

## Direct Costs of Parkinson's disease in Brazil: A case study

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Received: 09 Oct 2021,

Received in revised form: 30 Nov 2021,

Accepted: 08 Dec 2021,

Available online: 20 Dec 2021

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**Keywords—** Parkinson's disease, cost of illness, cost analysis, direct costs, health system

**Abstract—** The lack of studies on the costs of Parkinson's disease in Brazil makes it difficult to predict the costs of treatment, making it impossible to assess the economic impact on the individual's life and on health systems. The objective of this research is to estimate the average annual cost of treating Parkinson's Disease in users of the Public Health System in the city of Niterói (RJ), based on the variables Direct Cost. Methods: The research followed a mixed methods design, with qualitative and quantitative data, obtained through interviews with a sample of 54 patients and analyzed through non-parametric tests due to the rejection of the normality of the numerical data sets obtained. Results: Eleven direct cost variables were identified in the literature with the treatment of the disease, with 84.5% of the total cost concentrated in four of them (Drugs, Hospitalizations, Home Care and Auxiliary Treatments). There is a difference between patient costs and degree of illness for the public and private categories. Groups did not differ from each other when these costs were related to gender, race, education level and current employment. The mean total annual cost was estimated at R\$14,293.6 (US\$2,904.8) and calculated from the mean and 95% confidence interval obtained. Conclusion: The estimation of individual costs with the treatment proved to be of fundamental importance for understanding the total costs, and justifies the importance of studies in this area, both for patients and for managers involved in health care.

### I. INTRODUCTION

In recent years, the aging of the world population, the effects of urbanization and globalization, and the increased incidence of numerous diseases have contributed to the growth in patients with chronic conditions, such as neurological diseases<sup>1</sup>. The incidence of diseases is expected to increase as the population ages, once the aging process is a significant risk factor<sup>2</sup>. The

challenges of accurate diagnosis and delivery of health care profoundly impact individuals, their families and communities, the general health system, and governments<sup>3</sup>.

Parkinson's Disease (PD) stands out in this scenario, a chronic and degenerative neurological condition that mainly affects the elderly, causing important vital incapacities and reduced quality of life. This disease is the

second common neurodegenerative disease after Alzheimer's and the main burden on the patient, family, caregivers, and the healthcare system<sup>4,6</sup>.

Neurodegenerative diseases that are impossible to prevent, such as PD, are often associated with a significant burden borne by society, whether financial, social, or psychological<sup>7</sup>. As the disease progresses, patients face an increasing severity of symptoms, reflecting increased treatment costs<sup>8</sup>.

The financial impact of PD on society has been widely discussed in recent decades. Research has indicated a more significant effect in the current future, associated with an aging population<sup>9</sup>. However, the scarcity of studies on the cost of PD in Brazil makes it difficult to forecast and consequently assess the economic impact of PD on the individual's life and both in public and private health systems.

Determining the costs of disease makes it easier to understand its impact on society and health systems, assessing its degree of efficiency and understanding how the market tends to organize itself concerning specific values<sup>7</sup>. Therefore, based on the main Direct Cost variables, this case study aims to determine the average annual cost of PD treatment among patients who use the Public Health System in the city of Niterói (RJ, Brazil).

## II. MATERIALS AND METHODS

### *Participants*

A total of 59 patients were contacted between March and November 2020 during outpatient consultations at the Hospital Universitário Antônio Pedro (HUAP). Patients physically or cognitively incapable of answering the questionnaire ( $n = 5$ ) were excluded from the sample. All others consented to participate in the survey, resulting in a total of 54 interviews. Before participating in this study, participants signed a written consent after receiving a detailed explanation of all steps of the research protocol. According to the Declaration of Helsinki, all efforts were carried out and approved by the Research Ethics Committee of the Universidade Federal Fluminense (CAAE: nº 3.687.239.)

### *Study procedures*

The questionnaire, an instrument used for this research and consisting of 33 questions, was divided into three parts: 1) Introduction, with a presentation by the researchers and a brief contextualization of the theme and research objectives; 2) Screening, with questions that determine the profile of respondents; and 3) Identification of Treatment Costs, covering the variables: consultation, medication, auxiliary treatments,

hospitalization, home care, exams, transport, food, home adaptations, surgery, and special equipment. Such variables, identified in the literature on the subject, make up the Direct Costs with PD treatment. The Hoehn and Yahr Scale (HY – Degree of Disability Scale) was used to classify the stage of disease progression. In this modified format, the scale comprises seven steps of classification to assess the severity of PD and essentially encompasses global measures of signs and symptoms that allow classifying the individual as to the level of disability<sup>18</sup>. This was an observational, cross-sectional, retrospective study about disease prevalence that used a bottom-up approach (i.e., society's perspective). The time horizon used was six months. The study viewpoint was first carried out from the patient's perspective and then evaluated from the Public Health System prospect.

### *Cost calculation*

Costs were calculated for the period of 6-months before the interview and extrapolated to 1 year. The amounts are described in reais (\$) and US dollars (USD) using the Central Bank real-dollar quotation assessed on June 25, 2021. The research on the value of medical consultations and other cost variables that fall on the public health system was carried out in the database of the Ministry of Health of Brazil, known as the Sistema Único de Saúde (SUS; [www.datasus.gov.br](http://www.datasus.gov.br) and <http://sigtap.datasus.gov.br>). Access to the portal was carried out on January 16, 2021. At the research time, the average market price was estimated for patients who purchased medications in pharmacies or private drugstores. In cases where private therapists were hired, consultation fees were obtained directly from the professionals. For the other variables, the patient was asked immediately about the values practiced.

### *Statistical analysis*

The statistical description of the data was performed using tabular methods (statistical tables and summary tables of information). Absolute frequencies and proportions were used for categorical variables. For numerical variables, data were expressed as means accompanied by the standard deviation (as mean  $\pm$  SD). The normality of cost distribution, assessed by the Kolmogorov-Smirnov test, was rejected. Therefore, the Mann-Whitney U test was applied to independent samples, while the Wilcoxon signed-rank to paired samples. For more than two independent groups, the Kruskal-Wallis test was performed. The generalized linear model with gamma probability distribution and log link function were used to estimate the total annual cost. A significance level of 0.05 (5%) was adopted and the analyzes were conducted using R<sup>19</sup>.

### III. RESULTS

#### Descriptive analyses

Demographic and clinical characteristics of the participants are detailed in Table 1. Males represent 64.8% of the sample, 50% of patients had completed elementary school, 88.9% were retired, 48.1% of patients had an HY

score of 3 or 4 and 82.6% of patients had the disease for more than 5 years. The average monthly income of the sample was R\$ 4,460.4 (US\$ 906.52) and median R\$ 2,455.00 (US\$ 498.92).

Table.1: Patient characteristics

<b>Gender, n (%)</b>	
Male	35 (64.8)
<b>Race, n (%)</b>	
White	28 (51.9)
Black	5 (9.3)
Multiracial	21 (38.9)
<b>Age (years), mean <math>\pm</math> sd</b>	69.2 $\pm$ 10.7
<b>Education level, n (%)</b>	
Never attended school	1 (1.9)
Completed grade school	27 (50.0)
Completed middle school/high school	15 (27.8)
Completed college	7 (13.0)
Completed graduate school	4 (7.4)
<b>Current employment, n (%)</b>	
Retired	48 (88.9)
<b>Income (US\$), mean <math>\pm</math> sd</b>	906.52 $\pm$ 792.38
<b>Hoehn and Yahr scale, n (%)</b>	
1	3 (5.6)
1.5	2 (3.7)
2	10 (18.5)
2.5	5 (9.3)
3	14 (25.9)
4	12 (22.2)
5	8 (14.8)
<b>Disease duration, (years), n (%)</b>	
$\leq 5$	9 (16.7)
6–14	28 (51.9)
$\geq 15$	17 (31.4)

The distribution of the total biannual cost of PD treatment, considering the direct cost variables, is described in Table 2. Although composed of patients undergoing treatment in

the public health system, the study sample presents values associated with costs distributed in two categories: private and public. Note that medications, hospitalizations,

caregiver services and secondary treatments account for 86.4% of the total costs. When analyzing the cost categories, it is possible to observe that, in private system,

the main expense is medications (37.3%). On the other hand, hospitalizations in the public health system stand out with 45.5% of total costs.

Table. 2: Distribution of the 6-months costs of PD treatment

Costs US\$ (%)	Categorie		Total
	Private	Public	
<b>Direct medical costs</b>			
Medical visits	0.0 (0.0)	1,100.9 (2.2)	1,100.9 (1.1)
Medications	17,987.2 (37.3)	20,452.4 (41.7)	38,439.6 (39.6)
Secondary treatments (*)	8,803.8 (17.9)	840.1 (1.7)	9,643.9 (9.7)
Surgery	0.0 (0.0)	3,707.2 (7.6)	3,707.2 (3.8)
Hospitalizations	0.0 (0.0)	22,346.7 (45.5)	22,346.7 (23)
Caregiver services	13,656.8 (28.4)	0.0 (0.0)	13,656.8 (14.1)
Medical exams	0.0 (0.0)	621.7 (1.3)	621.7 (0.6)
<b>Direct nonmedical costs</b>			
Transport	776.4 (1.6)	0.0 (0.0)	776.4 (0.8)
Food	3,901.9 (8.1)	0.0 (0.0)	3,901.9 (4.0)
Home adaptations	2,040.4 (4.2)	0.0 (0.0)	2,040.4 (2.1)
Equipment to facilitate locomotion	1,225.2 (2.5)	0.0 (0.0)	1,225.2 (1.3)
<b>Total</b>	<b>48,391.7 (49.6)</b>	<b>49,069.0 (50.4)</b>	<b>97,460.7 (100)</b>

Essential as a non-pharmacological therapy, secondary treatments (Physiotherapy, Speech Therapy, Occupational Therapy, Psychologist, Nutritionist and Physical Activity) had a high number of prescriptions among the investigated patients (79.6%). Nevertheless, 41.9% of patients indicated for these treatments did not undergo them due to difficulties scheduling these therapies in the public health system. Among the treatments most frequently prescribed, physiotherapy and physical activity stand out.

The variables medical visits and exams, vital for the treatment and monitoring of the evolution of PD, do not have a large representation in the total cost of treatment (1.1% and 0.6%, respectively). These variables had their costs added to the public health system, and this fact is justified due to the profile of the sample.

Other variables, such as home adaptations, equipment to facilitate locomotion and transport, presented interesting biases that hampered the calculation of their respective average costs, and this fact certainly underestimated the

total cost estimate with these variables.

#### *Inferential analyses*

The Figure 1 shows the distribution of the 6-months costs of degree of illness (HY scale) for both categories, private and public (SUS), as well as the *p values* associated with the hypothesis tests to verify whether there is a difference in the median costs between the groups.

Clearly, there is a difference between patient costs and degree of illness for the two categories (private -  $p = 0.021$ ; public -  $p = 0.023$ ). In private, there is a difference between the groups mild and severe stages (Figure 1A). Furthermore, there is no difference between the costs of each category in relation to each of the stages (Figure 1B).

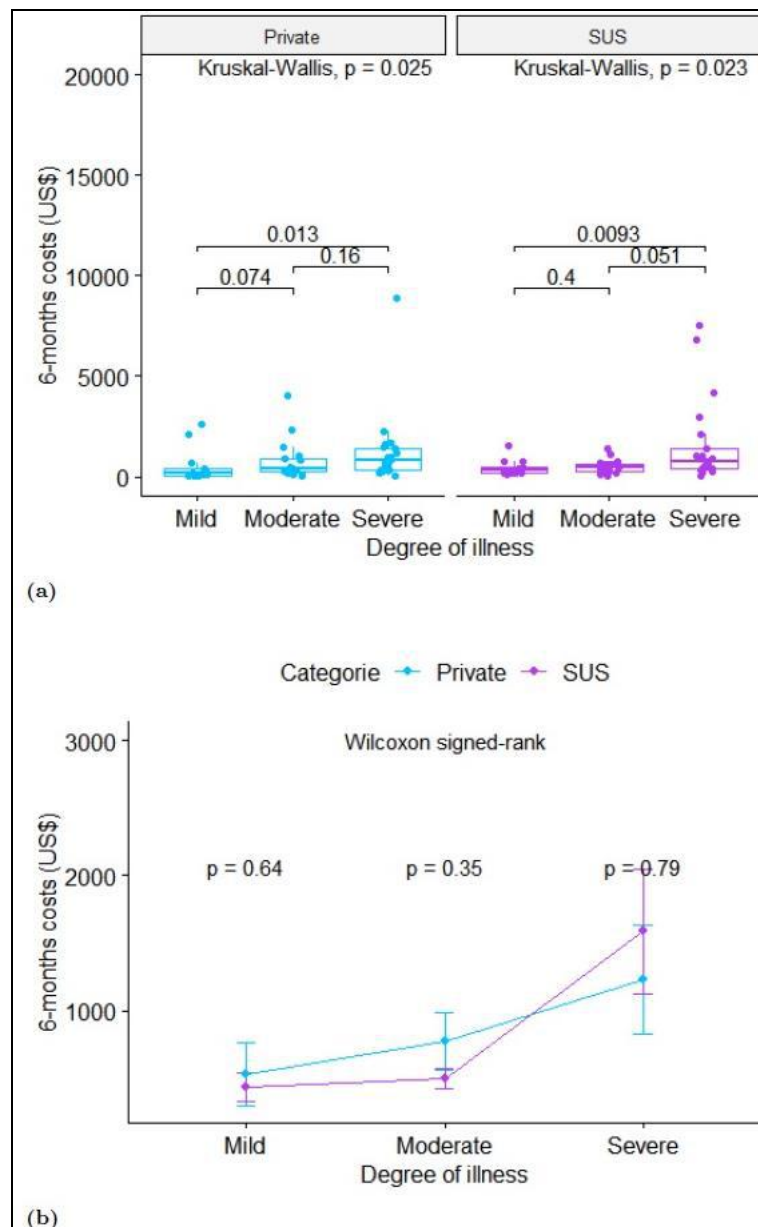


Fig.1: Comparison between degree of illness and 6-months costs.

(A) Comparison within each of the categories. (B) Comparison between categories.

Groups did not otherwise differ from each other when these costs were related with gender ( $p = 0.08$ ), race ( $p = 0.4$ ), education level ( $p = 0.68$ ) and current employment ( $p = 0.49$ ). Age was significant ( $p = 0.02$ ).

Table 3 shows the inferential analyses of the total annual cost of PD obtained by the generalized linear

model. A step-by-step variable selection process that searched for the variable combination that best explains the total cost was applied resulting in following variables: medical visits, transport, caregiver services, hospitalizations, surgery, secondary treatments, medical exams, stage of disease progression and age.



Table.3: Estimated means and 95% CIs for the total annual cost of Parkinson's disease (US\$)

Categories	Mean US\$ (95% CI)	Total Cost (%)
Private	1,532.1 (1,087.1- 1,977.0)	82,734.7 (52.8)
SUS	1,372.7 (958.5 -1,786.9)	74,127.8 (47.2)
<b>Total</b>	<b>2,904.8 (2,215.5 - 3,594.1)</b>	<b>156,862.5</b>

These data revealed an average annual cost of R\$14.293,6 (US\$2,904.8) per patient ranged from R\$10.901,9 (US\$ 2,215.5) to R\$17.685,4 (US\$3,594.1). Furthermore, the total costs associated with both categories are similar. There is no statistically significant difference between them ( $p = 0.52$ ).

#### IV. DISCUSSION

The cost analysis study carried out in this research is initially presented from the patient's perspective. The data collected in the quantitative phase was carried out through interviews with patients with PD who undergo treatment in the public health system. Using a retrospective time horizon of 6 months through the bottom-up valuation method and considering the prevalence and direct cost variables, an annual cost forecast for PD treatment was made.

Given the profile of the sample, it was expected that some variables, especially those related to direct medical cost, would concentrate their spending on public health system. In this research, we had 100% of the costs borne by public system in the variables medical visits, surgery, hospitalization and medical exams. This trend should not be extrapolated due to the size and characteristics of the sample. Still, they are an essential feature of this research and show an essential contribution in treating PD considering the adopted cost category (direct costs).

According to this scenario, it is possible to identify that 45.5% of the Direct Cost variables identified in the literature review and adopted in this study have their expenses (or part of them) added to the public health system. This represents a significant contribution to the patient's budget, given the socioeconomic characteristics of the studied sample.

Direct Costs comprise the total cost in health economic evaluation studies, being only part of this cost. Even so, its estimate is of fundamental importance for understanding total costs and justifies the importance of studies in this area, both for patients and for managers involved in health care.

In this case study, it was possible to diagnose that the allocation of public resources includes the main direct cost variables. Without estimating costs with the treatment of diseases, including PD, it is much more

challenging to decide on allocating resources for the treatment. In the study environment covered by this research, for example, it was possible to observe a high cost of medicines and hospitalizations subsidized by the public health system, which corroborates the data collected in the literature review. Cost studies with the treatment of chronic diseases, such as PD, are increasingly needed as a tool to support decision-making, aiming at the sustainability of health systems.

The estimated average annual cost of PD treatment revealed that 52.8% of these costs fall on the patient and 47.2% fall on the public health system. However, the non-parametric Wilcoxon test revealed that this difference does not have statistical significance.

In a study carried out in Australia, to estimate the annual cost of PD treatment from the perspectives of the family, the health system and society, Bohingamu Mudiyanse et al.<sup>6</sup> identified that two-thirds of the costs for the health system were related to Hospitalizations, with medical and pharmacological services contributing significantly to total costs. The study included 87 patients, assisted by the public and private health systems in that country. In another study, carried out in Singapore by Zhao et al.<sup>17</sup>, the authors identified pharmacotherapy and home care as the main components of the total cost. The sample consisted of 195 patients treated at the Center for Movement Disorders of the National Institute of Neurosciences in that country.

Medicines are the most frequently accumulated direct cost variable. Its expressive cost, which in this research represents 39.6% of the total direct cost, was also observed by Bovolenta et al.<sup>7</sup> in a study of a similar nature, carried out with patients also assisted by the public health system in the city of São Paulo. In this study, the authors identified that antiparkinsonian drugs accounted for 25% of the total cost (both drugs provided by the public health system and those purchased by patients) and 97% of direct medical costs. In a review study, Reese et al.<sup>13</sup> found that drug therapy accounted for 15% - 80% of total direct costs.

Hospitalizations have a high cost attributed to SUS due to the profile of the sample, composed of patients who undergo treatment for the disease in the public health system. No patient interviewed reported a case of admission to a private hospital. The high cost of admissions has already been identified in previous studies

on the cost of treating PD. However, the causes for such hospitalizations differ from the results found in this research.

A systematic review carried out by Koay et al.<sup>12</sup> pointed out as the main reasons for acute hospital admission in patients with PD falls, deterioration of motor and non-motor symptoms of the disease, cardiovascular events and infections, especially urinary tract infections and respiratory. The incidence of falls is also cited by Fundament et al.<sup>11</sup> and by Fletcher et al.<sup>10</sup> as being primarily responsible for cases of hospitalization among patients with PD.

In this research, the leading cause of hospitalizations cited by patients was surgeries performed to treat PD. Although only 11 patients underwent surgical interventions, some had postoperative complications and needed to extend the length of hospital stay, which significantly increased the costs with this variable. Besides that, 31.5% of the patients suffered some falls, but none resulted in hospitalization or need for medical care.

The high cost of formal home care, as demonstrated in the literature<sup>4,6,7</sup>, falls directly on the patient since the public health system does not offer this type of service. Although reported by only five patients (9.3% of the sample), it is the third variable with the highest accumulated cost distribution identified in this research. The association between age and stage of disease progression had already been identified by Dodel et al.<sup>16</sup> and von Campenhausen et al.<sup>15</sup> and Martinez-Martín et al.<sup>14</sup> in previous studies, where the authors identified an increase in costs directly proportional to the degree severity of PD.

## V. CONCLUSION

Considering this scenario, it is possible to identify that 45.5% of the Direct Cost variables identified in the literature review and adopted in this study have their expenses (or part of them) added to the public health system. This represents a significant contribution to the patient's budget, given the socioeconomic characteristics of the studied sample. Direct Costs make up the total cost in health economic evaluation studies, being only part of this cost. Even so, its estimate is of fundamental importance for understanding total costs, and justifies the importance of studies in this area, both for patients and for managers involved in health care. In this case study, it was possible to diagnose that the allocation of resources in the public health system includes the main direct cost variables. Without estimating costs with the treatment of diseases, including PD, it is difficult to decide on the allocation of resources for treatment.

Cost studies with the treatment of chronic diseases, such as PD, are increasingly needed as a tool to support decision-making, aiming at the sustainability of health systems. As proposals for further research, the replication of this study in a larger sample is suggested, with the possibility of generalizing the results. The possibility of comparing this cost analysis between patients treated in the public and private sectors is warranted. An analysis of the economic impacts of treatment, through a study of the costs of the same or similar nature, would also be interesting in the context of supplementary health.

## ACKNOWLEDGEMENT

"This study was financed in part by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior - Brasil (CAPES) - Finance Code 001"

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