



City Research Online

City, University of London Institutional Repository

Citation: Enoch, J., Dickinson, C. & Subramanian, A. (2021). What support is beneficial for caregivers of individuals with vision impairment?. *Clinical and Experimental Optometry*, 104(4), pp. 455-470. doi: 10.1080/08164622.2021.1878814

This is the accepted version of the paper.

This version of the publication may differ from the final published version.

Permanent repository link: <https://openaccess.city.ac.uk/id/eprint/25792/>

Link to published version: <https://doi.org/10.1080/08164622.2021.1878814>

Copyright: City Research Online aims to make research outputs of City, University of London available to a wider audience. Copyright and Moral Rights remain with the author(s) and/or copyright holders. URLs from City Research Online may be freely distributed and linked to.

Reuse: Copies of full items can be used for personal research or study, educational, or not-for-profit purposes without prior permission or charge. Provided that the authors, title and full bibliographic details are credited, a hyperlink and/or URL is given for the original metadata page and the content is not changed in any way.

City Research Online:

<http://openaccess.city.ac.uk/>

publications@city.ac.uk

REVIEW

What support is beneficial for caregivers of individuals with vision impairment? A scoping review.

Authors

Jamie Enoch, MSc*

Christine Dickinson, PhD†

Ahalya Subramanian, PhD*

* Division of Optometry and Visual Science, School of Health Sciences, City, University of London, London, UK.

† Division of Pharmacy & Optometry, University of Manchester and the Manchester Academic Health Sciences Centre, Manchester, UK.

Running title

Support for caregivers of individuals with vision impairment

Key words: caregivers, low vision, parents, support, vision impairment

Previous studies and reviews have documented the stress and challenges that may be associated with providing informal care for individuals with vision impairment (IVI). This scoping review was therefore conducted in order to synthesise published literature about forms of support which may benefit the informal caregivers of both adults and children with vision impairment (VI), and to identify research gaps in the support available for this population.

A systematic literature search was carried out using CINAHL, Medline, PsycINFO and PsycARTICLES, followed by citation tracking.

A total of 23 published studies met the eligibility criteria and were included in the review. The included studies focused on: exploring caregiver support needs (8/23); novel interventions supporting caregivers of IVI (10/23); evaluating usual care (2/23); and exploring how treatment for IVI directly impacts the caregiver (3/23).

Overall, support for caregivers of IVI is a relatively new research topic, with no eligible studies identified before 1999. Twelve of the 23 studies (52%) focussed on support for caregivers of adults with VI, while 11 (48%) focussed on support for caregivers of children with VI.

The studies illustrate that support groups may generally help to improve caregivers' knowledge and awareness of VI, although benefits for emotional wellbeing are more modest. Support interventions for parents of children with VI appear to reduce stress effectively; however, evidence regarding the value of interventions for caregivers of adults with vision impairment is less clear, partly due to small samples and a lack of standardised, comparable outcome measures. Caregivers often express a need for better information about the IVI's condition, even when information is apparently available. Further research is required comparing the benefits of different support modalities for caregivers of people with VI over longer follow-up periods.

Background

Worldwide, an estimated 285 million people are living with sight loss (i.e. are considered blind or partially sighted),¹ and this may challenge their ability to live independently. Many of them receive help and support from informal caregivers including friends and family,² which may facilitate adjustment and adaptation to changes in vision.

It is challenging to estimate the number of caregivers of individuals with vision impairment (IVI) across the world, particularly given continual changes in the caregiver population and lack of clarity regarding the definition of 'caregiver'. However, 17% of the population in England over 16 are caregivers and 20% of caregivers in England support someone living with a sensory impairment (including, but not limited to, vision impairment (VI)).³ This would suggest an estimated 3.4% (17% multiplied by 20%) of the English population care for someone with a sensory impairment. That percentage may be even greater in other regions of the world, such as South Asia and Sub-Saharan Africa, where prevalence of VI⁴ and hearing impairment⁵ is higher than in Western Europe.

While caregiving may have positive aspects for both for the IVI, the caregiver and society,⁶ it can often be stressful and thus negatively impact upon the caregiver. For example, in one 2009 study, approximately 35% of family caregivers of adults with VI were found to be at risk of clinical depression.⁷

Although caregiver distress has been acknowledged in a number of diseases such as stroke,⁸ cancer,⁹ and dementia,¹⁰ it has only emerged relatively recently as an area of concern in VI. A 2017 review documented the multiple negative emotional, economic and relational impacts of informal caregiving for people with VI.¹¹ There is clear evidence that as eye disease progresses and vision becomes more impaired, the use of formal and informal care increases.¹²⁻¹⁴

It is also important to consider the resources, strategies and interventions that may help to alleviate potential adverse aspects of caregiving. Some attention has been paid to improving support for caregivers of IVI. For example, Gopinath et al. are developing a multi-modal support intervention aiming to reduce distress among the family caregivers of people living with VI resulting from age-related macular degeneration (AMD).¹⁵

Many articles discussing caregiver distress and quality-of-life (QoL) refer to the need for improved caregiver support. For example, a 2018 review¹⁶ summarising research on QoL of parents of children with VI, states:

“Parents require better and more extensive information and guidance to understand the diagnosis of their children’s condition, become aware of the available resources and services, and receive support on how to manage and adjust to the situation”.

However, a systematic review on family functioning and VI¹⁷ acknowledged that little is known about how helpful support interventions and services are to caregivers of IVI. In fact, there is limited research on support that is currently available and tailored to the unique needs of caregivers of IVI; how much caregivers of IVI are accessing any available support; and what the barriers and facilitators may be to access.

This scoping review therefore explored the scientific literature regarding the support services, resources and interventions available to caregivers of IVI.

Rationale

Previous reviews have focussed on limited aspects of support for caregivers of IVI, particularly in the case of parents or carers of babies, infants and/or children with VI. For example, in 2019 Yesilkaya and colleagues published a systematic scoping review exploring barriers and facilitators to the implementation of interventions for children with VI, but also for their parents/guardians and educators.¹⁸ An earlier, more narrow systematic review considered interventions to foster more secure attachment and improved interaction between children with VI and their parents.¹⁹ While some studies included in these reviews were relevant to the present review, this review deals specifically with studies where support services or interventions for caregivers of IVI are the central focus of the research.

The purpose of scoping reviews is to “identify knowledge gaps, scope a body of literature, clarify concepts or to investigate research conduct”.²⁰ A scoping review typically addresses a broader research question, as in this case, where

the populations of interest were heterogeneous; including caregivers, who may be different kinds of family relatives or friends, and IVI (who may be adults or children; have a range of underlying ophthalmic and/or neurological conditions causing VI; and whose VI may range from mild to severe). This inevitably creates challenges in generalising about the caregiver experience, given especially that the nature of caregiving relationships (e.g. the spouse of an older adult with VI versus the parent of a child with VI) significantly influences the kind of support the caregiver needs. However, as a pragmatic decision we have chosen to include studies focussing on support for caregivers both of adults and children with VI, because an initial search showed a very small number of studies considering the various relevant domains of caregiver support. We also considered that comparing the support available could be instructive and illuminate shared features of support found to be beneficial, while being mindful of the differing relational context.

Furthermore, grey literature and conference abstracts, which would not necessarily be suitable for inclusion in a systematic review, were included. Indeed, a scoping review aims to encompass all available evidence regardless of quality, and therefore does not require a formal quality assessment or risk of bias assessment (as in a traditional systematic review).

The PRISMA extension for scoping reviews checklist was used to ensure high-quality reporting.²¹

Key definitions

Support

There are many different definitions and taxonomies of support in the health and social science literature.¹⁵ Here, 'support' was considered in a broad psychosocial sense to include the five macro-level categories of support defined by Cutrona and Suhr,²³ which are described in Table 1.

[Table 1 to go here]

Caregivers

In this review, 'caregiver' refers to a family member, friend or neighbour who may provide regular (informal) care or support, and who may or may not live with the IVI. Broadly, caregiver(s) may be considered (after Larizza et al., 2011)²⁴ as a person/people to whom the IVI may regularly turn for help or support. Teachers, educators or professional carers were not considered as 'caregivers' for the purpose of this review.

It is important to note that many relatives and friends who regularly provide support for children or adults with VI do not identify with the term 'caregiver', seeing themselves first and foremost as parents, spouses, children, siblings or friends.²⁵ For the purposes of this article, we use this imperfect term 'caregiver' as shorthand in order to encompass a broad group of individuals with different relational ties to the person with VI.

Objectives

This scoping review considered the support required by and available to caregivers of IVI. The following questions guided the review:

1. Which resources, strategies and interventions may specifically help and support caregivers of IVI?
2. Which factors facilitate or hinder caregivers of IVI benefiting from support that is apparently available?

Methods

Eligibility criteria

Types of participants

Studies and grey literature reports were eligible if they involved caregivers of either IVI or people with eye disease that may cause VI.

Generic caregiver support interventions (e.g. befriending), or interventions specifically trialled with caregivers of people with other diseases such as dementia, could feasibly yield benefits for caregivers of IVI. However, the scope of the review was limited to studies involving caregivers of IVI.

Studies that included caregivers as participants but solely explored outcomes or experiences relevant to the participants with VI were not included.

Types of intervention(s)/phenomena of interest

The emphasis of the review was on exploring a broad range of support strategies, services or interventions for caregivers of IVI, although studies considering caregivers' unmet support needs were also included. Studies on caregiver distress that provided broad recommendations for caregiver support, for instance in the concluding paragraphs, but did not primarily focus on caregiver support were not formally included. Nonetheless, to ensure a comprehensive review, the implications of these studies are shown in the Supplement.

Interventions which benefit care recipients may have positive "spillover" effects for caregivers.²⁶ However, these were not considered unless caregiver health or wellbeing was a key phenomenon of interest. For example, a study on the effectiveness of services for deaf-blind children²⁷ was not considered eligible; for whilst the study considers the child in the context of their family, support for the other family members is not a primary focus.

Types of studies

All quantitative and qualitative research designs were eligible. Relevant grey literature reports or unpublished doctoral theses were included alongside studies published in peer-reviewed journals.

Types of outcomes

The aim of this review was to map literature on support for caregivers of IVI and consider which types of outcomes are currently being measured in the research. Outcomes therefore included: changes in levels of caregiver strain; healthcare professional awareness of and response to caregiver distress; quality of life measures; measures of mental and/or physical health and wellbeing; or caregivers' perspectives on support services elicited through qualitative research.

Search strategy and information sources

A three-step search strategy was employed.²⁴ An initial limited search of relevant databases was undertaken, followed by an analysis of the text words contained in the title ("caregiv*", "carer", "caring", "support", "visual impair*"), and of the index terms used to describe relevant articles.

A second search using all identified keywords and index terms (for example, terms relating to specific conditions which may cause VI, e.g. "macular disease") was then undertaken across all included databases. Searches were run on the MEDLINE, CINAHL, PsycINFO and PsycARTICLES databases. Full search terms for each database are shown in Appendix 1. The search results included grey literature, such as conference proceedings.

A third step entailed searching the reference lists of all included reports and articles for additional studies. Studies published in English, French and Spanish (the first author's languages) were considered for inclusion in this review. Studies dating from any time were eligible for inclusion.

Data analysis

Data were extracted to record characteristics of included studies and the key information pertinent to the review questions (e.g. study population; methodology; key findings). As this was a scoping review, conducted to provide a broad picture of the existing literature on this topic, a formal assessment of methodological quality was not undertaken.

DRAFT

Results

Number of studies found, retrieved and included

Searches were run on 12 July 2019 and yielded 2,174 results (to which 8 further studies were added through reference list searching). Of these, 131 were automatically removed as duplicates. This left 2,043 studies to screen using title and abstract, of which 1,941 were excluded and 102 articles were assessed for full-text eligibility. Common grounds for exclusion were a lack of focus on *support* for caregivers of IVI, even if caregivers participated in the study; or a focus on the IVI, where the caregiver's perspective was only relevant insofar as it provided information about the IVI's experience. Ultimately, 23 full-text studies were selected for inclusion.

Searches were re-run on 20 July 2020, and no new studies were identified for inclusion in the main review. However, one study was added to the Supplement.

The study selection process is shown in a PRISMA flow diagram in Figure 1.

[Figure 1 to go here]

Trends

Twenty-one of 23 included publications (91%) were from peer-reviewed journals; one was a grey literature report, and one was a PhD thesis.

Of the 23 publications, the first was published in 1999, with eight published before 2010, and 15 between 2010-2019 (Figure 2).

[Figure 2 to go here]

Five studies (22%) were wholly qualitative, while 16 (70%) were primarily quantitative (with some elements of qualitative data analysis, e.g. free-text responses on otherwise quantitative surveys). Two studies (9%) were descriptive reports of services for caregivers.

The most common type of quantitative study design was interventional, including 3 randomised controlled trials (RCTs) and 4 pre-test/post-test designs. Three were cross-sectional survey studies, two were protocols for future RCTs,

and one was a longitudinal cohort study. Three of the studies were mixed-methods evaluations of improvements to services (Figure 3).

[Figure 3 to go here]

Twelve of the 23 studies (52%) were focussed on caregivers of adults with VI, while 11 (48%) were focused on caregivers of infants, children or adolescents with VI.

Six of the studies (26%) were based in the United States, five each in United Kingdom and Australia, and one each in Brazil, Canada, China, France, India, Iran, and Japan (Figure 4).

[Figure 4 to go here]

In terms of the thematic focus, eight studies considered caregiver needs for services or interventions. Ten were time-limited interventions to support caregivers (including completed studies, and protocols for ongoing trials). Two of the included studies described existing support services for caregivers. Three studies focussed expressly on how services for IVI or people with eye disease impact the caregiver. The subsequent overview of study findings is organised according to these thematic domains. Key details of included studies are summarised in Table 2.

[Table 2 to go here]

1. Studies exploring caregiver support needs

Eight studies elicited the views of caregivers on their unmet needs for support. Many of these studies also consider the individual with VI's needs, since many IVI lack support and this in turn impacts upon the caregiver. Overall, current research tends to focus more on how the caregiver can support the IVI with aspects of living with VI, rather than on support for the caregiver *per se*.

i. The heterogeneous needs of caregivers

The included studies clearly illustrate that caregivers of IVI are a heterogeneous group, and that the support required may depend on the characteristics of the IVI (their disease type, its severity, and their level of VI) as well as the caregivers' baseline circumstances. Satisfaction with care has been found to be higher among parents whose children have mild VI or isolated ophthalmic conditions, compared with more severe VI ($P = 0.003$) or multiple impairments ($P = 0.02$).²⁹ Comorbidities affecting older adults with VI clearly influence the support their caregivers require. For example, caregivers of individuals with both VI and dementia have highlighted respite services and specific advice on managing visual hallucinations as necessary forms of support.³⁰ In contrast, the caregivers in Shtein et al.'s study played a limited role in the support of their loved one with glaucoma, unless the disease progressed to a late stage..³¹

Even among caregivers of individuals with the same eye disease, caregivers express highly varying levels and kinds of unmet needs for themselves and the care recipients.³² In a survey of parents of children with cortical or cerebral VI, participants were asked what services should be - but are not currently - provided, a key example of what Cutrona and Suhr describe as tangible assistance.²³ Forty-two percent of participants believed their children were receiving all necessary services; second to this, 29% of parents believed sensory integration would benefit their child with CVI; and 24% highlighted an unmet need for orientation and mobility services. Some of these divergences in caregiver opinion may be attributable to the child's age, with caregivers perceiving more unmet needs once their child began school and difficulties arose obtaining necessary modifications and support.

Socio-demographic factors and dynamics within the familial or social support system around the IVI may also influence caregivers' support needs. Rahi et al. (2005) show that parents' support needs may vary according to ethnicity and their caregiving role (whether primary or secondary carer).²⁹ For instance, these authors found that primary carers, who are generally more responsible for helping their child access services, showed a greater need for information than secondary carers.

ii. Balancing support for caregivers and IVI

Involving caregivers in vision rehabilitation programmes may help improve the caregiver's understanding of low vision. This involvement may provide them with an opportunity to meet others in similar positions, and gave them an opportunity to express their feelings and learn coping strategies. However, from the perspective of vision rehabilitation professionals, a potential challenge is that family members or friends could shift the focus of discussions to their needs and thus potentially detract attention from the clients' concerns.³³ Indeed, it is important to note that adults with VI may have different views from their relatives about the value of education and information about the condition, both in terms of what kinds of information are helpful and how information is communicated.³¹ Additionally, caregivers may struggle to openly disclose their needs for information or support in the presence of the person with VI.²⁴ This suggests that interventions focused on simultaneously supporting both adults with VI and caregivers need to build in careful planning and facilitation, in order to balance meeting clients' and caregivers' shared and divergent needs.

One potential solution may be to offer separate groups for caregivers and IVI, running in parallel with crossover sessions where concerns common to both parties are discussed.³⁴ This may provide IVI and their sighted partners a dedicated space to each explore and express their feelings openly with peers. Qualitative feedback suggests partners value an opportunity to be open about feelings of anger, guilt and anxiety that may accompany caregiving for an individual with vision impairment. Sharing these emotions with peers may in turn enable better understanding and constructive conversations with their partners living with VI. Thus a separate group for caregivers may provide a valuable avenue for both esteem support and network support (Table 1).

iii. Complexities of informational support

Research with parents of children recently diagnosed with VI found that the greatest unmet need among these parents was for better general information, about their child's condition, social services, education and support networks.²⁹ Yet information supply in itself may not be perceived by caregivers as being particularly useful; for example, even caregivers who theoretically have regular access to up-to-date information about macular disease can still feel they are receiving limited information.³⁵ One study found that higher information provision for parents of deafblind children was a significant predictor of the parents' decreased family quality-of-life.^{36, 37} This suggests that even though many caregivers report being happy about receiving information, the content of the information may not be effectively meeting their needs. This may be the case particularly if information is overwhelming, or seen to predict negative outcomes for the child. The author suggests that information must be easy to navigate, and adopt a positive portrayal of children with disabilities.

Although health professionals are often seen as sources of helpful information, caregivers themselves may sometimes feel that onus falls on them to educate and inform health professionals. For example, in a survey of parents of children with cortical or cerebral VI, 51% of participants had received no information beyond a diagnosis, and 69% of participants reported undertaking their own research to gain the necessary information about their child's condition.³²

Taken together, the studies suggest that information is crucial for caregivers; however information should be personalised, meaningful, and presented in a clear, engaging way that avoids overwhelming the caregiver. It has been suggested that informational needs may be effectively met when one key worker works with families to signpost to other services, help navigate the care system, and provide support with the complex issues that families may encounter at the time of diagnosis.³⁸

2. Novel interventions supporting caregivers of IVI

i. Interventions for caregivers of adults with VI

There is some evidence from small studies exploring support interventions for caregivers of adults with VI, which integrate elements of informational, emotional, esteem, and network support (Table 1). One US-based programme, investigated by Cimarolli and colleagues, involved a support group for partners/spouses of IVI,³⁹ run across nine sessions and based upon a discussion guide.⁴⁰ Another study by Doorey et al. evaluated a self-management programme of six weekly sessions for caregivers of IVI, called *Care for the Carers*, developed at Curtin University of Technology.⁴¹ A third study evaluated an Australian group-based self-management programme for caregivers (*Living with Low Vision (LLV)*), which involved eight weekly sessions.²⁴

In the study by Cimarolli et al., caregivers found the most helpful aspect to be learning from others about how to adapt and cope with the situation.³⁹ Following the programme, caregivers showed an increase in: understanding what the partner can see ($P=0.02$) and do ($P=0.006$); certainty about ways to help ($P=0.001$), when to offer help ($P=0.001$), and about how much help to offer ($P=0.001$); and ability to talk openly ($P=0.035$). Participating caregivers also showed a reduction in: role captivity (i.e. feeling trapped; $P=0.002$); and their self-reported number of major issues ($P=0.011$). However, there were no changes to more global psychological or relational outcomes such as depression ($P=0.19$). The authors suggest that the lack of improvements on these broader indicators of wellbeing could be explained by the intervention being geared towards providing education and support, and also by the ceiling effect of a high level of wellbeing in pre-programme measurements. This latter factor arguably limits the generalisability of the programme, given that depression and stress may be prevalent among many caregiver groups.¹¹

In Doorey et al.'s study, eleven participants evaluated the programme, giving a mean satisfaction score of 9.0/10.⁴¹ Information was seen as relevant (mean rating 8.9/10), important (8.75) and well-delivered (8.71). However, post-intervention improvements to the psychological outcomes were relatively small, with only self-efficacy showing a statistically significant improvement. QoL and resilience showed small but not significant improvements. Notably, only eight participants completed QoL and self-efficacy measures, and only seven completed the resilience measure. This small sample size may partially explain the modest improvements, and demonstrates the importance of evaluating these interventions with larger samples.

In Larizza et al.'s study, the LLV programme increased awareness of low vision aids and practical strategies, but there was no statistically significant improvement in understanding of low vision, confidence, self-efficacy and emotional wellbeing.²⁴ Participants' qualitative feedback rated the LLV programme as relevant and helpful. The authors suggest the statistical null effect may stem from caregivers already having relatively high confidence and self-efficacy, since many of them were receiving support at the time of enrolment (as in Cimarolli et al.'s study).

Larger studies are currently being conducted in formal, registered RCT's, which will address some of the concerns in the studies above about low sample size. In Australia, a large multi-modal support intervention is currently being trialled in a formal RCT involving 360 caregivers of people with AMD.¹⁵ The support provided to caregivers in the intervention group includes mail-delivered cognitive behavioural therapy modules, telephone-delivered group counselling sessions, and education about available resources to help with coping, such as financial benefits and respite services. In France, Pozet et al. have created a protocol for their longitudinal study which will aim to recruit 1684 informal caregivers of people living with AMD.⁴² A RCT will be nested in the longitudinal study, exploring whether an intervention by a social worker provides efficient and effective support to caregivers.

ii. Interventions for caregivers of children with VI

Of five studies identified which evaluated interventions for parents or carers of children with VI, three used the Parenting Stress Index (PSI) as a key outcome measure. In one study using the PSI, a life skills training programme for mothers with blind children aged 7 to 12 years was evaluated in Iran.⁴³ The authors found that immediately after the programme, there was a significant reduction in mothers' parenting stress in the intervention group ($P < 0.001$). In another study with the PSI in the UK, Dale et al. found that using an early intervention based on the Developmental Journal for babies and young children with visual impairment (DJVI) showed better child and parent outcomes than when children received other forms of home support.⁴⁴ Specifically regarding PSI scores, there was a significant reduction in parenting stress for the DJVI group compared to those receiving other support (P

= 0.02). In a third study, Chen et al. conducted a RCT in China of an interactive education programme about congenital cataracts for parents of children with the condition, to evaluate parental stress, comprehension of disease information, and parental satisfaction.⁴⁵ The programme, which involved active learning strategies such as audience response systems and online group activities, showed large improvements over conventional care (where only oral information is given), increasing disease-related medical knowledge and reducing anxiety, at both 6-month and 12-month follow-up. There were moderate negative correlations between medical knowledge and parental anxiety, as measured by the PSI ($r = -0.357$) and Ocular Treatment Index ($r = -0.472$), at 12-month follow-up.

In a UK-based intervention, a mixed-methods evaluation assessed the views of parents of children with VI towards paediatric services, comparing the satisfaction level of parents who had received support from a Community Link Team (CLT) with those who had not.³⁸ Parents receiving CLT support benefitted from increased information, access to services, emotional and social support, and meeting with parents in similar situations. Qualitative feedback attested to the breadth of support offered by the CLT key-worker, including personalised emotional support, navigating the care system, applying for housing support, and linking to other families. A designated “quiet room” space at the clinic was found to be particularly beneficial, as was early contact between the CLT worker and the family (at the first outpatient consultation).

In a Canadian intervention study, McConnell considered how parents evaluated a programme which was designed to support parents of adolescents with VI, and the adolescents themselves, with career planning.⁴⁶ The parents noted that the programme encouraged them to dedicate time to career planning with their child, and that the programme had increased their child’s willingness to discuss careers with them openly. Parents’ feedback was positive about the programme’s structure, and they believed that the programme improved their children's self-understanding, self-confidence, motivation, preparation, and planning regarding career decisions.

3. Evaluating routine care for families of children with VI

As opposed to studies in the previous sections, which were seeking to understand unmet caregiver needs or to trial novel interventions, some studies considered more descriptively how routine health services currently operate to support the families of children with VI.

The geography and organisation of health services facilitates or hinders access to tangible caregiver support in certain contexts. This is illustrated particularly clearly when contrasting service provision for children with VI in two Brazilian cities.⁴⁷ One city hosts a cohesive care centre supporting both children and their caregivers, while the other, lacking such a centre, provides more fragmented, disjointed support. Problems highlighted by caregivers in both cities were inadequate communication between health services and the caregiver, and a lack of referrals to social and cultural organisations which could provide more everyday support. In both cities, caregivers depend heavily upon their family, social networks or faith-based organisations. This echoes the disparities evident in the US context, where Jackel et al. report that 42% of participating parents of children with CVI have received all necessary services, while 35% have received no support at all.³²

In the case of infants with VI (between 0-3 years), the use of specialised orientation and mobility (O&M) services increased children's O&M proficiency and parents' understanding of how their child's perceived limitations could be overcome.⁴⁸ This in turn helped parents to adopt a more "positive, informed attitude".

4. Studies exploring how treatment for IVI or people with eye disease directly impacts the caregiver

Three studies were identified which directly considered how caregivers are affected by treatments or interventions for the person with vision impairment.

Two studies considered the effects of anti-vascular endothelial growth factor (VEGF) on caregivers' wellbeing. In one study, Gohil et al. used a cross-sectional design to explore which factors affected 'caregiver burden' among 250 caregivers of patients receiving ranibizumab therapy for neovascular AMD (nAMD).⁴⁹ This study showed that most caregivers report receiving little or no support from healthcare

providers. Eighty-two percent reported being 'not at all' satisfied with the caregiver support given, while 82.4% were 'not at all' satisfied with the practical advice given. The most common forms of healthcare support received were having their needs as caregiver assessed (40.8% reported this 'often' happened), while 35.6% of caregivers received help to understand nAMD. Nonetheless, in both these cases, caregivers were more likely to report having received no assessment of their needs (46.8%) and no help to understand nAMD (49.2%). Overall, the study suggests a high unmet need for information and support among caregivers. However, regarding care satisfaction, as measured by the Client Satisfaction Questionnaire (CSQ-8), caregivers had relatively high levels of satisfaction (28.4 ± 4.1 out of a possible 32) compared with the AMD patients (15.7 ± 1.8). In another study involving 72 pairs of people with nAMD and caregivers, Hanemoto et al. evaluated how different kinds of anti-VEGF treatment regimens affect caregiver burden.⁵⁰ It was found that a 'treat-and-extend' (T&E) regimen reduced patients' hospital visits, and economic productivity losses were reduced in the T&E versus the care as needed (PRN) group. Despite time and money savings among the T&E dyads, there were limited reductions in caregiver burden (as assessed by the Burden Index of Caregivers or BIC-11). The authors suggest this may be because caregivers enrolled in the study had a low level of caregiver burden at study entry. Additionally, they contend that caregiver burden as measured by BIC-11 may be influenced more by the caregiver's subjective perception of the patient's visual acuity than by the patient's actual acuity. Therefore, while the T&E regimen did not yield clear emotional support benefits for caregivers in this study, it did appear to provide material, tangible assistance (Table 1) for caregivers, in terms of time and money savings.

There were fewer studies exploring how treatment for children with VI may benefit parents. In a rare example, Gothwal and colleagues, in a conference report, found that the QoL of caregivers of children with primary congenital glaucoma (PCG) was significantly higher 6 to 8 weeks following the child with PCG undergoing combined trabeculotomy-trabeculectomy.⁵¹ On an overall question rating QoL, 71% rated their QoL as poor or very poor before the procedure, compared with 20% after ($P < 0.0001$).

Discussion

Twenty-three studies met the eligibility criteria and were included. This scoping review demonstrates that exploring and responding to the support needs of caregivers of IVI is a relatively recent and burgeoning research area, with no relevant studies identified prior to 1999.

A search of clinical trial registries also demonstrated that there are four ongoing registered trials with secondary outcomes that explore how the trial intervention benefits the caregivers of IVI (Table 3). Furthermore, two included articles were protocols for RCTs (see Results, section 2.iii) which between them will involve over 2000 caregivers of people with AMD. Thus there is an encouraging trend towards trials evaluating the wellbeing of caregivers of IVI as an important primary or secondary outcome. Furthermore, in contrast to other included studies, the two protocols for RCTs are both seeking to measure the cost-effectiveness of interventions and thus consider the potential for their scale-up.

[Table 3 to go here]

[Table 4 to go here]

Table 4 displays a summary of results from across the 23 studies. These findings however must be qualified, in light of the studies' limitations. In particular, studies evaluating programmes for caregivers of adults with VI generally had small sample sizes and relatively high wellbeing levels at study entry, thereby reducing the potential for statistically significant improvements in wellbeing.^{24,39,41} There is a need for studies with greater statistical power that explore impacts on caregivers over a longer time-frame. Indeed, it could be instructive to learn from interventions focused on supporting parents of infants, children and adolescents with VI,⁴³⁻⁴⁵ which measured and demonstrated positive outcomes at 12 month follow-up. These studies with parents or carers of children with VI consistently use the Parenting Stress Index (PSI), which helpfully allows for inter-study comparability.

The findings that groups for caregivers of adults with VI are most effective for improving knowledge and awareness of VI are aligned with the broader caregiver support literature in other areas. Using Cutrona and Suhr's framework (Table 1),²³ it

appears that for adults with VI, such time-limited group interventions provide better informational than emotional support. This is somewhat consistent with a meta-analysis of 78 interventions to support caregivers of older adults, which found that interventions had a greater impact on improving knowledge and skills than on reducing psychological outcomes such as caregiver burden and depression.⁵² At the same time, it is important to consider individual differences in benefits derived from various forms of caregiver support (which may be challenging to demonstrate quantitatively). For example, qualitative work shows that forms of tangible assistance such as respite care may help certain caregivers regain a sense of “normality” and “freedom”, even though large-scale quantitative studies often show only modest benefits.⁵³ Indeed, the importance of respite (and other “tangible assistance” forms of support)²³ for caregivers of IVI was emphasised in some of the qualitative studies in this review,^{30,35} but not formally evaluated. The challenge of comparing benefits of support across studies is compounded by the lack of specific questionnaires for caregivers of people with VI. Instead, the included studies use generic questionnaires for caregivers (such as the Client Satisfaction Questionnaire-8 or the Parenting Stress Index) which may not capture aspects of caregiving unique to VI. Future research could consider the potential utility of questionnaires tailored to the needs of informal caregivers of IVI overall, or alternatively, distinct questionnaires for caregivers of children with VI and caregivers of adults with VI.

The included interventions were structured either to support individual caregivers (or families) of IVI,^{38,42,44} or more commonly to support caregivers in groups.^{15,24,39,41,43,45,46} Especially for caregivers of older adults with VI, the group-based model was predominant. In Cimarolli et al.’s study, participants found the most useful aspect of the intervention to be the opportunity to learn from others about how to adapt to the situation and cope with different problems.³⁹ This could be considered as a combination of informational and network support in Cutrona and Suhr’s framework.²³ It implies that meeting supportive peers and realising there are other caregivers in similar situations may be a particularly valuable underlying aspect of these interventions, which aligns with findings in caregiving research in dementia.⁵⁴ This may be explained by social identity theory, which posits that individuals’ wellbeing benefits when they are part of a group that they find psychologically meaningful and receive social support in that context.⁵⁵ Nonetheless, there is

evidence from the broader caregiver support literature that individual-level interventions, such as respite care for informal caregivers, may be more effective than group-level interventions.⁵⁶ This kind of individual-level support may be available to caregivers of IVI through contact with supportive eye care professionals and broader health, social, voluntary and faith-based services, as well as from relatives and friends. However no studies were identified evaluating this kind of support, perhaps because of the complexity of studying these more personalised, *ad hoc* forms of support, in contrast to discrete, structured programmes. Another explanation may be that one-to-one forms of support appear to be less cost effective than groups, although health economic evidence is mixed regarding the cost savings of group- over individual-level interventions.⁵⁷ Indeed, measuring the cost-effectiveness of support for caregivers of IVI would be valuable, and is an important component of the large-scale ongoing RCTs included in this review.^{15,42}

Several conceptual issues emerge which pose a challenge to identifying the most beneficial forms of support for caregivers of IVI. A note of caution that may apply across several studies included in this review is the potential to find a discrepancy between caregivers objectively receiving support and their subjective feeling of being supported. For example, Gohil et al. (2015) found that caregivers generally had high levels of satisfaction with support services, despite reporting receiving limited support.⁴⁹ This exemplifies the complexity of measuring “support” and disentangling the different informational, tangible and emotional dimensions. Indeed, support for caregivers is a highly nuanced concept, which is defined, operationalised and measured differently across studies. For example, no two intervention studies in the review trialled the same support programme, and different modes of support were rarely compared systematically. Inevitably, approaches and resources to support caregivers will differ significantly depending on the caregiver’s and care recipient’s circumstances, as well as their broader socioeconomic context, which may facilitate or hinder access to and benefit from support. The organisation and resourcing of the health and social care system will also clearly influence how support is delivered and sustained. Therefore, while this review identified a breadth of studies conducted across the world that may be instructive for other contexts, it is important to generate evidence which is sufficiently tailored to the health and social care system of interest. An additional complex issue is defining the ‘caregiver’ in

need of support, since many relatives or friends of people with chronic health conditions resist identifying as a carer or caregiver.^{25,58} Thus future research should carefully consider how caregiver support is framed, in order to engage groups who may not identify with the notion of ‘caring for’ an IVI.

The review suggests some clear gaps in the knowledge base around supporting caregivers of IVI. Firstly, no studies were identified specifically considering how forms of tangible assistance such as assistive technologies or low-vision aids for IVI may reduce stress among caregivers. Consideration of how assistive technologies may support older adults and their caregivers more generally is an emerging research area,⁵⁹ and it would be valuable to explore how new aids which can facilitate daily activities and mobility in the home environment may benefit caregivers alongside IVI themselves. Secondly, caregivers of people with many different chronic conditions or disabilities have been found to benefit from internet-based interventions providing professional and/or social support from peers.⁶⁰ However, no studies were identified in the present review exploring online-delivered support for caregivers of IVI, and this may be an avenue for future research. Thirdly, studies focussing on caregivers of adults with acquired vision loss tended to mostly focus on neovascular or “wet” AMD.^{15,35,42,49} Given that a significant component of support programmes involves education about the relevant condition and the specifics of how vision is affected, it may be useful to explore support for caregivers of people living with other acquired ocular or neurological conditions.

There are limitations to this review’s methodology, and potential issues around the narrow focus of its scope. Firstly, many studies exploring generic support for caregivers of people with a chronic illness or disability, or other diseases, were not included in this review but could yield useful, relevant findings for caregivers of IVI. Secondly, while this review was limited to studies exploring support for caregivers of IVI, the findings of studies documenting and measuring aspects of caregiver strain and distress may themselves help identify the most appropriate kind of support for the relevant population. The Supplement shows a number of the most relevant studies on caregiver burden and distress. These studies generally consider the implications for improving caregiver support at the end of their Discussion section. In general terms, they often advocate for more education, problem-solving

support, counselling and mental health support, and recommend healthcare or rehabilitation professionals meaningfully involving caregivers in decision-making.

In spite of these limitations, this review shows that research is increasingly exploring different modes of support for caregivers of IVI, in light of convincing evidence (see Supplement) of elevated distress that is prevalent among caregivers of IVI. Furthermore, some recent studies were identified which are primarily concerned with treatment or care of populations with vision impairment but are also including outcomes clearly focused on caregiver/parent/relative wellbeing. In common to many studies which investigate interventions, there are methodological and conceptual issues in researching caregiver support. These include: the inherent complexity and variety of individual caregivers' needs; the need for a regular, long-term commitment from research participants in structured programmes; and the difficulty of standardising "support" delivery and measurement in a way that is comparable across studies and contexts. Furthermore, studies often show a discrepancy between quantitative outcome measures and the subjective experience of caregivers,^{24,38,41} indicating profound differences between individual caregivers in which kinds of support are experienced as helpful or meaningful. Therefore, while studies are clear that structured, additional forms of support generally yield some benefit for caregivers of IVI, future research could valuably compare different modes of support (e.g. informational, emotional, tangible) for specific caregiver groups. Development of specific questionnaires or patient-reported outcome measures for caregivers of IVI – either one single measure, or distinct measures for caregivers of children and caregivers of adults with VI - could potentially help to better compare the benefits of different caregiver support modalities.

Conflicts of interest

None to declare.

Acknowledgements

The authors would like to acknowledge funding from the Eleanor Peel Trust. This review has been conducted as part of a study funded by the Eleanor Peel Trust titled: "Caring for the carers - what support exists for caregivers of people who are visually impaired: An exploratory study" led by Dr Ahalya Subramanian and Professor Christine Dickinson.

DRAFT

Appendix 1: Search terms

CINAHL (EBSCO)

- support OR (MH "Support, Psychosocial") OR (MH "Support Groups") OR (MH "Emotional Support (Saba CCC)") OR (MH "Coping Support (Saba CCC)") OR (MH "Labor Support") OR (MH "Caregiver Support") OR (MH "Social Support (Iowa NOC)") OR (MH "Social Support Index") OR (MH "Support Group (Iowa NIC)") OR (MH "Spiritual Support (Iowa NIC)") OR (MH "Young Adult Social Support Index") OR (MH "Bereavement Support (Saba CCC)") OR "support" OR (MH "Norbeck Social Support Questionnaire") OR (MH "Family Support (Iowa NIC)") OR (MH "Emotional Support (Iowa NIC)") OR (MH "Decision-Making Support (Iowa NIC)") OR (MH "Caregiver Support (Iowa NIC)") OR (MH "Financial Support")
AND
- caregiver OR caregiv* OR care givers OR carer OR family caregivers OR spouse caregivers OR caring OR (MH "Caregiver Burden") OR (MH "Caregiver Attitudes") OR (MH "Risk for Caregiver Role Strain (NANDA)") OR "caregiver" OR (MH "Caregiver Performance: Direct Care (Iowa NOC)") OR (MH "Caregiver Performance: Indirect Care (Iowa NOC)") OR (MH "Caregiver Physical Health (Iowa NOC)") OR (MH "Caregiver Role Strain (Saba CCC)") OR (MH "Caregiver Support") OR (MH "Family Caregiver Status (Iowa NOC)") OR (MH "Caregiver-Patient Relationship (Iowa NOC)") OR (MH "Caregiver Well-Being (Iowa NOC)") OR (MH "Caregiver Support (Iowa NIC)") OR (MH "Caregiver Stressors (Iowa NOC)") OR (MH "Caregiver Strain Index") OR (MH "Caregiver Role Strain (NANDA)")
AND
- visual impairment OR blind OR visually handicapped OR visually impaired OR low vision OR sight impairment OR sight impaired OR (MH "Deaf-Blind Disorders") OR (MH "Vision, Subnormal") OR (MH "Color Vision Defects") OR (MH "Vision Disorders") OR "vision loss" OR (MH "Glaucoma") OR "glaucoma" OR (MH "Macular Degeneration") OR (MH "Cataract") OR "cataract" OR (MH "Eye Diseases") OR (MH "Eye Diseases, Hereditary") OR

(MH "Diagnosis, Eye") OR "eye disease" OR (MH "Diabetic Retinopathy") OR "diabetic retinopathy" OR (MH "Blindness, Cortical") OR (MH "Blindness")

MEDLINE (OVID)

- (MH "Caregivers") OR "caregiver" OR caregiver or carer or family member or relative or informal caregiver
- (MH "Vision Disorders") OR (MH "Visually Impaired Persons") OR (MH "Education of Visually Disabled") OR "visual impairment" OR (MH "Blindness") OR "blindness" OR (MH "Blindness, Cortical") OR (MH "Deaf-Blind Disorders") OR (MH "Color Vision Defects") OR (MH "Vision Disorders") OR (MH "Glaucoma") OR "glaucoma" OR (MH "Macular Degeneration") OR (MH "Wet Macular Degeneration") OR (MH "Corneal Dystrophies, Hereditary") OR (MH "Geographic Atrophy") OR "macular" OR (MH "Macular Edema") OR (MH "Vitelliform Macular Dystrophy") OR (MH "Retinal Perforations") OR (MH "Optic Nerve Injuries") OR (MH "Optic Nerve Diseases") OR (MH "Optic Neuropathy, Ischemic") OR (MH "Optic Atrophy, Hereditary, Leber") OR "optic neuropathy" OR (MH "Cataract") OR "cataract" OR (MH "Eye Diseases") OR "eye disease" OR (MH "Diabetic Retinopathy") OR "diabetic retinopathy" OR (MH "Leber Congenital Amaurosis")
- (MH "Social Support") OR (MH "Psychosocial Support Systems") OR "support" OR (MH "Self-Help Groups") OR support OR supporting OR aid OR assistance OR help OR guidance OR resource

PsycINFO and PsychARTICLES (EBSCO)

- caregiver OR family member OR relative OR informal caregiver OR carer OR DE "Caregiver Burden" OR DE "Caregivers" OR DE "Respite Care"
- DE "Optic Neuritis" OR DE "Amblyopia" OR DE "Glaucoma" OR DE "Eye Disorders" OR DE "Vision Disorders" OR visual impairment OR blind OR visually handicapped OR visually impaired OR low vision OR sight impairment

- support OR supporting OR aid OR assistance OR help OR guidance OR resource OR advice OR DE "Social Support" OR DE "Support Groups"

DRAFT

References

1. Organization WH. Visual impairment and blindness Fact Sheet N 282. Retrieved June. 2014;21:2016.
2. Reinhardt JP. The importance of friendship and family support in adaptation to chronic vision impairment. *Journals Gerontol Ser B Psychol Sci Soc Sci*. 1996;51:P268–78.
3. Carers UK. Facts about carers [Internet]. 2019. Available from: https://www.carersuk.org/images/Facts_about_Carers_2019.pdf
4. Bourne RRA, Flaxman SR, Braithwaite T, et al. Magnitude, temporal trends, and projections of the global prevalence of blindness and distance and near vision impairment: a systematic review and meta-analysis. *Lancet Glob Heal* [Internet]. 2017 Sep 1;5:e888–97. Available from: [https://doi.org/10.1016/S2214-109X\(17\)30293-0](https://doi.org/10.1016/S2214-109X(17)30293-0)
5. Olusanya BO, Neumann KJ, Saunders JE. The global burden of disabling hearing impairment: a call to action. *Bull World Health Organ*. 2014;92:367–73.
6. Tarlow BJ, Wisniewski SR, Belle SH, et al. Positive aspects of caregiving: Contributions of the REACH project to the development of new measures for Alzheimer's caregiving. *Res Aging*. 2004;26:429–53.
7. Bambara JK, Owsley C, Wadley V, et al. Family caregiver social problem-solving abilities and adjustment to caring for a relative with vision loss. *Invest Ophthalmol Vis Sci* [Internet]. 2008/12/05. 2009 Apr;50:1585–92. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/19060279>
8. Rigby H, Gubitz G, Phillips S. A Systematic Review of Caregiver Burden following Stroke. *Int J Stroke* [Internet]. 2009 Aug 1;4:285–92. Available from: <https://doi.org/10.1111/j.1747-4949.2009.00289.x>
9. Bevans M, Sternberg EM. Caregiving burden, stress, and health effects among family caregivers of adult cancer patients. *Jama*. 2012;307:398–403.
10. Eppers L, Goodall D, Harrison BE. Caregiver burden among dementia patient

- caregivers: A review of the literature. *J Am Acad Nurse Pract* [Internet]. 2008 Aug 1;20:423–8. Available from: <https://doi.org/10.1111/j.1745-7599.2008.00342.x>
11. Kuriakose RK, Khan Z, Almeida DRP, et al. Depression and burden among the caregivers of visually impaired patients: a systematic review. *Int Ophthalmol*. 2017;37:767–77.
 12. Keeffe JE, Chou S-L, Lamoureux EL. The Cost of Care for People With Impaired Vision in Australia. *JAMA Ophthalmol* [Internet]. 2009 Oct 12;127:1377–81. Available from: <https://doi.org/10.1001/archophthalmol.2009.242>
 13. Marques AP, Macedo AF, Hernandez-Moreno L, et al. The use of informal care by people with vision impairment. *PLoS One* [Internet]. 2018 Jun 7;13:e0198631–e0198631. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/29879193>
 14. Schmier JK, Halpern MT, Covert D, et al. Impact of visual impairment on use of caregiving by individuals with age-related macular degeneration. *RETINA* [Internet]. 2006;26. Available from: https://journals.lww.com/retinajournal/Fulltext/2006/11000/IMPACT_OF_VISUAL_IMPAIRMENT_ON_USE_OF_CAREGIVING.12.aspx
 15. Gopinath B, Craig A, Kifley A, et al. Implementing a multi-modal support service model for the family caregivers of persons with age-related macular degeneration: a study protocol for a randomised controlled trial. *BMJ Open* [Internet]. 2017 Aug 1;7:e018204. Available from: <http://bmjopen.bmj.com/content/7/8/e018204.abstract>
 16. Lupón M, Armayones M, Cardona G. Quality of life among parents of children with visual impairment: A literature review. *Res Dev Disabil*. 2018;83:120–31.
 17. Bambara JK, Wadley V, Owsley C, et al. Family functioning and low vision: a systematic review. *J Vis Impair Blind*. 2009;103:137–49.
 18. Yesilkaya E, Best P, Byrne B, et al. The Barriers and Facilitators to the Implementation of Interventions for Children with Visual Impairments, Their

- Parents/Guardians or Educators: A Systematic Scoping Review. *Child Care Pract.* 2019;1–20.
19. van den Broek EGC, van Eijden AJPM, Overbeek MM, et al. A systematic review of the literature on parenting of young children with visual impairments and the adaptations for video-feedback intervention to promote positive parenting (VIPPP). *J Dev Phys Disabil.* 2017;29:503–45.
 20. Munn Z, Peters MDJ, Stern C, et al. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. *BMC Med Res Methodol* [Internet]. 2018;18:143. Available from: <https://doi.org/10.1186/s12874-018-0611-x>
 21. Tricco AC, Lillie E, Zarin W, et al. PRISMA extension for scoping reviews (PRISMA-ScR): checklist and explanation. *Ann Intern Med.* 2018;169:467–73.
 22. Helgeson VS. Social support and quality of life. *Qual life Res.* 2003;12:25–31.
 23. Cutrona CE, Suhr JA. Controllability of stressful events and satisfaction with spouse support behaviors. *Communic Res.* 1992;19:154–74.
 24. Larizza MF, Xie J, Fenwick E, et al. Impact of a low-vision self-management program on informal caregivers. *Optom Vis Sci.* 2011;88:1486–95.
 25. Enoch J, Dickinson CM, Subramanian A. What support do relatives or caregivers of people with vision impairment receive and require? A mixed-methods study. *Manuscr Prep.* 2020;
 26. Al-Janabi H, McLoughlin C, Oyebode J, et al. Six mechanisms behind carer wellbeing effects: A qualitative study of healthcare delivery. *Soc Sci Med* [Internet]. 2019;235:112382. Available from: <http://www.sciencedirect.com/science/article/pii/S0277953619303673>
 27. Watkins S, Clark T, Strong C, et al. The effectiveness of an intervener model of services for young deaf-blind children. *Am Ann Deaf.* 1994;139:404–9.
 28. Peters MDJ, Godfrey CM, Khalil H, et al. Guidance for conducting systematic scoping reviews. *Int J Evid Based Healthc.* 2015;13:141–6.
 29. Rahi JS, Manaras I, Tuomainen H, et al. Health services experiences of

- parents of recently diagnosed visually impaired children. *Br J Ophthalmol* [Internet]. 2005 Feb 1;89:213 LP-218. Available from: <http://bj.o.bmj.com/content/89/2/213.abstract>
30. Lawrence V, Murray J, ffytche D, et al. "Out of sight, out of mind": a qualitative study of visual impairment and dementia from three perspectives. *Int Psychogeriatrics* [Internet]. 2009;21:511–8. Available from: <https://www.cambridge.org/core/article/out-of-sight-out-of-mind-a-qualitative-study-of-visual-impairment-and-dementia-from-three-perspectives/97A2C1C165694FF43BC325EE263F8222>
 31. Shtein R, Newman-Casey P, Herndon L, et al. Assessing the Role of the Family/Support System Perspective in Patients With Glaucoma. *J Glaucoma*. 2016;25.
 32. Jackel B, Wilson M, Hartmann E. A survey of parents of children with cortical or cerebral visual impairment. *J Vis Impair Blind*. 2010;104:613–23.
 33. Rees G, Saw C, Larizza M, et al. Should family and friends be involved in group-based rehabilitation programs for adults with low vision? *Br J Vis Impair*. 2007;25:155–68.
 34. Sussman-Skalka C. Support group programs for partners of people with vision loss. *Generations*. 2003;27:98–101.
 35. Vukicevic M, Heraghty J, Cummins R, et al. Caregiver perceptions about the impact of caring for patients with wet age-related macular degeneration. *Eye* [Internet]. 2016;30:413–21. Available from: <https://doi.org/10.1038/eye.2015.235>
 36. Kyzar K. The relationship of perceptions of service and support adequacy to family quality of life for families of children with deafblindness. University of Kansas; 2010.
 37. Kyzar KB, Brady SE, Summers JA, et al. Services and supports, partnership, and family quality of life: Focus on deaf-blindness. *Except Child*. 2016;83:77–91.
 38. Rahi JS, Manaras I, Tuomainen H, et al. Meeting the needs of parents around

- the time of diagnosis of disability among their children: evaluation of a novel program for information, support, and liaison by key workers. *Pediatrics*. 2004;114:e477–82.
39. Cimarolli VR, Sussman-Skalka CJ, Goodman CR. “Program for Partners”: Support Groups for Partners of Adults with Visual Impairments. *J Vis Impair Blind*. 2004;98:90–8.
 40. Sussman-Skalka CJ. When Your Partner Becomes Visually Impaired: Helpful Insights and Tips for Coping [Internet]. Lighthouse International; 2002. Available from: http://li129-107.members.linode.com/downloads/publications/program_for_partners.pdf
 41. Doorey J, Douglas S, Viska L, et al. Care for the Carers: A Self-management program for carers of people with vision impairment [Internet]. Care for the carers. Centre for Research into Disability and Society within the Curtin Health ...; 2009. Available from: <http://hdl.handle.net/20.500.11937/24639>
 42. Pozet A, Lejeune C, Bonnet M, et al. Evaluation of efficacy and efficiency of a pragmatic intervention by a social worker to support informal caregivers of elderly patients (The ICE Study): study protocol for a randomized controlled trial. *Trials* [Internet]. 2016 Nov 3;17:531. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/27881145>
 43. Khooshab E, Jahanbin I, Ghadakpour S, et al. Managing parenting stress through life skills training: A supportive intervention for mothers with visually impaired children. *Int J community based Nurs midwifery*. 2016;4:265.
 44. Dale NJ, Sakkalou E, O’Reilly MA, et al. Home-based early intervention in infants and young children with visual impairment using the Developmental Journal: longitudinal cohort study. *Dev Med Child Neurol*. 2019;61:697–709.
 45. Chen H, Lin Z, Chen J, et al. The impact of an interactive, multifaceted education approach for congenital cataract on parental anxiety, knowledge and satisfaction: A randomized, controlled trial. *Patient Educ Couns* [Internet]. 2019; Available from: <http://www.sciencedirect.com/science/article/pii/S0738399119303830>

46. McConnell J. Parents, Adolescents, and Career Plans of Visually Impaired Students. *J Vis Impair Blind* [Internet]. 1999 Aug 1;93:498–515. Available from: <https://doi.org/10.1177/0145482X9909300803>
47. Barbieri MC, Broekman GVDZ, Souza ROD de, et al. Support network for families of children and adolescents with visual impairment: strengths and weaknesses. *Cien Saude Colet*. 2016;21:3213–23.
48. Dewald HP, Faris C, Borg KS, et al. Expanding the frontiers of orientation and mobility for infants and toddlers in New Mexico and Utah. *J Vis Impair Blind*. 2015;109:502–7.
49. Gohil R, Crosby-Nwaobi R, Forbes A, et al. Caregiver Burden in Patients Receiving Ranibizumab Therapy for Neovascular Age Related Macular Degeneration. *PLoS One*. 2015;10:e0129361–e0129361.
50. Hanemoto T, Hikichi Y, Kikuchi N, et al. The impact of different anti-vascular endothelial growth factor treatment regimens on reducing burden for caregivers and patients with wet age-related macular degeneration in a single-center real-world Japanese setting. *PLoS One*. 2017;12:e0189035.
51. Gothwal VK, Bharani S, Mandal AK. Impact of surgery on the quality of life of caregivers of children with congenital glaucoma. *Ophthalmology*. 2016;123:1161–2.
52. Sörensen S, Pinquart M, Duberstein P. How Effective Are Interventions With Caregivers? An Updated Meta-Analysis. *Gerontologist* [Internet]. 2002 Jun 1;42:356–72. Available from: <https://doi.org/10.1093/geront/42.3.356>
53. Ashworth M, Baker AH. “Time and space”: carers’ views about respite care. *Heal Soc Care Community*. 2000;8:50–6.
54. Haley WE, Brown SL, Levine EG. Experimental Evaluation of the Effectiveness of Group Intervention for Dementia Caregivers¹. *Gerontologist* [Internet]. 1987 Jun 1;27:376–82. Available from: <https://doi.org/10.1093/geront/27.3.376>
55. Haslam SA, Jetten J, Postmes T, et al. Social Identity, Health and Well-Being: An Emerging Agenda for Applied Psychology. *Appl Psychol* [Internet]. 2009 Jan 1;58:1–23. Available from: <https://doi.org/10.1111/j.1464->

0597.2008.00379.x

56. Knight BG, Lutzky SM, Macofsky-Urban F. A Meta-analytic Review of Interventions for Caregiver Distress: Recommendations for Future Research1. *Gerontologist* [Internet]. 1993 Apr 1;33:240–8. Available from: <https://doi.org/10.1093/geront/33.2.240>
57. Hoddinott P, Allan K, Avenell A, et al. Group interventions to improve health outcomes: a framework for their design and delivery. *BMC Public Health* [Internet]. 2010;10:800. Available from: <https://doi.org/10.1186/1471-2458-10-800>
58. Knowles S, Combs R, Kirk S, et al. Hidden caring, hidden carers? Exploring the experience of carers for people with long-term conditions. *Health Soc Care Community*. 2016;24:203–13.
59. Ben Mortenson W, Demers L, Fuhrer MJ, et al. Effects of a caregiver-inclusive assistive technology intervention: a randomized controlled trial. *BMC Geriatr* [Internet]. 2018 Apr 18;18:97. Available from: <https://www.ncbi.nlm.nih.gov/pubmed/29669536>
60. Guay C, Auger C, Demers L, et al. Components and outcomes of internet-based interventions for caregivers of older adults: systematic review. *J Med Internet Res*. 2017;19:e313.

Table 1. The five categories of support defined by Cutrona and Suhr (1992)

Type of support	Definition
Informational support	Advice, information or guidance from health and social care professionals, charities or informed peers. This could include referral to another professional, or signposting to helpful resources. It could also include education/training sessions about aspects of caregiving and/or visual impairment.
Tangible assistance	Practical support, which might include financial support (e.g. benefits or loans), respite care, or assistive technologies.
Esteem support	Positive enhancement of recipients' feelings about themselves and boosted confidence in their abilities. In Cutrona and Suhr's typology, this might include validation, or relief from feelings of guilt or self-blame about the situation.
Emotional support	Ranging from formal psychological therapy or counselling, to reassurance, listening, empathy or encouragement from family and friends.
Network support	A sense of belonging from access to, or the presence or availability of, supportive companions who share a similar experience. This category would include membership of a support group or an online forum (which itself could be a route to other kinds of support).

Table 2. Summary of main characteristics of included publications. For each category, studies involving caregivers of adults with VI are listed first (alphabetically by first author), followed by studies involving caregivers of children with VI.

<i>Studies exploring caregiver support needs (n = 8)</i>					
Author/year	Country of first author	Study design	Study population	Objective	Issue/intervention/outcome studied
Lawrence et al., 2009 ³⁰	UK	Qualitative	17 older adults with VI and dementia, 17 family caregivers, and 18 care professionals	Investigate the experiences and needs of older adults with VI and dementia, and their caregivers.	Issue: The lived experiences of people with dementia and serious VI, their family caregiver, and the care professionals with whom they have contact.
Rees et al., 2007 ³³	Australia	Qualitative	21 participants with low vision, and 64 vision rehabilitation professionals	Investigate the views of clients with low vision and vision rehabilitation professionals regarding the involvement of family and friends in group-based rehabilitation programmes.	Issue: advantages and disadvantages to involving “significant others” of people with low vision in rehabilitation groups.

Shtein et al., 2016 ³¹	United States	Qualitative	31 participants: 21 were family members/friends of glaucoma patients with poor vision; 10 were family members/friends of patients with good vision	Investigate the role of the family/friends support system for patients with glaucoma and their perspective on barriers to effective glaucoma management.	Issue: the role of the patients' support systems in the relationship between patient, doctor, and disease.
Sussman-Skalka, 2003 ³⁴	United States	Descriptive	Sighted partners of people with vision loss	Outline a programme developed specifically to address the needs of partners of people with VI.	Issue: different models of support groups for partners of people with vision loss, and caregiving issues raised by people with vision loss and their sighted partners.
Vukicevic et al., 2016 ³⁵	Australia	Qualitative	643 caregivers of people with neovascular (wet) AMD	Explore the perceptions of caregivers of persons with neovascular AMD in relation to the most important aspects of caring.	Issue: experiences of caring for someone with AMD, elicited with two open-ended questions: 1. Do you have any other comments about caring for someone with wet AMD that you believe are important for other people to know and understand?

					2. What are the three most important aspects of caring for someone with wet AMD for you?
Jackel et al., 2010 ³²	United States	Cross-sectional	80 parents of children with cortical or cerebral visual impairment (CVI)	Explore how a CVI diagnosis is received and supports that are provided after the diagnosis; the educational supports received by children with CVI; and the parents' perceptions of the supports that they and their children receive.	Issue: How do parents receive their children's diagnosis of CVI, and what supports are provided upon the diagnosis? What educational supports are children with CVI receiving? What do the parents of children with CVI feel about the supports that they and their children receive?
Kyzar, 2010 ³⁶	United States	Cross-sectional	227 families of children with deafblindness (between the ages of birth and 22)	Explore the relationship between families' perceptions of supports and services and family quality of life (FQOL) for families of children with deafblindness.	Outcome measure: Service Adequacy Scale for Families of Children who are Deafblind (SAS-DB) questionnaire. SAS-DB evaluates family members' perceptions of how effectively services (grouped into seven domains) have been supporting their child's and family's needs over the previous 12 months.

Rahi et al., 2005 ²⁹	UK	Mixed-methods service evaluation	Parents of 147 children recently diagnosed with VI	Investigate the health service experiences and needs of parents in the period around diagnosis of ophthalmic disorders in their children.	Outcome measures: 1. Client Satisfaction Questionnaire, to measure overall care satisfaction; 2. Measure of Processes of Care (MPOC), with five subscales assessing the processes (rather than content) of care.
<i>Novel interventions supporting caregivers of IVI (n = 10)</i>					
Author/year	Country of first author	Study design	Study population	Objective	Issue/intervention/outcome studied
Cimarolli et al., 2004 ³⁹	United States	Pre-test/post-test	32 individuals (average age, 69) who were living with partners with VI from acquired eye disease	Evaluate support groups for partners of adults with VI. The goals of these groups included alleviating stress and burdens, improving the sighted partners' understanding of the issues faced by the partners with VI, and enhancing communication.	Intervention: four different support group models, one with both the IVI and sighted partner, one in person with sighted partner only, one by telephone with sighted partner only, one self-directed with sighted partner only. Outcomes measured for sighted partner: Depressive symptoms; Life satisfaction;

					Relationship satisfaction; Understanding of what IVI could see; Role Captivity Scale score; Satisfaction with programme.
Doorey et al., 2009 ⁴¹	Australia	Mixed-methods service evaluation	11 participants in the <i>Care for the Carers</i> programme developed by Curtin University	Evaluate the content and delivery of the programme from the perspective of participants. A second aim was to evaluate the outcomes of the programme to determine if the programme was associated with improvements in perceived quality of life, knowledge self-efficacy in relation to vision loss, and resilience.	<p>Intervention: A six week psychoeducational group programme, with modules on: Introduction to Self-management and Understanding Vision Loss Orientation and Exploration; Making the Most of Remaining Vision and Using Other Senses; Orientation and Mobility (O&M); Taking Care of Ourselves; Exploring Emotional Issues; Resources for Caring – Now and in the Future.</p> <p>Outcomes:</p> <ol style="list-style-type: none"> 1. Participants' perceptions of the importance and relevance of the information in each session; the extent to which delivery of the information assisted their learning; and overall satisfaction with the session.

					2. Pre- and post-programme scores on: Two items measuring quality of life; the knowledge subscale of the Age Related Vision Loss Self Efficacy Scale, adapted for carers; and the Resilience Scale RS15.
Gopinath et al., 2017 ¹⁵	Australia	Trial protocol	360 caregiver–patient dyads (180 in each of the intervention and wait-list control groups)	Implement and evaluate an innovative, multi-modal support service programme that aims to empower family caregivers by improving their coping strategies, enhancing hopeful feelings such as self-efficacy and helping them make the most of available sources of social and financial support.	<p>Intervention: (1) mail-delivered cognitive behavioural therapy designed to improve psychological adjustment and adaptive coping skills; (2) telephone-delivered group counselling sessions allowing caregivers to explore the impacts of caring and share their experiences; and (3) education on available community services/resources, financial benefits and respite services.</p> <p>Outcomes: primary outcome is reduction in caregiver burden. Secondary outcomes include improvements in caregiver mental wellbeing, quality of life, fatigue and self-efficacy. Economic analysis will inform</p>

					whether this intervention is cost-effective, and if it is feasible to roll out this service on a larger scale.
Larizza et al., 2011 ²⁴	Australia	Pre-test/post-test	60 caregivers (mean (SD) age = 67.2 (14.8) years) of adults with low vision	Evaluate caregivers' experiences and outcomes following attendance at a patient-centred group-based self-management program called "Living with Low Vision" (LLV).	<p>Intervention: The LLV programme consisted of three-hourly sessions for eight consecutive weeks. The group discussed strategies to adapt, visual aids and assistive technologies, the emotional aspects of vision loss, O&M, problem solving and goal planning. The programme also included a self-help pack containing notes on each session and further resources (including contact details of relevant organisations).</p> <p>Outcomes: impact of the programme on single-item indicators that assessed caregiver levels of understanding of low vision, awareness of devices, aids and practical strategies. Questionnaires to assess: confidence to deal with low vision; self-efficacy; emotional wellbeing.</p>

Pozet et al., 2016 ⁴²	France	Trial protocol	1684 caregivers of people with AMD	Define profiles of caregivers of older patients with a diagnosis of AMD (along with other chronic diseases), generating longitudinal measures of quality of life, burden, depression, coping strategies, and social support. Additionally, evaluate the efficacy and efficiency of the implementation of a pragmatic intervention by a social worker to help informal caregivers, through a randomized interventional trial nested in the cohort.	Intervention: evaluate the effect of a pragmatic supportive intervention on caregivers, provided by a social worker and an information booklet (intervention arm) versus the control arm, where caregivers will only receive the information booklet, without social worker support. Outcomes: mental and physical health summary scores on the Medical Outcome Study Short Form (MOS SF)-36 at 1 year across the two groups. Secondary outcomes include inter-group comparison of: (1) mental and physical health summary scores of the MOS SF-36 at 2 years; (2) all health-related QoL dimensions of the MOS SF-36 at 1 year and at 2 years; (3) coping strategies; (4) Hospital Anxiety and Depression Scale scores; (5) Social Support Questionnaire SF 6; (6) Zarit Caregiver Burden Interview scores; (7) the incremental
-------------------------------------	--------	-------------------	---------------------------------------	---	--

					cost-effectiveness ratio of the social worker intervention.
Chen et al., 2019 ⁴⁵	China	RCT	177 parents of children with congenital cataract	Examine the impact of a patient education program for parents of children with congenital cataract on parental stress, comprehension of disease information, and parental satisfaction.	Intervention: a health education program with a multifaceted, interactive approach, involving a 2-hour lecture and 1-hour workshop using active learning strategies. Outcomes: Parenting Stress Index (PSI) and Ocular Treatment Index (OTI) scores.
Dale et al., 2019 ⁴⁴	UK	Longitudinal	54 infants with congenital disorders of the peripheral visual system (CDPVS) and profound-severe VI. 39 parents returned data.	Investigate the effects of home-based early intervention in children with severe visual impairment using the Developmental Journal for babies and young children with visual impairment (DJVI).	Intervention: use of the DJVI, an early childhood intervention programme for babies and children with severe VI (compared with those receiving “Other Support”). The DJVI includes a comprehensive developmental curriculum specifically focused on visual impairment disability needs, regular structured developmental monitoring and goal-setting, and activity guidance for parent-child everyday interaction and tasks.

					Outcomes measured for parent: Parenting Stress Index – Short Form scores; Family–Professional Partnership scale scores.
Khooshab et al., 2016 ⁴³	Iran	RCT	52 mothers with visually impaired children studying at an educational complex in Shiraz, Iran in 2013	Investigate the effect of life skills training (LST) program on parenting stress of mothers with children with VI aged 7 to 12 years.	Intervention: Life skills training, consisting of five 2-hour sessions across five consecutive weeks. Outcome: Parenting Stress Index scores, completed three times by the participants of both intervention and control groups (before, immediately after, and one month after the intervention).
McConnell, 1999 ⁴⁶	Canada	Mixed-methods service evaluation	20 visually impaired students and their parents	Describe/evaluate a model programme that included parents of students with VI in structured career planning exercises.	Intervention: A career development programme consisting of four manuals: a parents' guidance manual, an activity exploration workbook, a career-decision framework, and a planning manual. Outcomes measured: The Career Decision Scale's subscales about Career Certainty and Career Indecision; the Parent-Adolescent

					Communication scale; and the Career Salience Scale. Qualitative data regarding parents' views on the career development programme were also collected.
Rahi et al., 2004 ³⁸	UK	Mixed-methods service evaluation	79 families of visually impaired children (corrected acuity of 6/18 or worse in the better eye) in the pre-CLT group and 68 families in the post-CLT group	Report on the impacts of a novel, hospital-based, key worker service (Community Link Team [CLT]) at Great Ormond Street Hospital on the experiences of parents and the practices of healthcare professionals.	Intervention: The CLT was present during the first outpatient assessment by the consultant ophthalmologist and accompanied the family during other assessments performed during that visit. A dedicated room was used by the CLT members to spend time with each family after completion of the clinical assessments. The CLT members reiterated and/or clarified clinical information already provided, specifically advised the families about visual stimulation programs and the benefits and purpose of VI certification, and provided information about educational and social services. The same CLT member met the family at subsequent visits to the department and acted as the first point of contact for parents.

					Outcomes: Measure of Processes of Care (MPOC), specifically developed and used to assess parents' views of the degree to which health services for a range of childhood disorders are family-centred; the short form of the Client Satisfaction Questionnaire; qualitative data from in-depth interviews.
<i>Evaluating usual care (n = 2)</i>					
Author/year	Country of first author	Study design	Study population	Objective	Issue/intervention/outcome studied

Barbieri et al., 2016 ⁴⁷	Brazil	Qualitative	18 family members of children and adolescents with VI	Understand the interactions established between social support networks and families of children and adolescents with VI, in two different cities in the state of Sao Paulo, Brazil.	Issue: family members' access to other members of their own family, friends, spiritual and cultural activities, health services, government institutions, and philanthropic organizations as support networks.
Dewald et al., 2015 ⁴⁸	United States	Descriptive	Young children with VI and their families	Represent the perspectives of two states that have recognized the need for specialised O&M services in their early intervention programmes for young children with VI and their families.	Issue: the impact of O&M services on families of children with VI.

Studies exploring how treatment for IVI or people with eye disease directly impacts the caregiver (n = 3)

Author/year	Country of first author	Study design	Study population	Objective	Issue/intervention/outcome studied
--------------------	--------------------------------	---------------------	-------------------------	------------------	---

Gohil et al., 2015 ⁴⁹	UK	Cross-sectional	250 matched dyads of patients with AMD and their caregivers	Assess the caregiver burden and factors determining the burden in patients receiving ranibizumab therapy for neovascular AMD.	<p>Outcomes measured:</p> <ol style="list-style-type: none"> 1. Subjective caregiver burden, measured using caregiver reaction assessment scale; 2. Objective caregiver burden, determined by the caregiver tasks and level of care provided; 3. Factors predicting caregiver burden, including satisfaction and support provided by the healthcare service.
Hanemoto et al., 2017 ⁵⁰	Japan	RCT	71 pairs of patients with wet AMD and their caregivers	Evaluate how different kinds of anti-vascular endothelial growth factor (VEGF) treatment regimens affect caregiver burden.	<p>Intervention: the 'treat-and-extend' (T&E) regimen, which tailors the treatment to the patient's schedule, was compared with a treatment as needed or 'PRN' regimen.</p> <p>Outcomes measured: Burden Index of Caregivers (BIC-11), and depressive symptoms as assessed by the Centre for Epidemiologic Studies Depression scale.</p>

Gothwal et al., 2016 ⁵¹	India	Pre-test/ post-test	111 caregivers of children with unilateral (24%) or bilateral (76%) primary congenital glaucoma (PCG)	Investigate the changes in the QoL of caregivers of children with PCG after glaucoma surgery.	Outcome measured: Caregivers' scores on the Caregiver's Congenital Glaucoma Quality of Life (CarCGQoL) before and after surgery.
------------------------------------	-------	------------------------	---	---	--

Table 3. Clinical trials with outcomes relevant to caregivers of VIP

Trial centre	Trial name	Location	Caregiver outcomes measured	Web link
Children's Hospital Medical Center, Cincinnati	Remote Access: Cortical Visual Impairment	United States	<ul style="list-style-type: none"> Caregiver questionnaire responses regarding the number of sessions, the set up, and their child's improvement 	https://clinicaltrials.gov/ct2/show/NCT03957980
Johns Hopkins University	Improving Communication with Formal/Informal Caregivers among Older Adults with Dual Sensory Impairment: Feasibility of Hearing Intervention in a Low Vision Rehabilitation Clinic	United States	<ul style="list-style-type: none"> 12 item Zarit Burden Interview Questionnaire (ZBI-12) to assess burden of care before and after intervention Views on the feasibility of the intervention, elicited through semi-structured interviews 	https://clinicaltrials.gov/ct2/show/NCT03664349
Lawson Health Research Institute	Low-vision Rehabilitation Programme for Low-vision Patients and Care Givers	Canada	<ul style="list-style-type: none"> Health-related QoL Depression using the Patient Health Questionnaire (PHQ-9) Anxiety using Generalized Anxiety Disorder (GAD-7) Veterans Affairs Low Vision Visual Functioning Questionnaire (VA LV VFQ-48) <p>(Measures will be taken at the first study visit and after the low-vision rehabilitation programme.)</p>	https://clinicaltrials.gov/ct2/show/NCT03166072
University of Manchester	The SENSE-Cog Randomised Controlled Trial: Comparing	Cyprus France Greece	<ul style="list-style-type: none"> Mental wellbeing and quality of life, measured using 	http://www.isrctn.com/ISRC

	<p>individualised sensory intervention to standard care to improve quality of life in people with dementia and their companions</p>	<p>Ireland UK</p>	<p>General Health Questionnaire-12</p> <ul style="list-style-type: none"> • General mental and emotional health, measured using 12 item Short Form Survey • Depression and anxiety, measured using Hospital Anxiety and Depression Scale • Caregiving experience (relationships with the person with dementia), measured using Family Caregiving Role and Relationship Satisfaction Scale 	<p>TN17056 211</p>
--	---	-----------------------	--	--

DRAFT

Table 4. Summary of key findings from the 23 studies

<p>Caregivers of IVI are a highly heterogeneous group, whose needs will vary according to: the severity of visual impairment or eye disease; co-morbidities (e.g. learning disabilities; dementia); geographical location; caregiving role (e.g. primary vs secondary caregiver); and socio-demographic characteristics (e.g. ethnicity), among other factors.</p>
<p>Structured, time-limited group programmes may be particularly beneficial for improving caregivers' knowledge and awareness about visual impairment, and providing informational and network support. However, in terms of emotional support, they show more limited benefit for broader psychosocial outcomes such as life satisfaction, quality of life, resilience, or emotional wellbeing.</p>
<p>Support interventions should carefully weigh the relative benefits and risks of involving caregivers and adults with VI in the same group.</p>
<p>Structured, time-limited interventions supporting parents of children with VI can lead to long-term reductions in parenting stress, as measured by the Parenting Stress Index.</p>
<p>Interventions to improve the wellbeing of IVI show benefits for caregivers, but it is unclear how these benefits compare to dedicated modes of support for the caregiver.</p>
<p>Information <i>per se</i> is not automatically helpful or supportive for caregivers. It is important that informational support is personalised, meaningful, and presented in a clear, engaging way that avoids overwhelming the caregiver.</p>
<p>“Support” is a challenging concept to measure quantitatively, and the level of support received may not reflect how supported caregivers feel subjectively.</p>

Figures

Figure 1. PRISMA diagram showing study selection process

Figure 2. Included studies by year of publication

Figure 3. Study designs of included publications

Figure 4. Map showing number of studies by country

DRAFT