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Title	The economic impact of inherited retinal diseases: a systematic literature review
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Publication date	2024
Original Citation	Willmington, C., Murphy, A. and Kirby, A. (2024) 'The economic impact of inherited retinal diseases: a systematic literature review', 18th Pro Retina Research Colloquiu, Potsdam, Germany, 12- 13th April.
Type of publication	Conference item
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Download date	2026-11-05 23:39:10
Item downloaded from	https://hdl.handle.net/10468/17112



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THE ECONOMIC IMPACT OF INHERITED RETINAL DISEASES: A SYSTEMATIC LITERATURE REVIEW.

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INTRODUCTION

Affecting over 2 million people worldwide, inherited retinal disease (IRD) is considered one of the leading causes of blindness among the working-age population[1-2]. The recent market entry of Luxturna® to treat Leber Congenital Amaurosis (LCA) presents gene therapy as a promising option for IRD patients in the future. Furthermore, given the rarity of IRD and the high price of innovative medicines, it is essential to consider the current costs associated with IRD.

OBJECTIVES

To investigate the economic impact of inherited retinal diseases via a systematic literature review.

METHODS

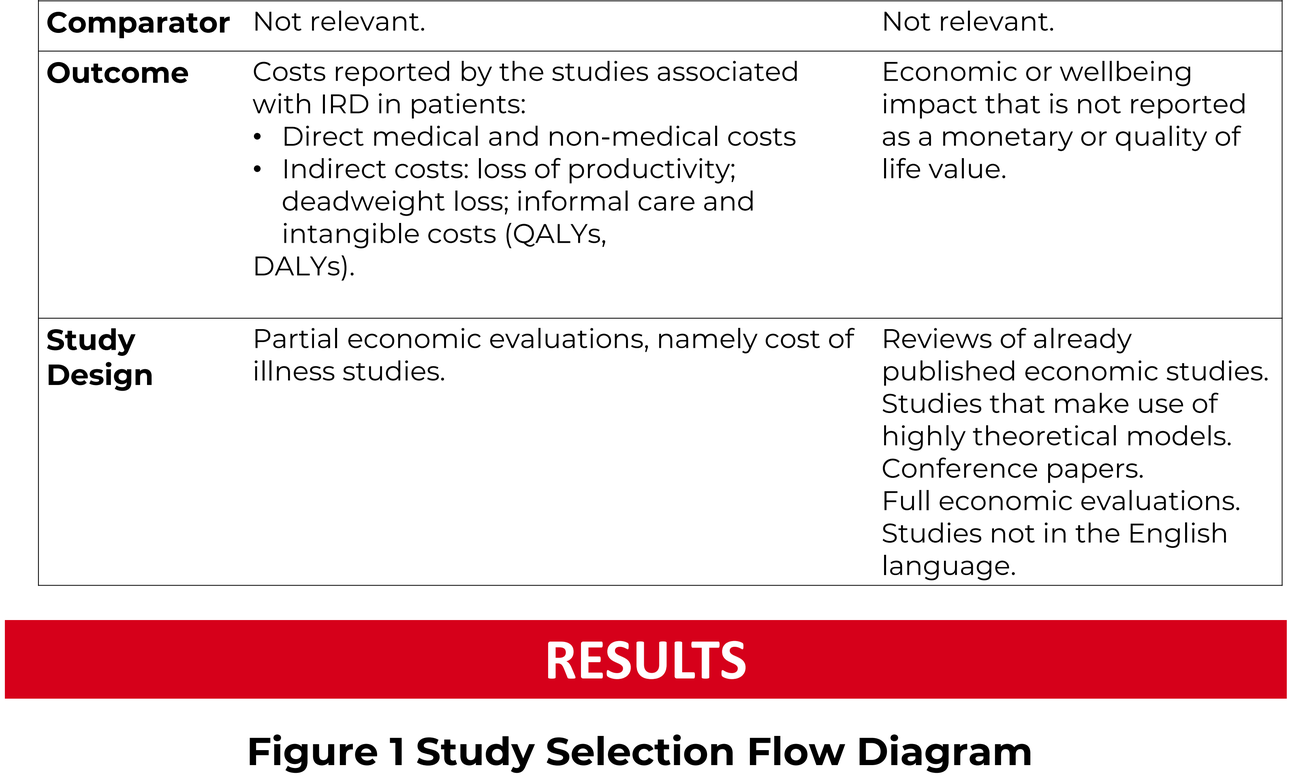
- The Medline, CINAHL, EconLit and Embase databases were searched from January 2000 to February 25th 2024.
- Search strategy guided by PICOS framework *(Table 1)*.
- Focus on economic studies estimating costs (direct as well as indirect) associated with IRD.
- Data synthesis and quality assessment conducted according to the PRISMA guidelines.

Table 1 PICOS Framework

Category	Inclusion Criteria	Exclusion Criteria
Population	Patients diagnosed with IRD and collaterally impacted individuals (i.e. caregivers). No restriction applied in terms of IRD type or patient age.	Patients with age-related retinal disease.
Intervention	Studies reporting the costs associated with IRD in patients.	Studies on screening strategies involving individuals without IRD.
Comparator	Not relevant.	Not relevant.
Outcome	Costs reported by the studies associated with IRD in patients: • Direct medical and non-medical costs • Indirect costs: loss of productivity; deadweight loss; informal care and intangible costs (QALYs, DALYs).	Economic or wellbeing impact that is not reported as a monetary or quality of life value.
Study Design	Partial economic evaluations, namely cost of illness studies.	Reviews of already published economic studies. Studies that make use of highly theoretical models. Conference papers. Full economic evaluations. Studies not in the English language.

RESULTS

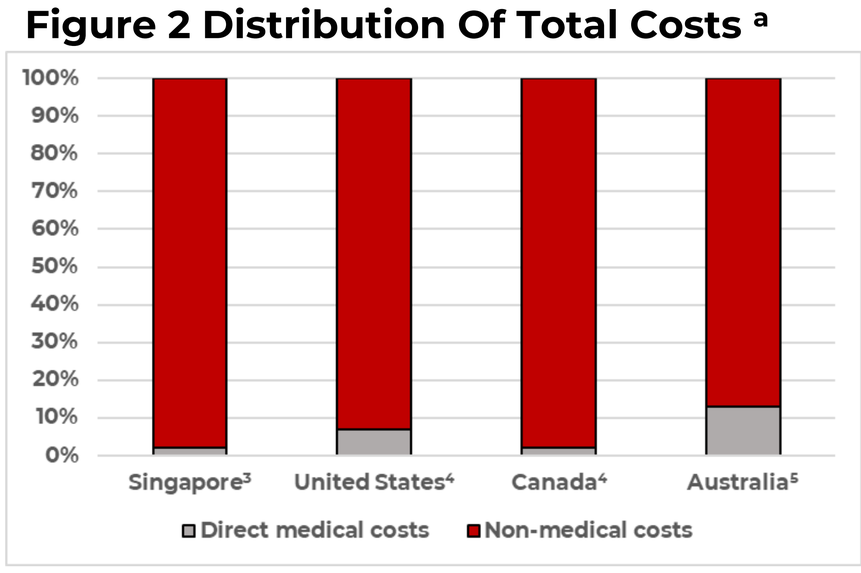
Figure 1 Study Selection Flow Diagram



Overview Of Search Results

- Nine studies were included in the review.
- Geographical distribution: North America (n=4), Asia (n=3), Europe (n=1) and Australia (n=1).
- IRDs of interest: Mix of IRDs (n=4), retinitis pigmentosa (n=2), Stargardt disease (n=1) and Choroideremia (n=1)
- Comparison of direct medical costs (inpatient, outpatient, and pharmacy services) across studies displayed on Figure 2.

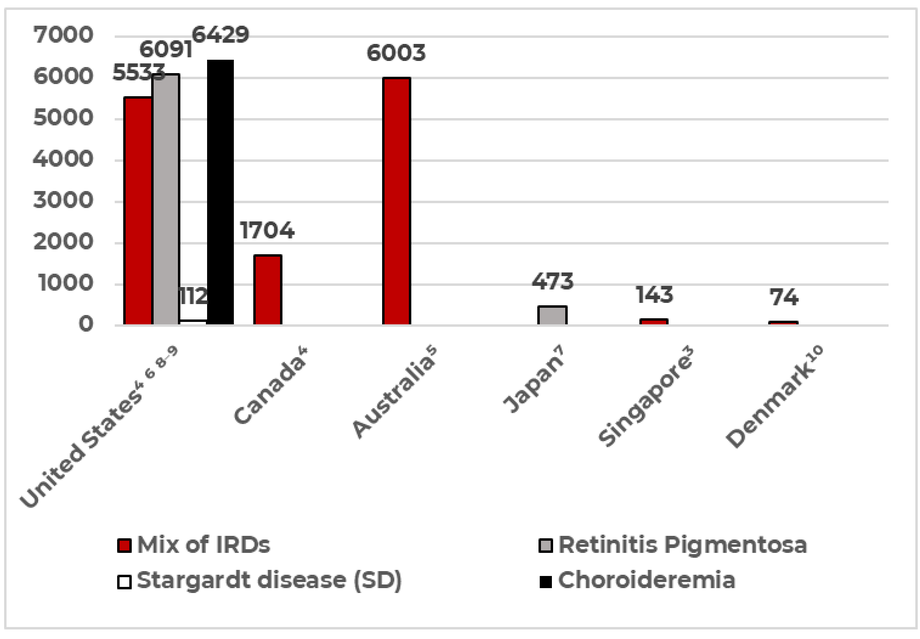
Figure 2 Distribution Of Total Costs [a]



Non-medical costs include:
- Carer costs
- Assistive technologies
- Productivity loss of patients
- Productivity loss of caregivers
- Informal care
- Loss of wellbeing
- Deadweight loss

[a] Only studies clearly reporting both medical and societal costs associated with IRD (n=3) were selected for this graph..

Figure 3 Direct Medical Costs (in USD per patient per year)[b]



[b] Studies clearly reporting medical costs associated with IRD (n=9) were selected for this graph..

CONCLUSIONS

- The studies varied in terms of size, methodology, as well as how costs were valued and reported.
- In studies where multiple cost components were reported, direct medical costs had the least impact on the total costs incurred as opposed to loss of productivity and quality of life, which had a much greater impact.
- The variation in the direct medical costs is in part reflective of how costs were reported, differences in burden of disease across IRDs, patient age, and healthcare provision from governments. Overall, direct medical costs tend to be lower in Asia and Europe than in North America and Australia.

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Funded by the European Union

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