Economic burden of retinitis pigmentosa and visual impairment in France: a Systematic Literature Review

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INTRODUCTION

Visual impairment is described as an insufficiency or a loss of image perceived by the eye, which can range from low vision to absolute blindness in one or both eyes.(1) In France, 207,000 people are considered blind or profoundly visually impaired.(2) Retinitis pigmentosa (RP) is a group of inherited genetic retinal diseases that can cause significant vision loss and potentially blindness.(3) The progressive genetic retinal disease is characterized by the degeneration of cone and rod photoreceptor cells and affects approximately 30,000 people in France.(2)

OBJECTIVE

The objective of this study was to conduct a systematic literature review of published papers on the cost and resource use associated with visual impairment, blindness, and more specifically RP in France. This project addresses the need to make an inventory of resource use and cost data (direct and indirect costs) available in RP.

METHODS

A systemic literature review aiming to assess the economic burden of RP and visual impairment in France was conducted. EMBASE and PubMed-Medline were searched from January 2000 to November 17th, 2022. Other relevant sources including congress proceedings were also searched for the 2019–2022 period. As a rare disease leading to visual impairment, paucity of evidence available on RP in France was expected. Therefore, the research was extended to any chronic ocular pathology associated with visual impairment.
Inclusion criteria are shown in **Table 1**

Table 1. Literature criteria based on PICOS principles

PICOS	Description
Population	Given the lack of literature for X-linked retinitis pigmentosa patients, we included French patients with visual impairment, excluding acute illnesses (e.g., acute uveitis, acute sight loss). Emphasis was placed on articles dealing specifically with X-linked retinitis pigmentosa.
Intervention	No restriction
Comparison	No restriction

Outcomes	• Resource consumption (hospitalizations, medical visits, etc.): type of resource consumed, frequency of occurrence, duration.
	• Associated costs: cost item, unit, and total costs, related to the care of patient and his entourage (societal perspective)
	  • Direct costs (treatment costs, diagnostic costs, etc.)
	  • Indirect costs (costs of carers, disabled allowances, etc.)

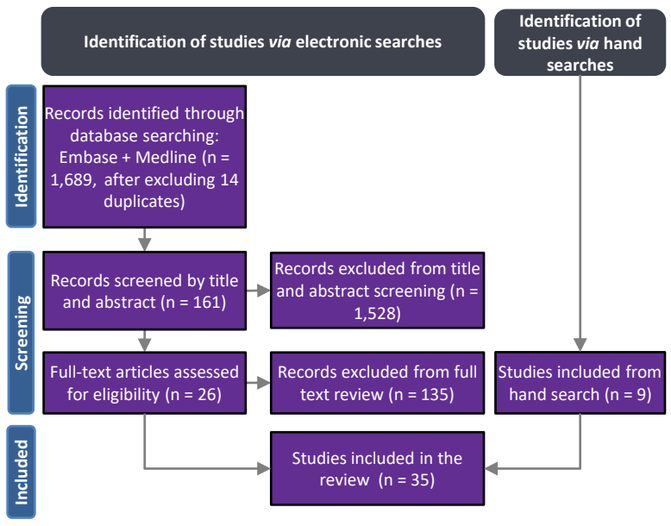
Studies	Economic evaluations (medico-economic analysis, cost-benefit analysis, cost minimization analysis, budget impact analysis); observational studies; clinical recommendations, systematic literature review.
Languages	English and French.
Time restriction	Articles published in 2000 and after.
Country	France.

- Two reviewers independently assessed the eligibility of identified citations via title/abstract and full-text screening using predefined selection criteria.
- Two reviewers independently extracted data from the included studies

RESULTS

Thirty-five articles of interest were identified, as shown in the flow diagram for study selection (**Figure 1).**

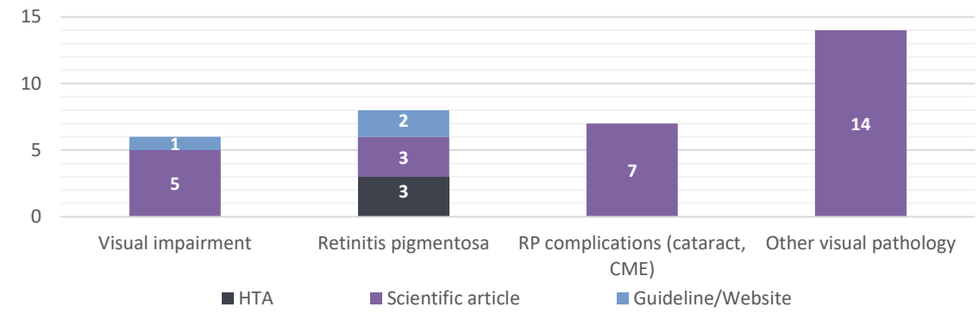
Figure 1. PRISMA Flow Diagram for Study Selection



Abbreviations: PRISMA: Preferred Reporting Items for Systemic Reviews and Meta-Analysis

The 35 considered papers were published between 2003 and 2022. The distribution of considered publications is displayed in **Figure 2.** Eight publications (~ 23%) focused on RP and furnished cost, resources use and employment data. Most reported outcomes were non-medical costs related to visual diseases. Six publications focused on cataract, which is one of the complications of RP. Pungor et al.(2022) was the unique paper that studied X-linked retinitis pigmentosa (XLRP) outcomes (10).

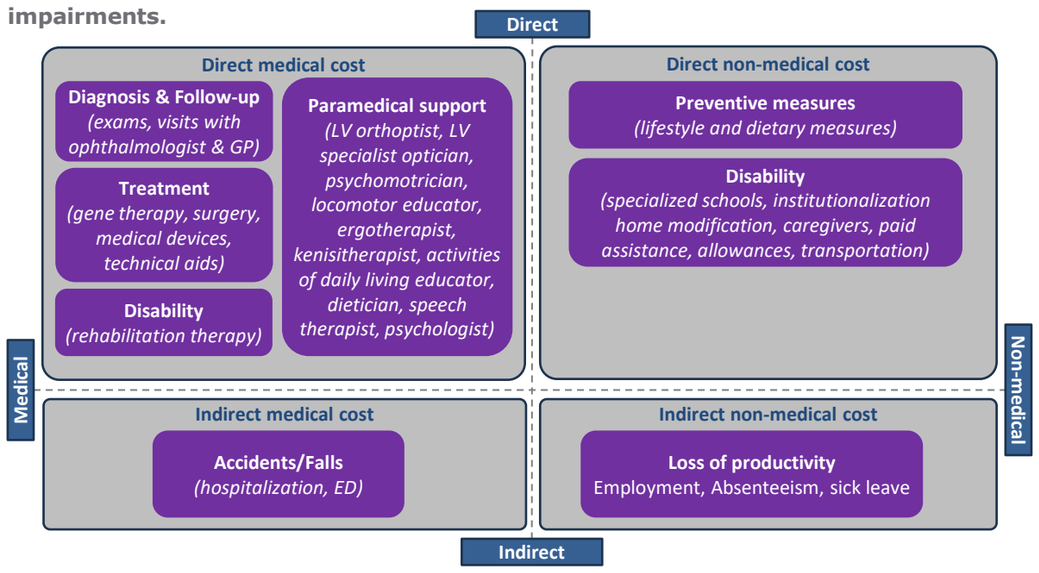
Figure 2. Distribution of publications by disease and category



Abbreviations: CME: Cystoid macular edema; HTA: Health technology assessment RP: Retinitis pigmentosa

Most identified studies have tended to focus on isolated cost components related to visual impairment, often failing to provide a comprehensive assessment of its overall economic impact. To illustrate, merely 13 publications (constituting 37% of the total) have undertaken a holistic analysis that considers both the medical and non-medical cost aspects associated with visual impairment. Among these, three publications exclusively examined the cost of transportation as part of non-medical costs. However, using inputs from all publications, a comprehensive list of cost components associated with vision impairment is presented in **Figure 3.** The main categories of costs associated with visual impairment identified in the review include medical costs (e.g., usual ones such as visits, exams and treatment but also paramedical support and technical aids) and non-medical costs mainly associated with disability (e.g., technical assistance, caregiver, home assistance, allowance, and loss of productivity). Non-medical cost accounted for most of the direct cost of visual impairment, especially in diseases such as RP, where few or no innovative treatments are available. (4-7). In the context of AMD, the direct non-medical expenses stands at 25.9%, 40.6%, and 49% in the studies conducted by Bandello, Cruess, and Bonastre, respectively. These studies involved the administration of costly medications such as verteporfin, which incurred annual expenses of 2080€ in Bandello and a unit cost of 1341€ in Cruess. (12-14) Conversely, in the studies conducted by Nordman and Thygesen, where expensive drugs were not factored in, direct non-medical costs represent a more substantial share of total direct costs at 73.8% and 83.7%, respectively. (15,16)

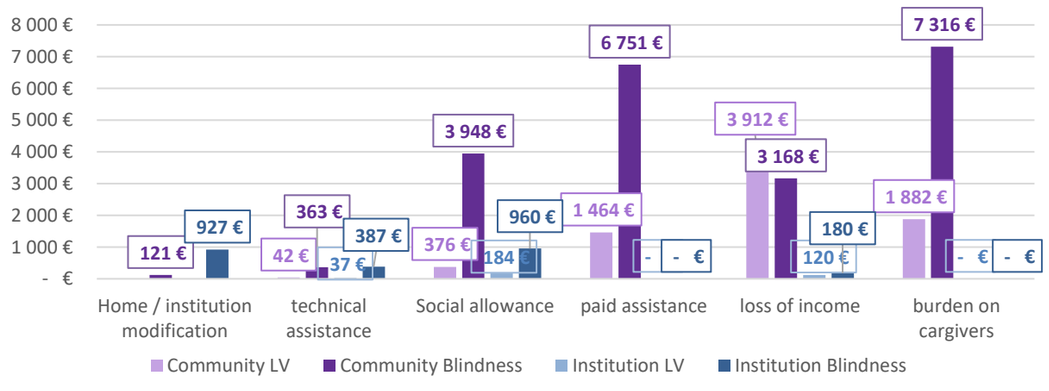
Figure 3. Medical and non-medical (direct and indirect) costs associated with visual impairments.



Abbreviations: ED- emergency department, GP- general practitioner; LV- low vision

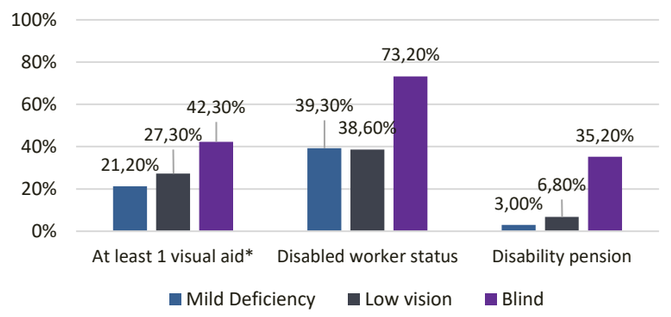
As non-medical costs are particularly important in the context of visual impairment, an emphasis is taken on non-medical costs from two surveys. These national surveys conducted in 1999 and 2000 by INSEE measured the costs and resource use associated with visual impairment (low vision and blindness) in France, both in patients living in the general community and those institutionalized.(4-7) **Figure 4** shows the excess cost of low vision and blindness compared to control (defined as subjects experiencing neither low vision or blindness), for patients living in institution and in the community. Costs associated with visual impairment were higher in patients with blindness compared to those with LV (15,679€ vs 7,242€ respectively)(4-7) while 48.0% and 29.7% of patients with RP have indicated to be affected by blindness and low vision, respectively.(8) Costs associated with visual impairment were also higher in patients living at home compared to institutions, as the main costs were the burden on caregivers and paid assistance (which do not apply to patients living in institutions) and as the institution cost was not directly reported in the survey (4-7).

Figure 4. Additional cost per annum per patient with low vision or blindness compared to control population(4-7)



Furthermore, a decrease in visual capacity due to the progression of RP was associated with a decreased level of autonomy, increased disability, and decreased employment, potentially associated with costs as demonstrated in available evidence from other ocular diseases, as shown in **Figure 5.** (8).

Figure 5. Impact of vision loss level on disability in RP



*Note: *Such as low vision rehabilitation, a white cane, or a guide dog*
Abbreviations: RP: Retinitis pigmentosa

DISCUSSION & CONCLUSION

In summary, our study underscores the need for further research to precisely quantify the economic burden of Retinitis Pigmentosa (RP) and visual impairment in general. RP and visual impairment carry a significant societal burden, driven mainly by non-medical costs, including disability expenses, paramedical support, and preventive measures. Our findings also reveal that the economic burden increases with the severity of visual impairment and that costs are substantial in individuals living in the community, particularly due to expenses related to paid assistance and caregivers. To address this broad economic impact, it is essential to adopt a more comprehensive perspective in cost-effectiveness analyses, moving beyond traditional approaches that may underestimate the true societal cost of visual impairment. Policymakers and healthcare stakeholders should prioritize holistic strategies to effectively alleviate this burden.

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