A patient-centric approach to cost of illness research: a case example of patient engagement to identify the hidden costs associated with blindness due to inherited retinal dystrophies

PRO139

Authors: Fiona Glen MA (hons) PhD,[1] Raisa Sidhu BA (hons) MSc,[2] Amanda Pulfer BA (hons),[1] Angela Rylands BSc CPsych PhD[1]

[1]OPEN VIE, Marlow, UK; [2]Novartis Pharmaceuticals UK Limited, Camberley, UK

Introduction

- The impact of technologies beyond direct health costs and benefits may be of interest to Health Technology Assessment (HTA) bodies, especially for highly specialised technologies.
- Cost of illness studies are commonplace but engagement of relevant stakeholders with experience of living with a disease (e.g. patients and carers) can help ensure that all relevant indirect variables have been considered and burden of disease is appropriately captured. This is particularly important for rare diseases where limited information is available and many costs may lie outside the health system.
- The aim of this study was to gather insights from patients and carers in order to identify a framework of costs (both financial and non-financial) associated with blindness due to inherited retinal dystrophies (IRD).

Methods

Phase 1: Advisory board with patients with IRD

- Five adult patients (all of working age) with retinitis pigmentosa (RP) were recruited via a patient organisation (Retina UK) to participate in a face-to-face advisory board.
- Patients underwent a facilitated, two-step concept elicitation exercise.[1]
- Firstly, they individually identified costs relevant for different patient age groups (patients under and over 18 years and their carers). Next, they participated in a group mapping exercise to discuss and confirm these costs.

Phase 2: Interviews with carers of patients with IRD

- The cost framework identified during Phase 1 was then explored and ratified via 1:1 market research telephone interviews with five adult carers of patients with RP (patients being cared for were aged 9-22 years).
- A semi-structured discussion guide was used for all interviews with questions focused on understanding experiences of IRD and testing the framework from the Phase 1 advisory board. Interviews were audio recorded and transcribed, with thematic analysis conducted using grounded theory principles.[2]

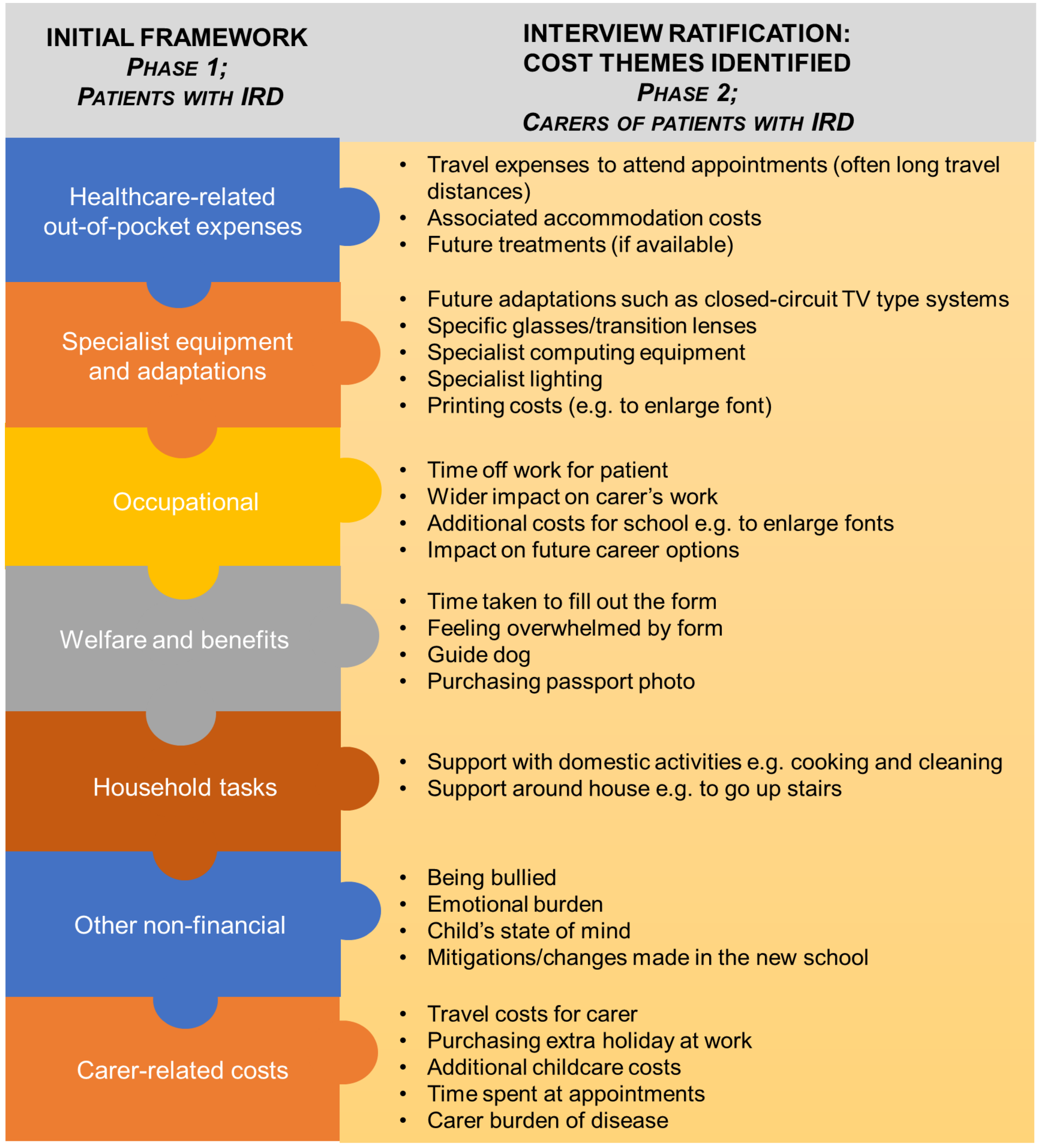
Results

- A framework of costs associated with IRD was developed based on discussion during the group mapping exercise conducted in Phase 1 (**Table 1**).
- In addition to financial costs, patients emphasised the wider impact of IRD on other aspects of health (e.g. mental health) resulting from a lack of treatment options and practical and emotional support, difficulty accessing welfare and benefits, loss of relationships, and financial hardship.
- All cost themes identified in Phase 1 were ratified in the 1:1 interviews in Phase 2 (**Figure 1**).

Table 1 Framework of costs identified during Phase 1

	Initial framework of costs associated with IRD
1	**Healthcare-related out-of-pocket expenses** e.g. the cost of travel to eye appointments.
2	**Costs associated with specialist equipment and adaptations** e.g. “high-vis” jackets; lightbulbs and computer technology to aid reading; tinted or thinned lenses or having to buy multiple pairs of glasses.
3	**Occupational costs** e.g. difficulty in finding work; taking time off for appointments; lack of resources for people with vision loss in organisations.
4	**Welfare and benefits** e.g. Disability Living Allowance (DLA) and patient burden related to DLA reassessments; limited training with white canes; requiring passport photos for ID cards.
5	**Household tasks** e.g. lack of confidence with cooking, cleaning and shopping resulting in assistance being required and people making unhealthy food choices.
6	**Other non-financial costs/ emotional burden.** The wider emotional and practical burden was emphasised; for example, the impact the condition has on their relationships. Children can often experience bullying which may lead to feelings of social isolation and can have a negative impact on personal growth.
7	**Costs experienced by carers** e.g. accompanying their loved ones to appointments using public transport.

Figure 1 Costs associated with blindness due to IRD from the perspective of patients and carers



INITIAL FRAMEWORK *PHASE 1;* *PATIENTS WITH IRD*	INTERVIEW RATIFICATION: COST THEMES IDENTIFIED *PHASE 2;* *CARERS OF PATIENTS WITH IRD*
Healthcare-related out-of-pocket expenses	• Travel expenses to attend appointments (often long travel distances) • Associated accommodation costs • Future treatments (if available)
Specialist equipment and adaptations	• Future adaptations such as closed-circuit TV type systems • Specific glasses/transition lenses • Specialist computing equipment • Specialist lighting • Printing costs (e.g. to enlarge font)
Occupational	• Time off work for patient • Wider impact on carer’s work • Additional costs for school e.g. to enlarge fonts • Impact on future career options
Welfare and benefits	• Time taken to fill out the form • Feeling overwhelmed by form • Guide dog • Purchasing passport photo
Household tasks	• Support with domestic activities e.g. cooking and cleaning • Support around house e.g. to go up stairs
Other non-financial	• Being bullied • Emotional burden • Child’s state of mind • Mitigations/changes made in the new school
Carer-related costs	• Travel costs for carer • Purchasing extra holiday at work • Additional childcare costs • Time spent at appointments • Carer burden of disease

- The Phase 2 interviews emphasised two significant areas of financial and non-financial burden:

a) **Opportunity loss**: opportunities in life, potential earnings and societal contributions are different for a sighted and non-sighted person

b) **Unpreparedness of society**: an overall lack of awareness in society for people with visual impairments and the fact IRD is an “invisible disability” meaning it is often assumed sight is “normal”.

Conclusions

- Incorporating patient and carer engagement provided a more holistic and patient-centric view of the costs and impact of blindness associated with IRD.
- These insights helped contextualise financial costs identified from the literature to inform quantitative analysis on the costs of blindness and provided a more in-depth understanding of the wider practical, emotional and societal impact of blindness, which will be used to inform future HTA submissions.

References [1]Trochim W. An introduction to concept mapping for planning and evaluation. Eval Program Plann. 1989;12:1–16. [2]Strauss A, Corbin J. Grounded theory methodology. Handbook of qualitative research. 1994;17:273-85.

Acknowledgements The authors are grateful to everyone who gave up their time to take part in the advisory board and interviews.

Disclosures FG, AP and AR are employees of OPEN VIE who were commissioned by Novartis UK to conduct the activities described in this poster as part of a wider project investigating the costs of blindness associated with IRD. RS is an employee of Novartis UK Ltd.

Poster presented at ISPOR Europe, Copenhagen, 6 November 2019
This study was sponsored by Novartis UK Ltd.

Presenter email address: fionaglen@openvie.com