

Economic And Humanistic Burden Of Visual Impairment And Blindness In Brazil: A Grey Literature Review

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Introduction

- Inherited retinal dystrophies (IRDs) are a group of diseases that lead to visual impairment (VI) or blindness¹.
- While studies on socio-economic costs of VI or blindness exist, the specific humanistic and economic burden resulting from IRDs has been largely overlooked.
- A grey literature review was conducted to understand and evaluate the humanistic and economic burden associated with VI from IRDs in Brazil that was not captured in peer-reviewed literature.
- The true extent of this burden is not fully quantified, due to the lack of available data from the private health insurance sector.

Methods

- A review of grey literature published between 2014 and 2019 was conducted by using a series of targeted keywords (IRD, patient, low vision, direct cost, productivity loss, etc.). The relevant data sources were conference abstracts, government publications, dissertations/theses, guidelines, policies and procedures.
- Where available, reference lists and cited articles within publications were also reviewed to identify further sources of interest.
- Since abstracts were often unavailable in grey literature documents, items' title, executive summary, or table of contents (whichever was available) were screened according to predetermined inclusion and exclusion criteria by native language reviewers **Table 1**.
- Relevant articles were selected, and the quality of each article was assessed using an evidence-grading system.

Table 1. Inclusion and exclusion criteria

	Inclusion criteria	Exclusion criteria
Population	Patients or caregiver of a patient with a clinical diagnosis of RP, LCA or other IRD, irrespective of any genetic mutation.	Patients (or caregivers) with diseases other than those described in the inclusion criteria
Outcomes	Economic burden of illness to affected patient OR caregiver including direct costs, indirect costs and intangible costs. Humanistic burden of disease to affected patient OR caregiver including the impact on quality of life, the impact on activities of daily living, treatment satisfaction or adherence.	Outcomes other than those described in the inclusion criteria
Publication date	Published 2014-2019	Published prior to 2014
Country: language	Brazil and Portuguese or English	Country and languages other than those described in the inclusion criteria

Results

Healthcare system and VI burden in Brazil

- Despite Brazil's public healthcare service, the Sistema Único de Saude (SUS) being free at the point of use, approximately 22% of residents pay for private health insurance²⁻³ **Figure 1**.
- The Brazilian Institute of Geography and Statistics (IBGE) reported that more than 6.5 million people had VI regardless of etiology in 2010⁴.
- Approximately, 3.4% of the local population per region had some form of VI, with the highest percentage in the north and north-eastern regions⁴ **Table 2**.
- Approximately, 55% of the population had not undergone genetic testing regarding their IRDs⁵.
- The Brazilian government has several national policies to protect the rights of those with disabilities, including VI⁶⁻⁹.

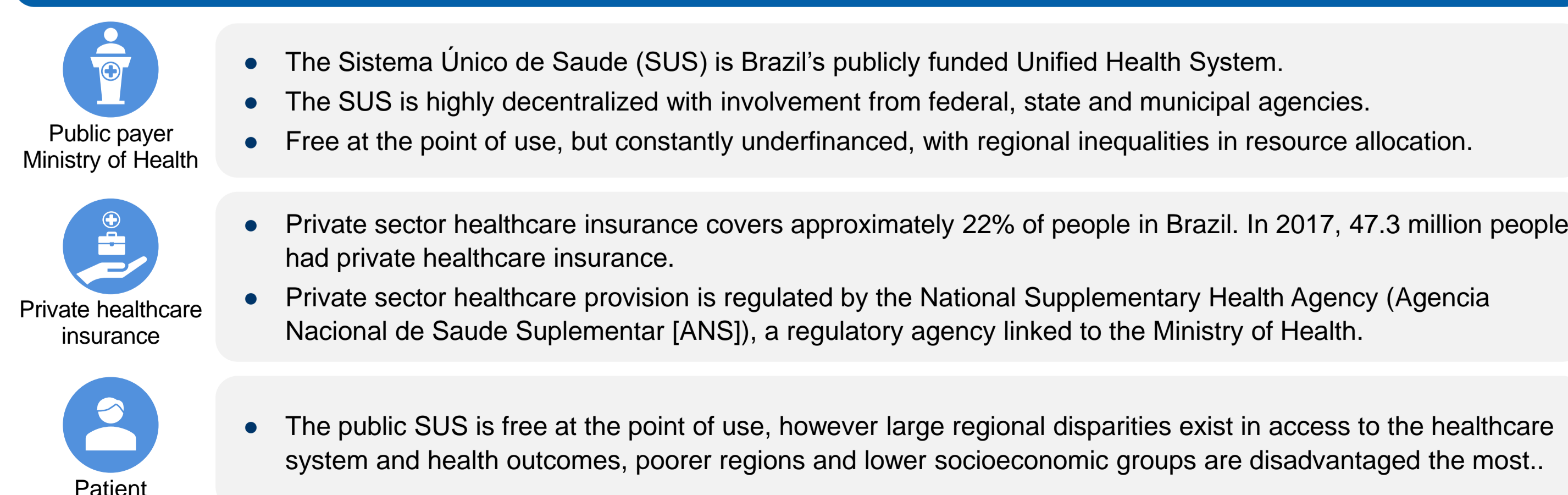
Economic burden of IRDs

- The Sistema de Informações Hospitalares do Sistema de Saude (SIH-SUS) hospital admissions database between 2014 and 2018, showed that the average cost for IRDs per adult patient was R\$ 2,785.73 per year¹⁰ **Table 3**.
- According to the SIH-SUS medical procedures to manage adverse events or comorbidities (eye inflammation, high intraocular pressure or cataracts), was calculated to cost R\$ 201 per patient¹⁰.
- However, the burden of VI and blindness to private health insurers and patient out-of-pocket expenditures was not documented in grey literature.
- The requirement for caregiver help increases with decreasing visual acuity, ranging between 18.4-94.1 hours per week¹¹.
- Indirect costs due to loss of productivity in adults with VI was estimated at R\$ 31,833.50 for 2017¹⁰ **Table 4**.
- Among the adult population (aged 18 to 65 years), social security (i.e., disability retirement) was estimated to be R\$ 11,976 per annum and loss of caregiver productivity was R\$ 680 per annum¹⁰ **Table 4**.
- The indirect economic impact on patients and caregivers with RP, LCA or other IRDs, including cessation of work or reduction of productivity caused by morbidity and mortality, was not documented in the grey literature.

Humanistic burden of IRDs

- The humanistic burden of IRDs has been described in a series of case reports, demonstrating the wide-reaching impact on patient's health-related quality of life (HRQoL), self-esteem and ability to find employment **Figure 2**.
- One study highlighted the lack of widely accepted and validated patient-reported outcomes (PROs) for measuring the impact of IRDs on HRQoL in Brazil¹².

In Brazil, there is a national universal healthcare system provided by the government however, this service is reported to be underfunded



ANS, Agencia Nacional de Saude Suplementar; SUS, Sistema Único de Saude. **References:** 2-3

Figure 1. Brazil's healthcare system and its key stakeholders

Table 2. Number of people with VI per region in Brazil

People with VI by region	Total (2010)	Population, %
North	574,823	3.6
Northeast	2,192,455	4.1
Southeast	2,508,587	3.1
South	866,086	3.2
Midwest	443,357	3.2
Total	6,585,308	3.4

VI, visual impairment

Table 3. Annual medical resource and assistive device costs

Resource	Cost (BRL, R\$)	Cost (USD, \$)*
Devices	204	47
Visual rehabilitation / occupational therapy	13	3
Outpatient care	48	11
Ophthalmological consultations	10	2
Hospital care	2,786	642

*Conversion rate of 1 BRL = 0.23043 USD, 2018. BRL, Brazilian Real; USD, United States Dollar; **References:** 10.

Table 4. Annual non-medical resource use by age group

Resource	Cost (BRL, R\$)	Cost (USD, \$)*
Loss of caregiver productivity	680	206
Social security	11,976	3620
Lost productivity	31,834	9623

*Conversion rate of 1 BRL = 0.23043 USD, 2018. BRL, Brazilian Real; USD, United States Dollar; **References:** 10

*Methodology: Patient sentiments were gathered from online media reports. Collecting patient perspectives in this way provides powerful insight into the patient voice; however, insights must be interpreted with caution as they represent a singular view at a cross-section in time.

RP, retinitis pigmentosa; VI, visual impairment. **References:** 13-14

Figure 2. Patient perspectives on VI*

Study limitations

- This grey literature review unveils important insights into the burden of VI, although must be interpreted with caution as a singular view is presented at a cross-section in time.
- Due to the lack of available data from the private health insurance sector, it must be acknowledged there is uncertainty surrounding the results and analytical insights obtained, therefore must be interpreted with caution.

Conclusions

- Approximately 3.4% of the Brazilian population have some form of VI.
- The humanistic burden of RP, LCA and other IRDs was described in a series of case reports demonstrating the wide-reaching impact on patient's quality of life, self-esteem and ability to find employment.
- However, due to the decentralized healthcare system in Brazil, the burden of VI and blindness to private health insurers was not quantified due to limited availability of data in grey literature.
- Few sources were identified describing the economic or humanistic burden explicitly associated with RP, LCA and other IRDs, therefore additional research focusing on these disease areas is warranted.
- Further research is needed to have a holistic view of burden from the health system and societal perspective.

Abbreviations

ANS, Agencia Nacional de Saude Suplementar; BRL, Brazilian Real; HRQoL, health-related quality of life; IRDs, inherited retinal dystrophies; LCA, leber congenital amaurosis; PROs, patient-reported outcomes; RP, retinitis pigmentosa; SUS, Sistema Único de Saude; VI, visual impairment; USD, United States Dollar

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