Insights from the epidemiology of cerebral palsy: navigating the advantages and limitations of registry vs. administrative health data

Received: 14 December 2023 | Accepted: 17 December 2023

DOI: 10.1111/ppe.13036

Paediatric and Perinatal Epidemiology WILEY

COMMENTARY

Insights from the epidemiology of cerebral palsy: Navigating the advantages and limitations of registry versus administrative health data

Sandra Julsen Hollung[1,2] | Joanne Given[3]

[1]Norwegian Quality and Surveillance Registry for Cerebral Palsy (NorCP), Vestfold Hospital Trust, Tønsberg, Norway

[2]Department of Clinical and Molecular Medicine, Norwegian University of Science and Technology, Trondheim, Norway

[3]Institute of Nursing and Health Research, Faculty of Life and Health Sciences, Ulster University, Belfast, Northern Ireland

Correspondence
Sandra Julsen Hollung. Norwegian Quality and Surveillance Registry for Cerebral Palsy (NorCP), Vestfold Hospital Trust, Tønsberg, Norway.
Email: sandra.julsen.hollung@siv.no

Epidemiological research provides insights into risk factors, prevalence and outcomes of cerebral palsy (CP), a lifelong motor disorder. It provides a foundation to monitor trends and evaluate the effectiveness of interventions and public health policy. There are different approaches to gathering these insights, including the use of population-based registries, administrative health databases and insurance claims data.

In this issue of *Paediatric and Perinatal Epidemiology*, Paget and colleagues[1] explored the differences between populations of children with CP identified through the New South Wales/Australian Capital Territory CP Register (gold standard) and New South Wales Admitted Patient Data Collection (hospital admission dataset). The authors compared socioeconomic, clinical characteristics and mortality of the children identified in these two data sources (supplemented with data from other registries/databases).[1]

Population-based CP registries are considered the gold standard because they (i) aim to capture all children with CP within a specified region to ensure completeness, often utilising multiple data sources; (ii) confirm the diagnoses around age 5–8 years using standardised diagnostic criteria to rule out another neurodevelopment disorder and ensure accuracy; and (iii) collect a comprehensive set of common data elements (CDE) using harmonising definitions and terminology around this age that are relevant to understanding the epidemiology, clinical characteristics and outcomes of CP.[2–4] This is essential to promote comparability to other CP populations to conduct meaningful analyses and draw accurate conclusions.

In their study, Paget et al. found that hospital admission data, using one or more admissions with G80 *International Classification of Disease*-10 AM diagnostic codes, had a sensitivity of 70% and a positive predictive value of 62% for identifying children with CP compared with the CP register, indicating moderate accuracy.[1]

Compared with population-based CP registries, children identified through the hospital admission data were more likely to have multiple chronic health conditions, reside in metropolitan areas and experience higher mortality rates.[1] These differences may represent the difficulties in making a clinical diagnosis, as CP is based on risk factors and signs of motor dysfunction, or the removal of the diagnosis at a later age.[1,5] Consequently, there remain some uncertainties in diagnosing CP. More recently, there has been a greater emphasis on early detection of CP, as early as less than 5 months of age (using specific guidelines), to improve outcomes.[5] In addition, in some cases, a child may be dependent on a CP diagnosis to receive access to timely multi-disciplinary supportive healthcare services. Improvements in the type and quality of administrative health data collected could improve the usefulness of data for both service evaluation and research purposes.[6] For example, the ability to record when a diagnosis is removed would prevent the inclusion of false positive cases, who in reality have another progressive condition, in CP populations created using administrative data as in the study by Paget et al.[1]

There exists a pressing need for further exploration into CP across the lifespan, not just during childhood. Paget et al. raised an interesting point that because their study was limited to children with CP, the applicability of the results to adults with CP is

uncertain.[1] They drew attention to the fact that there has been an increase in research on adults with CP, as evidenced by many epidemiological studies that have employed administrative health datasets to investigate various outcomes.[1] It is becoming increasingly common for CP registers to report on the epidemiology and outcomes of adults with CP through new data collection, given that the populations of individuals with CP have now reached adulthood. Some studies have reported that although adults with CP have high comorbidity rates, they lack access to the supportive healthcare services they had as children.[7] A recent article from the Northern Ireland Cerebral Palsy Register sheds light on the prevalence and clinical characteristics of adults living with CP, showing the complex nature of their needs and aiming to enhance supportive healthcare services.[8] Meanwhile, the Swedish CP Follow-up Programme and National Quality Registry (CPUP) have made significant advancements in their coverage, extending their registry in 2009 to include adults with CP residing in southern Sweden, followed by a nationwide expansion in 2019.[9] Their findings indicate that more than 90% of individuals with CP reach adulthood, with 8 out of 10 experiencing spasticity, contractures, misalignments, as well as complications related to balance and walking impairments, an increased risk of falling, pain, fatigue and dysphagia.[9] While the CPUP collects detailed information on adults with CP through the utilisation of the CPUP-adult assessment form, these valuable data remain absent from administrative health records. It would be highly beneficial to conduct research that compares administrative health data with CP registers, specifically focusing on adults with CP, as this would provide invaluable insights into this complex condition.

It should be noted that numerous studies examining CP primarily focus on populations residing in high-income countries. While there is limited data on CP registers in low- and middle-income countries, it is reported that the birth prevalence of CP is higher in these nations compared with high-income countries (3.4 versus 1.5 per 1000 live births).[10] There are also disparities in the aetiology of CP between high-income countries and low- and middle-income countries. Given that the majority of births occur in these less developed regions, it is crucial to incorporate data from these areas whenever feasible to obtain an updated understanding of CP.[10] However, a recent advancement in this field comes in the form of the Global Low- and Middle-Income Country CP Register (GLM-CPR), which was established as a collaborative effort between CP registers located in low- and middle-income countries across South Asia, South East Asia and Africa.[11] Epidemiological and clinical data were collected by these CP registers either by using the key informant method by local volunteers in the communities or when the children attended a health institution, using standardised data elements developed by the Surveillance of Cerebral Palsy in Europe and Australian CP Register.[11] The GLM-CPR has recently unveiled preliminary findings on risk factors for CP, such as elevated rates of home births and the scarcity of professional assistance during childbirth. Notably, post-neonatal CP was most often due to viral infections. The majority of children in these resource-limited settings had a severe type of CP (spastic bilateral) and level of gross motor dysfunction. Unfortunately, the CP registers were not able to identify all children within the respective regions needed to accurately determine prevalence estimates.[11]

While there are certainly limitations to using administrative health data, this remains an important source of data in many regions without CP registers. In addition, data linkage between CP registers and other datasets can enhance research outcomes by providing a more complete picture of health outcomes, education levels, employment and service utilisation, among others. Nevertheless, researchers must acknowledge the potential effects on sensitivity and positive predictive value when interpreting data on CP populations that are not obtained through the standardised methods employed by CP registers.

ACKNOWLEDGEMENTS

Open access publishing provided by the SIKT hybrid – Sykehuset i Vestfold HF agreement.

CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to disclose.

ORCID

Sandra Julsen Hollung https://orcid.org/0000-0002-7486-7454
Joanne Given https://orcid.org/0000-0003-4921-1944

REFERENCES

1. 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*. 2023:1-9.
2. Goldsmith S, McIntyre S, Smithers-Sheedy H, et al. An international survey of cerebral palsy registers and surveillance systems. *Dev Med Child Neurol*. 2016;58(Suppl 2):11-17.
3. Cans C, Dolk H, Platt MJ, Colver A, Prasauskiene A, Krägeloh-Mann IK. Recommendations from the SCPE collaborative group for defining and classifying cerebral palsy. *Dev Med Child Neurol*. 2007;49:35-38.
4. Weiskopf NG, Weng C. Methods and dimensions of electronic health record data quality assessment: enabling reuse for clinical research. *J Am Med Inform Assoc*. 2013;20(1):144-151.
5. Novak I, Morgan C, Adde L, et al. Early, accurate diagnosis and early intervention in cerebral palsy: advances in diagnosis and treatment. *JAMA Pediatr*. 2017;171(9):897-907.
6. McGuckin T, Crick K, Myroniuk TW, Setchell B, Yeung RO, Campbell-Scherer D. Understanding challenges of using routinely collected health data to address clinical care gaps: a case study in Alberta, Canada. *BMJ Open Quality*. 2022;11(1):e001491.
7. Peterson MD, Hurvitz EA. Cerebral palsy grows up. *Mayo Clin Proc*. 2021;96(6):1404-1406.
8. McConnell K, Livingstone E, Perra O, Kerr C. Population-based study on the prevalence and clinical profile of adults with cerebral palsy in Northern Ireland. *BMJ Open*. 2021;11(1):e044614.
9. Hägglund G, Alriksson-Schmidt A, Himmelmann K, Rodby-Bousquet E, Åstrand P. CPUP – from reactive to preventive treatment of cerebral palsy [Swedish]. *Lakartidningen*. 2023;120(23136):46-47.
10. McIntyre S, Goldsmith S, Webb A, et al. Global prevalence of cerebral palsy: a systematic analysis. *Dev Med Child Neurol*. 2022;64:1494-1506.
11. Jahan I, Muhit M, Hardianto D, et al. Epidemiology of cerebral palsy in low- and middle-income countries: preliminary findings from an international multi-centre cerebral palsy register. *Dev Med Child Neurol*. 2021;63(11):1327-1336.

AUTHOR BIOGRAPHIES

Sandra Julsen Hollung is a Health Informatics Specialist and Researcher for the Norwegian Quality and Surveillance Registry for Cerebral Palsy (NorCP) at the Vestfold Hospital Trust. She is also a guest researcher at the Norwegian University of Science and Technology, Department of Clinical and Molecular Medicine. Her research interests are risk factors and prevalence of cerebral palsy. NorCP has been a partner in the Surveillance of Cerebral Palsy in Europe since 2009, where she is a member of the steering committee and leader of the website and dissemination group.

Joanne Given is a Research Fellow in Epidemiology at the Institute of Nursing and Health Research at Ulster University. She works with EUROCAT congenital anomaly registry data and administrative health data across Europe. Her research interests include the validity and utility of administrative health data for research purposes, medication safety in pregnancy and morbidity in children with and without congenital anomalies. Dr. Given is currently a junior editor of *Paediatric and Perinatal Epidemiology*.

How to cite this article: Hollung SJ, Given J. Insights from the epidemiology of cerebral palsy: Navigating the advantages and limitations of registry versus administrative health data. *Paediatr Perinat Epidemiol*. 2023;00:1-3. doi:10.1111/ppe.13036