Spatial analysis of cerebral palsy in children and adolescents and its association with health vulnerability

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Abstract

Cerebral Palsy (CP) is commonly associated with low socioeconomic status. Use of spatial statistics and a Geographic Information Systems (GIS) are scarce and may contribute to the understanding of CP in a social context. To that end a spatial analysis of CP in children and adolescents was performed to analyze the association of CP with levels of vulnerability in a city (Aracaju, Sergipe) in north-eastern Brazil. In addition, an ecological study was conducted with data obtained from a population-based survey and secondary data. Exploratory spatial data analysis and linear regression were used. A total of 288 CP cases were identified, with a prevalence of 1.65/1,000 and differences among city neighbourhoods ranging from 0-4/1,000. The mean age of cases studied was 9 years 1 month, with a standard deviation of 5 years 2 months. Most study subjects with cerebral palsy (163) were male (56.4%). The distribution of CP in the study population was not homogeneous throughout the territory. Some areas had clusters, with more cases associated with areas of high vulnerability. Spatial data analysis using GIS was useful to gain an epidemiological understanding of CP distribution that can guide decision-making with respect to production, distribution, and regulation of health goods as well as services at the local level.

Introduction

The epidemiological characteristics of Cerebral Palsy (CP) have been studied using different perspectives and methods. This is due to the impact of CP on patient health, as it is the main cause of physical disability and permanent sequelae in children. However, many aspects of CP are not completely known and deserve further clarification (Graham *et al.*, 2016). The worldwide prevalence of CP is 2-3/1,000 live births and premature birth and low birth weight are among the main causes (McIntyre *et al.*, 2013; Oskoui *et al.*, 2013; Colver *et al.*, 2014). Researchers report that the association between lower Socio-Economic Status (SES) and low birth weight may increase CP risk (Solaski *et al.*, 2014). Another hypothesis is that postnatal lesions are more often associated with socioeconomic deprivation than with other factors (Dolk *et al.*, 2010). The use of composite indicators of deprivation or social disadvantage has been a methodological alternative for measuring SES (Dolk *et al.*, 2010; Oskoui *et al.*, 2016).

Geospatial analyses are commonly used for analyzing communicable diseases (Kirby *et al.*, 2017), with increasing use for non-communicable diseases and other health conditions such as pregnancy. Studies describing patterns of spatial distribution for congenital disabilities have aimed at understanding events, identifying risk areas, and formulating policies for prevention and care of disease in priority areas (Chi *et al.*, 2008; de Miranda *et al.*, 2014; Liao *et al.*, 2016). The use of spatial statistics and a geographical information systems (GIS) approach may contribute to the understanding of CP in a social context. This study aimed to perform a spatial analysis of CP in children and adolescents and to correlate the presence of CP with levels of health vulnerability in north-eastern Brazil.

Materials and Methods

Study Area

This ecological study used data obtained from a population-based survey between 2016 and 2017 in Aracaju, capital of the State of Sergipe in north-eastern Brazil. Aracaju has 39 neighbourhoods, 764 census tracts and no rural areas and occupies an area of 181.90 km² with an estimated population of 623,766 inhabitants and a population density of 3140.70 inhabitants/km² according to data from the Brazilian Institute of Geography and Statistics (IBGE) (IBGE, 2018). Primary health care services cover 93% of the population, with 44 healthcare units and 144 primary care teams.

Data collection

The data were collected between June 2016 and December 2017. The researchers provided training for key informants from the primary care health teams to identify addresses of children and adolescents with CP linked to each health service. After the training, the researchers performed house visits collecting data by applying a structured questionnaire developed by the researchers. The survey was also conducted in the only specialized public rehabilitation service, in order to reach the largest number of participants. Government data from the education and health sectors were also used. Data on children and adolescents with CP, mothers' names and dates of birth were cross-checked to avoid duplication. The flowchart in Figure 1 shows the data production process.

The research team collected the data in the homes of people with CP using a structured questionnaire developed by the researchers. Children and adolescents (0-18 years old) with CP who were diagnosed prior to the time of collection (CID10: G80.0-G80.9) were included. Those who did not live in the municipality and cases that were also present in the specialized data were excluded.

Statistical analysis

The period prevalence of CP was calculated (Braun *et al.*, 2015). We proposed a sample of 1,000 children and adolescents living in each neighbourhood to calculate the prevalence rate. Data of the residents were obtained from the IBGE (IBGE, 2018).

The Health Vulnerability Index (HVI) was considered in the analyses as a compound variable. Based on a previous study conducted in Brazil (Drachler *et al.*, 2014), the HVI was developed as a synthetic index using Principal Component Analysis that groups 10 variables related to 3 dimensions (Demographic, Socioeconomic, and Infrastructural). HVI scores were reported for each neighbourhood on a scale ranging from low vulnerability (0) to high vulnerability (1). The variable dimensions were the following:

Demography

i) Percentage of children under 5 years of age in the population;
ii) Percentage of brown or black people.

Socioeconomy

iii) Percentage of households with per capita income of up to 1/2 minimum wage;
iv) Average income of heads of household;
v) Percentage of women heads of household with a monthly nominal income of up to 1/2 minimum wage;
vi) Percentage of non-literate persons aged 8-15 years;
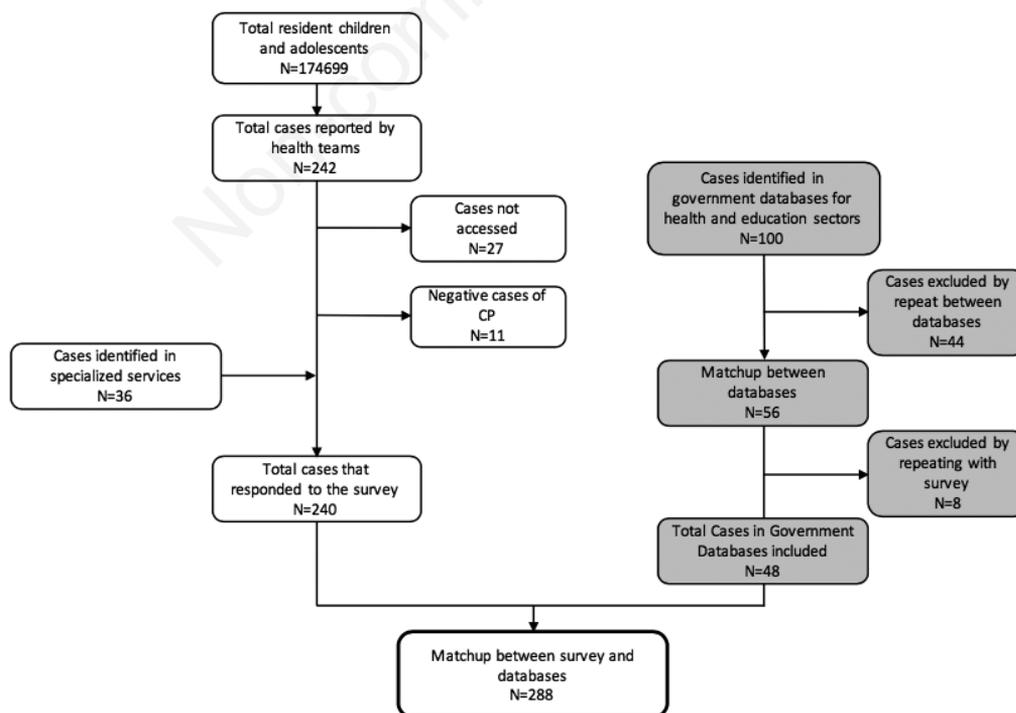
Figure 1. Flowchart of data production of children and adolescents with cerebral palsy in Aracaju, Sergipe, Brazil.

vii) Percentage of children living in households where none of the residents completed elementary education.

Infrastructure

viii) Percentage of households with water supply not connected to the distribution network;

ix) Percentage of households with inadequate sanitary sewage; 10. Percentage of households with no garbage collection service.

The Exploratory Analysis of Spatial Data (EASD) technique was used to describe spatial distributions (clusters or dispersions) in patterns of global and local spatial associations (Ministério da Saúde, 2007). An array of weights was created using the Queen Contiguity criterion among the 39 neighbourhoods analyzed by considering the neighbourhood of first order. The CP distribution by neighbourhood was determined using period prevalence rates and an empirical Bayesian model, whose estimator is a way of reducing the fluctuation associated with small samples. The weighting is performed using the average rate of neighbours (Anselin 2005).

The Global and Bivariate Local Moran's Index was used to identify the spatial correlation between CP and HVI, indicating the regions with characteristic patterns associated with geographical location. The Global Moran's Index may have positive values (between 0 and 1) indicating a direct correlation, negative values (between 0 and -1), indicating an inverse correlation, and a zero value indicating spatial independence (Anselin 2005). Moran's Local Bivariate analysis showed four types of spatial relationships (corresponding to the Moran scatter diagram) between the two variables proposed, considering a place unit and neighbouring units describing high prevalence of CP and high HVI as High-High (HH), high prevalence of CP and low HVI as High-Low (HL), low prevalence of CP and high HVI as Low-High (LH), and low prevalence of CP and low HVI as Low-Low (LL). Linear regression analysis was used to demonstrate the association between the prevalence of CP and the Health Vulnerability Score of neighbourhoods. Values with p<0.05 were considered significant.

The statistical analyses were carried out using QGIS software version 2.18.3 (Creative Commons Attribution ShareAlike 3.0 license CC BY-SA, Las Palmas, CA, USA) and GEODA software version 1.12 (Spatial Analysis Laboratory, University of Illinois, Urbana Champaign, USA). The cartographic base of the city of Aracaju was provided by IBGE (IBGE, 2018). The cartographic projection followed the SIRGAS 2000 geodetic reference system.

Results

Out of the total 174,699 children and adolescents living in the neighbourhoods under study, 288 with CP were included. The prevalence of CP was 1.65/1,000. The differences among city neighbourhoods varied from 0-4/1,000. The distribution of CP by neighbourhood according to gross prevalence rate (2A), and the prevalence rate reduced by the empirical Bayesian model (2B), is shown as choropleth maps (Figure 2). Visual examination of the maps showed a higher prevalence in the northern part of the city and lower prevalence in the central region. The mean age of the population with CP was 9 years, 1 month (SD = 5 years, 2 months). Table 1 categorizes the study population according to gender and age groups.

The results suggested that the areas of higher prevalence and

vulnerability are not randomly distributed. We observed a correlation between areas with a higher prevalence of CP and areas with higher HVI scores supported by the Global Bivariate Moran's test (I = 0.37, p = 0.001) revealed two HH cluster areas representing a higher prevalence of CP associated with a higher HVI score, one in the northern end of the city and another in the southern region (Figure 3). The neighbourhoods of the central region formed an LL cluster suggesting that the less vulnerable area had a lower prevalence of CP. An HL outlier was seen in the central region suggesting distancing from the neighbourhood. This was considered significant (p<0.05).

Linear regression analysis also suggested that increased HVI scores contributed to the increased CP prevalence (Figure 4). However, the coefficient of determination was low. There was no need to apply spatial regression techniques since the model met the normality assumptions as confirmed by the Jarque-Bera test (p = 0.51) and homoscedasticity as confirmed by the Breusch-Pagan (p = 0.11) and Koenker-Bassett (p = 0.07) tests. The Moran's Index and Lagrange multipliers for the residues were not significant (p>0.05), suggesting that there was no autocorrelation of regres-

Table 1. Distribution of children and adolescents with cerebral palsy according to gender and age.

Variable	No.	%
Sex		
Male	163	56.5
Female	125	53.5
Age (year)		
0 - 4	64	22.22
5 - 8	65	22.57
9 - 12	72	25.00
13 - 16	60	20.83
17 - 18	27	9.38
Total	**288**	**100.00**
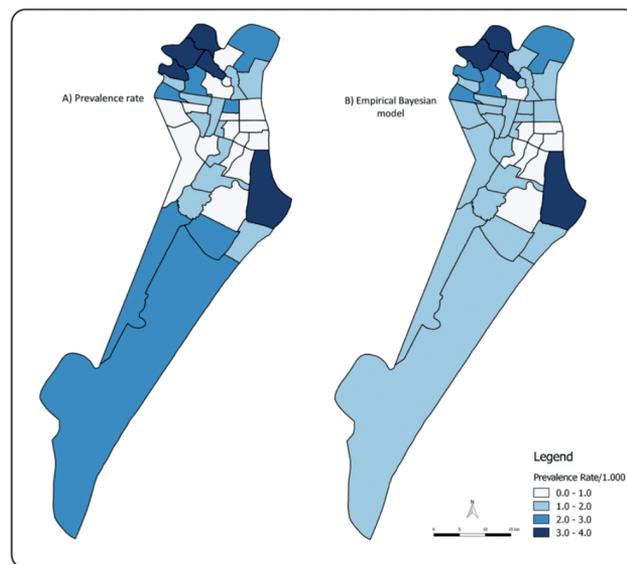
Figure 2. Maps representing the spatial distribution of cerebral palsy in Aracaju, Sergipe, Brazil, 2018 (A) and the gross prevalence rate per 1,000 (B). Prevalence rate reduced by the empirical Bayesian model.

sion residues among the neighbourhoods, indicating that the association in one neighbourhood occurred independently of the association in surrounding neighbourhoods.

Discussion

We found a lower CP prevalence rate (1.65/1,000) than in most other countries including high-income countries such as the USA (2.9/1,000) and in European countries (1.77/1,000) (Durkin *et al.*, 2016; Sellier *et al.*, 2016). A meta-analysis of the CP prevalence including 19 studies, with 16 surveys conducted in high-income countries, found an worldwide average CP prevalence of 2.11/1,000 live births (Oskoui *et al.*, 2013). Studies conducted in low and middle-income socioeconomic areas similar to Brazil also showed a higher CP prevalence. For example, a survey conducted in China, in which most of the participants were males, showed a prevalence of 2.8/1,000 children, and a population-based survey conducted in Uganda showed a prevalence of 2.9/1,000(Kakooza-Mwesige *et al.*, 2017; Tseng *et al.*, 2018). The low prevalence rate in the present study can be attributed to undiagnosed cases or a high mortality rate. It is unlikely that the emigration of patients with CP contributes to the prevalence rate since Aracaju City has more resources for treatment and rehabilitation than any other in the State. The differences within the city were relevant, since some neighbourhoods had a prevalence three to four times greater than others, revealing that the total prevalence rate is not a homogeneous indicator.

Exploratory analysis of the maps, spatial correlation tests and linear regression results confirmed that the spatial distribution of high vulnerability and that of CP were associated with spatial cluster formation, which shows that the prevalence of CP is associated with the contextual health inequities revealed by HVI. These results show similarity with two papers that found and association between CP and socioeconomic factors through composite indices of deprivation (Dolk *et al.*, 2006; Oskoui *et al.*, 2016). One of the studies, conducted in England, reported that the risk of post-neonatal CP was 86% higher in the poorest quintile of the population (Dolk *et al.*, 2010), while a Canadian study revealed that the use of the composite index improved the comparability of the findings and that socioeconomic contextual factors may imply greater risk and severity of CP (Oskoui *et al.*, 2016). This is supported by a study conducted in the north of England, aimed at reporting spatiotemporal grouping among CP cases, that found results consistent with exposure to maternal infections during the perinatal period, with such clusters found in densely populated areas (McNally and Colver, 2008).

Other studies associated health problems during gestation and low birth weight with areas of socioeconomic deprivation thereby demonstrating spatial dependence (Charreire and Combier, 2009; Chong *et al.*, 2013; de Miranda *et al.*, 2014; Insaf and Talbot, 2016). These investigations support the importance of this kind of research. Indeed, further investigation of certain spatial CP clusters might lead to a better understanding of other maternal and child health problems. The use of HVI was a relevant alternative to the traditional ways of measuring associations with socioeconomic status that focus only on income or education. HVI encompasses a set of demographics and socioeconomic and infrastructure variables that interact with one another and affect the daily life of people in complex ways.

Social vulnerability is associated with local inequalities,
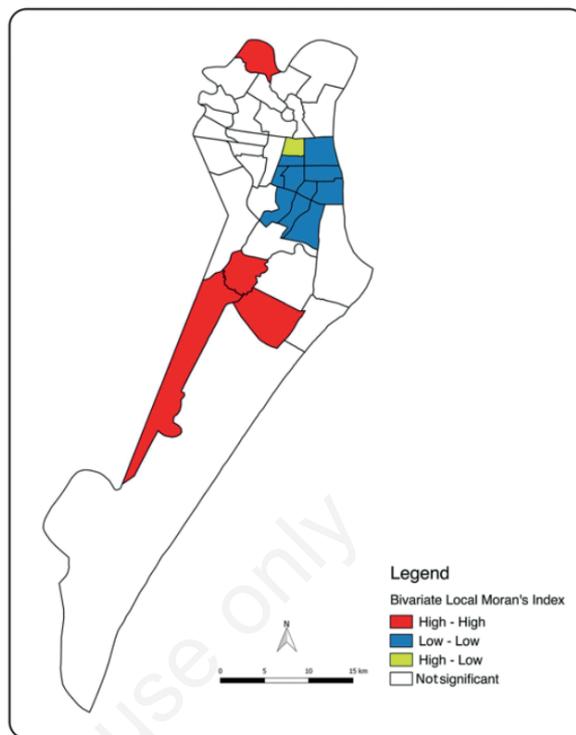
Figure 3. Prevalence of cerebral palsy and the Health Vulnerability Index based on bivariate Local Moran's Index by neighbourhood in Aracaju, Sergipe, Brazil.
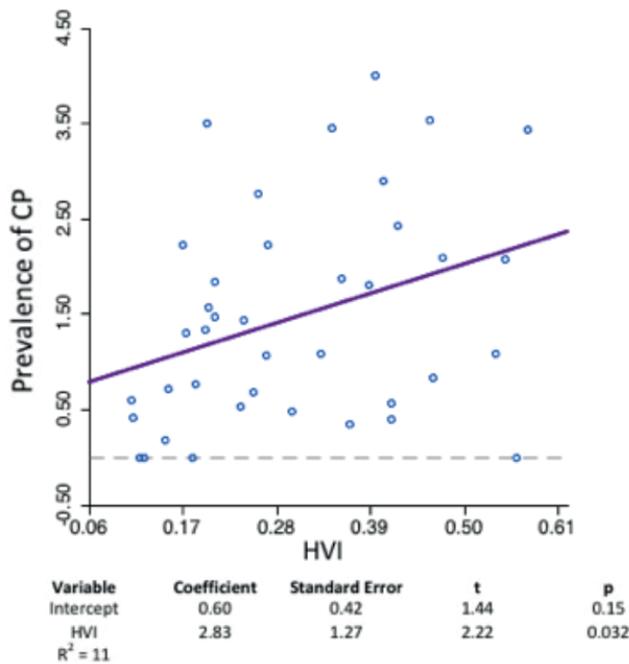
Figure 4. Prevalence rates of cerebral palsy and the Health Vulnerability Index based on linear regression scatter plot by neighbourhood in Aracaju, Sergipe, Brazil

including the characteristics of communities, the built environment, the level of urbanization, growth rates and economic vitality. These are social factors that influence or shape the susceptibility of various groups and their ability to respond (Cutter *et al.*, 2003). Socioeconomic aspects may influence the opportunities for education, housing, nutrition, health behaviour and healthcare. A study on health vulnerability states that human well-being is a result of the wider political, economic, social and ecological context and that structural inequities lead to disparities in individual health (Tallman, 2016). We must consider that inequities can lead to CP, just as CP can lead to inequities, since families may become impoverished as a result of the costs associated with this disability (WHO, 2011). Knowledge of the spatial distribution of health events has great relevance for cluster identification, formulation of hypothetical etiological factors, association with contextual environmental factors as well as issues related to socioeconomy and demography. According to Waldo Tobler's First Law of Geography (Tobler, 1970), "everything is related to everything else, but near things are more related than distant things."

Studies such as this can contribute to the formulation, implementation, and evaluation of interventions aimed at reducing CP's risk and impact factors. The effectiveness of interventions for the primary prevention of CP needs to be monitored in terms of reduction or exacerbation of socioeconomic inequalities (Dolk *et al.*, 2001). A systematic review of the economic aspects of CP states that prevention is the most cost-effective intervention measure (Shih *et al.*, 2018).

This study has some limitations. Despite being an association study, it did not specifically determine the interaction with risk factors or isolated aspects of individual exposure. However, one redeeming features of this study is that our sample representativeness was not restricted to participants who use rehabilitation services. It is known that the greatest impact in health caused by CP is felt by children who are not outpatients (Oskoui *et al.*, 2016). We also wish to emphasize that studies using spatial analysis as a tool to understand the epidemiology of CP are lacking.

Conclusion

Spatial data analysis using GIS is relevant to the epidemiological understanding of CP and can guide policymakers in decision-making for the production, distribution and regulation of health and services at the local level. One important finding is that the distribution of CP in the study population was not homogeneous throughout the territory as some CP clusters were associated with areas of high socioeconomic vulnerability.

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