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Direct healthcare costs for people with cerebral palsy in the Brazilian unified health system between 2015 and 2019

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ABSTRACT

Background: Estimate the costs of inpatient and outpatient care for people with Cerebral Palsy (CP) in Brazil.

Research design and methods: Health records of people with CP in the Hospital and Outpatient Information Systems of Brazil between 2015 and 2019 were analyzed. Variables analyzed were gender, age, ICD, Intensive Care Unit (ICU) use, total cost, and ICU cost. Costs were adjusted for inflation and converted to dollars. Linear regression analysis was performed to investigate the association between social and clinical variables and direct costs.

Results: A total direct cost of approximately $166 million to the National Health System was identified, with $7.08 million/year and $26.1 million/year of inpatient and outpatient costs, respectively. The healthcare was primarily for children up to 14 years of age. The ICD 'spastic quadriplegic CP' received the most attendance. Rehabilitation was responsible for 75% of the outpatient care, with physiotherapy standing out. Increased age, use of ICU, and the types of CP are related to increased cost.

Conclusions: Healthcare for people with CP produced expressive costs for the Brazilian public health system, mainly with outpatient procedures and rehabilitation, with children being the most attended. Estimating these costs assist in better resource allocation for more effective healthcare provision.

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Cost of illness; cerebral palsy; hospital cost; ambulatory care; costs and cost analysis

1. Introduction

Cerebral palsy (CP) refers to a group of developmental disorders of movement and posture due to a nonprogressive impairment of the immature brain [1] is the most common physical disability in childhood [2]. The prevalence in high-income countries has declined over the last decade [3], due to improvements in maternal and perinatal care [4]. In low- and middle-income countries (LMICs), the prevalence of CP is higher compared with high-income countries [3], mainly due to barriers to the adequate implementation of healthcare strategies focused on this population [5]. Although there are no studies on the prevalence of CP in Brazil, it is estimated to be similar to that of low- and middle-income countries.

Disabilities, such as neuromusculoskeletal impairments and activities and participation limitations, worsen throughout the lives of individuals with CP [6]. This aspect is relevant because it requires frequent medical assistance, medication, use of assistive technologies, and rehabilitation [7]. Thus, the diagnosis of CP, in addition to physical, psychological, and emotional issues, increases financial costs for the individual, their family, and society [8]. Current evidence related to economic assessments demonstrates that treatment costs and loss of productivity over the life of a person with CP can reach US $1.3 million [9]. Furthermore, a person with CP may have expenses 1.8 times greater than a person from the general population [9–11].

In Brazil, about 75% of the Brazilian population relies exclusively on the Brazilian public health system (Sistema Único de Saúde – SUS), and all services provided are processed by the information systems of the Brazilian Ministry of Health [12–16]. Studies aimed at understanding the economic burden of a certain health condition are useful in the decision-making process and for the formulation and prioritization of public policies and rational allocation of resources [15,16]. Previous studies carried out in Brazil on the costs of neonatal care showed that premature birth, one of the risk factors for CP, is related to high costs, especially for extreme preterm infants [17,18]. In Brazil, no cost studies were found on the health of people with CP.

The objective of this study was to estimate the direct costs arising from inpatient and outpatient care of people with CP in Brazil between 2015 and 2019. Our secondary aim was to

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Article highlights

- Considering the research literature, this is the first direct cost study considering Cerebral Palsy from the perspective of the public health system in Brazil.
- The study also explores an analytical component, as it analyzes the association of social and clinical factors that may be associated with inpatient and outpatient costs. We observed that age, type of CP and ICU admission increases the total cost of a person with CP.
- We observed that rehabilitation services were responsible for 75% of the outpatient care but less than half of the costs.
- Our study showed that adults receive a greater volume of care in hospital services and children in outpatient services, indicating how costs are distributed for Brazilian public health service, highlighting the importance of procedures focused on prevention and health promotion.

characterize the care provided and the costs according to the different clinical classifications of people with CP, in addition to investigating the association between a set of social and clinical variables and direct outpatient and inpatient costs.

2. Method

2.1. Study design

This is a prevalence-based cost-of-illness study that evaluates annual costs over a five-year period. It integrates both descriptive and analytical component from the perspective of the Brazilian public health system. Data from a national database were analyzed using a macro-costing approach, considering the direct health costs arising from health actions provided by SUS. As we used public secondary data available at https://datasus.saude.gov.br/, ethical approval was not required.

Cost-of-illness was based on the prevalence of CP, including the costs of all consultations for people with CP within public health services at various phases of the disease, regardless of the time of diagnosis. Costs were identified per year between 2015 and 2019. No data were analyzed after 2019 due to the COVID-19 pandemic, which substantially affected healthcare in Brazil [19].

2.2. Health condition

All eight CP codes according to the 10th edition of the International Classification of Diseases (ICD-10) were included. The codes 'Cerebral Palsy,' 'Other forms of CP' and 'Unspecified CP' were grouped into a group called 'CP without details' (eTable S1). Data from people with CP of all age groups were included. For the analysis, ages were divided into 18 groups: up to 1 year, 1–4, 5–9, 10–14, 15–19, 20–24, 25–29, 30–34, 35–39, 40–44, 45–49, 50–54, 55–59, 60–64, 65–69, 70–74, 75–79, and over 80 years.

2.3. Procedures for data extraction

Data were extracted from the Hospital Information System (SIH/SUS) and Ambulatory Information System (SIA/SUS) from January 2015 to December 2019 and were selected according to the ICD-10 recorded as the main diagnosis. Both systems are National database (DATASUS), publicly available (http://datasus.saude.gov.br/).

The SIH/SUS contains records of hospital admissions reimbursed by the Ministry of Health that happened in Brazilian public health hospitals or private hospitals that are National Health System (NHS) accredited hospitals. Based on the Hospital Admission Authorization (AIH) schedule, we extracted social data (sex, age), clinical data (ICD-10 and Intensive Care Unit (ICU) admission, total cost, and ICU cost. The total cost of each hospitalization is subdivided into hospital services and professional services. The amounts contained in the hospital services combine the costs arising from the daily rates, room rates, food, personal hygiene, bedside support, hospital materials, medication, and Auxiliary Diagnosis and Therapy Services as well as professional services (except doctors and dental surgeons). The costs contained in professional services include the professional services of doctors and dental surgeons.

The SIA/SUS contains information on the care, procedures, and treatments that were carried out by SUS health establishments in the outpatient setting, registered in the Ambulatory Procedures Authorization (APAC) schedule. Using the SIA, we extracted social data (sex, age), clinical data (ICD, outpatient procedure, and procedure professional), and total cost. The professional categories Physical Therapy, Occupational Therapy, and Speech Therapy were included in the cost analysis as rehabilitation. We also analyzed the costs of botulinum toxin procedures, diagnostic exams (magnetic resonance imaging, computed tomography, and radiography), as well as orthoses and special materials.

2.4. Data analysis

We used TABWIN software (version 4.1.5) to extract the data from the systems. An analysis at the individual level was not possible because the data in the systems are aggregated and linked to the hospital or ambulatory registration number, and not to the individual's name or identification; thus, there may be different procedures linked to the same registration, and also new registration numbers for the same individual. The amounts were extracted in Brazilian Real (R$) and adjusted for inflation according to the Consumer Price Index (IPCA) for 2019. Subsequently, the amounts were converted to US dollars using the World Bank's Purchasing Power Parity (PPP) exchange rate. Brazil's PPP in 2019 was 2.28 [20].

All data processing and analysis (e.g. descriptive, statistical, and regression analyses) were conducted using R software (version 4.2.1) [21]. We performed a descriptive analysis of the social, clinical, and cost data of inpatient and outpatient services, estimating median and interquartile range for numerical variables and frequency measures for categorical variables. The results are presented at the level of registration of hospital admissions or outpatient consultations, and not at the individual level. The processing of inpatient and outpatient data in the R software was done using the 'microdatasus' package [22].

We performed a linear regression analysis to investigate whether social and clinical variables explain the direct costs in the SIH/SUS and SIA/SUS. After confirming that residuals were not normally distributed in a Q-Q plot, a generalized

linear model (GLM) with gamma distribution (skewed data) was adopted to evaluate the association between certain predictive variables (i.e. independent) and the total direct cost (i.e. dependent variable) in each information system. The independent variables were age (in years), sex, ICD code, and ICU admission (yes or no). Records with zero cost data and incorrect age, in the SIA/SUS, were excluded, this data can influence the results of the regression and can reflect a no real relation of the variables. For the model estimation, we adopted the identity link function for the SIA/SUS and log link function for the SIH/SUS. The Goodness of fit was verified using the AIC (Akaike Information Criterion). The correlation matrix was analyzed and variables that were highly correlated ($r > 0.7$) were considered collinear. The results for the model with the log link were presented in odds ratio.

3. Results

Between 2015 and 2019, 43454 inpatient records and 5,677,939 outpatient visits related to CP were found to be the main diagnosis. We verified a total direct cost of approximately US$166 million in that period, with US$35.4 million (US $7.08 million/year) of inpatient costs and US$130.5 million (US $26.1 million/year) of outpatient costs (Table 1). Total inpatient and outpatient costs were higher for males, totaling US $20.6 million and US$73.2 million, respectively.

Approximately 89% of the inpatient costs were related to hospital services. There were 1,104 ICU admission records, costing approximately US$2.7 million. Of this total number of records, the majority (76.8%) were children up to 14 years old –25.3% were children up to one year.

In the hospital system, most hospitalizations were in the age groups of five to 14 years, and we observed an increase in total costs with advancing age up to the age group of 35–39 years (Figure 1). The most advanced age groups had higher cost medians than the lowest age groups, except for the age group up to one year (eTable S2). In outpatient care, approximately 60% of the records and 62% of the total cost were for children ages one to 14 years. We found a reduction in the number of visits as age increased (Figure 1), especially after the age group of 20–24 years. Figure 1 shows the total number of records and total hospital and outpatient costs by age group.

The number of records and total costs for each CP-related ICD are shown in Table 2. In about 54% of the inpatient admissions (23,521), the main diagnosis code was 'CP without details,' and in 31.3% of the hospitalizations, the code was 'spastic quadriplegic CP.' In outpatient visits, 41.7% of the records referred to the ICD 'spastic quadriplegic CP.'

In the context of rehabilitation, there were 4,259,173 outpatient visits of the specialties selected in this study, representing 75% of the total outpatient visits and 48% of the total outpatient cost in the period investigated. Physical therapy was the specialty with the highest share of costs (77.7%), followed by Speech Therapy (12.15%) and Occupational Therapy (10.1%). The total cost of physical therapy was US $63.5 million, which represents 35.4% of the total cost of outpatient care (Table 1).

There were 70,767 records of botulinum toxin procedures, with a total cost of approximately US$14.7 million (Table 1). This procedure generated an approximate cost of US$11.9 million for 'spastic quadriplegic CP,' US$2.19 million for 'spastic diplegic CP,' and US$570 thousand for 'spastic hemiplegic CP,' mainly in the age groups from 5 to 19 years.

We found 14,731 diagnostic test records, with a total cost of approximately US$1.01 million. The most frequently performed tests were hip radiography, computed tomography of the brain, and magnetic resonance imaging of the brain. Orthotics and specialty materials totaled a cost of approximately $52.2 million for 306,206 records, representing 40% of the total cost of outpatient services (Table 1).

The results of the regression analysis are presented in Table 3. In the hospital system, the male sex, increased age (in years), ICU admission, and the conditions 'spastic quadriplegic CP,' 'dyskinetic CP,' and 'ataxic CP' were associated with higher cost ($p < 0.001$). Regression analysis showed that ICU admission increases the total cost of a person with CP by 672% ($p < 0.001$), and between CP type, the higher cost was associated with 'ataxic CP,' 44% higher ($p < 0.001$). In the outpatient system, all variables were associated with higher costs, excluding 'ataxic CP.' The largest associations were in the types 'spastic quadriplegic CP' and 'dyskinetic CP.'

4. Discussion

The present study estimated the cost of healthcare for people with CP in Brazil. Our findings identified a total direct cost of approximately US$166 million, from 2015 to 2019, with inpatient and outpatient care of people with CP in the Brazilian public health system. The costs were higher for males and for 'spastic quadriplegic CP.' In the hospital system, children require more ICU admission, while people aged 25 years and older had a higher median cost. In outpatient care, the highest cost is related to rehabilitation, especially physical therapy, and availability of equipment and materials, such as orthoses. The need for ICU admission, advancing age, the male sex, and 'spastic quadriplegic,' 'spastic diplegic,' 'dyskinetic,' and 'ataxic' CP are associated with a higher cost of healthcare.

We found an annual expending of around US$ 33 million for patients with CP in Brazil. Putting the costs with CP into a Brazilian perspective, the total cost with CP represented between 0,035% and 0,025% of the total health expenditure for the Brazilian public health system in the analyzed years [23]. Even if the costs for CP seem low, they are similar to other conditions, such as prematurity [17], beyond being a condition with high prevalence and many limitations.

The literature reports a range of CP-related cost estimates in different countries [9]. It is important to note that the cost estimates are context-specific, and the studies assess the costs under different aspects, making it difficult to compare absolute values between studies. Studies that include the costs of lost patient productivity can account for about 80% of the societal costs [9,24]. In addition, the loss of family productivity and non-medical costs, such as transportation and special education, can also have a significant impact on total costs [9,24]. Such differences can be observed in the total cost

Table 1. Inpatient and outpatient costs for the healthcare of people with cerebral palsy within Brazil's public health system from 2015 to 2019 with median and interquartile interval per year.

	2015		2016		2017		2018		2019		Total 2015-2019	
Components	Total	Median (25%;75% percentile)	Total	Median (25%;75% percentile)	Total	Median (25%;75% percentile)	Total	Median (25%;75% percentile)	Total	Median (25%;75% percentile)	Total	Median (25%;75% percentile)
Inpatient service costs (in US$):												
Hospital service	6,554,790	1,013 (101 - 1,046.77)	6,382,445	953 (177 - 984)	6,402,248	925 (213 - 956)	5,910,096	892 (240 - 922)	6,348,264	855 (255 - 884)	31,597,844	884 (188 - 956)
Professional service	769,524	87 (47 - 89)	791,507	81 (58- 84)	742,068	79 (57 - 82)	678,166	76 (71 - 79)	767,223	73 (68 - 76)	3,748,490	79 (62 - 84)
Days in ICU	480,623	1,243 (497 - 4,226)	439,054	701 (233 - 2,105)	665,838	1,136 (454 - 4,316)	485,056	1,095 (438 - 3,285)	649,179	839 (419 - 2,992)	2,719,752	965 (419 - 3,285)
Inpatient Total	7,338,494	1,100 (174 - 1,136)	7,182,768	1,035 (216 - 1,069)	7,153,190	1,005.38 (266-1,038)	6,599,881	969 (314 - 1,001)	7,119,597	929 (313 - 960)	35,393,932	960 (242 - 1,038)
Outpatient service costs (in US$):												
Rehabilitation	10,959,482	3 (2- 11)	10,984,007	4 (2 - 10)	12,372,308	5 (2 - 10)	14,356,594	6.4 (2 - 9)	14,852,412	7 (2 - 9)	63,524,805	5 (2 - 9)
Diagnostic tests	232,501	50 (50 - 139)	225,027	47 (47 - 131)	214,689	46 (46 - 127)	179,188	44 (44 - 122)	161,454	42 (5 - 60)	1,012,861	47 (42 - 122)
Orthoses and special materials	10,313,804	72 (67 - 297)	9,347,447	64 (63 - 176)	11,023,324	62 (61 - 213)	11,069,290	60 (59 - 124)	10,515,091	57 (39 - 114)	52,268,958	66 (57 - 197)
Botulinum toxin	5,516,302	459 (0 - 918)	4,778,701	431 (0 - 863)	4,168,549	0 (0 - 419)	262,908	0 (0 - 0)	-	-	14,726,462	0 (0 - 419)
Others	3,865,640.82	11 (9 - 11)	4,341,908.32	10 (7 - 10)	4,252,779.08	10 (7 - 10)	3,965,728.32	08 (08 - 9)	3,731,922.75	7 (7 - 9)	20,157,979	9.51 (7 - 10)
Outpatient Total	27,209,691	7 (2 - 11)	26,387,942	7 (2 - 10)	27,850,667	7 (2 - 10,)	24,830,464	8 (2 - 9)	24,258,948	7 (2 - 9)	130,537,713	7.46 (2 - 10)

Abbreviations: ICU, Intensive Care Unit; US$, United States dollars. The median amounts and 25%; 75% percentiles refer to hospital admissions and outpatient consultations and are not per patient.

found between the studies. For example, one study estimated the annual cost of living at US$1.2 million per person with CP, including direct costs with special education and loss of productivity [9]. It is worth noting that the present study included only the direct health costs of the public health service and did not include the costs borne by the patient or productivity losses.

In the present study, most of the inpatient and outpatient records were related to males, who presented the highest total costs. Male sex occurs more frequently, probably owing to greater biological vulnerability, which has been explained by several factors such as a different brain organization [25,26] and genetic predisposition [27]. A higher prevalence of males with CP in healthcare was also found in other studies [11,24–29]. This higher prevalence may justify the association found in the regression analysis regarding the increase in the cost for males in the present study. Despite this association, this study and other studies show that sex alone is not a variable that influences cost at the individual level, but rather the severity of injury and severity of CP [10,29]. Sex is related to a greater number of cases, but the literature does not point to a relationship between sex and the severity of the PC injury [25].

In this study, the highest number of ICU admissions was in children up to 14 years of age. One of the main causes of hospitalizations, morbidity, and mortality in children with CP is respiratory system dysfunction [30]. Symptoms of respiratory dysfunction occur primarily at younger ages, which may explain the higher number of hospitalizations; as the age advances, the symptoms are more tolerable and recognized more quickly by the family, reducing the need for ICU admissions [31].

In addition to the higher number of ICU admissions, children up to 14 years of age have a higher total cost and number of hospital admissions and outpatient care than adults. A similar result was also found by a study conducted in South Korea [32]. The CP impacts child development and therefore interventions have a greater incidence in the early years [11] as gains in this phase are enhanced due to neural plasticity [33,34]. Interventions are carried out in greater quantity and frequency in childhood to prevent complications as well as provide improvement in functionality [32,33]. Consequently, this greater number of procedures and interventions generates a higher cost in early life, which may explain the results found in the present study. Another point to be considered is the life expectancy of people with CP which is lower than the general population and is reduced according to the degree of severity and as age advances; therefore, not all people with CP reach adulthood [7].

In general, we observed that adults with CP have a higher frequency of admission and cost in the hospital system compared to the outpatient service. In addition, regression analysis showed that, with each increase in age (in years), there is an increase in costs (for each age increment in years, the cost increases by 1%). Patients with CP tend to lose professional follow-up in health care in the transition from pediatric services to adult services and therefore usually seek care when a problem emerges or worsens, generating a higher cost [7]. Furthermore, with advancing age, there is a greater propensity

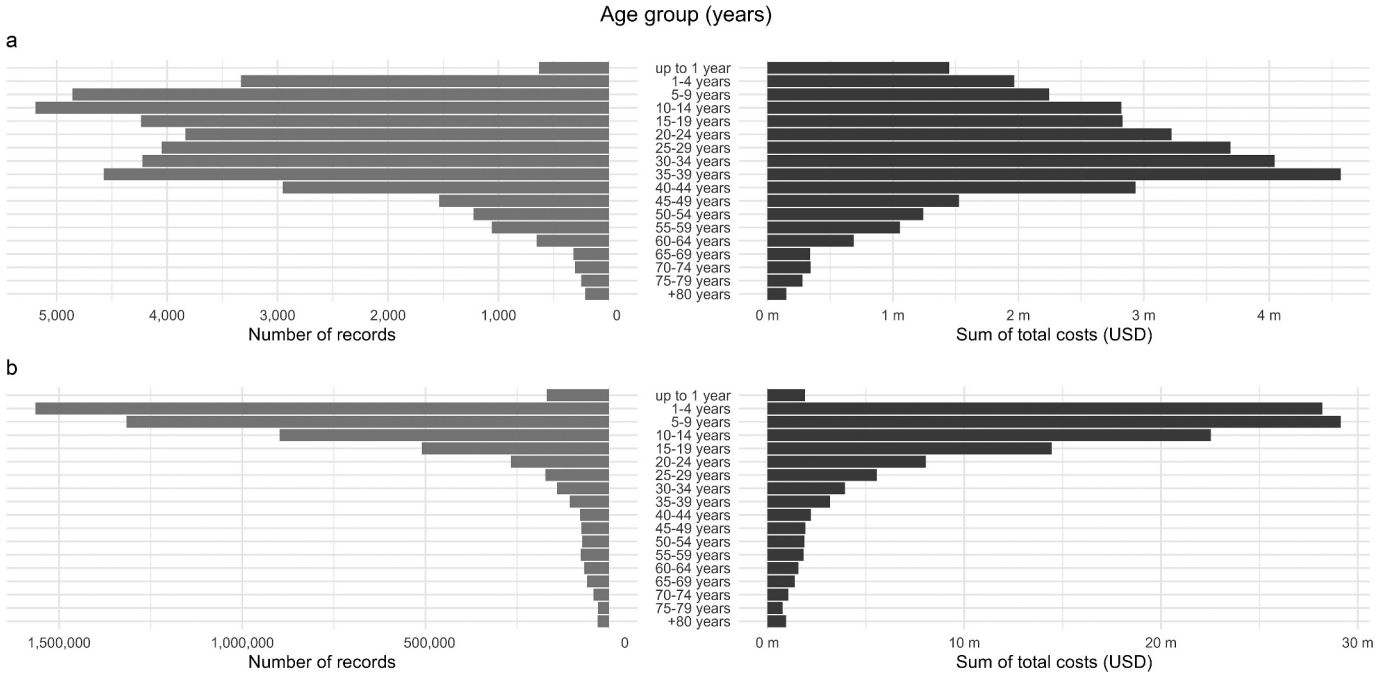

Figure 1. Number of records and Total Cost (USD) for people with Cerebral Palsy with Inpatient and Outpatient services, per age group, from 2015 to 2019.

Table 2. Inpatient and outpatient costs for the healthcare of people with cerebral palsy within Brazil's public health system from 2015 to 2019 with median and interquartile interval per ICD-10.

	Inpatient service costs (in US$):				Outpatient service costs (in US$):			
Condition:	Qty	%	Total cost	Median (25%;75% percentile)	Qty	%	Total cost	Median (25%;75% percentile)
Spastic quadriplegic CP	13,615	31.33	12,471,074	1,005 (929.1 - 1069.5)	2,367,269	41.69	60,794,546	7.27 (2.2 - 10.3)
Spastic diplegic CP	3,006	6.92	1,807,771	458 (163.2 - 1016.4)	533,814	9.40	10,331,331	8.08 (2.2 - 10.6)
Spastic hemiplegic CP	1,064	2.45	888,902	960 (230.9 - 1038.9)	255,364	4.50	3,937,237	7.75 (2.2 - 10.3)
Dyskinetic CP	1,114	2.56	1,032,890	1,005 (931.9 - 1069.5)	244,415	4.30	5,207,375	7.75 (2.2 - 10.3)
Ataxic CP	1,134	2.61	1,127,180	1,026 (960 - 1069.5)	44,350	0.79	621,455	7.21 (2.4 - 11.4)
CP without details	23,521	54.13	18,066,117	1,140 (170.5 - 1035)	2,232,727	39.32	49,645,773	7.75 (2.2 - 11.2)

Abbreviations: CP, Cerebral Palsy; Qty, Quantity; %, percentage. The median amounts and 25%; 75% percentiles refer to hospital admissions and outpatient consultations and are not per patient.

Table 3. Regression analysis of inpatient and outpatient costs based on the predictors sex (male/female), age (in years), ICU admission (yes/no), and ICD-10.

	Inpatient Care			Outpatient Care		
Variable	OR	95%CI	p-value	B	95%CI	p-value
Sex*						
Female	1	-				
Male	1.09	1.07; 1.10	<0.001	0.47	0.33;0.62	<0.001
Age	1.01	1.017; 1.018	<0.001	0.26	0.25; 0.26	<0.001
ICU▲	7.72	7.40; 8.05	<0.001			
ICD♦						
Spastic hemiplegic CP	1					
Spastic quadriplegic CP	1.31	1.25; 1.37	<0.001	9.85	9.59; 10.11	<0.001
Spastic diplegic CP	0.82	0.78; 0.86	<0.001	3.53	3.23; 3.84	<0.001
Dyskinetic CP	1.39	1.31; 1.47	<0.001	9.35	8.88; 9.82	<0.001
Ataxic CP	1.44	1.35; 1.52	<0.001	−1.17	−1.77; −0.57	<0.001
CP without details	0.98	0.94; 1.02	0.442	7.62	7.7; 7.87	<0.001

*The reference category was the female sex.
▲The reference category was 'no.'
♦The reference category was 'Spastic hemiplegic CP.'
Abbreviations: ICU, Intensive Care Unit; ICD, International Classification of Diseases; CP, Cerebral Palsy; 95%CI, 95% Confidence intervals; OR, Odds ratio.

for the appearance of other health conditions, such as fractures, need for surgery and heart diseases [32], leading to more hospital and emergency visits, highlighting a greater number of hospitalizations over outpatient care [28].

There was a predominance of consultations for people with spastic CP, followed by dyskinetic and ataxic CP. This distribution is similar to that presented by other studies carried out in other countries [7,35]. This result can be justified by the higher prevalence of spastic CP, which represents 80% of the people with CP [7,36]. Also, the spastic type generated higher costs compared to the other clinical types [11], and the 'spastic quadriplegic CP' is usually the most severe type, with greater complications and greater need for intervention [35]. Regression showed that the spastic quadriplegic, spastic diplegic, dyskinetic, and ataxic types of CP increase costs. These CP subtypes are associated with a greater number of comorbidities related to aspects such as speech, feeding, and seizure disorder and therefore greater severity of the condition [37]. One of the main demands of the spastic subtype is the management of hypertonia [35,38,39].

In fact, we found that the application of botulinum toxin, an intervention aimed to reduce hypertonia, prevent permanent misalignments and surgical interventions, and decreases pain [35,38,39], had a cost greater than 14 million dollars. An Australian and a Korean study evaluated the cost per patient with botulinum toxin treatment and estimated an increase in direct cost of AUD$793 and AUD$867 for patients with hemiplegia and diplegia, respectively [39]; and a cost of KRW 420,687 for the 0–6 years age group and KRW 303,775 for the 7–18 group [32]. Since the present study did not make an average estimate per individual, cost comparison with other studies is limited. However, if we consider the total cost over all records, we observe a mean of 208 dollars for the application of botulinum toxin, which seems to be a very low investment by the Brazilian public health system. Considering the importance and effectiveness of botulinum toxin in motor interventions and tone management for patients with CP, especially those with the spastic type [38], and analyzing the number of botulinum toxin applications found in this study, we can believe that the investment cost by SUS may not be sufficient for all those in need. This indicates that a greater allocation of resources could be warranted for this intervention.

In this study, we identified a significant number of ICD records that did not specify the clinical type of CP, which hinders an in-depth analysis of the specific costs related to the clinical type. It is worth mentioning that we were not able to carry out a cost analysis based on the severity of CP because the Brazilian public system does not record that data. The motor performance of the person with CP can be classified by the Gross Motor Function Classification System (GMFCS) into five levels, where level V represents greater functional severity. A Australian study showed the association between the general costs with health, rehabilitation, botulinum toxin, orthoses, mobility equipment, among others, and the GMFCS levels [9]. The GMFCS classification is more useful for directing investments and prevention actions than the clinical type. So, it is important that Brazilian health services adopt this classification in their medical records.

Rehabilitation accounted for almost half of the outpatient costs for the period analyzed in the present study. The person with CP has a high demand for outpatient services, mainly rehabilitation, due to the complexity of disabilities that progress throughout life [24,32]. Despite the relevance, each of the procedures performed by rehabilitation professionals' costs less than US$10, based on the amount refunded by the public health system. Physical therapy, occupational therapy, and speech therapy interventions have a great impact on improving functionality, including increased skills and greater participation of children in schools and of adults in the labor market [38].

Thus, the cost related to rehabilitation can be compensated with the reduction in disabilities and loss of productivity, suggesting that a greater investment in this area can improve independency and generate greater social participation for these patients and as result, reduce the total costs of CP [24,35]. A Korean study estimated the cost per person with rehabilitation between KRW 1,452,938 and KRW 2,023,308, age-dependent, not including costs with speech therapy and assistive devices [32]. Although it is a different way for estimating costs than the one presented in this study, the cost estimated for the Brazilian public health system with rehabilitation seems to be inferior, indicating a low expenditure on rehabilitation. More than 80% of health expenditure in Brazil is related to curative actions, medications, and medical supplies, with less than 20% of costs going toward prevention, health promotion and surveillance, long-term care, and rehabilitation. The share of rehabilitation care was around 1% of health spending for the period 2015–2019 in Brazil [23].

The cost of orthoses represented 40% of the total outpatient cost. Orthoses are used to prevent deformities and increase the level of activity of the person with CP [36]. Mobility devices, such as walkers and wheelchairs, increase independence in locomotion [40,41]. These devices are extremely necessary and recommended by the guidelines for people with CP [42]. Orthoses are among the most used therapies in the care of people with CP [35], and the greater the severity of the condition, the greater the number of devices in use [43].

As demonstrated, the costs of CP can vary according to the clinical and social differences between people with CP. Understanding how health costs are distributed and what resources are required to care for people with CP are important measures in health planning [11]. Estimating and knowing the prevalence of CP is also an important measure for planning, as it is known that risk factors associated with CP can be avoided with health actions, and thus the prevalence of CP could change [11]. This change would impact costs and estimates for future planning [11].

Considering health decision-making, future analyses of health care need to take the COVID-19 pandemic into account. This period, mainly in 2020 and 2021, changed health care in Brazil. Some health services, as they were not considered essential, were suspended for a period, reducing the availability of procedures by the SUS [19]. This affected health care for some conditions that require continuous care, and CP is one of the health conditions that may have been impacted during this period [19]. Health care may have changed during and after

this period in Brazil. In this context, these pre-pandemic cost data are extremely important, as this is the first cost analysis for this population in the country, and it can serve as baseline data for future comparisons in a post-pandemic scenario.

Looking forward, addressing these findings prompts critical reflections on the level of investment in CP healthcare services. Understanding these historical trends and current challenges is essential for informing future policy interventions and resource allocations aimed at improving healthcare equity and outcomes for individuals with CP.

In this context, we also propose the possibility of future studies that include predictive analyses regarding costs aiming to identify cost patterns for CP and projecting future scenarios, understanding that these projections can also be used in health decision-making processes. Another data that can also be included in future studies is data on indirect costs (e.g. lost productivity) related to this health condition.

4.1. Strengths

To the best of our knowledge, this is the first study to investigate the direct costs of CP generalizable to Brazil. The findings contribute to the knowledge of the distribution of costs between the social and clinical components investigated. Additionally, our findings contribute to the understanding of what factors are associated with inpatient and outpatient costs of healthcare for people with CP. The results have the potential to contribute to the development and practical implementation of public policies to improve the investment in healthcare, especially rehabilitation, to enhance the quality of life for individuals with CP. In addition, the results demonstrate the need for health services throughout the lives of these patients, highlighting the importance of implementing follow-up services for them.

It is also emphasized that preventive measures and health promotion efforts, particularly for pediatric patients, are important to mitigate future healthcare expenditures. Our analysis revealed that adults incurred the highest costs in hospital admissions, whereas children accounted for the largest share of outpatient costs.

4.2. Limitations of the study

The costs presented may be underestimated, considering the inherent limitations of secondary data from information systems in Brazil. For example, when extracting the data considering the ICD-10 code, procedures registered with other codes related to CP may not have been included. There is an inherent limitation to the completeness of records in health information systems. The available data did not allow a detailed analysis of the characteristics of individuals with CP treated within the public health system. The study did not include costs in the private system and costs in primary care.

5. Conclusion

Our findings demonstrated that the economic burden due to the management of individuals with cerebral palsy within the Brazilian public health system was considerable, totaling around US$166 million between 2015 and 2019, mainly for patients with spastic quadriplegic cerebral palsy. Outpatient care accounted for more than 75% of the total healthcare costs. We presented novel findings showing that inpatient and outpatient care costs were somewhat higher for individuals aged between 25 to 39 years and 5 to 14 years, respectively. Physical therapy interventions were widely adopted, representing the largest share of outpatient care costs. Regarding the PC type, 'spastic quadriplegic CP' type was the condition with the highest associated costs. Regression models demonstrated that advancing age, specific CP types (such as 'spastic quadriplegic,' 'dyskinetic,' and 'ataxic'), and ICU admissions led to increases in inpatient and outpatient costs.

These results are important for enhancing healthcare investment and service health assistance models to better meet the needs of CP patients. We emphasize the importance of more effectively allocated healthcare resources, ensuring comprehensive and equitable care for all individuals with Cerebral Palsy.

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Declaration of interest

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Author contributions

EB Freire and AM Toledo designed the study. We state that all authors had substantial contributions to the conception of the work, analysis, and interpretation of the data, and all authors have participated in drafting and revising it critically. All the authors agreed to be accountable for all aspects of the work and the work was approved by all the authors.

Data availability statement

All data generated or analyzed during this study are included in this published article. The datasets used during the current study are available from the corresponding author on reasonable request or provide link for the same.

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