ORIGINAL ARTICLE

Hospitalizations in School-Aged Children with Cerebral Palsy and Population-Based Controls

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ABSTRACT: ***Objective:*** To compare hospitalizations among children with cerebral palsy (CP) and healthy controls and to identify factors associated with hospitalizations in children with CP. ***Methods:*** This retrospective cohort study linked data from a provincial CP Registry and administrative health databases. The CP cohort was comprised of children born from 1999 to 2002. Age, sex, and region-matched controls were identified from administrative health databases. Mean differences, relative risk (RR), and 95% confidence intervals (CIs) were calculated. ***Results:*** A total of 301 children with CP were linked to administrative health data and matched to 6040 controls. Mean hospitalizations per child during the study period were higher in children with CP compared to controls (raw mean difference (RMD) 5.0 95% CI 4.7 to 5.2) with longer length of stay (RMD 2.8 95% CI 1.8 to 3.8) and number of diagnoses per hospitalization (RMD 1.6 95% CI 1.4 to 1.8). Increased risk of hospitalization was observed in non-ambulant children with CP (RR 1.12 95% CI 1.01 to 1.22) compared to ambulant children and among those with spastic tri/quadriplegic CP compared to other CP subtypes (RR 1.15, 95% CI 1.05 to 1.27). Feeding difficulties (RR 1.20 95% CI 1.13 to 1.27), cortical visual (RR 1.22 95% CI 1.13 to 1.32), cognitive (RR 1.16 95% CI 1.04 to 1.30), and communication impairment (RR 1.26 95% CI 1.10 to 1.44) were associated with increased hospitalizations. ***Conclusions:*** Children with CP face more frequent, longer hospital stays than peers, especially those with a more severe CP profile. Coordinated interdisciplinary care is needed in school-aged children with CP and medical complexity.

RÉSUMÉ : Comparaison entre les taux d'hospitalisation d'enfants d'âge scolaire atteints de paralysie cérébrale et de témoins représentatifs de la population. ***Objectif*** **:** Comparer les taux d'hospitalisation d'enfants atteints de paralysie cérébrale (PC) à ceux de témoins en santé ; identifier les facteurs associés à l'hospitalisation des enfants atteints de PC. ***Méthodes*** **:** Cette étude rétrospective de cohorte a lié entre elles des données tirées d'un registre provincial de la PC et des données administratives du domaine de la santé. Notre cohorte de sujets atteints de PC a inclus des enfants nés entre 1999 et 2002. Les témoins appariés en fonction de l'âge, du sexe et de la région de résidence ont été identifiés au moyen de données administratives du domaine de la santé. Nous avons ensuite calculé les écarts moyens (EM), le risque relatif (RR) et les intervalles de confiance (95 %). ***Résultats*** **:** Au total, 301 enfants atteints de PC ont été liés à des données administratives du domaine de la santé et appariés à 6040 témoins. Au cours de la période d'étude, les taux moyens d'hospitalisation par enfant se sont avérés plus élevés chez les enfants atteint de PC en comparaison avec les témoins (écart moyen brut [EMB] 5,0 ; IC 95 % 4,7-5,2). La durée de leur séjour était en outre plus élevée (EMB 2,8 ; IC 95 % 1,8-3,8) alors qu'ils étaient davantage susceptibles de recevoir un plus grand nombre de diagnostics par hospitalisation (EMB 1,6 ; IC 95 % 1,4-1,8). Un risque accru d'hospitalisation a également été observé chez des enfants non-ambulatoires atteints de PC (RR 1,12 ; IC 95 % 1,01-1,22) en comparaison avec des enfants ambulatoires. Ce risque est aussi apparu accru chez des enfants atteints de PC spastique impliquant une forme de triplégie ou de quadriplégie en comparaison avec les autres sous-types de PC (RR 1,15 ; IC 95 % 1,05-1,27). Enfin, tant des difficultés à se nourrir (RR 1,20 ; IC 95 % 1,13-1,27), des troubles visuels corticaux (RR 1,22 ; IC 95 % 1,13-1,32), des troubles cognitifs (RR 1,16 ; IC 95 % 1,04-1,30) que des troubles de la communication (RR 1,26 ; IC 95 % 1,10-1,44) ont été associés à des hospitalisations plus fréquentes. ***Conclusions*** **:** Les enfants atteints de PC, particulièrement ceux atteints des formes plus graves de cette affection, font donc face à des séjours à l'hôpital plus fréquents et plus longs si on les compare à des témoins. Il s'ensuit que des soins interdisciplinaires coordonnés sont nécessaires pour ces enfants dont les dossiers de santé sont complexes.

Keywords: Cerebral palsy, Pediatric neurology, Health services research

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Cerebral palsy (CP) encompasses a spectrum of permanent, nonprogressive motor disorders, resulting from congenital or acquired disturbances to the developing brain. It is the leading cause of physical impairment in children with an estimated prevalence of 2.0 per 1000 births.[1] Heterogeneity is seen in many aspects of CP, from risk factors to functional outcomes and a range of potential comorbidities such as epilepsy, feeding difficulties, orthopedic complications, and respiratory compromise.

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Given the chronic nature of CP and its impact on function and associated comorbidities by definition, it is unsurprising that there is a substantive cost associated with medical, rehabilitation, and educational services for affected children. In 2000, the estimated cost of CP in Canada was $381.8 million and was predominantly due to costs associated with morbidity and mortality, as opposed to direct healthcare costs.[2] Furthermore, a 1997 survey of pediatric discharges from US hospitals found that children with CP had longer lengths of stay, higher total charges, and more diagnoses and procedures per admission.[3] Studies drawing from population-based service registries suggest a clear temporal trend of increasing survival in children with CP well into adulthood, thus increasing the hospitalization burden on these individuals.[4] An improved understanding of patient- and disease-specific factors influencing the healthcare utilization of children with CP can better orient coordinated care models to target these risk factors, with the ultimate goal of reducing morbidity, hospitalization burden, and overall costs. Coordinated care models have been shown to reduce the use of inpatient care services and decrease costs in children with complex care needs.[5] However, data specific to children with CP are lacking in the current literature.

The primary goal of this study was to compare healthcare utilization of school-aged children with CP to peers without CP using population-based data. We hypothesized that children with CP have a higher number of admissions, with longer lengths of stay than peer controls. The secondary goal was to identify sociodemographic and clinical factors predictive of healthcare utilization in children with CP. We hypothesized that increased risk of hospitalization would be seen in children of lower socioeconomic status, with more severe CP subtypes, and comorbidities.

Methods

Study Design and Setting

This retrospective data linkage study was conducted at the Research Institute of the McGill University Health Centre. Ethical approval was obtained from the Research Ethics Board (REB) of the McGill University Health Centre and the REBs of participating institutions. Approval was also obtained from the Commission d'accès a l'information (CAI) to access nominal data for linkage purposes.

Data Sources

The *Registre de la paralysie cérébrale du Québec* (hereafter called the Registry) is a population-based Registry of children with CP from Quebec. The Registry spans 6 out of Quebec's 17 administrative health regions and represents half of the province's pediatric population, including urban, suburban, and rural regions. To be enrolled in the Registry, children must be at least 2 years of age and meet international consensus criteria for CP.[6] Follow-up is obtained at 5 years of age when possible. The Registry captures a comprehensive profile of each participant, including sociodemographic data, prenatal, perinatal, and neonatal CP risk factors, associated comorbidities, and functional outcomes. The CP cohort included in this study comprised registered cases born between January 1, 1999 and December 31, 2002 with a definite diagnosis of CP at 5 years of age. This cohort was selected as it represents the longest follow-up period in the registry. Participant data were linked to administrative data from birth to December 31, 2012. All participants were 10–13 years of age at the end of the study period. Functional and CP phenotypic data extracted from the Registry included CP subtype, gross motor function (Gross Motor Classification System (GMFCS), categorized as I–III (ambulant with or without the support of aids) and IV–V (non-ambulant)), and comorbidities (cognitive impairment, visual impairment, and sensorineural auditory impairment, feeding difficulties [gastrostomy and jejunostomy], communication difficulties, and epilepsy).

Children residing in Quebec for more than 3 months are registered with the *Régie de l'assurance maladie du Québec* (RAMQ) and are eligible for universal health coverage. The RAMQ records physician billing claims for reimbursement through a computerized database. Claims data include the primary diagnosis for each visit (using the International Classification of Diseases [ICD], Ninth Revision [ICD-9-CM]), the specialty of the physician submitting the claim, and patient sociodemographic information (date of birth, sex, and forward sortation area [i.e. the first three digits of their postal code]). The Quebec Ministry of Health records hospitalizations in the *Maintenance et exploitation des données pour l'étude de la clientèle hospitalière* database (MED-ÉCHO). Recorded data include date of hospitalization, length of hospitalization, primary hospitalization diagnosis (ICD-9-CM up to March 2006 and ICD Tenth Revision [ICD-10] since April 2006), and up to 30 secondary diagnoses per hospitalization. Age at CP diagnosis was derived from the RAMQ database and was defined as the age at the time of the first ICD code (ICD-9 343.x) for CP in the RAMQ database. The high specificity of the CP diagnostic code within this administrative health database has been previously established by our group.[7]

Data Linkage

A file containing Registry patient identification numbers, RAMQ insurance numbers, name, and sex was sent by registered mail to the RAMQ for data linkage. The resulting dataset was de-identified to protect patient confidentiality

Control Group

A large sample of population-based controls (general population) were selected from the RAMQ database from across the province of Quebec as children from the same age, gender, and administrative region cohorts who never received a diagnostic code of CP.

Sociodemographic Factors

For both the CP cohort and the peer control group, demographic and socioeconomic factors were sought and included for analysis. Region of residence was classified according to Statistics Canada's categorization of population centers (rural areas, small (1000–29,000), medium (30,000–99,999), and large (>100,000) population centers).[8] Socioeconomic status was also assessed using binary variables including eligibility for the public drug plan and eligibility of the child's parents for unemployment benefits. Material and social deprivation were calculated using the Pampalon deprivation index.[9] The deprivation index, which is based on data from dissemination areas – the smallest available geographic units in the Statistics Canada national census – has previously been used by our group in the population of children with CP.[10] The material deprivation index is calculated by integrating, in the population aged 15 years and over, the proportion of individuals without a high school diploma or equivalent, the employment-to-

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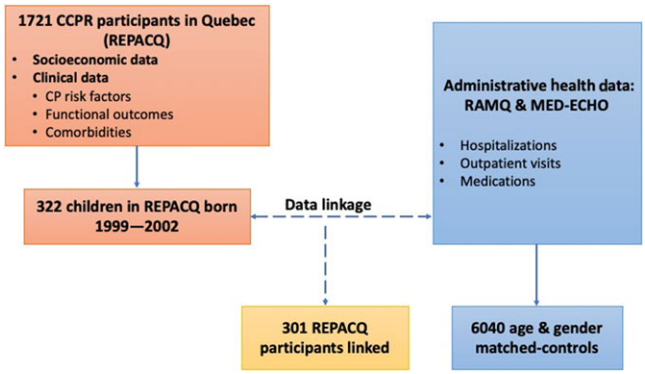

Figure 1: *Participant flow diagram.*

population ratio, and the average income. The social deprivation index, on the other hand, is calculated by integrating the proportion of individuals living alone, the proportion of the population who are separated, divorced, or widowed, and the proportion of single-parent families. Indices categorize participants into quintiles with those in the first quintile (Q1) being the most privileged and those in the fifth quintile (Q5) being the least privileged.

Outcomes

In order to assess use of inpatient services by the cohort of patients with CP, four categories of hospitalizations during the study period were used: total number of individuals with ≥1 hospitalization of any type, total number of individuals with ≥1 same-day admission, total number of individuals with ≥1 multi-day admission (less than 30 days), and total number of individuals with ≥1 prolonged admission (greater than 30 days). To compare the hospitalizations of children with CP to their healthy peers, the outcomes of interest were mean hospitalizations per patient during the study period, mean length of hospital stay, and mean number of diagnoses per hospitalization.

Data Analysis

All analyses were conducting using SAS 9.4 (SAS Institute Inc., Cary, North Carolina). Student's t-tests were used to compare continuous variables. Relative risk (RR) and 95% confidence intervals (CIs) were calculated where appropriate.

Results

A total of 1741 individuals were in the Quebec CP Registry, of which 322 met inclusion criteria for this analysis. Linkage with administrative health databases was possible for 96% of these children, resulting in a study sample of 301 children with CP (Figure 1). There were no differences in clinical profiles between children with CP whose Registry data were successfully or unsuccessfully linked to administrative health records. More than half of the children in the CP Registry cohort were male (56%) and the mean age at diagnosis was 3.6 years old (standard deviation [SD] 3.0 years). Spastic CP subtypes made up 74% of the REPACQ cohort, with spastic hemiplegia being the most common (28%). The majority of the CP cohort (60%) had a GMFCS of I–III. Communication difficulties were the most frequently reported comorbidity (54%) followed by epilepsy (34%) and cognitive impairment (32%) (Table 1).

Table 1: Profile of the CP cohort

	CP cohort n (%)
Total, n (%)	$n = 301$
Deprivation index (material)	
Q4–Q5	83 (27.6%)
Q1–Q3	193 (64.1%)
Missing	25 (8.3%)
Deprivation index (social)	
Q4–Q5	101 (33.6%)
Q1–Q3	175 (58.1%)
Missing	25 (8.3%)
Eligibility for the public drug plan	
Yes	171 (56.8%)
No	130 (43.1%)
Parental eligibility for unemployment benefits	
Yes	75 (24.9%)
No	96 (31.9%)
Missing	130 (43.2%)
Access to a primary care provider	
Yes	271 (90.0%)
No	13 (4.3%)
Missing	17 (5.7%)
Region	
Other	56 (18.6%)
Large urban population centre	228 (75.7%)
Missing	17 (5.6%)
GMFCS	
IV–V	70 (23.3%)
I–III	182 (60.5%)
Missing	49 (16.3%)
CP subtype	
Spastic tri/quadriplegia	82 (27.2%)
Other	170 (56.5%)
Unknown	49 (16.3%)
Epilepsy	
Yes	102 (33.9%)
No	152 (50.5%)
Unknown	47 (15.6%)
Feeding difficulties	
Yes	15 (5.0%)
No	235 (78.1%)
Unknown	51 (16.9%)
Cortical visual impairment	
Yes	54 (17.9%)
No	190 (63.1%)
Unknown	57 (18.9%)
Sensorineural auditory impairment	

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Table 1. *(Continued)*

	CP cohort n (%)
Yes	27 (9.0%)
No	217 (72.1%)
Unknown	57 (18.9%)
Cognitive impairment	
Yes	97 (32.2%)
No	113 (37.5%)
Unknown	91 (30.2%)
Communication difficulties	
Yes	162 (53.8%)
No	88 (29.2%)
Unknown	51 (16.9%)

Healthcare Use Compared to Peers

Overall, 84% of the CP cohort had at least one hospitalization during the study period, and 13% had at least one prolonged hospitalization of 30 days or more. Children with CP had a higher number of hospitalizations per child compared to general population controls (raw mean difference (RMD) 5.0 hospitalizations 95% CI 4.7 to 5.2), longer hospital stays (RMD 2.8 days 95% CI 1.8 to 3.8), and higher number of diagnoses per hospitalization (RMD 1.6 diagnoses 95% CI 1.4 to 1.8) (Table 2).

For children with CP, diseases of the nervous system (25%), respiratory system (23%), digestive tract (11%), and sensory organs (eyes and ears) (8%) were the most common primary hospitalization ICD-9-CM diagnosis categories (Table 3). Epilepsy made up 39% of all nervous system-related hospitalizations and 10% of all hospitalizations. The most common respiratory diagnostic codes were for influenza and pneumonia (26%), acute respiratory infections (18%), and chronic lower respiratory diseases, mostly asthma (16%). Diseases of the oral cavity, salivary glands, and jaws were the most commonly recorded digestive system issues requiring hospitalization (31%) (Supplemental Table 1). For peer controls, the most common reasons for hospitalizations were respiratory system-related disorders (34%) – including chronic adenoid or tonsil issues (35%), acute respiratory infections (22%), and asthma exacerbation (13%) – diseases of sensory organs (16%) – related to ear issues in most cases – and diseases of the digestive system (12%) (Supplemental Table 2). Nervous system-related admissions were less frequently encountered (2%).

Compared to controls, children with CP had a higher risk of admissions due to diseases of the nervous system (RR 10.15 95% CI 8.18 to 12.60) and congenital anomalies (RR 1.30 95% CI 1.04 to 1.64). Peer controls had a risk of admissions for respiratory system-related problems (RR 1.47 95% CI 1.33 to 1.62) and issues with sensory organs – eyes and ears – (RR 2.10 95% CI 1.76 to 2.52) (Table 3).

Factors Associated with Hospitalizations

Sociodemographic factors, which included the deprivation index, eligibility for the provincial public drug plan, parental eligibility for unemployment benefits, access to a primary care provider, and geographic region, were generally not predictive of hospitalization patterns, with the exception of the specific material deprivation index and eligibility for the public drug plan (Table 4). Indeed, less privileged children based on the material deprivation index had a higher risk of multiday hospitalizations less than 30 days (RR 1.22 95% CI 1.01 to 1.46) and those eligible for the provincial public drug plan had a higher risk of same-day admissions (RR 1.17 95% CI 1.02 to 1.41). With regard to other markers of low socioeconomic status (SES), some trends were observed but none met statistical significance. There was a trend toward decreased risk of prolonged admission of more than 30 days in patients with a high deprivation index; however, the RR for both the material and social deprivation indexes did not meet statistical significance ([RR 0.74 95% CI 0.35 to 1.58] and [RR 0.55 95% CI 0.26 to 1.18], respectively).

Risk of hospitalization of any type was higher for non-ambulatory children (RR 1.12 95% CI 1.01 to 1.22), those with spastic tri- or quadriplegic CP subtypes (RR 1.15 95% CI 1.05 to 1.27), as well as those comorbid feeding difficulties (RR 1.20 95% CI 1.13 to 1.27), communication impairment (RR 1.26 95% CI 1.10 to 1.44), cortical visual loss (RR 1.22 95% CI 1.13 to 1.32), and cognitive impairment (RR 1.16 95% CI 1.04 to 1.30). Children with these risk factors were also at risk of having multiday admission. Risk of having a prolonged admission (≥30 days) was increased in non-ambulant children (RR 2.08 95% CI 1.14 to 3.78), those with spastic tri- or quadriplegia (RR 2.59 95% CI 1.42 to 4.73) and those with feeding difficulties (RR 2.53 95% CI 1.15 to 5.55).

Interpretation

As hypothesized, our study shows that children with CP are hospitalized more frequently and for longer periods of time than their age-matched peers. Furthermore, patients with CP are noted to have a greater number of active hospitalization diagnoses than their peers, reflecting increased medical complexity. Their profile of hospitalization diagnoses is also quite different. Children with CP have a higher use of inpatient medical services than their peers, owing to the frequency, duration, and complexity of their medical admissions.

With regard to understanding the factors that underlie the increased risk of admission in patients with CP, we had hypothesized that children with more severe CP would have increased hospitalizations. This was indeed the case, with patient in our CP cohort exhibiting a more severe CP phenotype and more medical complexity being at a higher risk of frequent and prolonged hospitalizations; epilepsy and respiratory infections were the main drivers of this increased risk. We had previously shown that GMFCS status is strongly associated with CP subtype and comorbidity.[11] The primary reasons for admission in children with CP were central nervous system-related issues including epilepsy, followed by respiratory illnesses. We had also hypothesized that specific markers of low SES would increase the risk of hospitalization in patient with CP. This was indeed the case for some specific markers of low SES, including the material deprivation index and eligibility to the provincial public drug plan, which predicted the risk for multiday admissions and same-day admissions, respectively. Our study was likely underpowered to confirm other predictive socioeconomic factors that showed a trend but did not reach statistical significance, such as the parental eligibility for unemployment benefits, access to a primary care provider, and geographic localization. Of note, we did detect a

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Table 2: Healthcare utilization of CP and control cohorts

Healthcare utilization	CP cohort $n = 301$	Controls $n = 6040$	Raw mean difference	Raw mean difference 95% CI
*Hospitalizations per patient mean ± SD	5.6 ± 7.8	0.7 ± 1.4	5.0	4.7–5.2
*Length of hospital stay (days) mean ± SD	5.0 ± 8.8	2.2 ± 4.1	2.8	1.8–3.8
*Number of diagnoses per hospitalization mean ± SD	3.7 ± 2.1	2.1 ± 1.2	1.6	1.4–1.8

*Significant difference at the $p < 0.0001$ level.

Table 3: Comparison of primary diagnoses of CP and control cohorts

Most common primary diagnosis category for hospitalizations in CP cohort	CP cohort hospitalizations $n = 1699$ (% of all hospitalizations)	Control hospitalizations $n = 3825$ (% of all hospitalizations)	Relative risk (95% CI)
Nervous	424 (25.0%)	94 (2.5%)	10.15 (8.18–12.60)
Respiratory	391 (23.0%)	1296 (33.9%)	0.68 (0.62–0.75)
Digestive	189 (11.1%)	472 (12.3%)	0.90 (0.77–1.06)
Sense organs	129 (7.6%)	611 (16.0%)	0.48 (0.40–0.57)
Congenital anomalies	110 (6.5%)	190 (5.0%)	1.30 (1.04–1.64)
Other	456 (26.8%)	1169 (30.6%)	0.88 (0.80–0.96)

Table 4: CP cohort hospitalizations by patient characteristics

	≥1 Admission (any type) n (%)	≥1 Same-day admission n (%)	≥1 Multiday admission (<30 days) n (%)	≥1 Prolonged admission (≥30 days) n (%)
Total, n (%)	252 (83.7%)	228 (76%)	188 (62%)	39 (13%)
Deprivation index (material)				
Q4–Q5	72 (86.7%)	68 (81.9%)	58 (69.9%)	8 (9.6%)
Q1–Q3	160 (82.9%)	146 (75.6%)	111 (57.5%)	25 (13.0%)
Missing				
Relative risk (95% CI)[a]	1.05 (0.94–1.16)	1.08 (0.95–1.23)	1.22 (1.01–1.46)	0.74 (0.35–1.58)
Deprivation index (social)				
Q4–Q5	85 (84.2%)	83 (82.2%)	62 (61.4%)	8 (7.9%)
Q1–Q3	147 (84.0%)	131 (74.9%)	107 (61.1%)	25 (14.3%)
Missing				
Relative risk (95% CI)[a]	1.00 (0.90–1.11)	1.10 (0.97–1.24)	1.00 (0.82–1.21)	0.55 (0.26–1.18)
Eligibility for the public drug plan				
Yes	148 (86.5%)	138 (80.7%)	114 (66.7%)	23 (13.4%)
No	104 (80.0%)	90 (69.2%)	74 (56.9%)	16 (12.3%)
Relative risk (95% CI)	1.08 (0.98–1.20)	1.17 (1.02–1.34)	1.17 (0.98–1.41)	1.09 (0.60–1.98)
Parental eligibility for unemployment benefits				
Yes	68 (90.7%)	64 (85.3%)	55 (73.3%)	13(17.3%)
No	80 (83.3%)	74 (77.1%)	59 (61.5%)	10 (10.4%)
Relative risk (95% CI)	1.09 (0.97–1.22)	1.11 (0.96–1.28)	1.19 (0.97–1.5)	1.66 (0.77–3.58)
Access to a primary care provider				
Yes	241 (84.9%)	220 (77.5%)	179 (63.0%)	38 (13.4%)
No	11 (64.7%)	8 (47.1%)	9 (52.9%)	1 (5.9%)
Relative risk (95% CI)	1.31 (0.92–1.87)	1.64 (0.99–2.74)	1.19 (0.75–1.88)	2.27 (0.33–15.58)

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Table 4. *(Continued)*

	≥1 Admission (any type) n (%)	≥1 Same-day admission n (%)	≥1 Multiday admission (<30 days) n (%)	≥1 Prolonged admission (≥30 days) n (%)
Region				
Other	47 (83.9%)	42 (75.0%)	34 (60.7%)	9 (16.1%)
Large urban population centre	192 (84.2%)	178 (78.1%)	142 (62.3%)	25 (10.9%)
Missing				
Relative risk (95% CI)[b]	1.00 (0.88–1.13)	0.96 (0.81–1.13)	0.97 (0.77–1.23)	1.47 (0.72–2.96)
GMFCS				
IV–V	64 (91.4%)	57 (81.4%)	59 (84.2%)	16 (22.9%)
I–III	149 (81.9%)	138 (75.8%)	97 (53.3%)	20 (11.0%)
Missing				
Relative risk (95% CI)[c]	1.12 (1.01–1.22)	1.07 (0.93–1.23)	1.58 (1.33–1.87)	2.08 (1.14–3.78)
CP subtype				
Spastic tri/quadriplegia	76 (92.7%)	69 (84.1%)	68 (82.9%)	20 (24.4%)
Other	137 (80.6%)	126 (74.1%)	88 (51.8%)	16 (9.4%)
Unknown				
Relative risk (95% CI)[d]	1.15 (1.05–1.27)	1.85 (1.00–1.29)	1.60 (1.34–1.91)	2.59 (1.42–4.73)
Epilepsy				
Yes	88 (86.3%)	85 (83.3%)	68 (66.7%)	17 (16.7%)
No	126 (82.9%)	111 (73.0%)	89 (58.6%)	19 (12.5%)
Unknown				
Relative risk (95% CI)	1.04 (0.94–1.16)	1.14 (1.00–1.30)	1.14 (0.94–1.38)	1.33 (0.73–2.44)
Feeding difficulties				
Yes	15 (100.0%)	14 (93.3%)	15 (100.0%)	5 (33.3%)
No	196 (83.4%)	179 (76.2%)	139 (59.1%)	31 (13.2%)
Unknown				
Relative risk (95% CI)	1.20 (1.13–1.27)	1.23 (1.05–1.43)	1.69 (1.52–1.88)	2.53 (1.15–5.55)
Cortical visual impairment				
Yes	53 (98.1%)	48 (88.9%)	43 (79.6%)	10 (18.5%)
No	153 (80.5%)	140 (73.7%)	108 (56.8%)	24 (12.6%)
Unknown				
Relative risk (95% CI)	1.22 (1.13–1.32)	1.21 (1.06–1.37)	1.40 (1.17–1.68)	1.47 (0.75–2.87)
Sensorineural auditory impairment				
Yes	24 (88.9%)	22 (81.4%)	18 (66.7%)	2 (7.4%)
No	181 (83%)	165 (76%)	131 (60%)	32 (15%)
Unknown				
Relative risk (95% CI)	1.07 (0.92–1.23)	1.07 (0.88–1.30)	1.10 (0.83–1.47)	0.50 (0.13–1.98)
Cognitive impairment				
Yes	89 (91.8%)	80 (82.5%)	74 (76.3%)	17 (17.5%)
No	89 (78.8%)	82 (72.6%)	58 (51.3%)	12 (10.6%)
Unknown				
Relative risk (95% CI)	1.16 (1.04–1.30)	1.14 (0.98–1.32)	1.49 (1.20–1.84)	1.65 (0.83–3.28)
Communication difficulties				
Yes	148 (91.4%)	136 (84.0%)	112 (69.1%)	25 (15.4%)
No	64 (72.7%)	58 (65.9%)	44 (50.0%)	11 (12.5%)
Unknown				
Relative risk (95% CI)	1.26 (1.10–1.44)	1.27 (1.08–1.50)	1.38 (1.10–1.75)	1.23 (0.64–2.39)

[a]Q4–Q5 vs. Q1–Q3; [b]other vs. large urban population; [c]GMFCS IV–V vs. I–III; [d]Spastic triplegia vs. all other CP subtypes.

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trend toward decreased risk of prolonged (more than 30 days) admissions in patients with high material and social deprivation index, which would go against our initial hypothesis. However, both did not reach statistical significance and it is difficult to elaborate on clinical significance given the small numbers for that specific calculation. This would need to be reassessed in a larger cohort. The findings of this study are aligned with those from other international CP Registers. This supports the notion that the results of this study are generalizable to other similar jurisdictions. In the data linkage study from the CP Register of Victoria, Australia, CP severity as assessed using the GMFCS was associated with more admissions, with the most frequent primary reason being respiratory illness.[12] Furthermore, results from their study showed an increased risk of hospitalization among children with feeding difficulties and epilepsy. Whereas the current study did identify an increased risk of hospitalization in children with CP and comorbid feeding difficulties, the same cannot be said about comorbid epilepsy. Although a trend toward increased risk of hospitalizations in patients with CP and comorbid epilepsy was observed, our study may have been underpowered to detect a difference.

Our study's strengths include a well-defined population-based cohort of children with CP capturing the phenotypic spectrum and successful linkage with administrative health databases. Although several countries such as Norway and Sweden have instituted unique identifiers to allow seamless data linage for population health studies, this infrastructure is not in place in the province of Quebec. We were, however, able to successfully link 96% of the children in our CP cohort using the date of birth, gender, postal code, and RAMQ identification number.

The findings of this study have good generalizability to similar areas where multidisciplinary teams care for patients with CP. Although exact pathologies leading to hospitalizations may vary depending on local environmental and clinical contexts, the overall findings of increased inpatient healthcare use by patients with CP – especially those with a more complex clinical profile – are likely to be representative of North American populations.

One of the limitations of this study is the reliance on governmental administrative data, which may lack in specificity. However, using the Registry to identify children with CP, our determination of the two groups is highly specific.[7] The Registry also contains exhaustive information about the included patients, which reduces the risk of bias in this regard. Furthermore, the linkage between the population-based Registry and the administrative health databases enables us to increase the accuracy of the data that were retrieved in both cases. The reliance on ICD-9-CM codes for analysis of admission diagnoses is also a limitation, as hospitalization summary sheets may overlook some important details of the admission. Despite this caveat, important data can nonetheless be extracted. More precise administrative health databases unfortunately do not exist in the province of Quebec. Our study may also lack the power to detect differences in hospitalization rates because of a low number of patients in some specific categories of analysis.

We also note that mean age at diagnosis was surprisingly high in our cohort of CP patients (3.6 years old SD 3.0 years). Unfortunately, this study was not powered to compare children with an early diagnosis of CP with those who were diagnosed later in life. Previous literature has demonstrated that early diagnosis and early intensive interventions improve outcomes with regard to motor function, cognition, communication, and rates of CP-associated complications.[13,14] We would need to further explore the barriers to timely diagnosis of CP in our jurisdiction. The hospitalization burden in our study population was driven by medical complexity, epilepsy, and respiratory illness. Outpatient complex care clinical programs are emerging to meet the needs and improve the health outcomes of children with medical complexity who would benefit from coordinated care.[15] Exploring the impact of these programs on hospitalization rates of children with CP with varying levels of medical complexity would be an area of interest for future studies.

Conclusion

This study demonstrates the feasibility of linking a CP Registry to administrative health databases. It provides evidence to researchers and clinicians regarding the healthcare utilization of children with CP, as well as factors that increases the burden of hospitalization (both in frequency and duration) in these patients. It also highlights potential areas of focus to improve health services and coordinated care models for children with CP in Quebec. Improving interdisciplinary outpatient care for children with CP and medical complexity can potentially reduce their hospitalization burden. Preventive respiratory care for children with CP and improved epilepsy management and education could also potentially reduce their use of inpatient medical services.

Longer term data on these children as they transition to adolescence and adulthood will be of interest to explore if healthcare utilization patterns differ in this patient population once transition to adult care occurs. Other areas of interest for future direction include the influence of timely diagnosis and intervention on the hospitalization burden of children with CP, as well as barriers that delay access to appropriate diagnostic services and therapies.

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Statement of Authorship

MO led the study design and conceptualization, data acquisition, interpretation of data, and drafting and revising the manuscript. OF contributed to the interpretation of data and drafting and revising the manuscript. PN contributed to the data analysis and interpretation and drafting and revising the manuscript. MD contributed to the data analysis and revision of the manuscript. LK, NP, and MS contributed to the acquisition of data, interpretation of the data, and revision of the manuscript for intellectual content

Disclosures

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Supplementary Material

To view supplementary material for this article, please visit https://doi.org/10.1017/cjn.2020.199.

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