

# Epidemiological characteristics of cerebral palsy in children and adolescents in a Brazilian northeast capital

*Características epidemiológicas da paralisia cerebral em crianças e adolescentes em uma capital do nordeste brasileiro*

*Características epidemiológicas de la parálisis cerebral en niños y adolescentes de una ciudad del Nordeste de Brasil*

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**ABSTRACT** | This study aimed to describe the prevalence of cerebral palsy among children and adolescents, its subtypes, associated comorbidities, and socioeconomic characteristics of families. This is a cross-sectional epidemiological study conducted from a population-based survey on cerebral palsy among children and adolescents in the city of Aracaju, Sergipe, Brazil. Results: Based on guardians' answers to the proposed questionnaire, we obtained data on 240 children and adolescents with cerebral palsy in Aracaju. The period prevalence of cerebral palsy was 1.37 per 1,000 people. Some neighborhoods showed a prevalence three to four times higher than that found to the overall city, indicating that the total prevalence is not a homogeneous indicator. Most participants were male (56.25%), self-declared as mixed-race or Black (67.50%), and aged 8.56 years on average. Bilateral spastic was the most prevalent cerebral palsy subtype (45.42%), and epilepsy was the most common comorbidity (48.33%) along with intellectual disability. Household monthly income was \$ 252.87. Our results indicate that children and adolescents with cerebral palsy are mostly from minoritized groups, Black or mixed-race, and live in extreme poverty.

**Keywords** | Cerebral Palsy; Epidemiology; Social Determinants.

**RESUMO** | O objetivo deste estudo foi descrever a prevalência de paralisia cerebral entre crianças e adolescentes, seus subtipos, as possíveis comorbidades e as características socioeconômicas das famílias. Foi realizado um estudo epidemiológico do tipo transversal a partir de um inquérito de base populacional sobre a paralisia cerebral em crianças e adolescentes na cidade de Aracaju (SE), Brasil. O estudo obteve informações sobre 240 crianças e adolescentes com paralisia cerebral a partir das respostas a um questionário feitas por seus responsáveis. Foi encontrada a prevalência de período de 1,37 em cada mil. Alguns bairros possuem prevalência de três a quatro vezes maior, revelando que a taxa de prevalência total não é um indicador homogêneo. A maioria dos participantes foi do sexo masculino (56,25%), de raça/cor declarada como parda ou preta (67,50%), sendo que a média de idade foi de 8,56 anos. A paralisia cerebral de tipo espástica bilateral foi a mais frequente (45,42%) e a comorbidade referida na maioria dos casos foi a epilepsia (48,33%). A renda familiar mensal correspondia a \$252,87 dólares. O estudo revelou que as crianças e adolescentes com paralisia cerebral são, em grande parte, pertencentes a minorias sociais, de raça/cor parda ou preta, e suas famílias vivem na linha da extrema pobreza.

**Descritores** | Paralisia Cerebral; Epidemiologia; Determinantes Sociais.

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**RESUMEN** | El objetivo de este estudio fue describir la prevalencia de parálisis cerebral en niños y adolescentes, sus subtipos, las posibles comorbilidades y las características socioeconómicas de las familias. Se realizó un estudio epidemiológico transversal a partir de una encuesta poblacional sobre parálisis cerebral en niños y adolescentes de la ciudad de Aracaju, Brasil. El estudio obtuvo información de 240 niños y adolescentes con parálisis cerebral con base en las respuestas de sus padres a un cuestionario. Se encontró la prevalencia de período de 1,37 por mil. Algunos barrios tienen una prevalencia de tres a cuatro veces mayor, lo que revela que la tasa de prevalencia total no es un indicador homogéneo.

La mayoría de los participantes eran varones (56,25%), de raza/color declarado parda o negra (67,50%), con un promedio de edad de 8,56 años. La parálisis cerebral espástica bilateral fue la más frecuente (45,42%), y la comorbilidad reportada en la mayoría de los casos fue la epilepsia (48,33%). El ingreso familiar mensual correspondió a 252,87 dólares. El estudio reveló que los niños y adolescentes con parálisis cerebral son mayoritariamente de minorías sociales, raza/color parda o negra, y sus familias viven en extrema pobreza.

**Palabras clave** | Parálisis Cerebral; Epidemiología; Determinantes Sociales.

## INTRODUCTION

Cerebral palsy (CP) is the most common cause of physical disability in childhood. The structural and functional changes resulting from CP may generate great emotional, financial, and social impacts to individuals, families, and communities affected by the condition. This occurs because CP incurs considerable costs throughout the lives of affected individuals due to the need for continued healthcare, rehabilitation, education, and social interventions. Children and young people with CP have significantly worse health conditions<sup>1</sup> and meeting their needs within vulnerable communities with limited resources poses a great challenge<sup>2</sup>.

According to a systematic review of the international literature, the prevalence of CP is 2.11 per 1,000 live births and the best-known risk factors are placental abnormalities, congenital malformations, low birthweight, meconium aspiration, emergency cesarean section, birth asphyxia, neonatal infections and seizures, respiratory distress syndrome, and hypoglycemia<sup>3,4</sup>.

We found no epidemiological studies indicating the prevalence of CP in Brazil, but international researchers are deeply committed to provide information on its prevalence, trends, subtypes, causes, comorbidities, functionality, and associations with the socioeconomic characteristics of individuals affected by the disease. Understanding CP characteristics is essential to plan, implement, and evaluate prevention and treatment policies and programs.

Information technology and CP records in monitoring systems are commonly used by developed countries

to collect data on disease surveillance and different epidemiological approaches<sup>5-9</sup>. In the absence of CP records in low-income countries, as is the case in Brazil, researchers are conducting population studies with relevant information to the scientific community and their regions.

Thus, this article aims to describe the prevalence of CP among children and adolescents, its subtypes, associated comorbidities, and socioeconomic characteristics of affected families based on data from a northeastern Brazilian capital.

## METHODOLOGY

This is a cross-sectional epidemiological study based on a morbidity survey on cerebral palsy (CP) among children and adolescents applied in primary healthcare in the city of Aracaju (SE), Brazil.

### Study area

The study was conducted in the capital of Sergipe, Aracaju, which occupies an area of 181.90 km<sup>2</sup>. According to the Brazilian Institute of Geography and Statistics (IBGE)<sup>10</sup>, the municipality has an estimated population of 623,766 inhabitants and a population density of 3,140,70hab/km<sup>2</sup>. Aracaju comprises 39 neighborhoods and no rural areas.

Primary healthcare services cover 93% of the municipality population, including 44 health services and 144 primary care teams.

## Data production

Data was collected between June 2016 and December 2017 by an active search from key informants, members of primary care teams of all health units in the city. For qualifying data collection on children and adolescents with CP, the research team voluntarily provided training on all health units of the city. To reach as many participants as possible, the survey was also conducted in the only public secondary health service in the municipality, reference for people with CP. The research team collected data in the homes of people affected by CP using a structured questionnaire developed by the researchers.

The study included children and adolescents aged between 0 and 18 years, affected by cerebral palsy, and with medical diagnosis recorded as G80.0-G80.9 according to the International Statistical Classification of Diseases and Related Health Problems (ICD). Users of the rehabilitation services of Aracaju not residents of the municipality territory were excluded.

The following variables were observed: gender, age, ethnicity, country of birth, reported causes, presumed moment of involvement, place of delivery, type of delivery, gestational age, birth weight, hospitalization after birth, area of residence, family income, guardian's education level and work situation, and social security benefit.

The period prevalence of CP was calculated<sup>11</sup> using the denominator of 1,000 children and adolescents living in each neighborhood. Information on residents were obtained from IBGE<sup>10</sup>.

Data underwent a descriptive analysis with measures of central tendency and dispersion. Absolute and relative frequencies were also verified. Analyses were conducted using the R-studio functions of the R software (R Foundation for Statistical Computing, 2020), which waives a specific statistical package.

CP spatial distribution within neighborhoods was described using the exploratory spatial data analysis (ESDA) in the 2.18.3 *QGIS* software (Open Source Geospatial Foundation, 2016). The cartographic base of the city of Aracaju was provided by IBGE, and the

cartographic projection corresponds to the Geocentric Reference System for the Americas (SIRGAS2000).

## RESULTS

Based on guardians' answers to the proposed questionnaire, we obtained data on 240 children and adolescents with cerebral palsy in the municipality of Aracaju. Based on the population of 174,699 residents of the same age group reported by the Brazilian demographic census, the period prevalence of individuals affected by CP was 1.37 per 1,000 inhabitants. However, we verified differences in the prevalence of neighborhoods reaching up to 4 per thousand. Figure 1 shows the choropleth maps with CP distribution by neighborhoods according to the crude prevalence rate per 1,000 inhabitants (1A) and the prevalence rate smoothed by the empirical Bayesian model (1B). The maps indicate that the northern part of the city (peripheral region) presents a higher prevalence, whereas the central region presents a lower prevalence.

Most participants were male, self-declared mixed-race or Black, born in Aracaju, and aged between 0 and 4 years (Table 1). Mean age was 8.56 years old (SD=5.76).

Bilateral spastic cerebral palsy was the most prevalent, but over 30% of the respondents are unaware of the type of CP due to insufficient information, as medical reports indicated the diagnosis exclusively by G80.0 ICD code. Epilepsy was the most commonly reported comorbidity, followed by intellectual disability. According to respondents, clinical complications during childbirth are the most frequent causes of CP and perinatal brain injuries were the most reported (Table 2).

Within the study sample, most deliveries were vaginal and occurred in the hospital environment, resulting of full-term pregnancies with children of birthweight above 2,500g. According to the questionnaires, most children had to be hospitalized soon after birth (Table 3).

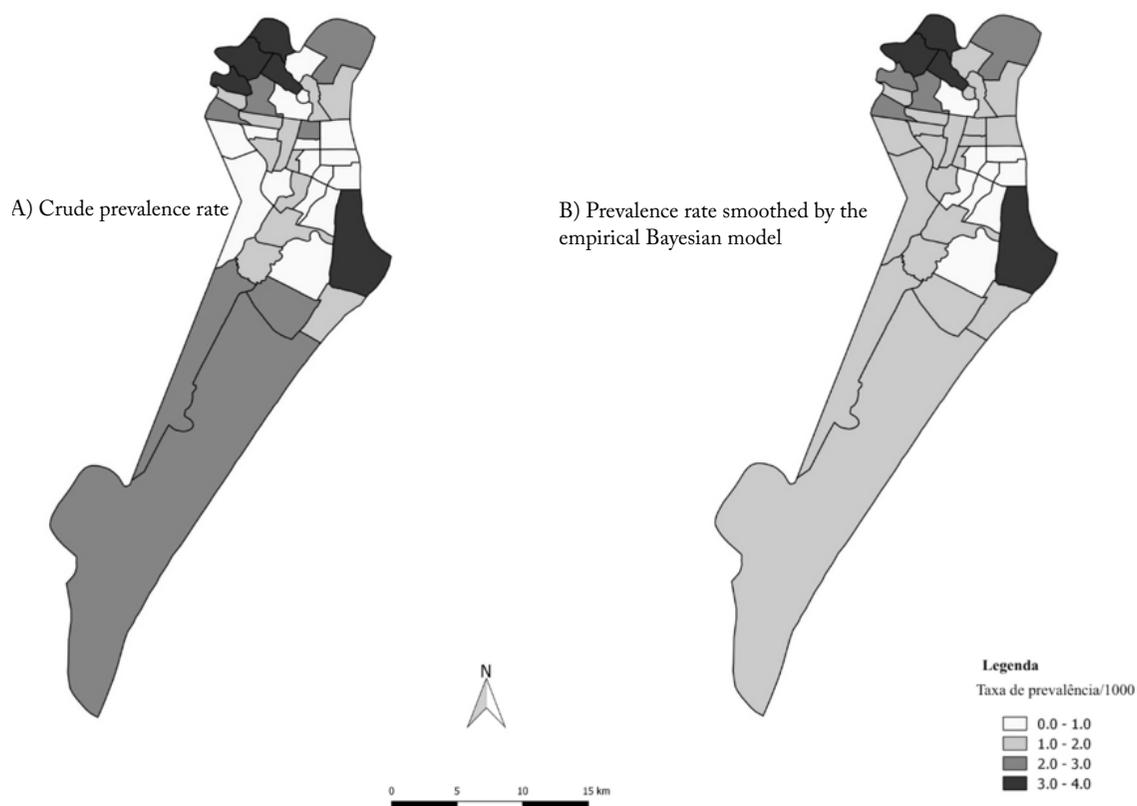


Figure 1. Spatial distribution of the prevalence of cerebral palsy in Aracaju neighborhoods (SE), Brazil, 2017

Table 1. Demographic characteristics of children and adolescents with cerebral palsy

Variables	n	%
Gender		
Male	135	56.25
Female	105	43.75
Ethnicity		
Asian	16	6.67
White	61	25.42
Indigenous	1	0.42
Black/mixed-race	162	67.50
Place of birth		
Aracaju	218	90.83
Other cities in Sergipe	18	7.50
Other states	4	1.67
Age group		
0-4 years	71	29.58
5-8 years	49	20.42
9-12 years	49	20.42
13-16 years	46	19.17
17-18 years	25	10.42
Total	240	100

Table 2. Description of cerebral palsy subtypes, comorbidities, reported causes, and presumed moment of injury

Variables	n	%
Type of cerebral palsy		
Bilateral spastic	109	45.42
Unilateral Spastic	46	19.17
Dyskinetic	5	2.08
Ataxic	7	2.92
Unknown	73	30.42
Reported comorbidities*		
Visual impairment	72	30.00
Hearing impairment	22	9.17
Intellectual disability	81	33.75
Behavioral disorders	51	21.25
Epilepsy	116	48.33
Reported cause		
Congenital disorders	64	26.67
Complications during pregnancy	34	14.17
Clinical complications during childbirth	67	27.92
Lack of support during childbirth	16	6.67
Childhood diseases or traumas	30	12.50
Unknown	29	12.08

(continue)

Table 2. Continuation

Variables	n	%
Presumed moment of injury		
Prenatal	81	33.75
Perinatal	102	42.50
Postnatal	33	13.75
Unknown	24	10.00
Total	240	100

\*Participants could report more than one comorbidity.

Table 3. Birth characteristics of children and adolescents with cerebral palsy

Variables	n	%
Place of delivery		
Hospital	230	95.83
Home	1	0.41
Unknown	9	3.75
Type of delivery		
Vaginal	123	51.25
Cesarean section	108	45.00
Unknown	9	3.75
Gestational age		
Preterm	74	30.83
Full-term	143	59.58
Postterm	11	4.58
Unknown	23	9.58
Birthweight		
< 1,000 g	4	1.67
≥ 1,000 g and < 1,500 g	12	5.00
≥ 1,500 g and < 2,500 g	42	17.50
≥ 2,500 g	117	48.75
Unknown	65	27.08
Postpartum hospitalization		
Yes	125	52.58
No	96	40.00
Unknown	19	7.92
Total	240	100

Data on family income were collected according to the minimum wage set in Brazil in 2016, which corresponded to \$252.87. The sum of household members living with the analyzed children and adolescents totaled 987 people, an average of 4.11 people per household. The monthly income of most studied families was equivalent to one minimum wage. The education level of most guardians ranged between nine and twelve years, corresponding to secondary education, and the predominant work activity was that of homemaker, which includes the care provided to the person with CP. Most participants received the social security benefit. According to the Brazilian government criteria, for families to be able to receive the benefit, the monthly household income must be equivalent to ¼ of the minimum wage (Table 4).

Table 4. Socioeconomic characteristics of families

Variáveis	n	%
Household income in minimum wages		
Up to 1 minimum wage	140	58,33
2	55	22,92
3	24	10,00
4	11	4,58
5 minimum wages or more	10	4,17
Guardian's education level in years		
0-4	30	12,5
5-8	62	25,83
9-12	94	39,17
13-18	54	22,5
Guardian's work situation		
Unemployed	40	16,67
Informal job	8	3,33
Formal job	17	7,08
Civil servant	50	20,83
Autonomous worker	25	10,42
Homemaker	100	41,67
Social security benefit		
Yes	172	71,67
No	68	28,33
Total	240	100

## DISCUSSION

The prevalence of cerebral palsy (CP) in our study population was lower than that reported by studies conducted in low-income countries, such as Uganda; middle-income countries, as China and Turkey; or high-income countries in Europe<sup>9,12-14</sup>. We verified relevant differences in prevalence within Aracaju, since some neighborhoods in the northern part (peripheral region) of the city showed a prevalence three to four times higher. This finding indicates that the total prevalence is not a homogeneous indicator and that this physical disability in childhood may be associated with social inequities.

A study conducted in northern England proposed a spatiotemporal clustering between CP cases and found consistent results that associate the disease onset with exposures to perinatal maternal infections in more densely populated areas<sup>15</sup>.

A systematic literature review reported significant associations between socioeconomic status and CP risk even when considering birthweight and gestational age as confounding variables<sup>16</sup>. Providing spatial information such as those addressed in our study may help preventing risk factors for CP – such as low birthweight, maternal infections, congenital malformations, and prenatal care difficulties – and mitigate their unwanted outcomes<sup>15,17</sup>.

To provide a better understanding on such prevalence difference, further trend analyses should be conducted grounded on our study. We do not attribute the low prevalence found in our study to limitations in case screening, given that primary services coverage in Aracaju is based on delimited territories with which primary care teams – the key informants to access the families – are well familiarized.

Most study participants are from Aracaju, and migration does not seem to interfere with the local prevalence of CP. However, the underdiagnosis and mortality rate of children with mild CP may interfere with the prevalence of the condition, so that future studies on the subject should conduct mortality analyses, given that the disease occurs chiefly among children aged between 0 and 12 years, making up more than 70% of the study population.

We verified a great ethnic disparity in CP, where mixed-race and Black individuals were more commonly affected. This finding is corroborated by other studies performed in different contexts<sup>18</sup>. A retrospective cohort conducted in California, US, found that Black babies were 29% more likely to have CP than white babies<sup>19</sup>. Another study assessing three areas of the United States found the risk of spastic CP to be more than 50% higher for Black children<sup>20</sup>. The studies do not fully explain racial disparities, but the increased risk of low birthweight among Black children is a hypothesis for the higher prevalence of CP within this group, as well as their highest socioeconomic vulnerability, which interferes with access to health services.

A study conducted in Uganda and another in Norway likewise verified a higher prevalence of bilateral spastic CP (45% and 49%, respectively)<sup>21,22</sup>. Corroborating our results, these studies found epilepsy and learning disabilities to be the most common comorbidities. A systematic review of the literature on CP in low-income countries found outpatient studies to present increased rates of spastic quadriplegia<sup>23</sup>.

According to the informants, most cases result from complications during childbirth, with injury presumably occurring in the perinatal moment. This finding alerts to eventual complications during pregnancy and delivery, such as the care of mother and birth asphyxia. A study conducted in Turkey reported that prenatal risk factors for CP were more frequently observed in upper-class groups, whereas perinatal factors were more common among lower-class groups<sup>14</sup>.

The predominance of full-term children weighing more than 2,500 g in our study should be interpreted with

caution, as prematurity and low birthweight are the largest risk factors for CP<sup>2,4</sup>. A study considered low birthweight and prematurity as mediators of the association between socioeconomic factors and CP<sup>24</sup>. Reported cases with typical gestational age and birthweight were considerably frequent, which may also indicate the occurrence of perinatal events. Surveys conducted in China and Korea with children born with 2,500 g and CP found similar results<sup>13,25</sup>, suggesting that injuries occurred during childbirth strongly contribute to this profile.

Data on household income indicates that most of our study population (nearly 60%) has a daily per capita income of two dollars, consistent with extreme poverty according to the World Bank<sup>26</sup>. A study conducted in Taiwan found family income to be strongly associated with the prevalence of CP<sup>27</sup>. Other studies conducted in high-income countries reported an association between socioeconomic gradients and CP<sup>16,17,28</sup>. The evidence gathered for low-income populations demonstrate that CP prevention must implicate, above all, coping with poverty and social inequalities. CP is the most common cause of physical disability in childhood and entails high costs for both families and health systems<sup>29</sup>. In this sense, confronting poverty and social differences is also to confront social determinants of health and one of the causes of the disease.

A study based on two cohorts conducted in Denmark and Norway verified a strong downward trend in CP incidence as guardians' education level increased, given that this factor may potentially influence lifestyle and health behaviors<sup>30</sup>. Most guardians who participated in our study have years of schooling equivalent to secondary education, which presupposes that they are capable of seeking care and adopting behaviors that do not harm the health of pregnant women. However, other socioeconomic characteristics suggest less social opportunities, interfering with the prevalence of CP.

Most guardians within our study sample were either unemployed or homemaker, exposing how women and families become economically vulnerable due to CP. The functional difficulties associated with physical disabilities and comorbidities demand an excessive load of housework, preventing caregivers from searching for paid jobs.

Our study has some limitations inherent to the cross-sectional design, such as the possibility of memory bias and difficulties in determining statistically how variables interact for the desired outcome. Yet, these difficulties were mitigated by the questionnaire quality and the sample size, as we sought to assess as many people with CP living in

Aracaju as possible, so that the study data may suggest relevant associations despite their descriptive nature.

This research advantages are related to its population-based design, which enabled us to cover the entire territory of the city and assess not only people who attend outpatient rehabilitation services. Our results provide key information for formulating, implementing, and evaluating public policies, besides serving as a basis for comparisons with studies conducted in other countries and with historical series from other places in Brazil.

## CONCLUSION

Cerebral palsy (CP) was less prevalent within our study population when compared to other countries. Epilepsy was the most common reported comorbidity, followed by intellectual disability. Most children and adolescents with CP stem from vulnerable social minoritized groups, are of mixed-race or Black, and their families live in extreme poverty. Our study contribute to monitoring future national research, and comparing them with data from national and international studies.

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