of populations using health services and classified by type: inpatient, emergency department, or outpatient. One author (S.P.) extracted data from each of the included studies, and accuracy of data extraction was checked by

Table 1 Characteristics and Quality Assessment of Studies Included in the Systematic Review

Study Details	Setting	Study Period	Population (N)	Age Range (y)	Outcome Types	Data Source (Outcome)	Risk of Bias Assessment Scales and Items: Newcastle Ottawa Scale								A	D
Cohort Studies							Represen-tativeness	Non-exposed Cohort	Exposure Ascertainment	Outcome Timing	Controls	Outcome Assessment	Follow-up Duration	Adequacy of Follow-up	Missing Outcomes	Statistics
Abdullahi 2020[18]	Australia	1983-2010	1663	0-27	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Blackmore 2018 & 2020[19,20,*]	Australia	2010-2016	482	1-26	IP, ED	Administrative health data	★	★	★	★	★	★	★	★	★	★
Carter 2020[22]	UK	2004-2014 (IP) 2010-2014 (OP)	1684	0-24	IP, OP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Kim 2014[24]	South Korea	2007	10659	1-18	IP, OP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Meehan 2017a[25,†]	Australia	2007-2014	1748	0-18	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Meehan 2017b[26,†]	Australia	2007-2014	1748	0-18	ED	Administrative health data	★	★	★	★	0	★	★	★	★	★
Morgan 2020[28]	Australia	2013-2017	551	0-80	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★
Reid 2018[30,†]	Australia	2007-2014	1748	0-18	IP, ED	Administrative health data	★	★	★	★	★	★	★	★	★	★
Yang 2020[32]	Taiwan	2000-2013	3234	0-18	IP	Administrative health data	★	★	★	★	0	★	★	★	★	★

Cross-sectional studies							Newcastle Ottawa Scale: Representativeness	Sample size	Response rate	Exposure ascertainment	Confounding controlled	Outcome assessment	Statistics	A Missing outcomes
Boulet 2009[21]	United States	1997-2003	305	3-17	OP	Self-report	★	★	★	★	★	0	★	★
Fortuna 2018[23]	United States	2011-2014	229	18 - >59[§]	OP, ED	Medical records	★	★	★	★	0	★	★	★
Meehan 2016[27]	Australia	2014	350	5-19	OP	Self-report	★	★	★	0	0	★	★	★
Pons 2017[29]	France	2010-2012	282	18->39[§]	OP	Self-report	★	★	★	★	0	0	★	★
Roquet 2018[31]	France	2010-2013	512	2->39[§]	OP	Self-report	★	★	★	★	★	0	★	★
Whitney 2019[3]	US	2016	5555	18-64	IP, OP, ED	Administrative health data	★	★	★	★	★	★	★	★
Young 2007[33,‡]	Canada	1999-2002	1064	9-32	OP	Administrative health data	★	★	0	★	0	★	★	★
Young 2011[34,‡]	Canada	1999-2002	1064	9-32	IP	Administrative health data	★	★	0	★	0	★	★	★

Abbreviations: A, Agency for Healthcare Research and Quality[15]; D, Downs and Black[14]; ED, emergency; IP, inpatient; OP, outpatient.
* Blackmore 2018 and 2020 limited to inpatient admissions, emergency department attendances due to respiratory causes.
† Studies[25,26,30] derived from same population.
‡ Studies[33,34] derived from same population.
§ Upper age limit unspecified beyond detail given.

a second author (K.O., S.G., or S.M.). Any differences were resolved through review and discussion.

Where possible, data were transformed into proportions and odds ratios calculated to aid comparison and a measure of effect size. Meta-analysis using random effects models was performed to provide pooled odds ratios (OR) and 95% confidence intervals (CI) of health service utilization from 2 or more comparable studies using Review Manager (RevMan) (version 5.3. Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014).

Results

Figure 2 presents the PRISMA flowchart of study selection. We identified 18 articles that described 17 studies for inclusion in the review.[3,18-34] The results of 1 study were reported in an original article[19] and a follow-up letter[20]; both articles were included but described as 1 study for the purpose of this review. Of the 17 studies, 5 reported different aspects of health service utilization for the same population.[25,26,30,33,34] Articles were published between 2006 and 2020, with two-thirds (12 studies, 71%) published during or after 2017 (see table 1).

Studies reported on research conducted in Australia (n=7), the United States (n=3), Canada (n=2), France (n=2), the United Kingdom (n=1), South Korea (n=1), and Taiwan (n=1). Half of the studies (9, 53%) were cohort studies and the remaining 8 were research of cross-sectional design. Seven studies reported exclusively about research on children with CP; 3 reported exclusively about research on adults with CP, and 7 reported on research about both children and adults. The sample size in each study ranged between 229 and 10,659 individuals with CP. The quality of the included studies was generally high (see table 1).

Articles reported research examining health service utilization in inpatient (10 studies), outpatient (9 studies), and emergency department (5 studies) settings. Research measured health service use over varied time periods from 1-27 years; most (12 studies) used administrative data and medical records to measure utilization; and 4 studies used self-report. Sixteen studies reported on health service utilization of any cause. One study[19,20] included only respiratory causes of hospital admissions (over 2 periods) and emergency department attendances and was considered separately.

Overall health service utilization

Determinants

The 16 studies described 26 determinants related to health service use (tables 2 and 3). The most common determinants examined were age (7 studies), sex (4 studies), Gross Motor Function Classification System (GMFCS) level[35] (6 studies), and epilepsy (4 studies). Determinants were matched to Andersen behavioral model characteristics (individual predisposing, individual enabling, individual need, and contextual), with most (n=15) determinants matched to "individual need" characteristics.

Individual predisposing factors

Younger children had more inpatient admissions[32] and outpatient attendances[24] than older children (see table 2). Younger children also represented a greater proportion of total inpatient admissions[25] and emergency department attendances[26] than older children (see table 2). Adults had 50% lower odds of inpatient admission than youth (OR, 0.5; 95% CI, 0.38-0.67)[33] and represented a lower proportion of inpatient admissions than children (table 3).[28] Comparison of outpatient use between adults and children was mixed depending on specialty (see table 3).[31] Most studies identified no association between sex and inpatient admissions (see table 2).[3,22,28,32,33] One study reported lower rates of outpatient attendance in men (OR 0.39; 95% CI 0.32-0.48) (see table 3),[22] but rates of health service use between sexes was similar in another.[3]

Individual enabling factors

The effect of parental employment status and private health insurance on outpatient health care utilization was explored in an Australian study (see table 2).[27] Full-time employment and access to private health insurance were both reported to be associated with increased use of private pediatric medical specialists.[27]

Individual need factors

A total of 15 individual need characteristics (perceived and evaluated) were identified. Persons who primarily used a wheelchair for mobility (GMFCS IV-V; compared with GMFCS I-III) had increased health service use across inpatient,[22,25] outpatient,[22,27,29] and emergency department[26] settings (see tables 2 and 3). Meta-analysis of 2483 children from 2 studies[22,25] showed children functioning at GMFCS levels IV-V having increased odds of hospital admission (pooled OR, 2.53; 95% CI, 1.28-5.02) (supplemental fig 1, available online only at http://www.archives-pmr.org/). Children with a comorbidity of epilepsy had consistently higher odds of inpatient admissions (OR, 2.55; 95% CI, 1.85-3.52),[25,32] outpatient (public) pediatric medical specialist appointments (OR, 2.74; 95% CI, 1.51-4.96),[27] and emergency department attendances (OR, 2.86; 95% CI, 2.27-3.59) (see table 3).[26] Children with a gastrostomy[25-27] and an intellectual[18,30] and/or additional developmental disability[21] (eg, autism, attention-deficit disorder) also had increased likelihood of health service use across settings (see tables 2 and 3).

Contextual factors

Few studies examined the effect of contextual factors on health service utilization (see table 2). Some regions of residence, larger hospital, and better provision of continuity of care were reported to be associated with reduced inpatient admission rates in a Taiwanese study[32]; and regional versus metropolitan location was associated with increased likelihood of private outpatient use in an Australian study.[27]

Respiratory health service utilization

Diseases of the respiratory system were identified as a leading cause of inpatient admissions across age ranges in multiple settings.[25,28,36] Determinants of respiratory inpatient admissions and emergency department attendances was the focus of a study[19,20] based in Western Australia that included some determinants (n=8) not examined in other studies (frequent respiratory symptoms, smoking, scoliosis, oropharyngeal dysphagia, respiratory symptoms with meals, snoring, 2 or more courses of antibiotics in previous year, previous hospitalization). This study found respiratory admissions over 5 years were associated with gross motor severity and similar comorbidities (eg, epilepsy, gastroesophageal reflux disease) as (general) inpatient admissions and emergency department attendances. A higher rate of hospital admission was reported for GMFCS V (compared with GMFCS I), with an incident rate ratio (IRR) of 80 (95% CI, 28.4-192.5), oropharyngeal dysphagia (IRR, 33.6; 95% CI, 17.7-63.7), and

Table 2 Determinants of Health Service Utilization Evaluated and Number of Studies Investigating and Reporting an Association

			Inpatient		Outpatient		Emergency	
Andersen Model Characteristic	Determinant	Total Articles (N)	Number Reporting Association	Direction of Association	Number Reporting Association	Direction of Association	Number Reporting Association	Direction of Association
Predisposing								
Demographic	Age	7						
	- Younger child vs older child	3	1/1	↑	2/2	↑		
	- Child vs adult	3	2/2	↑	1/1	Varied		
	- Younger adult vs older adult	1			0/1	NA	0/1	NA
	Men	4	1/4	NA	1/1	↓		
Social structure	Foreign-born mothers from high-income countries	1	1/1	↓				
	Two-parent household	1			0/1	NA		
	Parental undergraduate degree or higher	1			0/1	NA		
Enabling								
Assets	>=1 parent in full-time employment	1			1/1	↑ (Pr)		
	Private health insurance	1			1/1	↑ (Pr)		
Need								
Perceived	Reported good health	1			0/1	NA		
Evaluated disability	GMFCS IV-V	6	2/2	↑	3/4	↑	1/1	↑
Severity	Bilateral topography	1	1/1	↑				
	Dyskinetic Motor Type	1	1/1	↑				
Evaluated comorbidity	ID	2	1/2	↑			1/1	↑
	DD	1			1/1	↑	1/1	↑
	Epilepsy	4	2/2	↑	1/1	↑	1/1	↑
	Gastrostomy	3	1/1	↑	1/1	↑	1/1	↑
	GORD	1	1/1	↑				
	Nasogastric tube use	1	1/1	↑				
	ITB Pump	2	1/1	↑			1/1	↑
	Asthma	1	0/1	NA				
	PTL & SGA	1	1/1	↓				
	Perinatal complication	1	0/1	NA				
	Pneumonia	1	1/1	↑				
Contextual								
Place of Residence	Place of residence	2	1/1	Varied	1/1	↑ (Pr)		
	Urbanization level	1	0/1	NA				
Health Service Factors	High continuity of care	1	1/1	↓				
	Smaller hospital	1	1/1	↑				

NOTE. ↑Most studies report exposure is associated with increased HSU.
↓Most studies report exposure is associated with decreased HSU.
NA = Nonapplicable: studies do not support association with HSU.
Varied: studies suggest varied associations with HSU.
Abbreviations: COB, country of birth; DD, developmental disability; GMFCS, Gross Motor Function Classification System; GORD, gastro-esophageal reflux disease; ID, intellectual disability; ITB, intrathecal baclofen; Pr, private; PTL, pre-term labor; SGA, small for gestational age.

epilepsy (IRR, 25.6; 95% CI, 13.3-49.4). Previous respiratory hospital admission was also associated with higher rates of inpatient admission (IRR, 29.4; 95% CI, 11.2-77.5), and emergency department attendance (IRR, 11.8; 95% CI, 5.6-24.7).

Discussion

During the last decade there has been an increase in research examining health service utilization in people with CP. To our knowledge, this is the first systematic review to examine the factors that influence rates of health service utilization in this population. Our main findings were evidence of associations between CP severity, comorbidities, and age with health service utilization across settings. We found health service utilization across inpatient, outpatient, and emergency department settings to be increased in children with CP with more severe gross motor function limitations (GMFCS IV-V) and with associated comorbidities including intellectual disability, epilepsy, and gastrostomy-use. We also found limited research investigating and reporting on socioeconomic "individual enabling" factors and health system "contextual" determinants.

These findings are perhaps not surprising given that the management of comorbidities is a main focus of health services and the known association between CP severity and comorbidities.[2] However, they highlight the important role that outpatient health services play in the preventive management of complications and associated conditions of severe CP. Furthermore, they emphasize the necessity of ensuring equitable access to services to minimize unplanned health care use[6] and poor health outcomes.

We found evidence that age across the lifespan influences health service utilization for people with CP. Studies in our review

Table 3 Reported Association Between Andersen Model Determinants and Health Service Utilization by Health Setting

Determinant	Setting	Association(Odds Ratio, 95% Confidence Interval)	Source
Predisposing			
Age 5-9 y (vs older child)	OP	2.32 (1.32-4.07)	Meehan 2016[27,*]
Age (adult vs child)	IP	0.50 (0.38-0.67)	Young 2007[33]
	OP (PMR)	0.29 (0.19-0.45)	Roquet 2018[31]
	OP (psych)	6.42 (3.31-12.47)	Roquet 2018[31]
	OP (neuro)	0.67 (0.46-0.97)	Roquet 2018[31]
Sex (male)	OP	0.39 (0.32-0.48)	Carter 2020[22]
Enabling			
≥1 parent in full-time employment	OP (private)	4.33 (2.68-6.99)	Meehan 2016[27]
Private health insurance	OP (private)	3.07 (1.94-4.87)	Meehan 2016[27]
Need			
GMFCS IV-V	IP	1.82 (1.41-2.33)	Carter 2020[22]
	IP	3.63 (2.51-5.25)	Meehan 2017a[25]
	OP	1.41 (1.12-1.77)	Carter 2020[22]
	OP	2.34 (1.44-3.80)*	Meehan 2016[27]
	OP (PMR)	1.75 (1.08-2.83)	Pons 2017[29]
	ED	3.27 (2.57-4.17)	Meehan 2017b[26]
Bilateral topography	IP	1.63 (1.28-2.07)	Meehan 2017a[25]
Dyskinetic motor type	IP	2.20 (1.13-4.27)	Meehan 2017a[25]
Developmental disability	OP	11.89 (6.77-20.87)	Boulet 2009[21]
	ED	4.44 (1.87-10.51)	Boulet 2009[21]
Epilepsy	IP	2.55 (1.85-3.52)	Meehan 2017a[25]
	OP	2.74 (1.51-4.96)	Meehan 2016[27]
	ED	2.86 (2.27-3.59)	Meehan 2017b[26]
Gastrostomy	IP	6.65 (3.37-13.1)	Meehan 2017a[25]
	OP	3.99 (1.75-9.08)	Meehan 2016[27]
	ED	4.52 (3.15-6.48)	Meehan 2017b[26]
ITB	IP	7.27 (0.99-53.53)	Meehan 2017a[25]
	ED	4.2 (1.6-11.03)	Meehan 2017b[26]
Contextual			
Region of residence (regional vs metropolitan)	OP (private)	1.64 (1-2.7)	Meehan 2016[27]

Abbreviations: ED, emergency department; FT, full-time; GMFCS, Gross Motor Function Classification System; IP, inpatient; ITB, intrathecal baclofen; OP, outpatient; neuro, neurologist; PMR, physical medicine and rehabilitation physician; psych, psychiatrist.
* Meehan 2016[27] is GMFCS III-V.

suggested inpatient admissions reduce with age during childhood and adulthood, and outpatient health service utilizations change in the rates and types of outpatient health services attended between children and adults. The reasons for this are likely to involve both individual and contextual factors, including differences between how pediatric and adult health systems are structured. This may be explained, at least in part, by differences in the goals of the pediatric and adult systems, with the pediatric health system guiding the developing child to optimal physical functioning and participation, compared with the adult health system focus on treating existing health conditions. Similar findings have been described in health service utilization research exploring CP subtypes and severities.[37-40] Most children with CP now live into adulthood[41] and develop additional health problems as adults,[3,42] and many report their health needs are not met during this time.[40,43] There is a need for the health system to adapt to meet this need, with particular emphasis around the time of transition from pediatric to adult health services to ensure that young people with CP do not fall out of the system[39] and greater capacity for adults with CP throughout the lifespan. More research is needed to improve understanding of how health service utilization changes as adults with CP grow older and to support any changes to the health system.

We found less research that has explored the effect of individual enabling factors, such as socioeconomic status, on health service utilization, other than a study that suggests an association between indicators of socioeconomic status with (self-reported) private outpatient service attendance.[27] Socioeconomic disadvantage has been shown to be associated with both CP severity (nonambulatory status, moderate intellectual disability, or greater and more severe comorbidities)[44] and decreased used of health services in other childhood populations.[45,46] Research in broader disability groups have supported inequities of access to health service based around income.[47,48] Children who rely on public funding (Medicaid and Children's Health Insurance Program) may be denied access to some outpatient services or wait longer for treatment.[49] A greater understanding of the socioeconomic factors that influence health service access in CP and the consequences of this can support improvements in health service and support advocacy for structural reforms for funding of children's health care.[50] Studies have shown that simple measures such as continuity of care,[32] integration of clinical care and research data collection,[51] and telehealth services[52] can improve access and health outcomes for children with CP.

Understanding the health service utilization literature and applying its findings is challenging given the complexity of the

data. This review draws attention to the influence that CP severity and comorbidities have on health service use and highlights the need for further research about predisposing and enabling factors that investigate equity of access.

Study limitations

Although the quality of the included studies was generally high, our review was limited in its restriction to high income countries in that differences in the structure and funding of health systems between countries may affect the generalizability of some of the findings. For example, in countries with "user pay" or less universal or established health systems, availability and access to health care for people with CP may be reduced. Our eligibility criteria also excluded studies that focused on particular subgroups of the CP population that would be relevant when considering health service utilization for that subgroup. After protocol amendment, we excluded studies that only reported therapy service utilization; this type of health service provides an important source of support for children with CP; further research that explores determinants of therapy service utilization would be valuable. Some included studies also used self-reported health service use or survey design that may lead to recall bias[53]; however, given this was only a minority of studies, we believe our findings are less likely to be affected by this. We have not reported all measures of health service use described in the studies we have reviewed, such as inpatient admission duration[18,24,25,28,33,34] and subtypes of inpatient admission (eg, emergency, elective).[25,30] These may also be important measures of health service utilization to consider.

Conclusions

Understanding the determinants that influence health service utilization is an important first step toward improving equity of access for people with CP. We identified evidence that health service utilization is associated with age and "individual need" but less evidence to support how "individual predisposing" or "individual enabling" factors such as socioeconomic status might influence access. Improving this understanding should be a research priority, to guide the development of new models of care that aim to provide equitable access to the increasing number of interventions aimed at improving the health and wellbeing of people living with CP.

Keywords

Cerebral palsy; Health services; Health services accessibility; Rehabilitation

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Supplementary Table 1 Search Strategies

(a) MEDLINE

Search Term	Results
1. exp Cerebral Palsy/	20275
2. cerebral palsy.tw	21516
3. 1 or 2	26842
4. exp health services/sn	201371
5. exp emergency medical services/	137120
6. exp ambulatory care facilities/	54325
7. exp hospital departments/	180659
8. exp hospitals, pediatric/	13081
9. exp health services accessibility/	108785
10. (use* or utili* or attend* or present* or admi*) adj2 (department* or health care* or service* or hospital*)).tw	146253
11. 4 or 5 of 6 or 7 or 8 or 9 or 10	640611
12. 3 and 11	551

(b) Embase

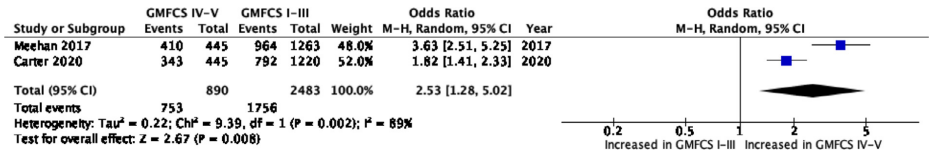
Search Term	Results
1. exp cerebral palsy/	37792
2. "cerebral palsy".tw	31515
3. 1 or 2	40812
4. health service/	160198
5. exp emergency health service/	99067
6. exp outpatient care/	36038
7. exp hospital department/	23185
8. exp outpatient department/	78742
9. exp general hospital/ or exp geriatric hospital/ or exp isolation hospital/ or exp non profit hospital/ or exp pediatric hospital/ or exp public hospital/ or exp teaching hospital/	262011
10. exp ambulatory care/ or exp integrated health care system/ or exp secondary health care/ or exp tertiary health care/	158544
11. ((use* or utili* or attend* or present* or admi*) adj2 (department* or health care* or service* or hospital*)).tw.	224415
12. 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11	943095
13. 3 and 12	1986
14. limit 13 to human	1730

(c) PsycInfo

Search Term	Results
1. exp Cerebral Palsy/	5377
2. cerebral palsy.tw.	7463
3. 1 or 2	7752
4. [exp health services/sn]	0
5. exp emergency medical services/	0
6. exp ambulatory care facilities/	0
7. exp hospital departments/	0
8. exp hospitals, pediatric/	0
9. exp health services accessibility/	0
10. ((use* or utili* or attend* or present* or admi*) adj2 (department* or health care* or service* or hospital*)).tw.	45084
11. 4 or 5 or 6 or 7 or 8 or 9 or 10	45084
12. 3 and 11	71

Supplementary Table 2 Risk of bias and quality assessment

Source	Question	Domain Addressed*
Cohort studies		
Newcastle-Ottawa Scale — Cohort (Wells 2000)	Representativeness of the exposed cohort	Selection
	Selection of the non-exposed cohort	Selection
	Ascertainment of exposure	Exposure
	Comparability of cohorts on the basis of design or analysis	Selection
	Assessment of outcome	Outcome
	Was follow-up long enough for outcomes to occur	Loss to follow up
	Adequacy of follow up of cohorts	Loss to follow up
AHRQ - Viswanathan 2013	Are any important primary outcomes missing from results?	Selective reporting
Downs & Black 1998	Were the main outcome measures used accurate (valid and reliable)?	Outcome
	Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?	Confounding
	Were the statistical tests used to assess the main outcomes appropriate	Analysis
Cross-sectional studies		
Newcastle-Ottawa Scale — Cohort (Herzog 2013)	Representativeness of the sample	Selection
	Sample size	N/A
	Non-respondents	Selection
	Ascertainment of the exposure	Exposure
	The subjects in different outcome groups are comparable, based on the study design or analysis. Confounding factors are controlled	Confounding
	Assessment of the outcome	Outcome
	Statistical test	Analysis
AHRQ - Viswanathan 2013	Are any important primary outcomes missing from results?	Selective reporting



Supplementary Figure 1 Forest plot of GMFCS and inpatient admissions

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10.2 Chapter 4

10.2.1 Statement of Contribution

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The co-authors made the following contributions to the manuscript:

- Simon Paget conceived and designed the study, performed all statistical analyses, lead data interpretation, drafted the initial manuscript and lead subsequent revisions of the manuscript.
- Sarah McIntyre contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Samantha Lain contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Shona Goldsmith contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Natasha Nassar contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

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ORIGINAL ARTICLE

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A comparison of cohorts of children with cerebral palsy from a population register and hospital admission data: A data linkage study

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A commentary based on this article appears on pages 31-33.

Abstract

Background: Administrative health data, such as hospital admission data, are often used in research to identify children/young people with cerebral palsy (CP).

Objectives: To compare sociodemographic, clinical details and mortality of children/young people identified as having CP in either a CP population registry or hospital admission data.

Methods: We identified two cohorts of children/young people (birth years 2001–2010, age at study end or death 2 months to 19 years 6 months) with a diagnosis of CP from either (i) the New South Wales (NSW)/Australian Capital Territory (ACT) CP Register or (ii) NSW hospital admission data (2001–2020). Using record linkage, these data sources were linked to each other and NSW Death, Perinatal, and Disability datasets. We determined the sensitivity and positive predictive value (PPV) of CP diagnosis in hospital admission data compared with the NSW/ACT CP Register (gold standard). We then compared the sociodemographic and clinical characteristics and mortality of the two cohorts available through record linkage using standardised mean difference (SMD).

Results: There were 1598 children/young people with CP in the NSW/ACT CP Register and 732–2439 children/young people with CP in hospital admission data, depending on the case definition used. The sensitivity of hospital admission data for diagnosis of CP ranged from 0.40–0.74 and PPV 0.47–0.73. Compared with children/young people with CP identified in the NSW/ACT CP Register, a greater proportion of those identified in hospital admission data (one or more admissions with G80 case definition) were older, lived in major cities, had comorbidities including epilepsy, gastrostomy use, intellectual disability and autism, and died during the study period (SMD > 0.1).

Conclusions: Sociodemographic and clinical characteristics differ between cohorts of children/young people with CP identified using a CP register or hospital admission data. Those identified in hospital admission data have higher rates of comorbidities and death,

suggesting some may have progressive conditions and not CP. These differences should be considered when planning and interpreting research using various data sources.

KEYWORDS
cerebral palsy, data linkage, health administrative data, patient registries, hospital admission data, register

1 | BACKGROUND

Cerebral palsy (CP) is a common childhood health condition and a leading cause of physical disability. CP represents an 'umbrella diagnosis' under which a broad range of aetiological pathways result in the disruption of early brain development, and cause disorders of movement and posture that can differ in phenomenology, topography, and functional impact.[1] There are currently no specific treatments that target the underlying brain lesion that causes CP, and the condition is lifelong, with substantial health, social, and economic impact, both at an individual and societal level.[2] No biomarkers exist to support the diagnosis of CP, and there are barriers to early diagnosis, with 50% of cases diagnosed after 1 year of age and a false positive diagnosis rate of up to 5%.[3]

Epidemiological research has an important role in improving understanding of CP, including reporting trends in prevalence, aetiology, health outcomes, and health care utilisation. Recent CP epidemiological research has tended to use one of two data sources to define CP populations. Firstly, an increasing number of countries and jurisdictions have developed CP population registers that collect (sociodemographic and clinical) data from people with CP.[4] Research driven by CP registers has been central in documenting the recent decline in birth prevalence of CP in high income countries,[5] and improved life expectancy.[6] Secondly, other research studies have used administrative health data, i.e., data that are routinely generated during patient encounters with the health system, often for financial or clinical purposes.[7] Types of administrative health data have included hospital admission data,[8–14] outpatient visits,[8,12–14] and insurance claims[10,15] and research using this approach has described prevalence,[10,15] patterns of health service utilisation[9,11] and comorbidities.[14,16] Administrative health data are an attractive data source for researchers where CP registers are not available,[10] as they can often be used to identify large cohorts in relatively timely and cost-effective ways, and with little loss to follow-up. However, because the data have been collected for administrative/financial purposes, there is a greater need to assess their validity for research.[17] In addition, administrative health data mostly do not include clinical descriptions (e.g. predominant motor disorder, topography, comorbidities) that are typically available in CP registers. People with CP are often identified in administrative health data using case definitions based on (individual or patterns of) the International Classification of Diseases (ICD-9, ICD-10) codes for CP. Published studies have used a number of different case definitions;[8–15] the impact of chosen case definition on clinical differences in the population studied is unknown.

Studies that have compared CP registers and administrative health data have suggested that administrative health data is only moderately sensitive to a diagnosis of CP, and less sensitive in those born term or with hemiplegia (potentially influenced by less routine follow-up and/or lesser severity).[12] A Norwegian study found administrative health data to misdiagnose CP in 14% of cases.[18] We hypothesized that there are clinically relevant differences between populations of children with CP derived from administrative health data and CP registers, and that these differences may influence research findings. The aims of this study were to compare the sociodemographic and clinical characteristics of cohorts of children with CP identified from administrative

Synopsis

Study Question

How do cohorts of children with cerebral palsy (CP) derived from a CP population register or hospital admission data compare?

What is Already Known

Administrative health data, such as hospital admission data, are often used to identify populations of children with CP. There are limited studies that compare these populations with CP population registers.

What this Study Adds

Compared with a CP population register, hospital admission data had a sensitivity between 0.40 and 0.74 and positive predictive value between 0.47 and 0.73 depending on case definition. Hospital admission data included a higher proportion of children with comorbidities and disease severity, and higher rates of death. This suggests that hospital admission data may potentially include children with degenerative conditions other than CP.

health data (hospital admission data) and a CP register and to determine the differences between cohorts for a key health outcome (mortality).

2 | METHODS

2.1 | Study population and data sources

We conducted this study in New South Wales (NSW), Australia's most populous state with an estimated population of 8.3 million inhabitants, using data from NSW and from neighbouring Australian Capital Territory (ACT) (population 460,000; geographically a small region within NSW borders).[19] There are an estimated 37,000 people with CP in Australia,[20] with a rate that is declining, from 2.1/1000 live births in the mid-1990s to a current low of 1.5/1000 live births (from birth year 2016).[21]

Children and young people with a diagnosis of CP were identified from two data sources (i) the New South Wales (NSW)/Australian Capital Territory (ACT) CP Register (NSW/ACT CPR) and/or (ii) the NSW Admitted Patient Data Collection (APDC). Children and young people were included if born between 1st January 2001 and 30th June 2010 to enable a minimum 10 year follow-up in available administrative health datasets. Children were aged between 2 months and 19 years 6 months at study end or time of death.

The NSW/ACT CPR is a population-based register with multiple ascertainment strategies of individuals with CP who were born or live in NSW or ACT. Families are given the option to opt out of the NSW/ACT CPR (we estimate 120 (7%) children in the age range of this study), and for those that do not opt-out, a minimum data set of sociodemographic, aetiological, and clinical data are collected and used for reporting and research purposes. Data are collected by trained NSW/ACT CPR staff in a standardised way when children are first clinically diagnosed. Cases are verified and clinical information is obtained from multiple sources, including tertiary children's hospitals, community-based services, and through cross-reference to health professionals and medical records. A second data collection is performed for each participant at/after the age of 5 years to confirm diagnosis and comorbidities, and where the diagnosis is still being questioned by treating health professionals, register staff 'keep following' until a definitive decision is made. The NSW/ACT CPR is considered to be complete (population-based) for birth years 2001 to 2016. The more recent birth years are generally not complete until 5 years after birth, i.e. the 2018 birth year data will be complete in early 2024. The NSW/ACT CPR was designated as the gold standard for CP diagnosis/description for comparison to health administrative data.

The APDC contains data for all (inpatient, day or overnight) admissions to public and private hospitals in NSW and contains details of the admission, including dates, up to 50 diagnoses classified using the International Classification of Diseases Australian Modification (ICD10-AM) and up to 50 procedures classified using the Australian Classification of Health Interventions (ACHI) (8th edition). Many ACT residents use hospitals in NSW to support their health needs.

BOX 1 Coding of cerebral palsy using the International Statistical Classification of Diseases and Related Health Problems, Tenth Revision Australian Modification (ICD10-AM).

G80	Cerebral palsy
G80.0	Spastic cerebral palsy
G80.1	Spastic diplegia
G80.2	Infantile hemiplegia
G80.3	Dyskinetic cerebral palsy
G80.4	Ataxic cerebral palsy
G80.8	Other cerebral palsy
G80.9	Cerebral palsy, unspecified
G81	Hemiplegia
G82	Paraplegia and tetraplegia
G83	Other paralytic syndromes

Additional clinical data were available for many of these children through data linkage with other datasets. Perinatal data were obtained from the NSW Perinatal Data Collection, which contains data relating to all births in public and private hospitals and homebirths in NSW. Information about developmental disabilities was obtained from the NSW Family and Community Services (FACS) Disability Dataset, which includes basic diagnostic information from all publicly funded providers of disability services (including case management, therapy, and early intervention services). Deaths of individuals were identified through the NSW Registry of Births, Deaths, and Marriages (BDM) Death Registrations and Cause of Death Unit Record File. Datasets were linked by the New South Wales (NSW) Centre for Health Record Linkage using a probabilistic approach to matching personal identifying information including an automated blocking algorithm and machine learning techniques for assigning weights to different information types.[22]

2.2 | Comparison of CP cohorts derived from the CP Register and APDC

Children and young people with CP in the APDC were identified using hospital discharge dates between 1st January 2001 and 30 June 2020, and applying case definitions based on the relevant ICD10-AM code: G80 (Box 1). We also included a case definition that included other codes: G81 (hemiplegia), G82 (paraplegia and tetraplegia), and G83 (other paralytic symptoms) as this was used in one study identified (Box 1).[8] Various case definitions were compared based on those identified as used in recent studies (Table S1).[8–15] This included the use of one or more ICD10-AM CP diagnosis codes, codes recorded in specific principal or additional fields, in single or multiple admissions, and with and without age restrictions. Children with only birth admissions were excluded from the analysis.

We compared sociodemographic characteristics of children and young people with CP identified from either the NSW/ACT CPR or from the APDC, focussing on the most common case definition of CP in the literature, one or more admissions with G80 code (henceforth APDC-G80) (Figure S1). Postcode of residence and statistical areas were used to determine geographical remoteness and socioeconomic disadvantage. Socioeconomic disadvantage was derived from the Indices of Relative Socioeconomic Disadvantage (IRD) and grouped into quintiles (quintile 1 being the most disadvantaged, and quintile 5 the least disadvantaged).[23] Geographical remoteness (metropolitan; regional; remote areas) was defined using the Australian Statistical Geography Standard.[24]

We then compared the clinical characteristics of the NSW/ACT CPR and APDC-G80 cohorts using variables obtained via data linkage (Figure S1). Gestation at birth (preterm/term) was available in the NSW/ACT CPR and Perinatal Data Collection. Clinical data (predominant motor type, Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS)) were available from the NSW/ACT CPR. The diagnosis of epilepsy was available in the NSW/ACT CPR and APDC. Epilepsy in the APDC was coded as yes/no and defined as one or more admissions with an ICD10 G40 diagnosis. Gastrostomy use (as a marker of severe difficulties with eating and drinking) was derived from the APDC. Gastrostomy was coded as yes/no and defined as one or more admissions with ACHI codes for gastrostomy insertion or revision (30375-07, 30481-00, 30482-00, 30483-00, 92073-00, 90302-00). Neurodevelopmental disorders (intellectual disability, autism) were derived from the NSW/ACT CPR and/or FACS. Deaths, age at death, and underlying cause of death were derived from the BDM. Some clinical characteristics were available in multiple data sources, for these the NSW/ACT CPR was defined as the gold standard for epilepsy and intellectual disability and Perinatal Data Collection for gestation at birth.

2.3 | Statistical analysis

Identification of children with CP was determined from the NSW/ACT CPR and using different case definitions from the APDC. We compared case definitions using the APDC with the NSW/ACT CPR as the gold standard using sensitivity and positive predictive value. We compared sociodemographic details of children with CP in the NSW/ACT CPR and APDC-G80 cohorts using standardised mean difference (SMD). We then identified clinical characteristics of the same cohorts using data linkage of multiple data sources using SMD. Differences in data sources were considered when the absolute standardised mean difference was greater than 0.1.[25] Where variables occurred in multiple data sources agreement was tested using Cohen's kappa statistic. To examine the impact of different data sources on outcomes, we compared proportions of deaths between groups, age at death (medians, interquartile ranges) and top three causes of death (based on ICD-10 chapters). We also compared the clinical characteristics of children identified in both NSW/ACT CPR and APDC G80 cohorts with those in NSW/ACT CPR cohort only using SMD. SAS v9.4 was used to identify individuals with CP and perform analyses and R package *tableone* to calculate SMD.

2.4 | Ethics approval

Ethics approval for the study was attained from the NSW Population and Health Services Research Ethics Committee (2019/ETH11532).

3 | RESULTS

A total of 1598 children and young people with a confirmed diagnosis of CP on the NSW/ACT CPR were identified (42.8% female). Of these, 97.3% ($n=1554$) had at least one (non-birth) hospital admission identified in the APDC during the study period. Of the children and young people who had no hospital admissions during the study period ($n=44$), 52% had spastic unilateral CP ($n=23$) and 75% were classified GMFCS levels I or II ($n=33$). All children who were identified as residing in ACT had at least one hospital admission during the study period.

We identified between 917 and 2439 children and young people with a diagnosis of CP recorded in the APDC using a range of ICD10-AM case definitions (Table 1). Sensitivity ranged from 0.40 (95% confidence interval (95% CI) 0.37, 0.42) (at least one G80 code before 4 years of age) to 0.74 (95% CI 0.72, 0.76) (at least one G80, G81, G82 or G83 codes). Positive predictive value ranged from 0.47 (95%CI 0.45, 0.49) (at least one G80, G81, G82 or G83 codes) to 0.73 (95% CI 0.70, 0.75) (at least one G80 code, principal diagnosis) (Table 1).

Comparison of sociodemographic characteristics of children and young people with CP identified in either the NSW/ACT CPR ($n=1598$) or APDC with common case definition of one or more admissions with G80 code (APDC-G80) ($n=1748$) is shown in Table 2. Compared with children identified in the NSW/ACT CPR, there was a higher proportion in the APDC-G80 cohort who were born in the earlier period (2001–2005, 56.2% vs. 50.5%) and who lived in major cities (65.0% vs. 54.8%) (SMD >0.1).

Data linkage showed $n=1082$ children and young people to be identified in both NSW/ACT CPR and APDC-G80 cohorts (67.7% of children in CPR, 61.9% of children in APDC-G80) (Figure 1). Comparison of clinical characteristics of children identified in the NSW/ACT CPR or APDC-G80 cohort using variables available through data linkage is shown in Table 3. When compared to children identified in the NSW/ACT CPR, a higher proportion of children and young people in the APDC-G80 cohort had comorbidities including epilepsy (31.2% vs. 26.3%), gastrostomy use (21.3% vs. 13.6%), intellectual disability (51.4% vs. 49.0%) and autism (14.9% vs. 11.0%) (SMD >0.1). Where clinical characteristics were available in multiple data sources, there was at least moderate agreement between sources (Table 4).

A higher proportion of children in the APDC-G80 cohort died during the study (8.1% vs. 2.9% in CPR) (SMD >0.1), and these deaths occurred at a median earlier age 6.3 years (interquartile range (IQR) 3.4–9.7 years), compared with 8.4 years (IQR 5.1–11.1 years) in the NSW/ACT CPR. The top three underlying causes of death was

TABLE 1 Sensitivity and positive predictive value of a diagnosis of cerebral palsy from the Admitted Patient Data Collection (APDC) compared with the NSW/ACT Cerebral Palsy Register (CPR), using various ICD10-AM diagnostic code case definitions for cerebral palsy.

ICD10-AM Diagnosis code case definition used in APDC	CP diagnosis in CPR and APDC (*n*)	CP diagnosis in APDC, not in CPR[a] (*n*)	CP diagnosis in CPR, not in APDC (*n*)	CP diagnosis in APDC (total) (*n*)	Sensitivity (95% CI)	PPV (95% CI)
	a	b	c	a+b	a/(a+c)	a/(a+b)
1 or more G80 codes	1082	666	472	1748	0.70 (0.67, 0.72)	0.62 (0.60, 0.64)
1 or more G80 codes after 2years age	1049	568	505	1617	0.68 (0.62, 0.70)	0.65 (0.63, 0.67)
1 or more G80 codes before 4years age	614	303	940	917	0.40 (0.37, 0.42)	0.67 (0.64, 0.70)
1 or more G80 codes, principal field	790	295	764	1085	0.51 (0.48, 0.53)	0.73 (0.70, 0.76)
1 or more G80 codes (top 3 fields)	1041	592	513	1633	0.67 (0.65, 0.70)	0.64 (0.61, 0.66)
2 or more plus G80 codes	854	362	700	1216	0.55 (0.53, 0.57)	0.70 (0.68, 0.73)
1 or more G80 or G81 codes	1142	1154	412	2296	0.74 (0.71, 0.76)	0.50 (0.48, 0.52)
1 or more G80, 81, 82 or 83 codes	1153	1286	401	2439	0.74 (0.72, 0.76)	0.47 (0.45, 0.49)

[a]There are approximately 120 children who are 'counted' in the CP Register prevalence analyses, that were not able to be linked, as they have 'opted off' the CP Register.

Abbreviations: CI, confidence interval; CP, cerebral palsy; ICD10-AM International Statistical Classification of Diseases and Related Health Problems, Tenth Revision, Australian Modification; PPV Positive Predictive Value; ICD10-AM codes: G80 cerebral palsy; G81 hemiplegia; G82 paraplegia, tetraplegia; G83 other paralytic syndromes.

similar between the groups (diseases of the nervous system, diseases of the respiratory system, congenital malformations).

Compared with children in the NSW/ACT CPR cohort but not in the APDC-G80 cohort, those identified in both cohorts were more likely to have spastic bilateral or dyskinetic motor types, be GMFCS IV-V and/or MACS IV-V and were more likely to have epilepsy and/or intellectual disability (Table S2).

4 | COMMENT

4.1 | Principal findings

Hospital admission data and a common case definition (one or more admissions with G80 code) had a sensitivity of 70% and a PPV of 62% for identifying CP in children compared with a gold standard of a population CP register (NSW/ACT CPR). Sensitivity varied from 0.40 to 0.74 and PPV varied from 0.47 to 0.73 when using other case definitions with hospital admission data. When compared with children identified in the NSW/ACT CPR, a higher proportion of children with CP identified in hospital admission data had comorbidities, lived in major cities, and had died during the study period.

4.2 | Strengths of the study

Strengths of this study include the use of data linkage of multiple datasets to assess differences between the CP register and hospital admission data cohorts, which supported a more comprehensive description of the populations. Our results can be used by researchers and research end-users alike when considering bias influencing the interpretation of epidemiological CP research.

4.3 | Limitations of the data

The main limitation of this study is the availability of diagnosis (ICD10-AM) codes only in the hospital admission data (APDC), and not in other data sources (e.g. outpatient, insurance claim) as in some other studies.[8,10,12–14] However, as almost all (97.3%) participants had at least one hospital admission, this is not likely to have influenced the results. The small number of children who have who have opted out of the NSW/ACT CPR may also have influenced the sensitivity and PPV rates that we have reported, although likely not substantially. Potential errors in coding that often exist in hospital admission data should also be considered; we have attempted to minimise these by including data from multiple admissions where available, and linkage with other data sources. Our study was also limited to children, with the oldest participant being 19 years old. It is therefore unclear whether our findings would be replicated in adults. Future research focused on adults with CP would be helpful to explore this further.

TABLE 2 Comparison of the sociodemographic characteristics of children and young people identified as having cerebral palsy in either the CP Register (CPR) or hospital admission data (APDC).

Sociodemographic characteristic	CPR ($n=1598$) n (%)	APDC[a] ($n=1748$) n (%)	SMD[b]
Sex			
Male	914 (57.2)	1003 (57.4)	.034
Female	684 (42.8)	744 (42.6)	
Birth years			
2001–2005	807 (50.5)	983 (56.2)	.115
2006–2010	791 (49.5)	765 (43.8)	
Index of relative social disadvantage			
1 (most disadvantaged)	290 (18.1)	323 (18.5)	.101
2	288 (18)	288 (16.5)	
3	315 (19.7)	304 (17.4)	
4	327 (20.5)	290 (16.6)	
5 (least disadvantaged)	290 (18.1)	239 (13.7)	
Unknown	88 (5.5)	304 (17.4)	
Remoteness			
Major city	876 (54.8)	1136 (65)	.182
Regional	625 (39.1)	526 (30.1)	
Remote	10 (0.6)	13 (0.7)	
Unknown	87 (5.4)	73 (4.2)	

[a]Cerebral palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis.

[b]Differences in data sources were considered where SMD > 0.1.

Abbreviations: APDC, Admitted Patient Data Collection; CPR, Cerebral Palsy Register; SMD, Standardised Mean Difference.

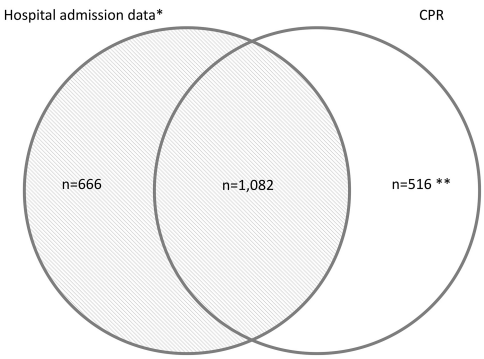

FIGURE 1 Overlap of children with a diagnosis of CP ascertained from either a CP register or hospital admission data. *Cerebral palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis. **Includes $n=44$ cases in CPR where no admission during study period, and $n=472$ cases in CPR where cases had one or more admissions but no admissions with G80 diagnosis. CPR NSW/ACT Cerebral Palsy Register.

4.4 | Interpretation

Understanding the differences between cohorts from administrative health data such as hospital admission data and a CP register is crucial for researchers to consider when planning studies, and for research end-users when interpreting study results. Our findings suggest that studies with populations derived from hospital admission data will include a population with more comorbidities and greater severity. These factors are known to be associated with greater health service utilisation[26] and adverse health outcomes[27,28] and results based on such populations are likely to overstate these (negative) outcomes. We also noted that children with CP identified from hospital admission data were more likely to be older than those from a CP register. This may be due to these children having a longer lived period to experience a hospital admission. Researchers should ensure a long enough period to capture health care episodes (such as hospital admissions) when using health administrative data such as hospital admission data to ascertain their population.

Our findings also highlight that many (27%–53%) CP cases defined using administrative health data are not found in a population-based CP register. Some of these cases may represent families who have chosen to not have their details included in a CP register (opted out) but are still counted for prevalence estimates (we estimate $n=120$ (7.5%) cases in our study). Some may represent cases of CP missed by a CP register, with a recent Norwegian study reporting up to 60% of cases of CP ascertained in administrative health data but not in a CP population registry were correct.[18] Other cases may represent known complexities in diagnosis of CP, including definitions based on age at injury (post-neonatal causes), children who would 'lose' a CP diagnosis following clinical review, when another, progressive condition is identified (false positives)[17] or those described as 'at risk

TABLE 3 Comparison of the clinical characteristics of children and young people ascertained using data linkage to various data sources based on identification of cerebral palsy using either CP Register or hospital admission data (APDC).

Data source used to identify clinical characteristic	Clinical characteristic	Data source used to identify cerebral palsy		
		CPR *n* (%)	APDC[a] *n* (%)	SMD[b]
CPR		1598 (100)	1082 (61.9)	
	Epilepsy			
	Yes	420 (26.3)	338 (31.2)	0.128
	No or resolved	865 (54.1)	564 (52.1)	
	Unknown	313 (19.6)	180 (16.6)	
	Intellectual disability			
	Yes	783 (49)	556 (51.4)	0.048
	No	610 (38.2)	395 (36.5)	
	Unknown	205 (12.8)	131 (12.1)	
PDC		1331 (83.3)	1357 (77.6)	
	Gestation at birth			
	Preterm	488 (36.7)	448 (33)	0.133
	Term	842 (63.3)	908 (66.9)	
APDC		1554 (97.2)	1748 (100)	
	Gastrostomy use			
	Yes	211 (13.6)	373 (21.3)	0.152
	No	1343 (86.4)	1375 (78.7)	
FACS		1413 (88.4)	1454 (83.2)	
	Autism			
	Yes	156 (11)	217 (14.9)	0.178
	No	1257 (89)	1237 (85.1)	
BDM	Death			
	Yes	47 (2.9)	142 (8.1)	0.228
	No	1551 (97.1)	1606 (91.9)	

[a]Cerebral Palsy case definition based on hospital admission data (APDC) where an individual had one or more admissions with a recorded ICD10-AM G80 diagnosis.

[b]Differences in data sources was considered where SMD > 0.1.

Abbreviations: APDC, Admitted Patient Data Collection; BDM, Registry of Births Deaths and Marriages; CPR, New South Wales/Australian Capital Territory Cerebral Palsy Register; FACS, Family and Community Services dataset; PDC, Perinatal Data Collection; SMD, Standard Mean Difference.

TABLE 4 Agreement between clinical variables ascertained from either a cerebral palsy register (CPR) or hospital admission data (APDC) in children with cerebral palsy.

Clinical variable	Source dataset 1	Source dataset 2	Kappa (95% CI)
Epilepsy	CPR	APDC	0.70 (0.66, 0.75)
Prematurity	CPR	PDC	0.93 (0.91, 0.96)
Intellectual disability	CPR	FACS	0.44 (0.39, 0.50)

Abbreviations: APDC, Admitted Patient Data Collection; CI, confidence interval; CPR, New South Wales/Australian Capital Territory Cerebral Palsy Register; FACS, Family and Community Services dataset; PDC, Perinatal Data Collection.

of CP' who have no discernible functional impairment at the age of 5 years when a diagnosis of CP might be confirmed, and overlap with other neurodevelopmental disorders.[29,30]

There are limited studies that have compared CP population registers and administrative health data. Comparison of the APDC with the CPR in our study was similar to that found in a recent study based on inpatient and outpatient visits in Quebec, when using a similar case definition.[12] A study that used a broader case definitions (e.g. including G81, G82 or G83 ICD-10AM codes) is likely to have included a larger proportion of individuals who are not in CP population registers.[8]

In recent years, there has been a substantial increase in research about adults with CP,[31] including several epidemiological studies that have used administrative health datasets to explore a range of outcomes.[13,14] Many (younger) CP registers may not have sufficient data on adults with CP at the present time, and in this group administrative health data may be particularly useful. Researchers should be aware of impact of case definitions on sensitivity and PPV when using administrative data to ascertain CP and choose case definitions that best meet the needs of the research question.

Where available, the use of CP registers to define a research population offer substantial advantages over using administrative health data, and data linkage can further enhance their value for research. CP registers often include condition-specific clinical data including motor type, markers of functional severity (e.g. GMFCS), and comorbidities such as developmental disabilities which are often not captured or incompletely captured in more general administrative health data. Data linkage of CP registers with other data sets can enable a comprehensive picture of health and education outcomes and service utilisation. Data linkage research of large datasets may enable the identification of patterns of treatment outcomes, supporting health care providers to develop more personalised treatment programs such as medication or therapy interventions.

5 | CONCLUSIONS

In summary, this study has shown that populations of children with CP derived from CP registers and hospital admission data are likely to differ in severity and associated comorbidities. The chosen method for population ascertainment should be considered when interpreting the results of epidemiological research.

AUTHOR CONTRIBUTIONS

SP, SM and NN conceived and designed the study. NN and SMc obtained access to the data. SP performed the analysis. SP, SM, SL, SG and NN contributed to data interpretation. SP drafted the initial manuscript. All authors made substantial contributions to the analysis, plan, data interpretation and critically revised the manuscript.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the NSW Ministry of Health, but restrictions apply to data availability, which were used under licence for the current study and therefore are not publicly available.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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10.3 Chapter 5

10.3.1 Statement of Contribution

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The co-authors made the following contributions to the manuscript:

- Simon Paget conceived and designed the study, performed all statistical analyses, lead data interpretation, drafted the initial manuscript and lead subsequent revisions of the manuscript.
- Sarah McIntyre contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Shona Goldsmith contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Katarina Ostojic contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Jane Shrapnel obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Francisco Schneuer contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Mary-Clare Waugh contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Maria Kyriagis contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Natasha Nassar contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

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ORIGINAL ARTICLE

Non-attendance at outpatient clinic appointments by children with cerebral palsy

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Aim: To determine factors that influence non-attendance at outpatient clinics by children with cerebral palsy (CP).

Method: This was a retrospective cohort study of 1395 children with CP (59.6% male; born 2005 to 2017) identified from the New South Wales (NSW)/Australian Capital Territory CP Register, who had scheduled appointments at outpatient clinics at two NSW tertiary paediatric hospitals between 2012 and 2019. Associations between sociodemographic, clinical, and process-of-care factors and non-attendance were examined using multivariate logistic regression with generalized estimating equations.

Results: A total of 5773 (12%) of 50 121 scheduled outpatient days were not attended. Non-attendance increased over time (average increase 5.6% per year, 95% confidence interval [CI]: 3.7–7.3). Older children aged 5 to 9 years (adjusted odds ratio [aOR] 1.11; 95% CI: 1.02–1.22) and 10 to 14 years (aOR 1.17; 95% CI: 1.03–1.34), socioeconomic disadvantage (aOR 1.29; 95% CI: 1.11–1.50), previous non-attendance (aOR 1.38; 95% CI: 1.23–1.53), and recent rescheduled or cancelled appointments (aOR 1.08; 95% CI: 1.01–1.16) were associated with increased likelihood of non-attendance.

Interpretation: One in eight outpatient appointments for children with CP were not attended. Non-attendance was associated with increasing age, socioeconomic disadvantage, previous non-attendance, and recent rescheduled or cancelled appointments. Identifying specific barriers and interventions to improve access to outpatient services for these groups is needed.

Cerebral palsy (CP) is a neurodevelopmental condition characterized by a permanent disorder of movement and posture attributed to non-progressive disturbances in early brain development.[1] CP is the most common cause of physical disability in childhood, with a birth prevalence of approximately 2.0 per 1000 live births in most high-income countries,[2] although prevalence has declined in recent birth years in Australia.[3] For many children with CP, the motor disorder is accompanied by neurological disorders (e.g. epilepsy), diseases of other body systems (e.g. respiratory, digestive system),[4] and musculoskeletal deformities[5,6] (e.g. scoliosis, hip displacement) that further complicate their health.

Abbreviations: ACT, Australian Capital Territory; NAP, Non-admitted patient; NSW, New South Wales; SCHN, Sydney Children's Hospitals Network.

The long-term management of CP and its associated health conditions and complications is conducted by health services,[7] with most management occurring in outpatient settings. As children with CP often have complex health needs, specialty medical and surgical outpatient services are generally centralized in children's hospitals and frequently involve multidisciplinary teams including medical, nursing, and allied health professionals.

Non-attendance at scheduled outpatient appointments is recognized as a major issue across the health care system and health conditions. At a patient level, non-attendance may represent a missed opportunity for early diagnosis of a health-related problem, or the initiation of an intervention to improve the health or well-being of their child. In children with neurological conditions, it may result in increased, unplanned health care use such as emergency department presentations.[8] At a health service level, non-attendance is recognized to increase health care costs, decrease services' effective capacity, and add to waiting times for consultations and procedures.[9]

In this context, understanding factors associated with non-attendance at outpatient appointments is important to enable the identification of strategies to improve attendance and health outcomes for children with CP.[10] A systematic review of non-attendance across patient groups identified multiple factors that are likely to be relevant, including elements related to the individual (younger age, lower socioeconomic status, history of previous non-attendance) and those related to the clinic (e.g. specialty type) and service.[9] Given the complexity and diversity of CP, other aspects, such as the severity of CP and comorbidities, may also be important. We aimed to explore the factors associated with non-attendance by children with CP at specialty outpatient clinics located at two locations across a tertiary children's hospital network.

What this paper adds

- Twelve per cent of scheduled appointments for children with cerebral palsy are not attended.
- Proportions of appointments not attended has increased over the last decade.
- Increasing age and socioeconomic disadvantage increase the likelihood of non-attendance.
- Previous non-attendance and recent cancelled or rescheduled appointments increase the likelihood of further non-attendance.

METHOD

Study population and data sources

We conducted a retrospective cohort study of children with CP, born from 2005 to 2017, who attended outpatient clinics at two children's hospitals in New South Wales (NSW) that provide services for children in NSW and the Australian Capital Territory (ACT). Children with CP were identified from the NSW/ACT CP Register (n=1764), a population-based database with multiple ascertainment strategies. The Register contains details of individuals with CP who were born or live in NSW or the ACT, including demographic and clinical (motor type, severity of CP, presence of comorbidities) information. For each child, corresponding information was ascertained on outpatient appointments scheduled at either of two tertiary paediatric hospitals in metropolitan Sydney: Sydney Children's Hospital, Randwick and the Children's Hospital at Westmead (as part of the Sydney Children's Hospitals Network [SCHN]) between 1st January 2012 and 31st December 2019. This time frame was chosen because data before 2012 were incomplete due to changes in the data collection processes. Outpatient data were obtained from the SCHN non-admitted patient (NAP) administrative data collection. SCHN NAP data is based on two sources: data documenting scheduled outpatient appointments and patient-level clinician activity including demographic information, clinical specialty type, location, attendance/non-attendance, and clinician discipline. The accuracy of the NAP data collection is ensured as it is a statutory with the NSW Ministry of Health mandating the collection and reporting of patient level non-admitted activity for all clinical and/or therapeutic services provided or contracted by NSW Health. Outpatient clinics at both hospitals are provided under a government universally funded system (either state-funded or Medicare) without a fee to the patient, typically 8am to 5pm, Monday to Friday.

Study outcomes

The main study outcome was frequency of scheduled outpatient appointments categorized as attended or not attended. The data available in the SCHN NAP did not discriminate between appointments rescheduled or cancelled by the hospital for administrative reasons (e.g. staff being unavailable) and those rescheduled by families. Scheduled outpatient appointments were categorized based on clinical specialty (see Table S1) and health care professionals seen were categorized by discipline (medical/dental, nursing, allied health, other). To adjust for varied scheduling practices (e.g. some specialties scheduled multiple appointments with health care professionals of different disciplines on the same day), scheduled outpatient appointments were converted to outpatient days. At each (attended) outpatient day, a child could be reviewed by different clinical specialties and seen by multiple health care professionals of different disciplines. A flow diagram presenting an overview of the study processes including study exclusions is shown in Figure S1.

Patient sociodemographic, clinical, and process of care factors

Patient sociodemographic and clinical factors were collected from the CP Register and SCHN NAP, and included

demographic information on age at appointment, sex, preferred language, and country of birth. Patients' postcode of residence was used to estimate socioeconomic disadvantage and geographical remoteness. Socioeconomic disadvantage was measured with reference to the general population, using the Index of Relative Socioeconomic Disadvantage and categorizing into quintiles (quintile 1 being the most disadvantaged and quintile 5 being the least disadvantaged).[11] Geographical remoteness was defined using the Australian Statistical Geography Standard, which categorizes populated localities as major cities, inner/outer regional, and rural/remote areas) based on ease of access to services via road network.[12] Clinical variables included Gross Motor Function Classification System (GMFCS) classification (dichotomized into levels I–III [ambulant] and IV–V [non-ambulant]),[13] predominant motor type (grouped into spastic, dyskinetic, and other [ataxia, hypotonia, those identified as 'early and at risk' of CP]), and the presence of comorbidities of epilepsy and intellectual disability (dichotomized as 'yes' or 'no').

Process-of-care factors were identified using NAP data. Recent multidisciplinary team care was defined as review by two or more health care professionals from different disciplines at the previous outpatient day (visit). Recent experience of care coordination was defined as review by two or more different clinical specialties at the previous outpatient day. Recent non-attendance was defined as non-attendance at the previous outpatient day. Appointments that were rescheduled or cancelled were also identified. Recent rescheduled or cancelled appointments were defined as one or more rescheduled/cancelled appointment in the previous 6 months. Where there was no previous recorded appointment (e.g. at the first scheduled outpatient day during the study period), these process-of-care factors were classified as 'no'.

Statistical analysis

Children who were scheduled to attend clinic appointments at either of the two hospitals on at least one occasion (n=1395) were described in terms of their demographic and clinical features. Proportions, counts, and rates of scheduled appointments by specialty type were compared. Characteristics of children reviewed by major specialties and proportions of scheduled outpatient appointments by age group were compared using χ^2 tests. Associations between patient factors and non-attendance were assessed using univariate and multivariate logistic regression including date of appointment, child sociodemographic and clinical factors, and process-of-care measures. Multivariate analyses were conducted using generalized estimating equations and an exchangeable correlation structure to account for repeated outpatient attendances by the same child. Analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC, USA). The study was approved by the SCHN human research ethics committee (2019/ETH11829).

RESULTS

We identified 1395 children from the NSW/ACT CP Register who had one or more outpatient appointment scheduled during the study period (Table 1). Of these children, 831

TABLE 1 Characteristics of 1395 children with cerebral palsy with scheduled outpatient appointments, 2012 to 2019

Demographic/clinical factor	*n* (%)
Sex	
Male	831 (59.6)
Female	564 (40.4)
Country of birth	
Australia	1298 (93.7)
Overseas	88 (6.3)
Preferred language	
English	1214 (91)
Other	120 (9)
Remoteness	
Major cities of Australia	941 (67.7)
Inner regional Australia	399 (28.7)
Outer regional Australia	42 (3)
Remote Australia	7 (0.5)
State/territory of residence	
New South Wales	1323 (95.9)
Australian Capital Territory	57 (4.1)
IRSD quintile	
1 (most disadvantaged)	274 (19.7)
2	210 (15.1)
3	275 (19.8)
4	288 (20.7)
5 (least disadvantaged)	342 (24.6)
GMFCS level	
I–III	998 (74.5)
IV–V	342 (25.5)
Predominant motor type	
Spastic	1010 (73.6)
Dyskinetic	192 (14.0)
Other	171 (12.5)
Intellectual disability	
Yes	645 (46.2)
No	515 (36.9)
Not reported	235 (16.8)
Epilepsy	
Yes	394 (28.2)
None or resolved	796 (57.1)
Not reported	205 (14.7)

Abbreviations: GMFCS, Gross Motor Function Classification System; IRSD, Index of Relative Socioeconomic Disadvantage.

(59.6%) were male; most (1340; 96.1%) lived either in major cities or inner regional areas.

There was a total of 50 121 scheduled outpatient days during 2012 to 2019; each child had a median of 4.8 (interquartile range [IQR)] 2.0–7.9) scheduled appointments per year. There was variation in the frequency and involvement of different specialties (Table 2). Most children were reviewed one or more times by rehabilitation medicine (82.2%), allied health (78.9%), and neurology/neurosurgery (55.6%) clinics. These clinics were also the most frequently attended (Table 2). There were differences between the groups of children reviewed by different specialties (Table S2). Compared with children without each respective comorbidity, children with epilepsy (odds ratio [OR] 6.28; 95% confidence interval [CI]: 4.68–8.44) and intellectual disability (OR 3.06; 95% CI: 2.41–3.90) were substantially more likely to be seen in neurology/neurosurgery clinics; children with non-ambulant CP were more likely to be reviewed in orthopaedic clinics (OR 3.69; 95% CI: 2.84–4.79) (Table S2). There were also differences in specialty scheduled outpatient days between age groups (Table S3). The 0 to 4-year age group attended 54% of neurology outpatient days (compared with 40% of total outpatient days) and the 10 to 14-year age group attended 11% of neurology outpatient days (compared with 15% of total outpatient days). In contrast, the 0 to 4-year age group attended 18% of orthopaedic outpatient days while the 10 to 14-year age group attended 30% of orthopaedic outpatient days.

Most (*n*=44 348, 88.5%) scheduled outpatient days were attended, with children seen by a single provider in about half (51.1%) of all attended outpatient days (Fig. 1). Multidisciplinary team care was provided in 39.4% of attended outpatient days and most involved a doctor and an allied health professional (47.7%), doctor and nurse (23.1%), or doctor, nurse, and allied health professional (24.4%) (Fig. 1). Care coordination of multiple specialty appointments occurred in 19.9% (*n*=8813) of all attended outpatient days.

A total of 5773 (11.5%) scheduled outpatient days were not attended (Table 2). The rate of non-attendance increased on average by 5% per year (OR 1.05; 95% CI: 1.04–1.07) from 11.5% in 2012 to 14.2% in 2019. The rate of increase was similar among the 0 to 4-year age group (OR 1.04; 95% CI: 1.01–10.07), 5 to 9-year age group (OR 1.04; 95% CI: 1.01–1.06), and 10 to 14-year age group (OR 1.08; 95% CI: 1.01–1.16) (Fig. 2).

The association between sociodemographic, clinical, and process-of-care factors and non-attendance is shown in Table 3. After adjusting for all factors, increased likelihood of non-attendance was associated with older age children 5 to 9 years (aOR 1.11; 95% CI: 1.02–1.22) and 10 to 14 years (aOR 1.17; 95% CI: 1.03–1.34) and greater socioeconomic disadvantage (Index of Relative Socioeconomic Disadvantage quintile 1: aOR 1.29; 95% CI: 1.11–1.50 and Index of Relative Socioeconomic Disadvantage quintile 2: aOR 1.50; 95% CI: 1.29–1.76) (Table 3). There was no statistical evidence of an association between clinical variables, such as GMFCS level and predominant motor type, and likelihood of non-attendance. Recent experience of multidisciplinary team or coordinated care was not associated with the likelihood of non-attendance. However, children with recent rescheduled or cancelled appointments (aOR 1.08; 95% CI: 1.01–1.16) or previous non-attendance (aOR 1.38; 95% CI: 1.23–1.53) had increased odds of non-attendance.

DISCUSSION

Non-attendance at outpatient clinics for children with CP is a little-researched area. We found non-attendance to be associated with four factors: increasing age, socioeconomic disadvantage, previous non-attendance at an outpatient clinic, and recent cancellation or rescheduling of an appointment. Non-attendance was not associated with area of residence, CP severity, nor the presence of major comorbidities. Non-attendance was also not associated with recent multidisciplinary team or coordinated care. Rates of non-attendance increased during the study period.

Outpatient clinics are the dominant model through which the health system provides support for the management of

TABLE 2 Number and proportion of children with cerebral palsy attending scheduled outpatient appointments and non-attendance by specialty group, 2012 to 2019

Specialty group	Children attending outpatient clinics *n* (%)	Frequency of outpatient days *n* (%)	Number of scheduled appointments/year mean (SD)	Scheduled appointments not attended *n* (%)
Allied health	1100 (78.9)	19 008 (37.9)	2.2 (2.7)	2187 (11.5)
General medicine	297 (21.3)	1863 (3.7)	0.2 (0.9)	416 (22.3)
Rehabilitation medicine	1147 (82.2)	14 918 (29.8)	1.8 (2.1)	1651 (11.1)
Neurology/neurosurgery	776 (55.6)	4982 (9.9)	0.7 (1.6)	469 (9.4)
Other medical specialty	776 (55.6)	6172 (12.3)	0.8 (2)	669 (10.8)
General surgery	305 (21.9)	1030 (2.1)	0.1 (0.5)	127 (12.3)
Orthopaedics	604 (43.3)	4433 (8.8)	0.5 (0.9)	655 (14.8)
Other surgical specialty	681 (48.8)	4559 (9.1)	0.6 (1)	604 (13.2)
Medical imaging	625 (44.8)	4620 (9.2)	0.6 (1.2)	214 (4.6)
Total	1395 (100)	50 121 (100)	6.1 (6.1)	5773 (11.5)

Numbers and percentages do not sum to totals as children may attend multiple specialty clinics.

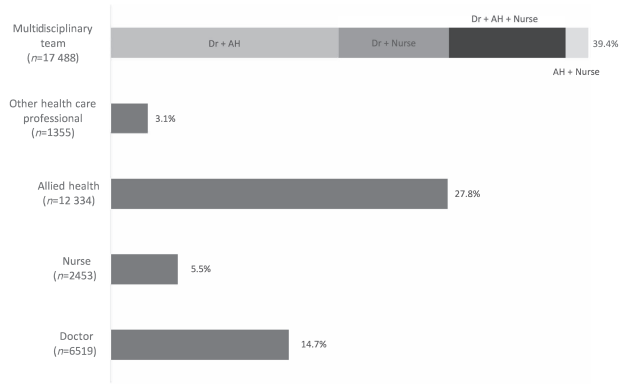

FIGURE 1 Proportion of outpatient days attended by children with cerebral palsy (by health care professional discipline). The health care professional involved was unknown in 4199 (9.5%) outpatient days; all groups are mutually exclusive. AH, allied health

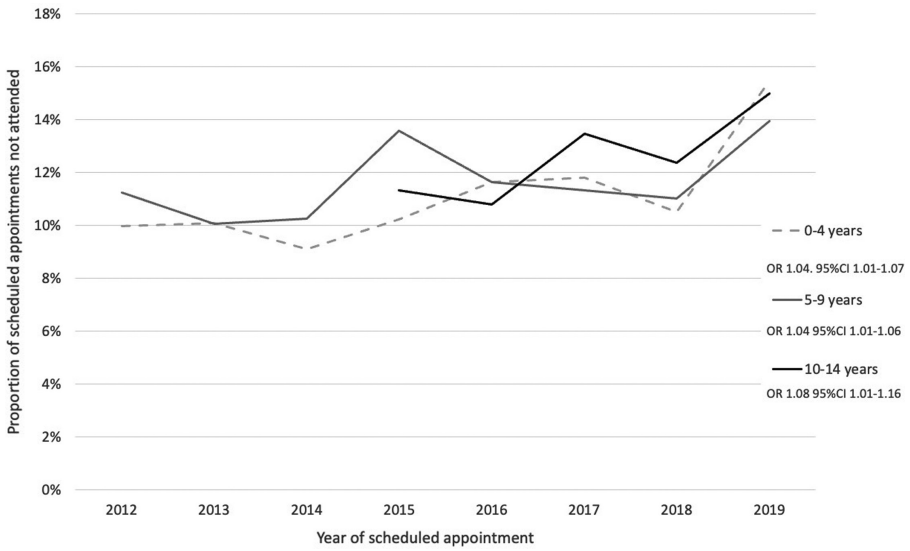

FIGURE 2 Proportion of scheduled outpatient clinic appointments not attended by year and age group in children with cerebral palsy at a children's hospitals network

chronic health conditions. Non-attendance at outpatient clinics can, therefore, have important consequences for children with CP. Not attending an outpatient clinic appointment means a child misses an opportunity to receive timely (and evidence-based) health interventions and/or engage in health surveillance and education. This may result in their using unplanned heath care (e.g. emergency departments) to support their needs,[8] which can contribute over time to worse health outcomes. Our results suggest that children at greater socioeconomic disadvantage, who are already known to have higher rates of CP severity, intellectual disability, and comorbidities,[14] are also inequitably exposed to these risks. It is encouraging that patients of overseas birth and non-English speaking backgrounds, or those from regional or remote areas were not associated with non-attendance.

That non-attendance increases with age also requires further investigation. While this may represent changing priorities as children grow older, greater need for young-person engagement, or reduced perceived need, some health conditions associated with CP are known (for the most part) to

TABLE 3 Association between sociodemographic, clinical, and process-of-care factors with non-attendance at outpatient clinics for children with cerebral palsy

Sociodemographic, clinical, and process-of-care factors	Univariate OR (95% CI)	Multivariate OR (95% CI)
Sociodemographic factors		
Year of appointment	1.05 (1.04–1.07)	1.04 (1.02–1.06)
Sex		
Male	1.04 (0.94–1.15)	1.01 (0.92–1.12)
Female	Reference	Reference
Age		
0–4 years	Reference	Reference
5–9 years	1.22 (1.12–1.34)	1.12 (1.03–1.23)
10–14 years	1.45 (1.3–1.63)	1.19 (1.04–1.35)
Country of birth		
Australia	Reference	Reference
Overseas	1.11 (0.92–1.34)	1.06 (0.8–1.27)
Preferred language		
English	Reference	Reference
Other	1.08 (0.92–1.26)	0.98 (0.83–1.16)
IRSD quintile		
1 (most disadvantaged)	1.32 (1.13–1.53)	1.30 (1.12–1.52)
2	1.50 (1.28- 1.76)	1.52 (1.30–1.78)
3	1.20 (1.04–1.40)	1.20 (1.03–1.39)
4	1.12 (0.96–1.30)	1.13 (0.97–1.30)
5 (least disadvantaged)	Reference	Reference
Remoteness		
Major cities of Australia	Reference	Reference
Regional/remote	1.04 (0.92–1.16)	0.95 (0.85–1.05)
Clinical factors		
GMFCS level		
I–III	Reference	Reference
IV–V	1.11 (1.00–1.24)	1.08 (0.95–1.23)
Predominant motor type		
Spastic	Reference	Reference
Dystonic	0.94 (0.83–1.08)	0.92 (0.79–1.06)
Other	0.96 (0.82–1.12)	0.97 (0.82–1.14)
Intellectual disability		
Yes	1.12 (1.01–1.23)	1.09 (0.97–1.22)
No	Reference	Reference
Epilepsy		
Yes	0.99 (0.89–1.10)	0.92 (0.82–1.04)
No	Reference	Reference
Process-of-care factors		
Last appointment with multidisciplinary team care		
Yes	1.05 (0.99–1.12)	1.01 (0.94–1.07)
No	Reference	Reference
Last appointment with care coordination		
Yes	1.05 (0.97–1.14)	1.03 (0.95–1.12)
No	Reference	Reference
Last appointment not attended		
Yes	1.40 (1.26–1.56)	1.32 (1.17–1.48)
No	Reference	Reference
Recent cancelled or rescheduled appointment		
Yes	1.14 (1.06–1.22)	1.08 (1.01–1.16)
No	Reference	Reference

Abbreviations: CI, confidence interval; GMFCS, Gross Motor Function Classification System; IRSD, Index of Relative Socioeconomic Disadvantage; OR, odds ratio.

only become apparent with increasing age. Examples of this include scoliosis,[5] and cognitive (e.g. attention-deficit/hyperactivity disorder), affective, and anxiety disorders, which are also known to be more prevalent in children and adolescents with CP than other children.[15] Our results support this finding, for example the development of musculoskeletal problems indicated by increased use of orthopaedic services in older age groups. Care fragmentation among multiple specialties as children grow older and new priorities arise may also result in children missing important aspects of care that are not typically addressed by all specialties. Awareness of this issue and ensuring services are adapted to be sensitive to changing needs and age-appropriate is important.

Our findings are largely consistent with the research in non-attendance at outpatient clinics in children (with CP and other health conditions). The rate of non-attendance that we report is similar to that reported in a recent study of children with neurological conditions,[8] although the reported rate of non-attendance can vary substantially depending on setting. Studies in paediatric settings have suggested that factors relating to both individuals (e.g. sociodemographic factors, ethnicity, insurance status) and systems (e.g. waiting times for appointments, administrative error) are associated with non-attendance.[16,17] Studies of adults in outpatient[18] and primary care settings[19] have also identified social deprivation and age to be associated with non-attendance (with younger adults more likely to not attend than older adults), suggesting that our results may reflect broader trends and may be applicable to other childhood patient groups. The reasons that families do not attend outpatient clinic appointments has also been the subject of recent qualitative studies.[20,21] Common reasons reported included travel difficulties, competing priorities, and administrative issues (e.g. not receiving an appointment, difficulties in changing an appointment) that highlight the complexity that families face in balancing their child's health needs and other priorities.[22] These perspectives can help us reconsider non-attendance as a weakness in the model of care provided through outpatient clinics, in that they rely on face-to-face contact between a patient and health care professional at a

particular point in time. There is a need for increased acknowledgement that health care is not a 'one-size-fits-all' provision and personalizing health care delivery should sit alongside the agenda to personalize therapeutics. Strategies such as mHealth (e.g. SMS reminders[23]) and telemedicine may help support this agenda. The increased use of telemedicine during the COVID-19 pandemic has suggested that this is an acceptable alternative for many face-to-face consultations.[24] Our results also underpin the need for improved coordination of care and integration of speciality care with a child's local health care service and primary-care team, to ensure all opportunities to optimize health and development are pursued. The increasing rate of non-attendance that we identified makes these requirements time sensitive.

The strengths of our study include its size and the use of data from a CP register, which improved the precision of our study population and availability of clinical and sociodemographic descriptors, and the use of routinely collected administrative data. This is mandated by the NSW state government and ensured accurate estimation of attendance rates and service events. However, our methodology was not designed to explore the reasons that families did not attend, nor the reasons for the increase in non-attendance over time. Other limitations of our methodology include missing pertinent factors in our available data, including residency status. Our data also did not allow us to identify appointments cancelled by families prior to their appointment (distinct from those cancelled by the hospital). As others have identified,[25] this is another clinically important group, as they too represent a missed opportunity for health care.

Our study shows an association between non-attendance at outpatient appointments and socioeconomic disadvantage, increasing age, recent non-attendance, and cancelled or rescheduled appointments. These factors are readily identified and should be targeted when considering strategies to support families who may be experiencing difficulties with health care access. For example, clinicians can follow-up families who have missed appointments and consider alternative ways of supporting children's health where possible. Future studies to investigate barriers and facilitators for attendance to outpatient clinics and interventions to improve health care accessibility are warranted. This will enable the design and implementation of appropriate measures for uptake and access to care and services.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Flow diagram of inclusions and exclusions in study.

Table S1: Proportions of children and likelihood of scheduling for major specialty outpatient clinics in children with cerebral palsy, 2012 to 2019.

Table S2: Proportions of children and likelihood of scheduling for major specialty outpatient clinics in children with cerebral palsy, 2012 to 2019.

Table S3: Rates and proportions of scheduled outpatient days by age group and major specialty outpatient clinics in children with cerebral palsy, 2012 to 2019.

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10.4 Chapter 6

10.4.1 Statement of Contribution

Chapter 6 is (as of 16 September 2024) in peer review with *Journal of Child Neurology*.

The co-authors made the following contributions to the manuscript.

- Simon Paget conceived and designed the study, performed all statistical analyses, lead data interpretation, drafted the initial manuscript and lead subsequent revisions of the manuscript.
- Sarah McIntyre contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Amy von Huben contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Kirsty Stewart contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Tracey Williams contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Emma Maly contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Katrina Ford contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Sue Woolfenden contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Natasha Nassar contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

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10.5 Chapter 7

10.5.1 Statement of Contribution

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The co-authors made the following contributions to the manuscript:

- Simon Paget conceived and designed the study, performed all statistical analyses, lead data interpretation, drafted the initial manuscript and lead subsequent revisions of the manuscript.
- Sarah McIntyre contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.
- Francisco Schneuer contributed to the interpretation of results, including producing Figure 2 and Supplementary Figure 1 (Sankey diagrams) and reviewed and assisted with revision of the manuscript.

- Tan Martin contributed to the interpretation of results, including with a specific focus on the results of Aboriginal and/or Torres Strait Islander children and young people with CP, and reviewed and assisted with revision of the manuscript.

- Louise Sellars contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Heather Burnett contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Sophie Price contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

- Natasha Nassar contributed to the conception and design of the study, provided supervision, obtained access to the data, contributed to the interpretation of results and reviewed and assisted with revision of the manuscript.

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ORIGINAL ARTICLE

Outpatient encounters, continuity of care, and unplanned hospital care for children and young people with cerebral palsy

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Abstract

Aim: To describe the relationships between outpatient encounters, continuity of care, and unplanned hospital care in children/young people with cerebral palsy (CP).

Method: In this population-based data-linkage cohort study we included children/young people with CP identified in the New South Wales/Australian Capital Territory CP Register (birth years 1994–2018). We measured the frequency of outpatient encounters and unplanned hospital care, defined as presentations to emergency departments and/or urgent hospital admissions (2015–2020). Continuity of outpatient care was measured using the Usual Provider of Care Index (UPCI).

Results: Of 3267 children/young people with CP, most ($n = 2738$, 83.8%, 57.6% male) had one or more outpatient encounters (123 463 total encounters, median six outpatient encounters per year during childhood). High UPCI was more common in children/young people with mild CP (Gross Motor Function Classification System levels I–III, with no epilepsy or no intellectual disability), residing in metropolitan and areas of least socioeconomic disadvantage. Low UPCI was associated with four or more emergency department presentations (adjusted odds ratio [aOR] 2.34; 95% confidence interval [CI] 1.71–3.19) and one or more urgent hospital admissions (aOR 2.02; 95% CI 1.57–2.61).

Interpretation: Children/young people with CP require frequent outpatient services. Improving continuity of care, particularly for those residing in regional/remote areas, may decrease need for unplanned hospital care.

Cerebral palsy (CP) is the most common cause of physical disability in childhood and is defined by disorder(s) of movement and posture that arise from a disturbance of early brain development.[1] Comorbidities, such as epilepsy and respiratory and gastrointestinal diseases, are common in CP[2] and a frequent cause of (planned and/or unplanned) hospital admissions and emergency department presentations.[3,4] Children and young people with CP require hospital services substantially more than the general population,[5,6] with Gross Motor Function Classification System (GMFCS) levels IV to V, epilepsy, gastrostomy use, and intellectual disability associated with increased health service utilization.[7]

Outpatient services have an important role in the management of CP, providing early detection and preventive measures for health problems, therapy, medical interventions, and assessment for and planning of surgery.[8] Good outpatient management of CP and associated comorbidities may reduce adverse health outcomes. Missing outpatient appointments has been associated with urgent hospital admissions and emergency department presentations (unplanned

Abbreviations: NSW, New South Wales; UPCI, Usual Provider of Care Index.

hospital care) in children with CP and related neurological conditions.[9]

Access to health services is a key measure of health system performance. Health service access may be influenced by sociodemographic factors such as private health insurance, distance to major metropolitan hospitals, socioeconomic disadvantage, and parental employment status.[7,10] Continuity of care is a key measure of the quality of outpatient services.[11,12] Improved continuity of care has been associated with decreased hospital admission duration in children with CP,[13] and decreased hospital admissions and emergency department presentations in other childhood populations.[14–16]

Aboriginal and Torres Strait Islander peoples comprise 3.8% of the total Australian population.[17] Aboriginal and Torres Strait Islander Australians experience substantial health inequities, caused in part by the continuing effects of colonization and barriers to culturally appropriate and accessible health care. Addressing these health inequities is recognized as a major health care priority.[18] There is a lack of research exploring equity of access to health services for Aboriginal and Torres Strait Islander people living with CP.

Little is known about the relationships between outpatient encounters, quality of care, and unplanned hospital care utilization in people with CP. We aimed to: (1) explore the relationship between outpatient service utilization, continuity of care, and unplanned hospital care for children and young people with CP and (2) describe patterns of outpatient encounters for children and young people with CP and identify if there are variations related to sociodemographic and clinical factors, and for Aboriginal and Torres Strait Islander children and young people.

What this paper adds

- Many children with cerebral palsy use multiple and frequent outpatient services.
- Better continuity of care is associated with living in metropolitan and less socioeconomically disadvantaged areas.
- Outpatient service utilization reduces at the time of transition to adult services.
- High outpatient utilization is associated with unplanned hospital care.
- Decreased continuity of care is associated with unplanned hospital care.

METHOD

Population

We conducted a population-based record linkage cohort study of children and young people with CP using multiple administrative health data sets. The study population was derived from the New South Wales (NSW)/Australian Capital Territory Cerebral Palsy Register. We included all children with confirmed CP on the register at the time of linkage (20th January 2019), born between 1994 and 2018 and who were alive at the beginning of the study period (1st July 2015 to 30th June 2020).

Data sources

The NSW/Australian Capital Territory CP Register is a population-based database, with multiple ascertainment strategies, of individuals with CP who were born or live in NSW or Australian Capital Territory. Health data sets included the NSW Non-Admitted Patient Data Collection which contains data from all non-admitted (outpatient) encounters with clinical and/or therapeutic content provided by NSW health facilities; NSW Emergency Department Data Collection which contains data from presentations to NSW public hospital emergency departments; and NSW Admitted Patient Data Collection which contains data from all admissions to NSW hospitals (public and private). Deaths of individuals were identified through the NSW Registry of Births, Deaths, and Marriages Death Registrations. Data sets were probabilistically linked by the NSW Centre for Health Record Linkage.

Clinical and sociodemographic variables

Clinical CP variables derived from the NSW/Australian Capital Territory CP Register included predominant motor type, topography, GMFCS (categorized into GMFCS levels I–III and GMFCS levels IV–V), and the presence of epilepsy and/or intellectual disability. Sociodemographic variables of children included sex and maternal country of birth (dichotomized as born in Australia or overseas). Postcode of residence was used to determine geographical remoteness and socioeconomic disadvantage. Socioeconomic disadvantage was derived from the Indices of Relative Socioeconomic Disadvantage and grouped into quintiles (quintile 1 most disadvantaged and quintile 5 least disadvantaged).[19] Geographical remoteness (metropolitan; regional/remote areas) was defined using the Australian Statistical Geography Standard.[20]

Aboriginal and Torres Strait Islander status

Children and young people who identify as Aboriginal and/or Torres Strait Islander were identified by applying the 'enhanced reporting of Aboriginality' algorithm[21] to 'self-reported Aboriginality' as captured in the NSW Admitted Patient Data Collection and NSW Emergency Department Data Collection. It is important to acknowledge that being of Aboriginal and/or Torres Strait Islander background is not considered itself as a risk factor; rather our focus is the recognized systemic health inequities that Aboriginal and Torres Strait Islander people face.

Study measures

To evaluate outpatient service utilization, outpatient clinic encounters between 1st July 2015 and 30th June 2020 (5 years) were identified for each child and young person. Each child was dichotomized as having any outpatient encounters or not. The frequency of outpatient encounters during the study period was not normally distributed, with substantial positive skew. The total number of outpatient encounters for each child was categorized into tertiles (no use, tertile 1 ['low use'], tertile 2 ['medium use'], tertile 3 ['high use']). Continuity of care was defined using the Usual Provider of Care Index (UPCI).[22] We designated each child a usual provider of care by identifying the specialty service (acknowledging location and specialty type, e.g. rehabilitation, general medicine, neurology) with whom they had the highest frequency of outpatient encounters during the study period. The UPCI was then defined as the proportion of total outpatient encounters conducted with the usual provider of care. We decided to calculate UPCI only for those children who had five or more outpatient encounters during the study period; those with less than five (including none) encounters ($n=813$) were assigned a UPCI score of zero. Multidisciplinary clinics were categorized to the primary service specialty. We categorized continuity of care at an individual level into three categories, a priori and consistent with recent studies:[15,23] low continuity of care (UPCI <0.4), medium continuity of care (UPCI 0.4–0.69), and high continuity of care (UPCI ≥0.7). Outpatient encounters were also evaluated by hospital type (paediatric/metropolitan/regional), mode of delivery (in-person/virtual care [including telephone]), professional grouping (medical/allied health and nursing/procedural/diagnostic), and specialty groups.

Unplanned hospital care was defined as: (1) any presentation to an emergency department; and (2) urgent hospital admission because of an emergency presentation. The frequency of emergency department presentations was highly skewed and was therefore categorized into clinically and numerically meaningful groups defining none, one, two to three, and four or more presentations. Principal diagnosis for urgent hospital admissions was also identified based on the International Classification of Diseases, Revision 10, Australian Modification (ICD10-AM) and categorized according to disease chapters. Where the principal diagnosis was classified as either cerebral palsy (ICD10-AM G80.0–G80.9) or 'Symptoms, signs, and abnormal clinical and laboratory findings' (ICD10-AM R00–R99), the next diagnosis code was used.

Statistical analysis

Initial analysis of the frequency of outpatient encounters, emergency department presentations, and urgent hospital admissions highlighted these data to be skewed and they were therefore described using non-parametric measures (medians, interquartile ranges [IQR]). Clinical and sociodemographic variables were compared for those who had and had not accessed each type of health service using contingency tables and χ^2 tests where a p-value lower than 0.05 was considered statistically significant. Rates of outpatient encounters, emergency department presentations, and urgent hospital admissions were calculated as time-at-risk and reported as per 100 person years. Correlation between continuous variables (e.g. UPCI and frequency of outpatient encounters) was examined using Pearson's rank correlation coefficient.

The association between outpatient encounters and continuity of care with emergency department presentations and urgent hospital admissions was evaluated using multivariable multinomial (emergency department presentations) and binomial (urgent hospital admissions) logistic regression models adjusted for clinical and sociodemographic variables. For the logistic regression models, only children who completed the whole study period (5 years; $n=3058$) were included, and where appropriate variables were dichotomized (present/not present or unknown). Associations between outpatient encounter or UPCI tertiles and unplanned hospital care was described as odds ratios with 95% confidence intervals (CI). Stratified analysis by sociodemographic factors and clinical severity, and Aboriginal and/or Torres Strait Islander status was conducted to explore any potential differences in the association by specific subgroup. Analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC, USA). The study was approved by the NSW Population and Health Services Research Ethics Committee (2019/ETH11532), including a waiver of consent for participants, and the Aboriginal Health and Medical Research Council of NSW Human Research Ethics Committee (1861/21).

RESULTS

Of 3267 children and young people with CP, we identified 2738 (83.8%) who attended at least one outpatient encounter over the 5-year study period (Table 1). The youngest child was in their first month of life at their first encounter and the oldest person was 26 years at their last encounter. A higher proportion of those with at least one outpatient encounter lived in metropolitan areas (58.6% vs 52.3%), had predominant dyskinetic motor type (11.6% vs 5.7%), and/or epilepsy (27.1% vs 18.3%) (Table 1).

There was a total of 123 463 outpatient encounters during a total of 13 376 person years, with a median rate of 620 per 100 person years (IQR 240–1200). The median number of encounters per year for each individual was stable through childhood (median = 6) and declined into adulthood (median = 3) (Figure 1). For children aged 0 to 18 years, 65.5% of outpatient encounters occurred at children's hospitals and 18% of outpatient encounters occurred using virtual care (Table 2). Half of outpatient encounters (48.1%) were coordinated with at least one other encounter that day (28.3% two, 12.0% three, and 7.8% four or more encounters). Almost all encounter types were either medical (49%) or allied health

TABLE 1 Outpatient encounters and continuity of care for children and young people with cerebral palsy.

Demographic, clinical factor, and Aboriginal status	Total	Outpatient encounters			Level of continuity of care (UPCI)			
		Persons with 1+ encounter *n* (%)	Persons with no encounters *n* (%)	*p*	Low (UPCI <0.4) *n* (%)	Medium (UPCI 0.4–0.69) *n* (%)	High (UPCI ≥0.7) *n* (%)	*p*
Total	3267 (100)	2738 (83.8)	529 (16.2)		1625 (49.7)	942 (28.8)	700 (21.4)	
Sex				0.056				0.9
Male	1903 (58.2)	1576 (57.6)	327 (61.8)		952 (58.6)	543 (57.6)	408 (58.3)	
Female	1364 (41.8)	1162 (42.4)	202 (38.2)		673 (41.4)	399 (42.4)	292 (41.7)	
Maternal country of birth				0.896				<0.001
Australia	2215 (73.6)	1860 (73.5)	355 (73.8)		1151 (76.7)	607 (69.1)	457 (72.2)	
Overseas	796 (26.4)	670 (26.5)	126 (26.2)		349 (23.3)	271 (30.9)	176 (27.8)	
Aboriginality				0.016				0.22
Aboriginal	262 (8)	234 (8.5)	28 (5.3)		126 (7.8)	87 (9.2)	49 (7)	
Not Aboriginal	3005 (92)	2504 (91.5)	501 (94.7)		1499 (92.2)	855 (90.8)	651 (93)	
Remoteness				0.013				<0.001
Metropolitan areas	1851 (57.6)	1597 (58.6)	254 (52.3)		764 (50.5)	803 (51)	598 (63.5)	
Regional/remote areas	1361 (42.4)	1128 (41.4)	233 (47.9)		750 (49.5)	771 (49)	343 (36.5)	
IRD quintile				0.326				<0.001
1 (most disadvantaged)	591 (18.4)	518 (19)	73 (15)		286 (18.2)	188 (20)	117 (16.8)	
2	623 (19.4)	526 (19.3)	97 (20)		325 (20.6)	174 (18.5)	124 (17.8)	
3	705 (22)	591 (21.7)	114 (23.5)		360 (22.9)	217 (23.1)	128 (18.4)	
4	669 (20.8)	563 (20.7)	106 (21.8)		333 (21.2)	185 (19.7)	151 (21.7)	
5 (least disadvantaged)	623 (19.4)	527 (19.3)	96 (19.8)		269 (17.1)	177 (18.8)	177 (25.4)	
GMFCS level				<0.001				<0.001
I–III	2141 (65.5)	1758 (64.2)	383 (72.4)		1037 (63.8)	563 (59.8)	541 (77.3)	
IV–V	768 (23.5)	679 (24.8)	89 (16.8)		386 (23.8)	275 (29.2)	107 (15.3)	
Unknown	358 (11)	301 (11)	57 (10.8)		202 (12.4)	104 (11)	52 (7.4)	
Motor type				<0.001				<0.001
Spastic unilateral	1070 (35.3)	860 (26.4)	210 (42.5)		508 (34.3)	259 (29.4)	303 (45.5)	
Spastic bilateral	1381 (45.6)	1157 (35.5)	224 (45.3)		694 (46.8)	420 (47.7)	267 (40.1)	
Dyskinetic	322 (10.6)	294 (9.0)	28 (5.7)		134 (9)	124 (14.1)	64 (9.6)	
Ataxic	128 (4.2)	103 (3.2)	25 (5.1)		78 (5.3)	31 (3.5)	19 (2.9)	
Other	129 (4.3)	122 (3.7)	7 (1.4)		69 (4.7)	47 (5.3)	13 (2.0)	

TABLE 1 (Continued)

Demographic, clinical factor, and Aboriginal status	Total	Outpatient encounters			Level of continuity of care (UPCI)			
		Persons with 1+ encounter *n* (%)	Persons with no encounters *n* (%)	*p*	Low (UPCI <0.4) *n* (%)	Medium (UPCI 0.4–0.69) *n* (%)	High (UPCI ≥0.7) *n* (%)	*p*
Intellectual disability				<0.001				<0.001
Yes	1551 (47.5)	1321 (48.2)	230 (43.5)		777 (47.8)	505 (53.6)	269 (38.4)	
No	1222 (37.4)	998 (36.4)	224 (42.3)		580 (35.7)	298 (31.6)	344 (49.1)	
Unknown	494 (15.1)	419 (15.3)	75 (14.2)		268 (16.5)	139 (14.8)	87 (12.4)	
Epilepsy				<0.001				<0.001
Yes	840 (25.7)	743 (27.1)	97 (18.3)		408 (25.1)	299 (31.7)	133 (19)	
None or resolved	1728 (52.9)	1405 (51.3)	323 (61.1)		843 (51.9)	452 (48)	433 (61.9)	
Unknown	699 (21.4)	590 (21.5)	109 (20.6)		374 (23)	191 (20.3)	134 (19.1)	

P-values relate to differences between health service use groups by demographic, clinical factors, or Aboriginal status. Motor type 'Other' group combines hypotonia and early at risk. Totals may not sum where missing data <10%.
Abbreviations: GMFCS, Gross Motor Function Classification System; IRD, Index of Relative Socioeconomic Disadvantage; UPCI, Usual Provider of Care Index.

and nursing (48%) (Table 2). Rehabilitation (35%), neurology (14.5%), and general medicine (14%) were the most common medical specialty types.

Children and young people were reviewed by a median of seven (IQR 2–16) different specialty services during the 5-year period. Although representing one-quarter of the population, those in GMFCS levels IV to V had disproportionally high outpatient services use across different specialty types (Figure 2). Similar relationships were seen with major comorbidities (Figure S1). Two-thirds of children (65.7%) had an identified usual provider of care; most of these were based at a children's hospital (88.2%). Common usual provider of care specialty types were rehabilitation (47.9%), general medicine (14.6%), and neurology (15.5%). The median UPCI for children was 0.56 (IQR 0.40–0.78), indicating almost 60% of total outpatient appointments were conducted with their usual provider of care (Figure S2). Children receiving high levels of continuity of care (UPCI ≥0.7) were overrepresented in those with a mother born overseas, living in metropolitan areas, from less socioeconomic disadvantaged areas, spastic unilateral motor type, and neither epilepsy nor intellectual disability (Table 1). Outpatient utilization (total number of outpatient encounters) and UPCI were only weakly correlated (Pearson's rank correlation coefficient 0.12).

Two-thirds (65.9%) of children and young people had at least one emergency department presentation (median 3, IQR 1–6) and 33.4% had at least one urgent hospital admission (median 2, IQR 1–4) (Table S1). For children aged 0 to 18 years, 42.4% of urgent admissions occurred at children's hospitals. The most common principal diagnoses at admission were diseases of the respiratory system (30.1%) and diseases of the nervous system (17.8%), although in early adulthood injuries, diseases of the digestive system, and mental health disorders became more common (Table S2).

Outpatient utilization, continuity of care, and unplanned hospital care

Multinomial and binomial analysis of factors associated with emergency department presentations and urgent hospital admissions are shown in Table 3. Children and young people with high outpatient utilization had substantially increased odds of having four or more emergency department presentations (adjusted odds ratio [aOR] 5.00; 95% CI 3.62–6.92) and an urgent hospital admission (aOR 3.38; 95% CI 2.63–4.35). Compared with high continuity of care, low and medium continuity of care were also associated with increased odds of having four or more emergency department presentations (low: aOR 2.34; 95% CI 1.71–3.19; medium: aOR 1.63; 95% CI 1.21–2.20) and one or more urgent hospital admissions (low: aOR 2.02; 95% CI 1.57–2.61; medium: aOR 1.88; 95% CI 1.48–2.39). Stratified analysis using major sociodemographic and clinical factors and for Aboriginal and/or Torres Strait Islander people is shown in Figure 3 and Figure S3. These analyses suggested that associations between UPCI and unplanned hospital care persisted when limited to children with CP and markers of less

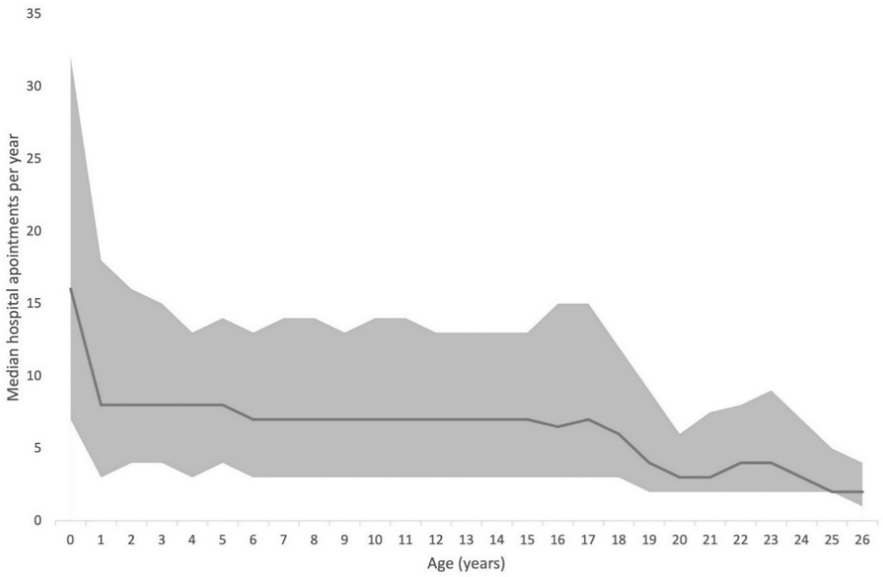

FIGURE 1 Median rate of hospital outpatient encounters by age for children and young people with cerebral palsy. The shaded area represents interquartile range.

TABLE 2 Comparison of frequency and proportions of outpatient encounters by age group for children and young people with cerebral palsy in 2015 to 2020.

		Facility location *n* (%)			Contact mode *n* (%)		Outpatient encounter type *n* (%)		
Age group, years	Total (*n*)	Paediatric	Metro	Regional/ rural	In person	Virtual care	Medical	Allied health and nursing	Diagnostic and procedural
0–4	27 340	15 933 (58.3)	6930 (25.3)	4477 (16.4)	22639 (82.8)	4701 (17.2)	12 685 (46.4)	14 203 (51.9)	161 (0.6)
5–9	36 195	24 687 (68.2)	4116 (11.4)	7392 (20.4)	29884 (82.6)	6311 (17.4)	19 076 (52.7)	16 385 (45.3)	181 (0.5)
10–14	33 057	22 746 (68.8)	4582 (13.9)	5729 (17.3)	27403 (82.9)	5654 (17.1)	15 592 (47.2)	16 798 (50.8)	110 (0.3)
15–19	22 062	12 548 (56.9)	4645 (21.1)	4869 (22.1)	17108 (77.5)	4954 (22.5)	11 356 (51.5)	9984 (45.3)	146 (0.7)
20+	4809	121 (2.5)	3194 (66.4)	1494 (31.1)	3841 (79.9)	968 (20.1)	2244 (46.7)	2050 (42.6)	95 (2)
Total	**123 463**	**76 035 (61.6)**	**23 467 (19)**	**23 961 (19.4)**	**100 875 (81.7)**	**22 588 (18.3)**	**60 953 (49.4)**	**59 420 (48.1)**	**693 (0.6)**

Totals may not add up to totals because of missing data.

severity (e.g. GMFCS levels I–III, no epilepsy, and/or no intellectual disability).

Aboriginal and/or Torres Strait Islander children with CP

Most (89%; $n = 234/262$) Aboriginal and/or Torres Strait Islander children and young people attended at least one outpatient encounter during the 5 years (rate 770 per 100 person years [IQR 300–1580]). During the same timeframe, 79.7% ($n = 209$) of Aboriginal and/or Torres Strait Islander children had at least one emergency department presentation and 44.3% ($n = 116$) had at least one urgent hospital admission. Almost half (47.3%, $n = 124$) of Aboriginal and/or Torres Strait Islander children had four or more emergency department presentations and they were more likely to live in regional or remote areas (76.4%) than those with fewer than four emergency department presentations (64.4%) ($p = 0.036$). Stratified analysis showed no statistical evidence of a relationship between UPCI and urgent admissions (OR low UPCI vs high UPCI 1.15; 95% CI 0.45–2.94) or four or more emergency department presentations (OR low UPCI vs high UPCI 1.95; 95% CI 0.62–6.13).

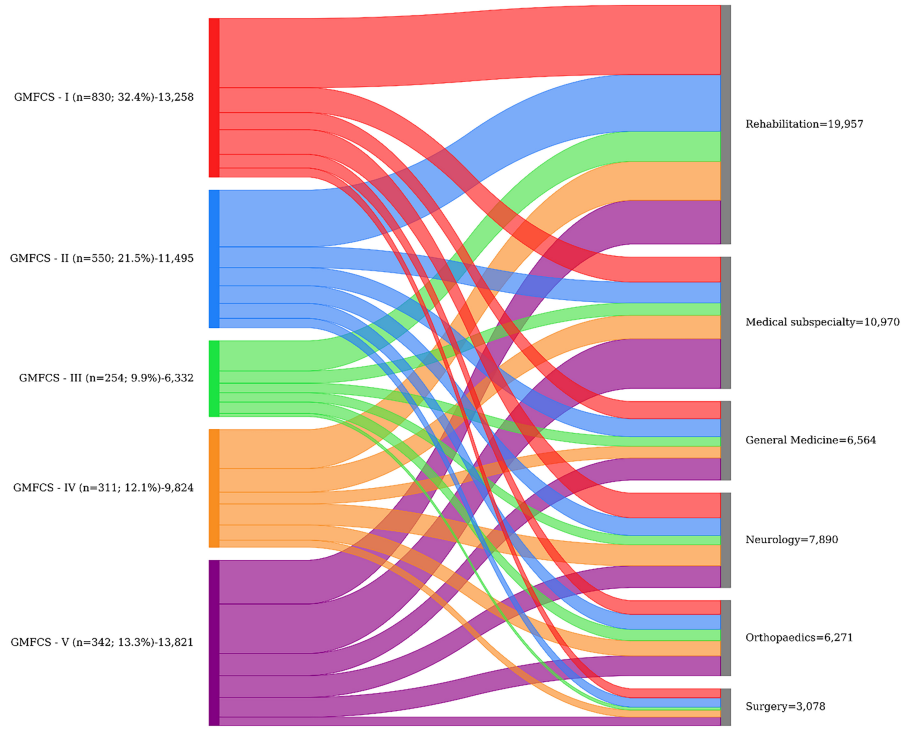

FIGURE 2 Major outpatient speciality use in children with cerebral palsy grouped by Gross Motor Function Classification System level.

TABLE 3 Univariable, multivariable, and multinomial logistic regression of outpatient factors associated with emergency department presentations and urgent hospital admissions in children and young people with cerebral palsy.

	Emergency department presentations[a]				Urgent hospital admissions[a]	
		Multivariable				
Outpatient factors	Univariable OR (95% CI)	1 presentation OR (95% CI)	2–3 presentations OR (95% CI)	4+ presentations OR (95% CI)	Univariable OR (95% CI)	Multivariable OR (95% CI)
Outpatient use						
No use	0.24 (0.19–0.30)	0.37 (0.27–0.52)	0.21 (0.14–0.3)	0.15 (0.09–0.23)	0.22 (0.15–0.33)	0.22 (0.15–0.34)
Low (1st tertile)	REF	REF	REF	REF	REF	REF
Medium (2nd tertile)	1.40 (1.14–1.72)	1.09 (0.81–1.47)	1.47 (1.10–1.97)	2.40 (1.76–3.27)	1.78 (1.43–2.21)	1.90 (1.48–2.43)
High (3rd tertile)	2.34 (1.88–2.93)	1.23 (0.88–1.72)	1.88 (1.36–2.58)	5.00 (3.62–6.92)	4.09 (3.31–5.07)	3.38 (2.63–4.35)
Usual Provider of Care Index						
Low	0.72 (0.60–0.87)	1.11 (0.81–1.52)	1.38 (1.02–1.87)	2.34 (1.71–3.19)	1.17 (0.95–1.44)	2.02 (1.57–2.61)
Medium	1.68 (1.35–2.09)	1.35 (1.00–1.81)	1.3 (0.97–1.73)	1.63 (1.21–2.20)	2.48 (1.98–3.09)	1.88 (1.48–2.39)
High	REF	REF	REF	REF	REF	REF

Multivariable models adjusted for demographic variables (sex, age, country of birth, Index of Relative Socioeconomic Disadvantage, residential location), Aboriginal and/or Torres Strait Islander background and clinical variables (Gross Motor Function Classification System, motor type, intellectual disability, epilepsy).

[a]Odds ratios presented compared with no emergency department presentations and no urgent hospital admissions respectively.

Abbreviations: CI, confidence interval; OR, odds ratio; REF, reference group.

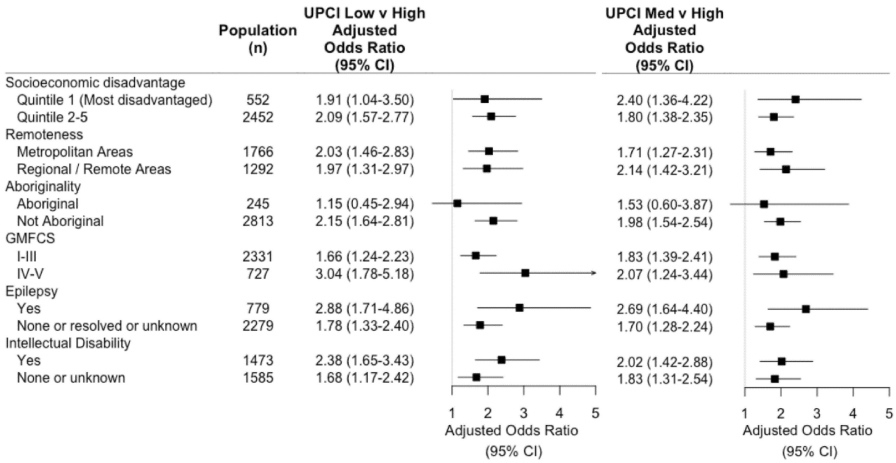

FIGURE 3 Association between continuity of care and urgent hospital admissions stratified by major sociodemographic and clinical factors and Aboriginal status. Abbreviations: CI, confidence interval; GMFCS, Gross Motor Function Classification System; UPCI, Usual Provider of Care Index.

DISCUSSION

This study is one of the first to examine outpatient service utilization in children and young people with CP at a population level, and the first epidemiological study of outpatient service utilization in Aboriginal and Torres Strait Islander children and young people with CP. Our results highlight that over 4 in 5 children with CP access outpatient services, with many using services frequently. Continuity of care was inequitably distributed, with a higher proportion of high continuity of care (high UPCI) for children living in metropolitan areas and in least socioeconomically disadvantaged areas. Less access to continuity of care was strongly associated with increased unplanned hospital care utilization. These associations persisted when analyses were restricted to those with less severe CP.

Our findings build on other studies that show the frequency and complexity with which children and young people with CP access the health system[24] and the associated carer burden.[25] Our findings emphasize the need for better continuity of care and care coordination, particularly in those with complex health needs. Our results suggest that continuity of care is less accessible for those living in the most socioeconomically disadvantaged and in regional and remote areas, reflecting the known challenges of recruiting and retaining health service staff in these areas.[26] In other CP populations, access to care coordination has also shown a socioeconomic gradient, with unmet need for care coordination increasing with decreasing household income.[27]

Recent Australian epidemiological research has highlighted that while 1 in 5 Aboriginal and Torres Strait Islander children with CP live in socioeconomically disadvantaged remote areas, most live in more populated areas.[28] While our study suggests that broadly access to outpatient services for Aboriginal and Torres Strait Islander children with CP is similar to that of the whole population, it is essential that a focus on equity of access is maintained, prioritizing the needs of those identified as having barriers to access to continuity of care and care coordination, and that care is provided in culturally appropriate ways.[29,30]

Continuity of care and interrelated concepts such as care coordination acknowledge the importance of the health care system working as a cohesive whole (integrated care). Continuity of care highlights the importance of stable relationships between patient and provider(s) and between providers within and between settings, and sharing of information and knowledge about patients, consistent with patients' needs and preferences.[23,31] Continuity of care is the more frequently used concept in research, likely as it is easier to measure.[32] Improved continuity of care has been associated with reductions in emergency department presentations[14,33] and hospitalizations[14,23,33] overall and for children with CP.[13] Coordinated care programmes that provide standardized systems of musculoskeletal evaluation for children with CP available in some countries[34,35] have been credited with decreasing incidence of musculoskeletal deformities.

Recent government reports have highlighted the need to support navigation of the health care system for families with children with complex needs and offered suggestions for improvement.[36] The COVID-19 pandemic has accelerated the uptake of virtual care, including for children with neurodevelopmental disabilities, and offers the potential to decrease the burden and costs associated with frequent outpatient visits for both families and the health system. There

is an opportunity to further develop virtual care consultations[37] to augment in-person health care and reduce travel and time lost from work, school, and family life. Similarly, better care coordination between speciality teams and hospitals, including development of integrated electronic health records,[38] use of care navigators, and outreach clinics can improve consumer experience of health care quality,[39] reduce travel,[40] and potentially reduce hospital admissions[41] and improve continuity of care.

During the last 10 years there has been an increased focus on CP as a lifelong health condition. Our findings highlight that outpatient service use declines during transition to adult services. Further work is needed in this area to understand the reasons for this decline and its impact. Our results also highlight that while diseases of the respiratory system continue to be a major cause of hospitalization in CP into adulthood,[42] early adulthood is associated with emergence of other health problems (e.g. mental health disorders)[43] that will likely require a change of focus for clinicians to ensure efforts for early detection/intervention during adolescence are successful.

Strengths of our study include the use of a large population-based data set. Using data linkage of a CP register with administrative health data enables ascertainment of CP severity and comorbidities, ensures complete follow-up, and minimizes selection bias observed when using administrative health data alone.[44] The broad health care setting including specialist children's hospitals and general hospitals also supports a more complete picture of outpatient health service utilization. Our study also has some limitations. In Australia, primary health care and some specialist care is also available in private settings and these data were not available. However, previous studies suggest that 80% of children have seen a general practitioner and just under 40% of children have seen a private paediatric medical specialist in the preceding year.[45] Access to National Disability Insurance Scheme-funded services may also influence health service utilization, and this should be a focus of future work. There was also 16% of children without any record of outpatient encounters. This may be due to those living near state borders who may have travelled interstate for their health care, or those who may be only accessing private services. There are known relationships between measures of severity (e.g. GMFCS, epilepsy, intellectual disability) and health service utilization. Associations between these and UPCI may have impacted our regression models, although our stratified analysis supports an association in less severe CP. There are also limitations in the use of continuity of care and application of the UPCI in the context of children with CP with multiple and complex health conditions where best care may be better achieved by seeing multiple providers of different specialties.[32] Future research should focus on the quality of care children and young people with CP receive and identifying best ways of measuring this quality.

In conclusion, our study highlights the importance of coordination of outpatient services for children and young people with CP. Establishing better continuity of care, preferentially prioritizing this for families residing in rural/remote and from most disadvantaged areas, may improve quality of care and decrease unplanned hospital care utilization for these groups. Achieving this will require a whole systems approach.

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CONFLICT OF INTEREST STATEMENT

The authors have stated that they have no interests that might be perceived as posing a conflict or bias.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from NSW/ACT Cerebral Palsy Register and NSW Ministry of Health, but restrictions apply to data availability, which were used under license for the current study and therefore are not publicly available.

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SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: Major outpatient speciality use in children with cerebral palsy.

Figure S2: Frequency of Usual Provider of Care Index for children and young people with cerebral palsy.

Figure S3: Association between continuity of care and emergency department presentations stratified by major sociodemographic and clinical factors and Aboriginal status.

Table S1: Comparison of proportions of emergency department attendances and urgent hospital admissions for children and young people with cerebral palsy in 2015 to 2020.

Table S2: Principal diagnosis for urgent hospital admissions for children and young people with cerebral palsy.

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