The Sight Loss and Vision Priority Setting Partnership (SLV-PSP): Overview and results of the research prioritisation process.

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The Sight Loss and Vision Priority Setting Partnership (SLV-PSP): Overview and results of the research prioritisation process.

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Abstract

Objectives: The Sight Loss and Vision Priority Setting Partnership aimed to identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians. These priorities can be used to inform funding bodiesdecisions and enhance the case for additional research funding.

Design: Prospective survey with support from the James Lind Alliance.

Setting: UK wide NHS and non-NHS.

Participants: Patients, carers and eye health professionals. Academic researchers were excluded. The survey was disseminated by patient groups, professional bodies, at conferences and through the media, and was available for completion online, by phone, by post and by alternative formats (Braille and audio).

Outcome measure: People were asked to submit the questions about prevention, diagnosis and treatment of sight loss and eye conditions that they most wanted to see answered by research. Returned survey questions were reviewed by a data analysis group and out of scope/duplicate/answered questions removed. The remaining questions went through an interim prioritisation process. The top priorities were established across eye disease categories at final workshops.

Results: 2,220 people responded generating 4,461 submissions. Sixty-five percent of respondents had sight loss and/or an eye condition. Following initial data analysis, 686 submissions remained which were circulated for interim prioritisation (excluding cataract and ocular cancer questions) to 446 patients/carers and 218 professionals. The remaining 346 questions were discussed at final prioritisation workshops to reach agreement of top questions per category.

Conclusions: The exercise engaged a diverse community of stakeholders generating a wide range of conditions and research questions. Top priority questions were established across 12 eye disease categories.

Keywords: Sight loss; Vision; Research; Priorities; Partnership; James Lind Alliance

Article summary

Strengths and limitations of this study:

- Wide ranging and comprehensive coverage
- Substantial response
- The hardest to reach and those with the least opportunity to be heard may indeed have not been heard
- Any such groups now feeling excluded should have an opportunity to redress this.

Introduction

In the UK it is estimated that almost 2 million people are affected by sight loss and this number is expected to double by 2050[1]. Currently 50% of sight loss in the UK is avoidable but there are also many unavoidable sight loss conditions. Whether it is to address childhood eye conditions or those affecting adults, research is needed to inform us about prevention, to develop new techniques for early diagnosis and to develop new and more effective treatments for many eye conditions.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP) was developed from earlier work by the Eye Research Group (ERG) of VISION 2020 UK whose mission is the elimination of avoidable sight loss and the amelioration of the effects of sight loss when it cannot be avoided[2]. The UK Vision Strategy sets out ways of addressing avoidable sight loss in the UK. There is much that can be done to improve the nation's eye health, and to eliminate avoidable sight loss. There are however gaps in the evidence base regarding eye health interventions and the best way to deliver services. Research is needed to support the UK Vision Strategy. In key areas research is urgently required to enable the Vision Strategy to be implemented in an evidence-based way that ensures efficient and effective development.

Despite on-going research in the UK and other countries, there are still many questions about the prevention, diagnosis and treatment of sight loss and eye conditions that remain unanswered. Given that resources for research are limited, it

is important for research funders to understand how patients, carers and eye health professionals prioritise these unanswered questions so that future research can be consolidated and targeted accordingly[3].

The purpose of this project was to undertake a comprehensive, UK-wide, survey of patients, carers and clinicians to identify research questions and priorities to inform decisions of funding bodies and enhance the case for additional research funding.

The priority setting process has been well established by the James Lind Alliance (JLA) (http://www.lindalliance.org/) which has supported partnerships on a range of topics since 2004. This partnership initiated by Fight for Sight, the UK's leading eye research charity was established with support, financial and in kind, from the College of Optometrists, the Royal College of Ophthalmologists, the NIHR Biomedical Research Centre for Ophthalmology, the RNIB, UK Vision Strategy and the Cochrane Eyes and Vision Group. A representative from the JLA convened meetings of the steering committee and provided independent chairmanship for this and the priority setting workshops. Their extensive experience in this process ensured no single voice exerted undue influence over the prioritisation process and that the views of patients, their carers and clinicians were paramount. The views of researchers with no clinical involvement with patients and views of commercial organisations were not included.

Methods and materials

The detailed methods for this prioritisation process have been described in detail elsewhere[4]. In brief, the process comprised five stages (figure 1).

Establishing the SLV-PSP

A steering committee and data assessment group comprising the authors of this article oversaw the process. Each member was responsible for contributing to and managing a part of the process and was selected for their expertise and association with eye research. The steering committee also included patient representatives and eye health professionals.In April 2012 an initial stakeholder meeting was held to engage the groups and organisations with member bases and community influence.

This was to ensure that the initial survey would be disseminated and completed by as many patients, relatives, carers and eye health professionals as possible in the UK.

Main survey

The SLV-PSP survey was launched on 1 May 2012 and was open until 31 July 2012. The aim of the survey was to identify patients', carers' and eye health professionals' unanswered questions about sight loss and eye conditions. The survey's primary question was "What question(s) about the prevention, diagnosis and treatment of sight loss and eye conditions would you like to see answered by research?"

Data analysis

Following closure of the survey, all submissions were examined. Out-of-scope submissions were removed including those not related to the topic and uncertainties better suited to social research. In-scope uncertainties were allocated into disease-specific groups and re-worded in PICO format (Population, Intervention, Comparison, Outcome). Searches were then undertaken to ascertain whether or not each uncertainty could be answered by an up-to-date systematic review.All unanswered uncertainties were then allocated to one of 12 eye disease categories, with duplicates removed and similar questions combined. The 12 categories were formed following discussions by the steering group on the most logical and pragmatic way to organise the data within the time and resources available.

Interim prioritisation

In order to start reducing the number of uncertainties, an interim prioritisation exercise was conducted over email and by post. Patients, carers and eye health professionals were invited to examine the long lists and then choose and rank 10 of the uncertainties.

Final prioritisation

The remaining uncertainties were ranked by patients, carers, relatives, organisation representatives and eye health professionals in one-day workshops facilitated by the JLA, using Nominal Group Technique – a mix of discussion and ranking. For each category, the top 10/11 questions were agreed.

Results

Main survey

In response to the survey, 2220 people generated 4461 submissions. Of these respondents, 17% identified themselves as healthcare professionals including primarily ophthalmologists, optometrists, orthoptists, ophthalmic nurses, opticians and people working in social care and rehabilitation (figure 2). Over 60% were people with sight loss or an eye condition. The average age of survey participants was 65.7 years old (range:16 months (proxy completion of survey by adult carer)to 105 years). Just under two thirds (62%) of respondents were female. The geographical split was: England 89%; Scotland 6%; Wales 4%; Northern Ireland 1%.

Data analysis

Following data analysis to remove duplicate/answered/out of scope uncertainties, 686 uncertainties remained. These were divided into twelve eye disease categories. Table 1 shows each category with the initial number of submissions received after the survey responses were submitted, the number of uncertainties sent to interim prioritisation, the number of participants at interim prioritisation and the number of uncertainties considered at the final prioritisation workshops.

Interim prioritisation

Respondents from the initial survey, organisations and eye healthcare professionals with expertise in the eye diseases in ten categories were contacted to provide interim priority rankings. The smaller number of questions asked in the categories relating to cataract and ocular cancer meant that an interim prioritisation exercise was not required for either category. A large response was received for the interim exercise, with input from 446 patients, carers and relatives plus 218 eye health professionals. Uncertainties accumulated scores based on rank and frequency, resulting in short lists of around 30 uncertainties per category which were taken to final workshops.

Final prioritisation

In April and May 2013, 12 final prioritisation workshops were held: one for each eye disease category. Balanced numbers of patients/carers/relatives and eye heath professionals participated. In total, 155 participants attended across all 12 workshops: 78 patients and 77 clinicians (table 2). The topicsdebated at each workshop comprised between 19-31 questions per category. Overall 153 questions about sight loss and vision were considered resulting in lists of 10/11priorities for each of the 12 categories:

Age-related macular degeneration

1. Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?
2. What is the cause of AMD?
3. How can AMD be prevented?
4. Are there ways of restoring sight loss for people with AMD?
5. Can the development of AMD be predicted?
6. What is the most effective way to detect and monitor the progression of early AMD?
7. What factors influence the progression of AMD?
8. Can a non-invasive therapy be developed for wet AMD?
9. Can dietary factors, nutritional supplements, complementary therapies or lifestyle changes prevent or slow the progression of AMD?
10. What are the best enablement strategies for people with AMD?

Cataract

1. How can cataracts be prevented from developing?
2. Can the return of cloudy or blurred vision after cataract surgery known as posterior capsule opacity (PCO) or secondary cataract be prevented?
3. How can cataract progression be slowed down?
4. What alternatives to treat cataracts other than cataract surgery are being developed?
5. What is the cause of cataract?
6. How can cataract surgery outcomes be improved?
7. How safe and effective is laser assisted cataract surgery?
8. Should accommodative lenses be developed for cataract surgery?

9. What is the best measure of visual disability due to cataract?
10. Can retinal detachment be prevented after cataract surgery?
11. What are the outcomes for cataract surgery among people with different levels of cognitive impairment (whatever the cause but including dementia, stroke, neurological conditions, head injuries)?

Childhood-onset disorders

1. How can cerebral visual impairment be identified, prevented and treated in children?
2. How can treatment for visual pathway damage associated with pre-term birth be developed?
3. How do we improve screening and surveillance from the ante-natal period through to childhood to ensure early diagnosis of impaired vision and eye conditions?
4. Can the treatment of amblyopia be improved to produce better short and long term outcomes than are possible with current treatments?
5. How can cataract be prevented in children?
6. What are the causes of coloboma and microphthalmia/anophthalmia and how can they be prevented?
7. Can vision be corrected in later life for people with amblyopia?
8. How can retinoblastoma be identified, prevented and treated in children?
9. Can better treatments for glaucoma in children be developed?
10. Can a treatment be developed to improve vision for people with albinism?

Corneal and external diseases

1. Can new therapies such as gene or stem cell treatments be developed for corneal diseases?
2. What is the most effective management for dry eye and can new strategies be developed?
3. Can treatments to save eye sight from microbial keratitis be improved?
4. How can the rejection of corneal transplants be prevented?
5. Can the outcomes of corneal transplantation be improved?
6. What causes keratoconus to progress and can progression be prevented?
7. Can non-surgical therapy be developed for Fuchs' corneal dystrophy?

8. Can corneal infections be prevented in high-risk individuals such as contact lens wearers?
9. What is the cause of keratoconus and can it be prevented?
10. What is the most effective management of ocular complications associated with Stevens Johnson Syndrome?
11. Can severe ocular surface disease in children, such as blepharokeratoconjunctivitis and vernal keratoconjunctivitis be managed better?

Glaucoma

1. What are the most effective treatments for glaucoma and how can treatment be improved?
2. How can loss of vision be restored for people with glaucoma?
3. How can glaucoma be stopped from progressing?
4. What can be done to improve early diagnosis of sight-threatening glaucoma?
5. What causes glaucoma?
6. What is the most effective way of monitoring the progression of glaucoma?
7. How can glaucoma patients with a higher risk to progress rapidly be detected?
8. Why is glaucoma more aggressive in people of certain ethnic groups, such as those of West African origin?
9. How can glaucoma be prevented?
10. Is there a link between treatment adherence and glaucoma progression and how can adherence be improved?

Inherited retinal diseases

1. Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?
2. How can sight loss be prevented in an individual with inherited retinal disease?
3. Is a genetic (molecular) diagnosis possible for all inherited retinal diseases?
4. What factors affect the progression of sight loss in inherited retinal diseases?
5. What causes sight loss in inherited retinal diseases?
6. What is the most effective way to support patients with inherited retinal disease?

7. Can the diagnosis of inherited retinal diseases be refined so that individuals can be given a clearer idea about their specific condition and how it is likely to progress?
8. What is the relationship between sight loss and mental health for people with inherited retinal diseases?
9. Would having a treatment for an inherited retinal disease preclude a patient from having another treatment?
10. With regard to inherited retinal diseases what is the role of pre-natal and pre-implantation diagnosis in helping parents make informed choices?

Neuro-ophthalmology

1. What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?
2. What are the most effective treatments and rehabilitation for optic neuropathies, e.g. Leber's hereditary optic neuropathy and anterior ischaemic optic neuropathy?
3. Can vision loss due to optic nerve diseases such as giant cell arteritis, Leber's hereditary optic neuropathy, optic neuritis and optic atrophy, be restored, for example through gene therapy and stem cell treatment?
4. What rehabilitation or treatment methods are most effective for vision loss following brain damage due to stroke, brain injury, cerebral vision impairment, tumours and dementias?
5. What is the most effective way to assess vision in patients with neurological visual impairment i.e. stroke, dementia and cerebral/cortical visual impairment?
6. Can the early stages of optic neuropathy be detected?
7. How can optic neuropathies be prevented, for example anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?
8. Can treatments be developed for visual field and ocular motility manifestations following stroke?
9. How can electronic devices improve or restore vision for people with optic neuropathies?

10. Can an alternative or new treatment be developed that will treat the sight loss caused by giant cell arteritis?

Ocular cancer

1. What can be done to help ocular cancer sufferers?
2. Can gene-based targeted therapies for ocular cancers be developed?
3. How can immunotherapy be used to fight metastatic ocular melanoma?
4. What are the most effective detection and screening methods for follow up to detect metastasis of ocular melanoma?
5. How can follow-up for ocular complications be managed in patients with ocular melanoma?
6. What is the best management of metastatic choroidal melanoma?
7. What activates choroidal melanoma metastasis in the liver after the primary melanoma has been treated?
8. Can adjuvant therapies be developed to treat ocular melanoma?
9. What are the causes of ocular cancer and how can they be prevented?
10. What is the most effective treatment for primary ocular melanoma?

Ocular inflammatory diseases

1. What are the most effective treatments for ocular and orbital inflammatory diseases?
2. What causes thyroid eye disease?
3. Can the severity of ocular and orbital inflammatory disease in an individual be predicted?
4. Is it possible to prevent further occurrences of retinal damage caused by toxoplasmosis?
5. What causes birdshot retinopathy?
6. Why does disease burn out in patients with ocular and orbital inflammatory diseases?
7. Can early detection methods be developed for ocular and orbital inflammatory diseases?
8. What medications best prevent the development of eye disease in Behcets?
9. What causes scleritis?
10. Can diet or lifestyle changes prevent uveitis from developing?

Refractive error and ocular motility

1. What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?
2. What is the cause of both congenital and acquired nystagmus?
3. How can the development of binocular vision in young children with squint and amblyopia be promoted, and would the same approach work in older individuals without inducing intractable diplopia?
4. Would correction of refractive error have a positive impact on early life learning and development?
5. Does early diagnosis of refractive error improve long-term prognosis and promote faster, more effective treatment?
6. What is the effect of congenital nystagmus on visual and emotional development?
7. What is the most effective treatment for exotropia and when should it be delivered?
8. How can the functional effects of surgical treatment for squint best be assessed?
9. Could the accurate testing of refractive error be made less dependent on a subjective response i.e. the person's own response?
10. How can myopia be prevented?

Retinal vascular diseases

1. What are the best methods to prevent retinopathy of prematurity?
2. How can sight loss from diabetic retinal changes be prevented and reduced?
3. What are the predictive factors for the progression to sight threatening diabetic eye disease?
4. Is there a way to improve screening of premature babies for retinopathy of prematurity?
5. Can an effective long lasting treatment for diabetic macular oedema, both ischaemic and non-ischaemic, be developed?
6. Can a retinal vein occlusion be predicted and prevented?
7. Can new non-invasive treatments be developed to slow down the progression of diabetic retinopathy?
8. What are the barriers that prevent diabetic patients having regular eye checks?

9. What rehabilitation programmes are best for the management of distorted vision from retinal diseases?
10. What is the efficacy and safety of anti-VEGF agents in the treatment of retinopathy of prematurity?

Vitreoretinal and ocular trauma

1. How can surgical techniques be improved to save sight for eyes damaged by injury?
2. How can the risk of losing sight for people with retinal detachment be reduced?
3. How can better interventions be developed that are effective in treating vitreous opacities/eye floaters?
4. What causes retinal detachment and can it be prevented?
5. Can more effective diagnostic tools be developed for assessing the vitreous and eye floaters?
6. Can a functioning prosthetic eye be developed to replace an eye damaged by injury?
7. How can epiretinal membrane/fibrosis be prevented or treated?
8. Can stem cells be used to regrow an eye or part of an eye?
9. What causes posterior vitreous detachment/vitreous syneresis?
10. Are there methods to prevent and improve the treatment of macular holes?

Discussion

About 50% of sight loss in the UK is currently avoidable[1]. Recognising this and the UK's pre-eminent position in eye research, the Vision 2020UK ERG considered that any UK Research Agenda must look at addressing unavoidable sight loss for it to be credible. The challenge was to produce a coherently constructed and constituted prioritised research agenda whose methods were clear and for which there had been inclusive and widespread consultation, where everyone with an interest had been offered the opportunity to contribute and be heard.

The SLV-PSP is unique because it sought the combined views of patients, carers and eye health professionals to identify uncertainties about the prevention, diagnosis and treatment of sight loss and eye conditions and prioritise them for research to

address[5]. It is rare that those with direct experience of conditions are able to influence the research agenda[3,5,6]. However, this SLV-PSP provided an extensive set of unanswered questions prioritised by patients, carers and eye health professionals across twelve categories of eye conditions. These questions addressed a broad range of eye conditions and considered issues of aetiology, prevention, screening, assessment and management. These may now be used to encourage researchers to investigate what is most important to these groups. Similar to other PSP processes, it is envisaged that research funders will be able to use the list to inform commissioned calls for research and identify which research applications to response mode funding opportunities can answer questions that these groups have agreed are a priority.

The SLV-PSP will also help to increase awareness of why research into sight loss and vision is necessary and important. It will be used to campaign for major funders to invest in sight loss and eye conditions, all of which are placing increased emphasis on researchers demonstrating how they have consulted and involved the public and patients in the process of developing their research.

These remain significant goals. For a sector with around 700 organisations to arrive at any kind of consensus for research priority areas, a process that was genuinely consultative, open and engaging to the individuals whose interests these organisations represent as well as at an organisational level was recognised as being critical. For a prioritisation exercise to be useful to the sector it needed to make sense to funders and statutory bodies with responsibilities and interests in these areas as well as to researchers. It was recognised that a prioritisation of research areas produced by a small group within the sector would not be credible and would never engage the support required for it to achieve the goals listed above.

Conclusions

Following a systematic process of national consultation and widespread survey of patients, carers and clinicians, 2220 individuals generated 4461 questions. Through a process of data analysis, interim prioritisation and final workshops, a top ten or eleven research questions have been identified for twelve categories of eye

conditions. This is the first time, to our knowledge, that an exercise like this has been carried out anywhere in the world for sight loss and vision. Not only is this the most wide ranging and ambitious James Lind Alliance priority setting partnership, it also engaged a diversity of participants and enabled them to reach consensus together. For the first time, we have a clear idea of what the consumers of eye research – the patients and the people who care for and treat them – believe research money should be spent on. It has provided a focus for research in sight loss and vision and it is intended that these priorities are used to inform funders, researchers, clinicians and the public.

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Competing interests: The authors declare that they have no competing interests.

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Figure 1 Flowchart of SLVPSP process

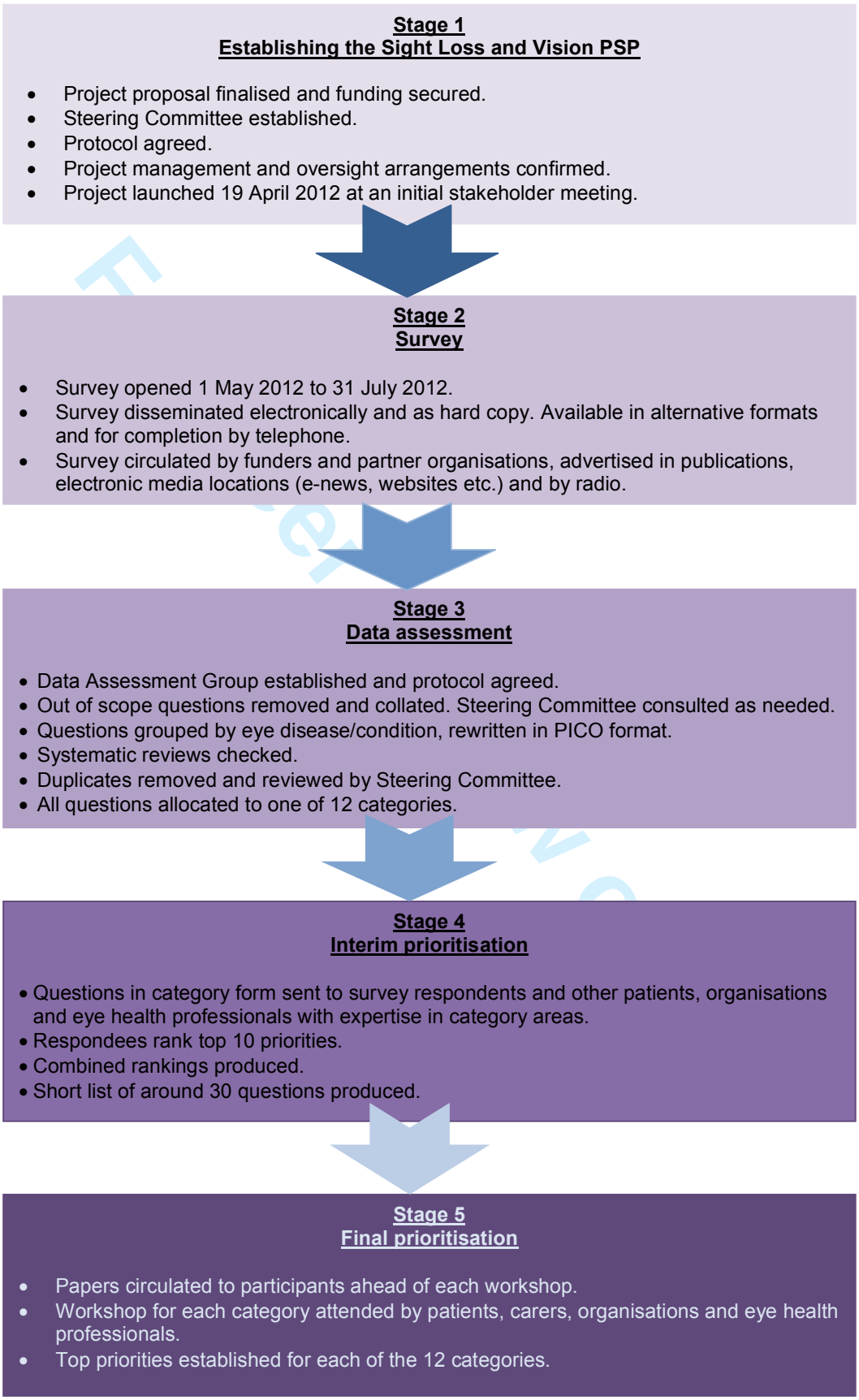

Figure 2 Background of respondents

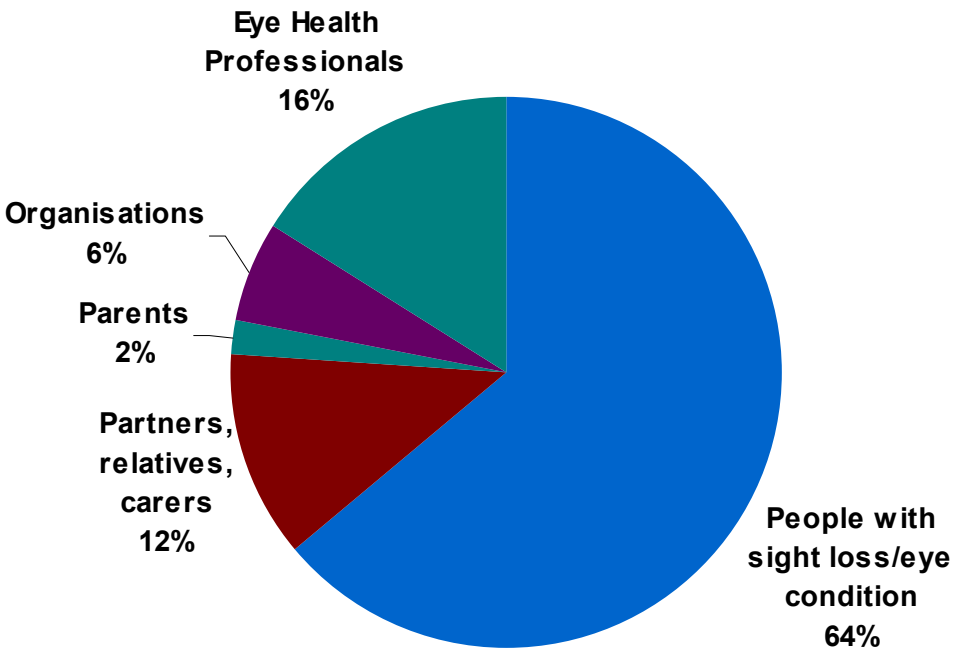

Table 1 Categories of eye condition

	Initial number of survey questions	Number of questions at interim prioritisation	Number of participants at interim prioritisation	Number of questions at final prioritisation
Age-related macular degeneration	763	43	101 PPI 25 Professionals	29
Cataract	191	27	Not required	27
Childhood-onset disorders	125	69	12 PPI 20 Professionals	30
Corneal and external diseases	292	93	25 PPI 38 Professionals	30
Glaucoma	1235	78	182 PPI 25 Professionals	30
Inherited retinal diseases	280	63	27 PPI 25 Professionals	30
Neuro-ophthalmology	125	43	15 PPI 21 Professionals	30
Ocular cancer	26	19	Not required	19
Ocular inflammatory diseases	472	66	27 PPI 21 Professionals	30
Refractive error and ocular motility	188	70	21 PPI 23 Professionals	31
Retinal vascular diseases	205	56	15 PPI 12 Professionals	30
Vitreoretinal and ocular trauma	265	59	21 PPI 8 Professionals	30

PPI: Inclusive of patients, carers, relatives, organisation representatives.

Professionals: Inclusive of eye health professions.

Table 2 Final workshop participants

Category	Total number of workshop participants	Number of patients, relatives, carers, patient groups and organisations	Number of eye health professionals
Age-related Macular Degeneration	17	9	8
Cataract	11	5	6
Childhood-Onset Disorders	16	7	9
Corneal and External Diseases	12	5	7
Glaucoma	17	9	8
Inherited Retinal Diseases	19	11	8
Neuro-ophthalmology	10	6	4
Ocular Cancer	10	6	4
Ocular Inflammatory Diseases	10	5	5
Refractive Error and Ocular Motility	12	5	7
Retinal Vascular Diseases	11	3	8
Vitreoretinal and Ocular Trauma	10	7	3
TOTAL	**155**	**78**	**77**

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Abstract

Objectives: The Sight Loss and Vision Priority Setting Partnership aimed to identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians. These priorities can be used to inform funding bodies' decisions and enhance the case for additional research funding.

Design: Prospective survey with support from the James Lind Alliance.

Setting: UK wide NHS and non-NHS.

Participants: Patients, carers and eye health professionals. Academic researchers were excluded solely from the prioritisation process. The survey was disseminated by patient groups, professional bodies, at conferences and through the media, and was available for completion online, by phone, by post and by alternative formats (Braille and audio).

Outcome measure: People were asked to submit the questions about prevention, diagnosis and treatment of sight loss and eye conditions that they most wanted to see answered by research. Returned survey questions were reviewed by a data assessment group. Priorities were established across eye disease categories at final workshops.

Results: 2,220 people responded generating 4,461 submissions. Sixty-five percent of respondents had sight loss and/or an eye condition. Following initial data analysis, 686 submissions remained which were circulated for interim prioritisation (excluding cataract and ocular cancer questions) to 446 patients/carers and 218 professionals. The remaining 346 questions were discussed at final prioritisation workshops to reach agreement of top questions per category.

Conclusions: The exercise engaged a diverse community of stakeholders generating a wide range of conditions and research questions. Top priority questions were established across 12 eye disease categories.

Keywords: Sight loss; Vision; Research; Priorities; Partnership; James Lind Alliance

Article summary

Strengths and limitations of this study:

- Wide ranging and comprehensive coverage
- Substantial response
- The hardest to reach and those with the least opportunity to be heard may indeed have not been heard
- Any such groups now feeling excluded should have an opportunity to redress this.

Introduction

In the UK it is estimated that almost 2 million people are affected by sight loss and this number is expected to double by 2050[1]. Currently 50% of sight loss in the UK is avoidable but there are also many unavoidable sight loss conditions. Whether it is to address childhood eye conditions or those affecting adults, research is needed to inform us about prevention, to develop new techniques for early diagnosis and to develop new and more effective treatments for many eye conditions.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP) was developed from earlier work by the Eye Research Group (ERG) of VISION 2020 UK whose mission is the elimination of avoidable sight loss and the amelioration of the effects of sight loss when it cannot be avoided[2]. The UK Vision Strategy sets out ways of addressing avoidable sight loss in the UK. There is much that can be done to improve the nation's eye health, and to eliminate avoidable sight loss. There are however gaps in the evidence base regarding eye health interventions and the best way to deliver services. Research is needed to support the UK Vision Strategy. In key areas research is urgently required to enable the Vision Strategy to be implemented in an evidence-based way that ensures efficient and effective development.

Despite on-going research in the UK and other countries, there are still many questions about the prevention, diagnosis and treatment of sight loss and eye conditions that remain unanswered. Given that resources for research are limited, it is important for research funders to understand how patients, carers and eye health professionals prioritise these unanswered questions so that future research can be consolidated and targeted accordingly[3].

The purpose of this project was to undertake a comprehensive, UK-wide, survey of patients, carers and clinicians to identify research questions and priorities to inform decisions of funding bodies and enhance the case for additional research funding.

The priority setting process has been well established by the James Lind Alliance (JLA) (http://www.lindalliance.org/) which has supported partnerships on a range of topics since 2004. This partnership initiated by Fight for Sight, the UK's leading eye research charity was established with support, financial and in kind, from the College of Optometrists, the Royal College of Ophthalmologists, the NIHR Biomedical Research Centre for Ophthalmology, the RNIB, UK Vision Strategy and the Cochrane Eyes and Vision Group. A representative from the JLA convened meetings of the steering committee and provided independent chairmanship for this and the priority setting workshops. Their extensive experience in this process ensured no single voice exerted undue influence over the prioritisation process and that the views of patients, their carers and clinicians were paramount. The views of researchers with no clinical involvement with patients and views of commercial organisations were not included in the prioritisation.

Methods and materials

The detailed methods for this prioritisation process have been described in detail elsewhere[4]. In brief, the process comprised five stages (figure 1). Our study did not require ethical approval or consent from participants. James Lind Alliance priority setting partnerships do not require ethical approval. Dissemination of the survey is via open communications through professional bodies, charities and related organisations. The survey is not undertaken through Higher Education Institutes or through NHS organisations and does not recruit NHS patients. The survey contains clear information on the aims of the priority setting partnership, how the process works and how data will be used. In addition, submission of questions is anonymised. For the workshops in which priorities were discussed and agreed, participants choose to voluntarily attend and consent is not required for this. We followed the ethical guidance for participation, information and evaluation from the James Lind Alliance guidebook (http://www.jlaguidebook.org/jla-guidebook.asp?val=56).

Establishing the SLV-PSP

A steering committee and data assessment group comprising the authors of this article oversaw the process. Each member was responsible for contributing to and managing a part of the process and was selected for their expertise and association with eye research. The steering committee also included patient representatives and eye health professionals. In April 2012 an initial stakeholder meeting was held to engage the groups and organisations with member bases and community influence. This was to ensure that the initial survey would be disseminated and completed by as many patients, relatives, carers and eye health professionals as possible in the UK.

Main survey

The SLV-PSP survey was launched on 1 May 2012 and was open until 31 July 2012. The aim of the survey was to identify patients', carers' and eye health professionals' unanswered questions about sight loss and eye conditions. The survey's primary question was "What question(s) about the prevention, diagnosis and treatment of sight loss and eye conditions would you like to see answered by research?"

Data analysis

Following closure of the survey, all submissions were examined. Out-of-scope submissions were removed including those not related to the topic and uncertainties better suited to social research. In-scope uncertainties were allocated into disease-specific groups and re-worded in PICO format (Population, Intervention, Comparison, Outcome). Searches were then undertaken to ascertain whether or not each uncertainty could be answered by an up-to-date systematic review. All unanswered uncertainties were then allocated to one of 12 eye disease categories, with duplicates removed and similar questions combined. Checks were also made to identify any on-going trials which might address the uncertainty. The 12 categories were formed following discussions by the steering group on the most logical and pragmatic way to organise the data within the time and resources available.

Interim prioritisation

In order to start reducing the number of uncertainties, an interim prioritisation exercise was conducted over email and by post. Patients, carers and eye health professionals were invited to examine the long lists and then choose and rank 10 of the uncertainties.

Final prioritisation

The remaining uncertainties were ranked by patients, carers, relatives, organisation representatives and eye health professionals in one-day workshops facilitated by the JLA, using Nominal Group Technique – a mix of discussion and ranking. For each category, the top 10/11 questions were agreed.

Results

Main survey

In response to the survey, 2220 people generated 4461 submissions. Of these respondents, 17% identified themselves as healthcare professionals including primarily ophthalmologists, optometrists, orthoptists, ophthalmic nurses, opticians and people working in social care and rehabilitation (figure 2). Over 60% were people with sight loss or an eye condition. The average age of survey participants was 65.7 years old (range:16 months (proxy completion of survey by adult carer)to 105 years). Just under two thirds (62%) of respondents were female. The geographical split was: England 89%; Scotland 6%; Wales 4%; Northern Ireland 1%.

Data analysis

Following data analysis to remove duplicate/answered/out of scope uncertainties, 686 uncertainties remained. These were divided into twelve eye disease categories. Table 1 shows each category with the initial number of submissions received after the survey responses were submitted, the number of uncertainties sent to interim prioritisation, the number of participants at interim prioritisation and the number of uncertainties considered at the final prioritisation workshops.

Interim prioritisation

Respondents from the initial survey, organisations and eye healthcare professionals with expertise in the eye diseases in ten categories were contacted to provide interim priority rankings. The smaller number of questions asked in the categories relating to cataract and ocular cancer meant that an interim prioritisation exercise was not required for either category. A large response was received for the interim exercise, with input from 446 patients, carers and relatives plus 218 eye health professionals. Uncertainties accumulated scores based on rank and frequency, resulting in short lists of around 30 uncertainties per category which were taken to final workshops.

Final prioritisation

In April and May 2013, 12 final prioritisation workshops were held: one for each eye disease category. Balanced numbers of patients/carers/relatives and eye health professionals participated. In total, 155 participants attended across all 12 workshops: 78 patients and 77 clinicians (table 2). The topics debated at each workshop comprised between 19-31 questions per category. Overall 153 questions about sight loss and vision were considered resulting in lists of 10/11priorities for each of the 12 categories (table 3). The questions addressed the broad topics of aetiology, prevention, identification and interventions with the number 1 questions as follows:

Age-related macular degeneration

1. Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?

Cataract

1. How can cataracts be prevented from developing?

Childhood-onset disorders

1. How can cerebral visual impairment be identified, prevented and treated in children?

Corneal and external diseases

1. Can new therapies such as gene or stem cell treatments be developed for corneal diseases?

Glaucoma

1. What are the most effective treatments for glaucoma and how can treatment be improved?

Inherited retinal diseases

1. Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?

Neuro-ophthalmology

1. What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?

Ocular cancer

1. What can be done to help ocular cancer sufferers?

Ocular inflammatory diseases

1. What are the most effective treatments for ocular and orbital inflammatory diseases?

Refractive error and ocular motility

1. What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?

Retinal vascular diseases

1. What are the best methods to prevent retinopathy of prematurity?

Vitreoretinal and ocular trauma

1. How can surgical techniques be improved to save sight for eyes damaged by injury?

Discussion

About 50% of sight loss in the UK is currently avoidable[1]. Recognising this and the UK's pre-eminent position in eye research, the Vision 2020UK ERG considered that any UK Research Agenda must look at addressing unavoidable sight loss for it to be credible. The challenge was to produce a coherently constructed and constituted prioritised research agenda whose methods were clear and for which there had been

inclusive and widespread consultation, where everyone with an interest had been offered the opportunity to contribute and be heard. As a result of this priority setting partnership we have established top ten lists of research questions for a range of eye conditions.

There are a number of strengths to this study. The SLV-PSP is unique because it sought the combined views of patients, carers and eye health professionals to identify uncertainties about the prevention, diagnosis and treatment of sight loss and eye conditions and prioritise them for research to address[5]. It is rare that those with direct experience of conditions are able to influence the research agenda[3,5,6]. The views of patients, carers and professionals were given equal merit. All submitted questions were evaluated independently and equally. Duplicate questions and out of scope questions were removed. We did not encounter particularly misunderstandings between lay persons and professionals or insufficient knowledge of the public. Open discussions occurred during the face-to-face workshops with good communication and facilitation to encourage respectful listening in accordance with James Lind Alliance guidelines. Questions could only be pooled if this was agreed by patients, carers and professionals. Where there was no agreement, the questions remained separate. Thus, any differing perspectives of priorities between participants were acknowledged. We did not aim to compare and contrast questions from patients, carers and professionals but to represent and act on all.

This SLV-PSP provided an extensive set of unanswered questions prioritised by patients, carers and eye health professionals across twelve categories of eye conditions. These questions addressed a broad range of eye conditions and considered issues of aetiology, prevention, screening, assessment and management. Importantly, the public were as likely to propose questions in relation to aetiology, assessment and management just as professionals were as likely to raise questions regarding impact of sight loss.

In addition to the strengths of our study, we identified a number of limitations. We did not request the views of 'pure' researchers (i.e. scientists with no current clinical practice) as these individuals are intentionally excluded from the priority setting process by the James Lind Alliance. This step is a key feature of the James Lind Alliance methodology in which the remit is to provide an opportunity for patients,

carers and clinicians to influence the research agenda. We acknowledge this is a different approach but do not consider their exclusion as a flaw in this process as we include clinical researchers who did take part alongside clinicians and the public.

These questions may now be used to encourage researchers to investigate what is most important to these groups. We do not know how our research questions compare to the prioritisation of research areas by scientists, government agencies or other organisation research funders. We are unaware of any systematic data collating such data. However, similar to other PSP processes, we have provided information on out research priorities openly to national funding organisations and it is envisaged that research funders will be able to use the list to inform commissioned calls for research and identify which research applications to response mode funding opportunities can answer questions that these groups have agreed are a priority. Furthermore, any questions or uncertainties not prioritised in this process were submitted to and are currently available on the UK Database of Uncertainties about the Effects of Treatments (UK DUETs). Thus individuals looking for uncertainties for their research can access such information direct from UK DUETs. This sharing of information contributes to the quality assurance process of avoiding waste in research.

The SLV-PSP will also help to increase awareness of why research into sight loss and vision is necessary and important. It will be used to campaign for major funders to invest in sight loss and eye conditions, all of which are placing increased emphasis on researchers demonstrating how they have consulted and involved the public and patients in the process of developing their research.

These remain significant goals. For a sector with around 700 organisations to arrive at any kind of consensus for research priority areas, a process that was genuinely consultative, open and engaging to the individuals whose interests these organisations represent as well as at an organisational level was recognised as being critical. For a prioritisation exercise to be useful to the sector it needed to make sense to funders and statutory bodies with responsibilities and interests in these areas as well as to researchers. It was recognised that a prioritisation of research areas produced by a small group within the sector would not be credible and would never engage the support required for it to achieve the goals listed above.

Conclusions

Following a systematic process of national consultation and widespread survey of patients, carers and clinicians, 2220 individuals generated 4461 questions. Through a process of data analysis, interim prioritisation and final workshops, a top ten or eleven research questions have been identified for twelve categories of eye conditions. This is the first time, to our knowledge, that an exercise like this has been carried out anywhere in the world for sight loss and vision. Not only is this the most wide ranging and ambitious James Lind Alliance priority setting partnership, it also engaged a diversity of participants and enabled them to reach consensus together. For the first time, we have a clear idea of what the consumers of eye research – the patients and the people who care for and treat them – believe research money should be spent on. It has provided a focus for research in sight loss and vision and it is intended that these priorities are used to inform funders, researchers, clinicians and the public.

Competing interests: The authors declare that they have no competing interests.

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Data sharing statement: All original data are held by Fight for Sight. Extra data is available from: http://www.library.nhs.uk/duets/SearchResults.aspx?catID=14501

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Table 1 Categories of eye condition

	Initial number of survey questions	Number of questions at interim prioritisation	Number of participants at interim prioritisation	Number of questions at final prioritisation
Age-related macular degeneration	763	43	101 PPI 25 Professionals	29
Cataract	191	27	Not required	27
Childhood-onset disorders	125	69	12 PPI 20 Professionals	30
Corneal and external diseases	292	93	25 PPI 38 Professionals	30
Glaucoma	1235	78	182 PPI 25 Professionals	30
Inherited retinal diseases	280	63	27 PPI 25 Professionals	30
Neuro-ophthalmology	125	43	15 PPI 21 Professionals	30
Ocular cancer	26	19	Not required	19
Ocular inflammatory diseases	472	66	27 PPI 21 Professionals	30

Refractive error and ocular motility	188	70	21 PPI 23 Professionals	31
Retinal vascular diseases	205	56	15 PPI 12 Professionals	30
Vitreoretinal and ocular trauma	265	59	21 PPI 8 Professionals	30

PPI: Inclusive of patients, carers, relatives, organisation representatives.

Professionals: Inclusive of eye health professions.

Table 2 **Final workshop participants**

Category	Total number of workshop participants	Number of patients, relatives, carers, patient groups and organisations	Number of eye health professionals
Age-related Macular Degeneration	17	9	8
Cataract	11	5	6
Childhood-Onset Disorders	16	7	9
Corneal and External Diseases	12	5	7
Glaucoma	17	9	8
Inherited Retinal Diseases	19	11	8
Neuro-ophthalmology	10	6	4
Ocular Cancer	10	6	4
Ocular Inflammatory Diseases	10	5	5
Refractive Error and Ocular Motility	12	5	7
Retinal Vascular Diseases	11	3	8
Vitreoretinal and Ocular Trauma	10	7	3
TOTAL	**155**	**78**	**77**

Table 3 Top ten lists per category

	Age-related macular degeneration	*Cataract*	*Childhood-onset disorders*	*Corneal and external diseases*	*Glaucoma*	*Inherited retinal diseases*
1	Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?	How can cataracts be prevented from developing?	How can cerebral visual impairment be identified, prevented and treated in children?	Can new therapies such as gene or stem cell treatments be developed for corneal diseases?	What are the most effective treatments for glaucoma and how can treatment be improved?	Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?
2	What is the cause of AMD?	Can the return of cloudy or blurred vision after cataract surgery known as posterior capsule opacity (PCO) or secondary cataract be prevented?	How can treatment for visual pathway damage associated with pre-term birth be developed?	What is the most effective management for dry eye and can new strategies be developed?	How can loss of vision be restored for people with glaucoma?	How can sight loss be prevented in an individual with inherited retinal disease?
3	How can AMD be prevented?	How can cataract progression be slowed down?	How do we improve screening and surveillance from the ante-natal period through to childhood to ensure early diagnosis of impaired vision and eye conditions?	Can treatments to save eye sight from microbial keratitis be improved?	How can glaucoma be stopped from progressing?	Is a genetic (molecular) diagnosis possible for all inherited retinal diseases?
4	Are there ways of restoring sight loss for people with AMD?	What alternatives to treat cataracts other than cataract surgery are being developed?	Can the treatment of amblyopia be improved to produce better short and long term outcomes than are possible with current treatments?	How can the rejection of corneal transplants be prevented?	What can be done to improve early diagnosis of sight-threatening glaucoma?	What factors affect the progression of sight loss in inherited retinal diseases?
5	Can the development of AMD be predicted?	What is the cause of cataract?	How can cataract be prevented in children?	Can the outcomes of corneal transplantation be improved?	What causes glaucoma?	What causes sight loss in inherited retinal diseases?
6	What is the most effective way to detect and monitor the progression of early AMD?	How can cataract surgery outcomes be improved?	What are the causes of coloboma and microphthalmia/anophthalmia and how can they be prevented?	What causes keratoconus to progress and can progression be prevented?	What is the most effective way of monitoring the progression of glaucoma?	What is the most effective way to support patients with inherited retinal disease?
7	What factors influence the progression of AMD?	How safe and effective is laser assisted cataract surgery?	Can vision be corrected in later life for people with amblyopia?	Can non-surgical therapy be developed for Fuchs' corneal dystrophy?	How can glaucoma patients with a higher risk to progress rapidly be detected?	Can the diagnosis of inherited retinal diseases be refined so that individuals can be given a clearer idea about their specific condition and how it is likely to progress?
8	Can a non-invasive therapy be developed for wet AMD?	Should accommodative lenses be developed for cataract surgery?	How can retinoblastoma be identified, prevented and treated in children?	Can corneal infections be prevented in high-risk individuals such as contact lens wearers?	Why is glaucoma more aggressive in people of certain ethnic groups, such as those of West African origin?	What is the relationship between sight loss and mental health for people with inherited retinal diseases?
9	Can dietary factors, nutritional supplements, complementary therapies or lifestyle changes prevent or slow the progression of AMD?	What is the best measure of visual disability due to cataract?	Can better treatments for glaucoma in children be developed?	What is the cause of keratoconus and can it be prevented?	How can glaucoma be prevented?	Would having a treatment for an inherited retinal disease preclude a patient from having another treatment?
10	What are the best enablement strategies for people with AMD?	Can retinal detachment be prevented after cataract surgery?	Can a treatment be developed to improve vision for people with albinism?	What is the most effective management of ocular complications associated with Stevens Johnson Syndrome?	Is there a link between treatment adherence and glaucoma progression and how can adherence be improved?	With regard to inherited retinal diseases what is the role of pre-natal and pre-implantation diagnosis in helping parents make informed choices?
11		What are the outcomes for cataract surgery among people with different levels of cognitive		Can severe ocular surface disease in children, such as blepharokeratoconjunctivitis and		

		impairment (all causes excluding dementia, stroke, neurological conditions, head injuries)?		vernal keratoconjunctivitis be managed better?		
	Neuro-ophthalmology	***Ocular cancer***	***Ocular inflammatory diseases***	***Refractive error and ocular motility***	***Retinal vascular diseases***	***Vitreoretinal and ocular trauma***
1	What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What can be done to help ocular cancer sufferers?	What are the most effective treatments for ocular and orbital inflammatory diseases?	What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?	What are the best methods to prevent retinopathy of prematurity?	How can surgical techniques be improved to save sight for eyes damaged by injury?
2	What are the most effective treatments and rehabilitation for optic neuropathies, e.g. Leber's hereditary optic neuropathy and anterior ischaemic optic neuropathy?	Can gene-based targeted therapies for ocular cancers be developed?	What causes thyroid eye disease?	What is the cause of both congenital and acquired nystagmus?	How can sight loss from diabetic retinal changes be prevented and reduced?	How can the risk of losing sight for people with retinal detachment be reduced?
3	Can vision loss due to optic nerve diseases such as giant cell arteritis, Leber's hereditary optic neuropathy, optic neuritis and optic atrophy, be restored, for example through gene therapy and stem cell treatment?	How can immunotherapy be used to fight metastatic ocular melanoma?	Can the severity of ocular and orbital inflammatory disease in an individual be predicted?	How can the development of binocular vision in young children with squint and amblyopia be promoted, and would the same approach work in older individuals without inducing intractable diplopia?	What are the predictive factors for the progression to sight threatening diabetic eye disease?	How can better interventions be developed that are effective in treating vitreous opacities/eye floaters?
4	What rehabilitation or treatment methods are most effective for vision loss following brain damage due to stroke, brain injury, cerebral vision impairment, tumours and dementias?	What are the most effective detection and screening methods for follow up to detect metastasis of ocular melanoma?	Is it possible to prevent further occurrences of retinal damage caused by toxoplasmosis?	Would correction of refractive error have a positive impact on early life learning and development?	Is there a way to improve screening of premature babies for retinopathy of prematurity?	What causes retinal detachment and can it be prevented?
5	What is the most effective way to assess vision in patients with neurological visual impairment i.e. stroke, dementia and cerebral/cortical visual impairment?	How can follow-up for ocular complications be managed in patients with ocular melanoma?	What causes birdshot retinopathy?	Does early diagnosis of refractive error improve long-term prognosis and promote faster, more effective treatment?	Can an effective long lasting treatment for diabetic macular oedema, both ischaemic and non-ischaemic, be developed?	Can more effective diagnostic tools be developed for assessing the vitreous and eye floaters?
6	Can the early stages of optic neuropathy be detected?	What is the best management of metastatic choroidal melanoma?	Why does disease burn out in patients with ocular and orbital inflammatory diseases?	What is the effect of congenital nystagmus on visual and emotional development?	Can a retinal vein occlusion be predicted and prevented?	Can a functioning prosthetic eye be developed to replace an eye damaged by injury?
7	How can optic neuropathies be prevented, for example anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What activates choroidal melanoma metastasis in the liver after the primary melanoma has been treated?	Can early detection methods be developed for ocular and orbital inflammatory diseases?	What is the most effective treatment for exotropia and when should it be delivered?	Can new non-invasive treatments be developed to slow down the progression of diabetic retinopathy?	How can epiretinal membrane/fibrosis be prevented or treated?
8	Can treatments be developed for visual field and ocular	Can adjuvant therapies be developed to treat ocular	What medications best prevent the development of eye disease in	How can the functional effects of surgical treatment for squint best	What are the barriers that prevent diabetic patients	Can stem cells be used to regrow an eye or part of an eye?

	motility manifestations following stroke?	melanoma?	Behcets?	be assessed?	having regular eye checks?	
9	How can electronic devices improve or restore vision for people with optic neuropathies?	What are the causes of ocular cancer and how can they be prevented?	What causes scleritis?	Could the accurate testing of refractive error be made less dependent on a subjective response i.e. the person's own response?	What rehabilitation programmes are best for the management of distorted vision from retinal diseases?	What causes posterior vitreous detachment/vitreous syneresis?
10	Can an alternative or new treatment be developed that will treat the sight loss caused by giant cell arteritis?	What is the most effective treatment for primary ocular melanoma?	Can diet or lifestyle changes prevent uveitis from developing?	How can myopia be prevented?	What is the efficacy and safety of anti-VEGF agents in the treatment of retinopathy of prematurity?	Are there methods to prevent and improve the treatment of macular holes?

Figure legends

Figure 1: Flowchart showing the steps of the process from Stage 1 when establishing the PSP through to Stage 5 at the final prioritisation.

Figure 2: Background of respondents showing that questions were largely received from people who have sight loss or an eye condition, but also including eye health professions, organisations, parents, family and carers.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP): Overview and results of the research prioritisation survey process.

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- Any such groups now feeling excluded should have an opportunity to redress this.

Introduction

In the UK it is estimated that almost 2 million people are affected by sight loss and this number is expected to double by 2050[1]. Currently 50% of sight loss in the UK is avoidable but there are also many unavoidable sight loss conditions. Whether it is to address childhood eye conditions or those affecting adults, research is needed to inform us about prevention, to develop new techniques for early diagnosis and to develop new and more effective treatments for many eye conditions.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP) was developed from earlier work by the Eye Research Group (ERG) of VISION 2020 UK whose mission is the elimination of avoidable sight loss and the amelioration of the effects of sight loss when it cannot be avoided[2]. The UK Vision Strategy sets out ways of addressing avoidable sight loss in the UK. There is much that can be done to improve the nation's eye health, and to eliminate avoidable sight loss. There are however gaps in the evidence base regarding eye health interventions and the best way to deliver services. Research is needed to support the UK Vision Strategy. In key areas research is urgently required to enable the Vision Strategy to be implemented in an evidence-based way that ensures efficient and effective development.

Despite on-going research in the UK and other countries, there are still many questions about the prevention, diagnosis and treatment of sight loss and eye conditions that remain unanswered. Given that resources for research are limited, it is important for research funders to understand how patients, carers and eye health professionals prioritise these unanswered questions so that future research can be consolidated and targeted accordingly[3].

The purpose of this project was to undertake a comprehensive, UK-wide, survey of patients, carers and clinicians to identify research questions and priorities to inform decisions of funding bodies and enhance the case for additional research funding.

The priority setting process has been well established by the James Lind Alliance (JLA) (http://www.lindalliance.org/) which has supported partnerships on a range of topics since 2004. This partnership initiated by Fight for Sight, the UK's leading eye research charity was established with support, financial and in kind, from the College of Optometrists, the Royal College of Ophthalmologists, the NIHR Biomedical Research Centre for Ophthalmology, the RNIB, UK Vision Strategy and the Cochrane Eyes and Vision Group. A representative from the JLA convened meetings of the steering committee and provided independent chairmanship for this and the priority setting workshops. Their extensive experience in this process ensured no single voice exerted undue influence over the prioritisation process and that the views of patients, their carers and clinicians were paramount. The views of researchers with no clinical involvement with patients and views of commercial organisations were not included in the prioritisation.

Methods and materials

The detailed methods for this prioritisation process have been described in detail elsewhere[4]. In brief, the process comprised five stages (figure 1). Our study did not require ethical approval or consent from participants. James Lind Alliance priority setting partnerships do not require ethical approval. Dissemination of the survey is via open communications through professional bodies, charities and related organisations. The survey is not undertaken through Higher Education Institutes or through NHS organisations and does not recruit NHS patients. The survey contains clear information on the aims of the priority setting partnership, how the process works and how data will be used. In addition, submission of questions is anonymised. For the workshops in which priorities were discussed and agreed, participants choose to voluntarily attend and consent is not required for this. We followed the ethical guidance for participation, information and evaluation from the James Lind Alliance guidebook (http://www.jlaguidebook.org/jla-guidebook.asp?val=56).

Establishing the SLV-PSP

A steering committee and data assessment group comprising the authors of this article oversaw the process. Each member was responsible for contributing to and managing a part of the process and was selected for their expertise and association with eye research. The steering committee also included patient representatives and eye health professionals. In April 2012 an initial stakeholder meeting was held to engage the groups and organisations with member bases and community influence. This was to ensure that the initial survey would be disseminated and completed by as many patients, relatives, carers and eye health professionals as possible in the UK.

Main survey

The SLV-PSP survey was launched on 1 May 2012 and was open until 31 July 2012. The aim of the survey was to identify patients', carers' and eye health professionals' unanswered questions about sight loss and eye conditions. The survey's primary question was "What question(s) about the prevention, diagnosis and treatment of sight loss and eye conditions would you like to see answered by research?"

Data analysis

Following closure of the survey, all submissions were examined. Out-of-scope submissions were removed including those not related to the topic and uncertainties better suited to social research. In-scope uncertainties were allocated into disease-specific groups and re-worded in PICO format (Population, Intervention, Comparison, Outcome). Searches were then undertaken to ascertain whether or not each uncertainty could be answered by an up-to-date systematic review. All unanswered uncertainties were then allocated to one of 12 eye disease categories, with duplicates removed and similar questions combined. Checks were also made to identify any on-going trials which might address the uncertainty. The 12 categories were formed following discussions by the steering group on the most logical and pragmatic way to organise the data within the time and resources available.

Interim prioritisation

In order to start reducing the number of uncertainties, an interim prioritisation exercise was conducted over email and by post. Patients, carers and eye health

professionals were invited to examine the long lists and then choose and rank 10 of the uncertainties.

Final prioritisation

The remaining uncertainties were ranked by patients, carers, relatives, organisation representatives and eye health professionals in one-day workshops facilitated by the JLA, using Nominal Group Technique – a mix of discussion and ranking. For each category, the top 10/11 questions were agreed.

Results

Main survey

In response to the survey, 2220 people generated 4461 submissions. Of these respondents, 17% identified themselves as healthcare professionals including primarily ophthalmologists, optometrists, orthoptists, ophthalmic nurses, opticians and people working in social care and rehabilitation (figure 2). Over 60% were people with sight loss or an eye condition. The average age of survey participants was 65.7 years old (range:16 months (proxy completion of survey by adult carer)to 105 years). Just under two thirds (62%) of respondents were female. The geographical split was: England 89%; Scotland 6%; Wales 4%; Northern Ireland 1%.

Data analysis

Following data analysis to remove duplicate/answered/out of scope uncertainties, 686 uncertainties remained. These were divided into twelve eye disease categories. Table 1 shows each category with the initial number of submissions received after the survey responses were submitted, the number of uncertainties sent to interim prioritisation, the number of participants at interim prioritisation and the number of uncertainties considered at the final prioritisation workshops.

Interim prioritisation

Respondents from the initial survey, organisations and eye healthcare professionals with expertise in the eye diseases in ten categories were contacted to provide interim priority rankings. The smaller number of questions asked in the categories relating to

cataract and ocular cancer meant that an interim prioritisation exercise was not required for either category. A large response was received for the interim exercise, with input from 446 patients, carers and relatives plus 218 eye health professionals. Uncertainties accumulated scores based on rank and frequency, resulting in short lists of around 30 uncertainties per category which were taken to final workshops.

Final prioritisation

In April and May 2013, 12 final prioritisation workshops were held: one for each eye disease category. Balanced numbers of patients/carers/relatives and eye health professionals participated. In total, 155 participants attended across all 12 workshops: 78 patients and 77 clinicians (table 2). The topics debated at each workshop comprised between 19-31 questions per category. Overall 153 questions about sight loss and vision were considered resulting in lists of 10/11priorities for each of the 12 categories (table 3). The questions addressed the broad topics of aetiology, prevention, identification and interventions with the number 1 questions as follows:

Age-related macular degeneration

1. Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?

Cataract

1. How can cataracts be prevented from developing?

Childhood-onset disorders

1. How can cerebral visual impairment be identified, prevented and treated in children?

Corneal and external diseases

1. Can new therapies such as gene or stem cell treatments be developed for corneal diseases?

Glaucoma

1. What are the most effective treatments for glaucoma and how can treatment be improved?

Inherited retinal diseases

1. Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?

Neuro-ophthalmology

1. What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?

Ocular cancer

1. What can be done to help ocular cancer sufferers?

Ocular inflammatory diseases

1. What are the most effective treatments for ocular and orbital inflammatory diseases?

Refractive error and ocular motility

1. What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?

Retinal vascular diseases

1. What are the best methods to prevent retinopathy of prematurity?

Vitreoretinal and ocular trauma

1. How can surgical techniques be improved to save sight for eyes damaged by injury?

Discussion

About 50% of sight loss in the UK is currently avoidable[1]. Recognising this and the UK's pre-eminent position in eye research, the Vision 2020UK ERG considered that any UK Research Agenda must look at addressing unavoidable sight loss for it to be credible. The challenge was to produce a coherently constructed and constituted prioritised research agenda whose methods were clear and for which there had been inclusive and widespread consultation, where everyone with an interest had been offered the opportunity to contribute and be heard. As a result of this priority setting partnership we have established top ten lists of research questions for a range of eye conditions.

There are a number of strengths to this study. The SLV-PSP is unique because it sought the combined views of patients, carers and eye health professionals to identify uncertainties about the prevention, diagnosis and treatment of sight loss and eye conditions and prioritise them for research to address[5]. It is rare that those with direct experience of conditions are able to influence the research agenda[3,5,6]. The views of patients, carers and professionals were given equal merit. All submitted questions were evaluated independently and equally. Duplicate questions and out of scope questions were removed. We did not encounter particularly misunderstandings between lay persons and professionals or insufficient knowledge of the public. Open discussions occurred during the face-to-face workshops with good communication and facilitation to encourage respectful listening in accordance with James Lind Alliance guidelines. Questions could only be pooled if this was agreed by patients, carers and professionals. Where there was no agreement, the questions remained separate. Thus, any differing perspectives of priorities between participants were acknowledged. We did not aim to compare and contrast questions from patients, carers and professionals but to represent and act on all.

This SLV-PSP provided an extensive set of unanswered questions prioritised by patients, carers and eye health professionals across twelve categories of eye conditions. These questions addressed a broad range of eye conditions and considered issues of aetiology, prevention, screening, assessment and management. Importantly, the public were as likely to propose questions in relation to aetiology, assessment and management just as professionals were as likely to raise questions regarding impact of sight loss.

In addition to the strengths of our study, we identified a number of limitations. We did not request the views of 'pure' researchers (i.e. scientists with no current clinical practice) as these individuals are intentionally excluded from the priority setting process by the James Lind Alliance. This step is a key feature of the James Lind Alliance methodology in which the remit is to provide an opportunity for patients, carers and clinicians to influence the research agenda. We acknowledge this is a different approach but do not consider their exclusion as a flaw in this process as we include clinical researchers who did take part alongside clinicians and the public.

These questions may now be used to encourage researchers to investigate what is most important to these groups. We do not know how our research questions compare to the prioritisation of research areas by scientists, government agencies or other organisation research funders. We are unaware of any systematic data collating such data. However, similar to other PSP processes, we have provided information on out research priorities openly to national funding organisations and it is envisaged that research funders will be able to use the list to inform commissioned calls for research and identify which research applications to response mode funding opportunities can answer questions that these groups have agreed are a priority. Furthermore, any questions or uncertainties not prioritised in this process were submitted to and are currently available on the UK Database of Uncertainties about the Effects of Treatments (UK DUETs). Thus individuals looking for uncertainties for their research can access such information direct from UK DUETs. This sharing of information contributes to the quality assurance process of avoiding waste in research.

The SLV-PSP will also help to increase awareness of why research into sight loss and vision is necessary and important. It will be used to campaign for major funders to invest in sight loss and eye conditions, all of which are placing increased emphasis on researchers demonstrating how they have consulted and involved the public and patients in the process of developing their research.

These remain significant goals. For a sector with around 700 organisations to arrive at any kind of consensus for research priority areas, a process that was genuinely consultative, open and engaging to the individuals whose interests these organisations represent as well as at an organisational level was recognised as being critical. For a prioritisation exercise to be useful to the sector it needed to make sense to funders and statutory bodies with responsibilities and interests in these areas as well as to researchers. It was recognised that a prioritisation of research areas produced by a small group within the sector would not be credible and would never engage the support required for it to achieve the goals listed above.

Conclusions

Following a systematic process of national consultation and widespread survey of patients, carers and clinicians, 2220 individuals generated 4461 questions. Through a process of data analysis, interim prioritisation and final workshops, a top ten or eleven research questions have been identified for twelve categories of eye conditions. This is the first time, to our knowledge, that an exercise like this has been carried out anywhere in the world for sight loss and vision. Not only is this the most wide ranging and ambitious James Lind Alliance priority setting partnership, it also engaged a diversity of participants and enabled them to reach consensus together. For the first time, we have a clear idea of what the consumers of eye research – the patients and the people who care for and treat them – believe research money should be spent on. It has provided a focus for research in sight loss and vision and it is intended that these priorities are used to inform funders, researchers, clinicians and the public.

Figure legends

Figure 1: Flowchart showing the steps of the process from Stage 1 when establishing the PSP through to Stage 5 at the final prioritisation.

Figure 2: Background of respondents showing that questions were largely received from people who have sight loss or an eye condition, but also including eye health professions, organisations, parents, family and carers.

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Table 1 Categories of eye condition

	Initial number of survey questions	Number of questions at interim prioritisation	Number of participants at interim prioritisation	Number of questions at final prioritisation
Age-related macular degeneration	763	43	101 PPI 25 Professionals	29
Cataract	191	27	Not required	27
Childhood-onset disorders	125	69	12 PPI 20 Professionals	30
Corneal and external diseases	292	93	25 PPI 38 Professionals	30
Glaucoma	1235	78	182 PPI 25 Professionals	30
Inherited retinal diseases	280	63	27 PPI 25 Professionals	30
Neuro-ophthalmology	125	43	15 PPI 21 Professionals	30
Ocular cancer	26	19	Not required	19
Ocular inflammatory diseases	472	66	27 PPI 21 Professionals	30

Refractive error and ocular motility	188	70	21 PPI 23 Professionals	31
Retinal vascular diseases	205	56	15 PPI 12 Professionals	30
Vitreoretinal and ocular trauma	265	59	21 PPI 8 Professionals	30

PPI: Inclusive of patients, carers, relatives, organisation representatives.

Professionals: Inclusive of eye health professions.

Table 2 **Final workshop participants**

Category	Total number of workshop participants	Number of patients, relatives, carers, patient groups and organisations	Number of eye health professionals
Age-related Macular Degeneration	17	9	8
Cataract	11	5	6
Childhood-Onset Disorders	16	7	9
Corneal and External Diseases	12	5	7
Glaucoma	17	9	8
Inherited Retinal Diseases	19	11	8
Neuro-ophthalmology	10	6	4
Ocular Cancer	10	6	4
Ocular Inflammatory Diseases	10	5	5
Refractive Error and Ocular Motility	12	5	7
Retinal Vascular Diseases	11	3	8
Vitreoretinal and Ocular Trauma	10	7	3
TOTAL	**155**	**78**	**77**

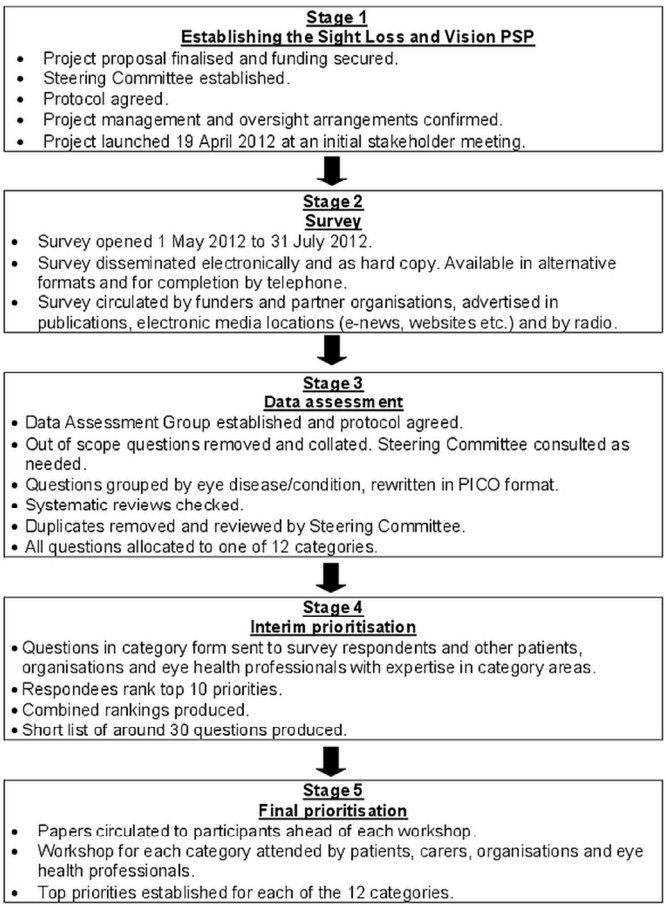
Table 3 Top ten lists per category

	Age-related macular degeneration	*Cataract*	*Childhood-onset disorders*	*Corneal and external diseases*	*Glaucoma*	*Inherited retinal diseases*
1	Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?	How can cataracts be prevented from developing?	How can cerebral visual impairment be identified, prevented and treated in children?	Can new therapies such as gene or stem cell treatments be developed for corneal diseases?	What are the most effective treatments for glaucoma and how can treatment be improved?	Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?
2	What is the cause of AMD?	Can the return of cloudy or blurred vision after cataract surgery known as posterior capsule opacity (PCO) or secondary cataract be prevented?	How can treatment for visual pathway damage associated with pre-term birth be developed?	What is the most effective management for dry eye and can new strategies be developed?	How can loss of vision be restored for people with glaucoma?	How can sight loss be prevented in an individual with inherited retinal disease?
3	How can AMD be prevented?	How can cataract progression be slowed down?	How do we improve screening and surveillance from the ante-natal period through to childhood to ensure early diagnosis of impaired vision and eye conditions?	Can treatments to save eye sight from microbial keratitis be improved?	How can glaucoma be stopped from progressing?	Is a genetic (molecular) diagnosis possible for all inherited retinal diseases?
4	Are there ways of restoring sight loss for people with AMD?	What alternatives to treat cataracts other than cataract surgery are being developed?	Can the treatment of amblyopia be improved to produce better short and long term outcomes than are possible with current treatments?	How can the rejection of corneal transplants be prevented?	What can be done to improve early diagnosis of sight-threatening glaucoma?	What factors affect the progression of sight loss in inherited retinal diseases?
5	Can the development of AMD be predicted?	What is the cause of cataract?	How can cataract be prevented in children?	Can the outcomes of corneal transplantation be improved?	What causes glaucoma?	What causes sight loss in inherited retinal diseases?
6	What is the most effective way to detect and monitor the progression of early AMD?	How can cataract surgery outcomes be improved?	What are the causes of coloboma and microphthalmia/anophthalmia and how can they be prevented?	What causes keratoconus to progress and can progression be prevented?	What is the most effective way of monitoring the progression of glaucoma?	What is the most effective way to support patients with inherited retinal disease?
7	What factors influence the progression of AMD?	How safe and effective is laser assisted cataract surgery?	Can vision be corrected in later life for people with amblyopia?	Can non-surgical therapy be developed for Fuchs' corneal dystrophy?	How can glaucoma patients with a higher risk to progress rapidly be detected?	Can the diagnosis of inherited retinal diseases be refined so that individuals can be given a clearer idea about their specific condition and how it is likely to progress?
8	Can a non-invasive therapy be developed for wet AMD?	Should accommodative lenses be developed for cataract surgery?	How can retinoblastoma be identified, prevented and treated in children?	Can corneal infections be prevented in high-risk individuals such as contact lens wearers?	Why is glaucoma more aggressive in people of certain ethnic groups, such as those of West African origin?	What is the relationship between sight loss and mental health for people with inherited retinal diseases?
9	Can dietary factors, nutritional supplements, complementary therapies or lifestyle changes prevent or slow the progression of AMD?	What is the best measure of visual disability due to cataract?	Can better treatments for glaucoma in children be developed?	What is the cause of keratoconus and can it be prevented?	How can glaucoma be prevented?	Would having a treatment for an inherited retinal disease preclude a patient from having another treatment?
10	What are the best enablement strategies for people with AMD?	Can retinal detachment be prevented after cataract surgery?	Can a treatment be developed to improve vision for people with albinism?	What is the most effective management of ocular complications associated with Stevens Johnson Syndrome?	Is there a link between treatment adherence and glaucoma progression and how can adherence be improved?	With regard to inherited retinal diseases what is the role of pre-natal and pre-implantation diagnosis in helping parents make informed choices?
11		What are the outcomes for cataract surgery among people with different levels of cognitive		Can severe ocular surface disease in children, such as blepharokeratoconjunctivitis and		

		impairment (all causes excluding dementia, stroke, neurological conditions, head injuries)?		vernal keratoconjunctivitis be managed better?		
	Neuro-ophthalmology	***Ocular cancer***	***Ocular inflammatory diseases***	***Refractive error and ocular motility***	***Retinal vascular diseases***	***Vitreoretinal and ocular trauma***
1	What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What can be done to help ocular cancer sufferers?	What are the most effective treatments for ocular and orbital inflammatory diseases?	What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?	What are the best methods to prevent retinopathy of prematurity?	How can surgical techniques be improved to save sight for eyes damaged by injury?
2	What are the most effective treatments and rehabilitation for optic neuropathies, e.g. Leber's hereditary optic neuropathy and anterior ischaemic optic neuropathy?	Can gene-based targeted therapies for ocular cancers be developed?	What causes thyroid eye disease?	What is the cause of both congenital and acquired nystagmus?	How can sight loss from diabetic retinal changes be prevented and reduced?	How can the risk of losing sight for people with retinal detachment be reduced?
3	Can vision loss due to optic nerve diseases such as giant cell arteritis, Leber's hereditary optic neuropathy, optic neuritis and optic atrophy, be restored, for example through gene therapy and stem cell treatment?	How can immunotherapy be used to fight metastatic ocular melanoma?	Can the severity of ocular and orbital inflammatory disease in an individual be predicted?	How can the development of binocular vision in young children with squint and amblyopia be promoted, and would the same approach work in older individuals without inducing intractable diplopia?	What are the predictive factors for the progression to sight threatening diabetic eye disease?	How can better interventions be developed that are effective in treating vitreous opacities/eye floaters?
4	What rehabilitation or treatment methods are most effective for vision loss following brain damage due to stroke, brain injury, cerebral vision impairment, tumours and dementias?	What are the most effective detection and screening methods for follow up to detect metastasis of ocular melanoma?	Is it possible to prevent further occurrences of retinal damage caused by toxoplasmosis?	Would correction of refractive error have a positive impact on early life learning and development?	Is there a way to improve screening of premature babies for retinopathy of prematurity?	What causes retinal detachment and can it be prevented?
5	What is the most effective way to assess vision in patients with neurological visual impairment i.e. stroke, dementia and cerebral/cortical visual impairment?	How can follow-up for ocular complications be managed in patients with ocular melanoma?	What causes birdshot retinopathy?	Does early diagnosis of refractive error improve long-term prognosis and promote faster, more effective treatment?	Can an effective long lasting treatment for diabetic macular oedema, both ischaemic and non-ischaemic, be developed?	Can more effective diagnostic tools be developed for assessing the vitreous and eye floaters?
6	Can the early stages of optic neuropathy be detected?	What is the best management of metastatic choroidal melanoma?	Why does disease burn out in patients with ocular and orbital inflammatory diseases?	What is the effect of congenital nystagmus on visual and emotional development?	Can a retinal vein occlusion be predicted and prevented?	Can a functioning prosthetic eye be developed to replace an eye damaged by injury?
7	How can optic neuropathies be prevented, for example anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What activates choroidal melanoma metastasis in the liver after the primary melanoma has been treated?	Can early detection methods be developed for ocular and orbital inflammatory diseases?	What is the most effective treatment for exotropia and when should it be delivered?	Can new non-invasive treatments be developed to slow down the progression of diabetic retinopathy?	How can epiretinal membrane/fibrosis be prevented or treated?
8	Can treatments be developed for visual field and ocular	Can adjuvant therapies be developed to treat ocular	What medications best prevent the development of eye disease in	How can the functional effects of surgical treatment for squint best	What are the barriers that prevent diabetic patients	Can stem cells be used to regrow an eye or part of an eye?

	motility manifestations following stroke?	melanoma?	Behcets?	be assessed?	having regular eye checks?	
9	How can electronic devices improve or restore vision for people with optic neuropathies?	What are the causes of ocular cancer and how can they be prevented?	What causes scleritis?	Could the accurate testing of refractive error be made less dependent on a subjective response i.e. the person's own response?	What rehabilitation programmes are best for the management of distorted vision from retinal diseases?	What causes posterior vitreous detachment/vitreous syneresis?
10	Can an alternative or new treatment be developed that will treat the sight loss caused by giant cell arteritis?	What is the most effective treatment for primary ocular melanoma?	Can diet or lifestyle changes prevent uveitis from developing?	How can myopia be prevented?	What is the efficacy and safety of anti-VEGF agents in the treatment of retinopathy of prematurity?	Are there methods to prevent and improve the treatment of macular holes?

Figure 1 Flowchart of SLVPSP process



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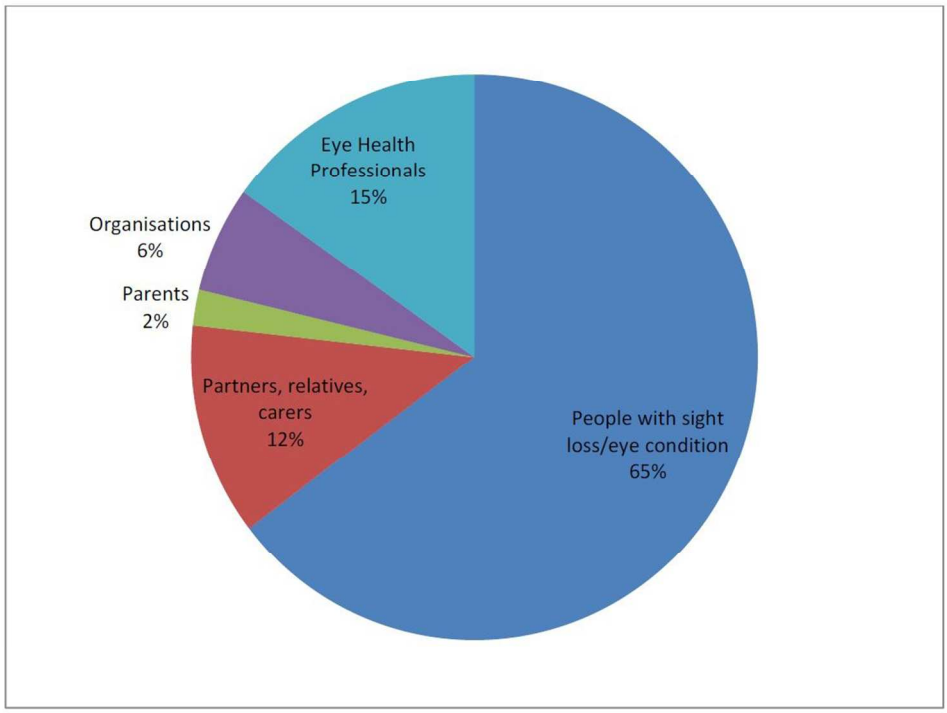

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The Sight Loss and Vision Priority Setting Partnership (SLV-PSP): Overview and results of the research prioritisation survey process.

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Abstract

Objectives: The Sight Loss and Vision Priority Setting Partnership aimed to identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians. These priorities can be used to inform funding bodies' decisions and enhance the case for additional research funding.

Design: Prospective survey with support from the James Lind Alliance.

Setting: UK wide NHS and non-NHS.

Participants: Patients, carers and eye health professionals. Academic researchers were excluded solely from the prioritisation process. The survey was disseminated by patient groups, professional bodies, at conferences and through the media, and was available for completion online, by phone, by post and by alternative formats (Braille and audio).

Outcome measure: People were asked to submit the questions about prevention, diagnosis and treatment of sight loss and eye conditions that they most wanted to see answered by research. Returned survey questions were reviewed by a data assessment group. Priorities were established across eye disease categories at final workshops.

Results: 2,220 people responded generating 4,461 submissions. Sixty-five percent of respondents had sight loss and/or an eye condition. Following initial data analysis, 686 submissions remained which were circulated for interim prioritisation (excluding cataract and ocular cancer questions) to 446 patients/carers and 218 professionals. The remaining 346 questions were discussed at final prioritisation workshops to reach agreement of top questions per category.

Conclusions: The exercise engaged a diverse community of stakeholders generating a wide range of conditions and research questions. Top priority questions were established across 12 eye disease categories.

Keywords: Sight loss; Vision; Research; Priorities; Partnership; James Lind Alliance

Article summary

Strengths and limitations of this study:

- Wide ranging and comprehensive coverage
- Substantial response
- The hardest to reach and those with the least opportunity to be heard may indeed have not been heard
- Any such groups now feeling excluded should have an opportunity to redress this.

Introduction

In the UK it is estimated that almost 2 million people are affected by sight loss and this number is expected to double by 2050[1]. Currently 50% of sight loss in the UK is avoidable but there are also many unavoidable sight loss conditions. Whether it is to address childhood eye conditions or those affecting adults, research is needed to inform us about prevention, to develop new techniques for early diagnosis and to develop new and more effective treatments for many eye conditions.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP) was developed from earlier work by the Eye Research Group (ERG) of VISION 2020 UK whose mission is the elimination of avoidable sight loss and the amelioration of the effects of sight loss when it cannot be avoided[2]. The UK Vision Strategy sets out ways of addressing avoidable sight loss in the UK. There is much that can be done to improve the nation's eye health, and to eliminate avoidable sight loss. There are however gaps in the evidence base regarding eye health interventions and the best way to deliver services. Research is needed to support the UK Vision Strategy. In key areas research is urgently required to enable the Vision Strategy to be implemented in an evidence-based way that ensures efficient and effective development.

Despite on-going research in the UK and other countries, there are still many questions about the prevention, diagnosis and treatment of sight loss and eye conditions that remain unanswered. Given that resources for research are limited, it is important for research funders to understand how patients, carers and eye health professionals prioritise these unanswered questions so that future research can be consolidated and targeted accordingly[3].

The purpose of this project was to undertake a comprehensive, UK-wide, survey of patients, carers and clinicians to identify research questions and priorities to inform decisions of funding bodies and enhance the case for additional research funding.

The priority setting process has been well established by the James Lind Alliance (JLA) (http://www.lindalliance.org/) which has supported partnerships on a range of topics since 2004. This partnership initiated by Fight for Sight, the UK's leading eye

research charity was established with support, financial and in kind, from the College of Optometrists, the Royal College of Ophthalmologists, the NIHR Moorfields Biomedical Research Centre, the RNIB, UK Vision Strategy and the Cochrane Eyes and Vision Group. A representative from the JLA convened meetings of the steering committee and provided independent chairmanship for this and the priority setting workshops. Their extensive experience in this process ensured no single voice exerted undue influence over the prioritisation process and that the views of patients, their carers and clinicians were paramount. The views of researchers with no clinical involvement with patients and views of commercial organisations were not included in the prioritisation.

Methods and materials

The detailed methods for this prioritisation process have been described in detail elsewhere[4]. In brief, the process comprised five stages (figure 1). Our study did not require ethical approval or consent from participants. James Lind Alliance priority setting partnerships do not require ethical approval. Dissemination of the survey was via open communications through professional bodies, charities and related organisations. The survey was not undertaken through Higher Education Institutes or through NHS organisations and does not recruit NHS patients. The survey contained clear information on the aims of the priority setting partnership, how the process works and how data will be used. In addition, submission of questions was anonymised. For the workshops in which priorities were discussed and agreed, participants choose to voluntarily attend and consent was not required for this. We followed the ethical guidance for participation, information and evaluation from the James Lind Alliance guidebook (http://www.jlaguidebook.org/jla-guidebook.asp?val=56).

Establishing the SLV-PSP

A steering committee and data assessment group comprising the authors of this article oversaw the process. Each member was responsible for contributing to and managing a part of the process and was selected for their expertise and association with eye research. The steering committee also included patient representatives

and eye health professionals. In April 2012 an initial stakeholder meeting was held to engage the groups and organisations with member bases and community influence. This was to ensure that the initial survey would be disseminated and completed by as many patients, relatives, carers and eye health professionals as possible in the UK.

Main survey

The SLV-PSP survey was launched on 1 May 2012 and was open until 31 July 2012. The aim of the survey was to identify patients', carers' and eye health professionals' unanswered questions about sight loss and eye conditions. The survey's primary question was "What question(s) about the prevention, diagnosis and treatment of sight loss and eye conditions would you like to see answered by research?"

Data analysis

Following closure of the survey, all submissions were examined. Out-of-scope submissions were removed including those not related to the topic and uncertainties better suited to social research. In-scope uncertainties were allocated into disease-specific groups and re-worded in PICO format (Population, Intervention, Comparison, Outcome). Searches were then undertaken to ascertain whether or not each uncertainty could be answered by an up-to-date systematic review. All unanswered uncertainties were then allocated to one of 12 eye disease categories, with duplicates removed and similar questions combined. Checks were also made to identify any on-going trials which might address the uncertainty. The 12 categories were formed following discussions by the steering group on the most logical and pragmatic way to organise the data within the time and resources available.

Interim prioritisation

In order to start reducing the number of uncertainties, an interim prioritisation exercise was conducted over email and by post. Patients, carers and eye health professionals were invited to examine the long lists and then choose and rank 10 of the uncertainties.

Final prioritisation

The remaining uncertainties were ranked by patients, carers, relatives, organisation representatives and eye health professionals in one-day workshops facilitated by the JLA, using Nominal Group Technique – a mix of discussion and ranking. For each category, the top 10/11 questions were agreed.

Results

Main survey

In response to the survey, 2220 people generated 4461 submissions. Of these respondents, 17% identified themselves as healthcare professionals including primarily ophthalmologists, optometrists, orthoptists, ophthalmic nurses, opticians and people working in social care and rehabilitation (figure 2). Over 60% were people with sight loss or an eye condition. The average age of survey participants was 65.7 years old (range:16 months (proxy completion of survey by adult carer)to 105 years). Just under two thirds (62%) of respondents were female. The geographical split was: England 89%; Scotland 6%; Wales 4%; Northern Ireland 1%.

Data analysis

Following data analysis to remove duplicate/answered/out of scope uncertainties, 686 uncertainties remained. These were divided into twelve eye disease categories. Table 1 shows each category with the initial number of submissions received after the survey responses were submitted, the number of uncertainties sent to interim prioritisation, the number of participants at interim prioritisation and the number of uncertainties considered at the final prioritisation workshops.

Interim prioritisation

Respondents from the initial survey, organisations and eye healthcare professionals with expertise in the eye diseases in ten categories were contacted to provide interim priority rankings. The smaller number of questions asked in the categories relating to cataract and ocular cancer meant that an interim prioritisation exercise was not required for either category. A large response was received for the interim exercise, with input from 446 patients, carers and relatives plus 218 eye health professionals.

Uncertainties accumulated scores based on rank and frequency, resulting in short lists of around 30 uncertainties per category which were taken to final workshops.

Final prioritisation

In April and May 2013, 12 final prioritisation workshops were held: one for each eye disease category. Balanced numbers of patients/carers/relatives and eye health professionals participated. In total, 155 participants attended across all 12 workshops: 78 patients and 77 clinicians (table 2). The topics debated at each workshop comprised between 19-31 questions per category. Overall 153 questions about sight loss and vision were considered resulting in lists of 10/11priorities for each of the 12 categories (table 3). The questions addressed the broad topics of aetiology, prevention, identification and interventions with the number 1 questions as follows:

Age-related macular degeneration

1. Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?

Cataract

1. How can cataracts be prevented from developing?

Childhood-onset disorders

1. How can cerebral visual impairment be identified, prevented and treated in children?

Corneal and external diseases

1. Can new therapies such as gene or stem cell treatments be developed for corneal diseases?

Glaucoma

1. What are the most effective treatments for glaucoma and how can treatment be improved?

Inherited retinal diseases

1. Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?

Neuro-ophthalmology

1. What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?

Ocular cancer

1. What can be done to help ocular cancer sufferers?

Ocular inflammatory diseases

1. What are the most effective treatments for ocular and orbital inflammatory diseases?

Refractive error and ocular motility

1. What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?

Retinal vascular diseases

1. What are the best methods to prevent retinopathy of prematurity?

Vitreoretinal and ocular trauma

1. How can surgical techniques be improved to save sight for eyes damaged by injury?

Discussion

About 50% of sight loss in the UK is currently avoidable[1]. Recognising this and the UK's pre-eminent position in eye research, the Vision 2020UK ERG considered that any UK Research Agenda must look at addressing unavoidable sight loss for it to be credible. The challenge was to produce a coherently constructed and constituted prioritised research agenda whose methods were clear and for which there had been inclusive and widespread consultation, where everyone with an interest had been offered the opportunity to contribute and be heard. As a result of this priority setting partnership we have established top ten lists of research questions for a range of eye conditions.

There are a number of strengths to this study. The SLV-PSP is unique because it sought the combined views of patients, carers and eye health professionals to identify uncertainties about the prevention, diagnosis and treatment of sight loss and eye conditions and prioritise them for research to address[5]. It is rare that those with direct experience of conditions are able to influence the research agenda[3,5,6]. The views of patients, carers and professionals were given equal merit. All submitted questions were evaluated independently and equally. Duplicate questions and out of scope questions were removed. We did not encounter particular misunderstandings between lay persons and professionals or insufficient knowledge of the public. Open discussions occurred during the face-to-face workshops with good communication and facilitation to encourage respectful listening in accordance with James Lind Alliance guidelines. Questions could only be pooled if this was agreed by patients, carers and professionals. Where there was no agreement, the questions remained separate. Thus, any differing perspectives of priorities between participants were acknowledged. We did not aim to compare and contrast questions from patients, carers and professionals but to represent and act on all.

This SLV-PSP provided an extensive set of unanswered questions prioritised by patients, carers and eye health professionals across twelve categories of eye conditions. These questions addressed a broad range of eye conditions and considered issues of aetiology, prevention, screening, assessment and management. Importantly, the public were as likely to propose questions in relation to aetiology, assessment and management just as professionals were as likely to raise questions regarding impact of sight loss.

In addition to the strengths of our study, we identified a number of limitations. We were unable to calculate a response rate for the survey because of the nature of its design and implementation. We did not request the views of 'pure' researchers (i.e. scientists with no current clinical practice) as these individuals are intentionally excluded from the priority setting process by the James Lind Alliance. This step is a key feature of the James Lind Alliance methodology in which the remit is to provide an opportunity for patients, carers and clinicians to influence the research agenda. We acknowledge this is a different approach but do not consider their exclusion as a flaw in this process as we include clinical researchers who did take part alongside clinicians and the public.

These questions may now be used to encourage researchers to investigate what is most important to these groups. We do not know how our research questions compare to the prioritisation of research areas by scientists, government agencies or other organisation research funders. We are unaware of any systematic data collating such data.

Various organisations in the sector have set out priorities for eye health and eye research in the past, for example Vision2020UK[7]. In addition, organisations representing the interests of patients/carers and eye health professionals took part in the process, both in promoting the survey and being directly involved in priority setting. Future work to review the SLV-PSP projects priorities with these organisations could be helpful in developing an understanding of how these new, patient and clinician led priorities can inform the sectors approach to commissioning research and focusing resources. Organisations in the sector are already working to review their organisational priorities with the SLV priorities, and have begun to invite researchers seeking funding to consider how their proposed research relates to the SLV priorities.

Similar to other PSP processes, we have provided information on our research priorities openly to national funding organisations and it is envisaged that research funders will be able to use the list to inform commissioned calls for research and identify which research applications to response mode funding opportunities can answer questions that these groups have agreed are a priority. Furthermore, any questions or uncertainties not prioritised in this process were submitted to and are currently available on the UK Database of Uncertainties about the Effects of Treatments (UK DUETs). Thus individuals looking for uncertainties for their research can access such information direct from UK DUETs. This sharing of information contributes to the quality assurance process of avoiding waste in research.

The SLV-PSP will also help to increase awareness of why research into sight loss and vision is necessary and important. It will be used to campaign for major funders to invest in sight loss and eye conditions, all of which are placing increased emphasis on researchers demonstrating how they have consulted and involved the public and patients in the process of developing their research.

These remain significant goals. For a sector with around 700 organisations to arrive at any kind of consensus for research priority areas, a process that was genuinely consultative, open and engaging to the individuals whose interests these organisations represent as well as at an organisational level was recognised as being critical. For a prioritisation exercise to be useful to the sector it needed to make sense to funders and statutory bodies with responsibilities and interests in these areas as well as to researchers. It was recognised that a prioritisation of research areas produced by a small group within the sector would not be credible and would never engage the support required for it to achieve the goals listed above.

Conclusions

Following a systematic process of national consultation and widespread survey of patients, carers and clinicians, 2220 individuals generated 4461 questions. Through a process of data analysis, interim prioritisation and final workshops, a top ten or eleven research questions have been identified for twelve categories of eye conditions. This is the first time, to our knowledge, that an exercise like this has been carried out anywhere in the world for sight loss and vision. Not only is this the most wide ranging and ambitious James Lind Alliance priority setting partnership, it also engaged a diversity of participants and enabled them to reach consensus together. For the first time, we have a clear idea of what the consumers of eye research – the patients and the people who care for and treat them – believe research money should be spent on. It has provided a focus for research in sight loss and vision and it is intended that these priorities are used to inform funders, researchers, clinicians and the public.

Competing interests: The authors declare that they have no competing interests.

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Data sharing statement: All original data are held by Fight for Sight. Extra data is available from: http://www.library.nhs.uk/duets/SearchResults.aspx?catID=14501 The additional unpublished data includes research questions not included in the final top ten lists of research priorities.

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Table 1 Categories of eye condition

	Initial number of survey questions	Number of questions at interim prioritisation	Number of participants at interim prioritisation	Number of questions at final prioritisation
Age-related macular degeneration	763	43	101 PPI 25 Professionals	29
Cataract	191	27	Not required	27
Childhood-onset disorders	125	69	12 PPI 20 Professionals	30
Corneal and external diseases	292	93	25 PPI 38 Professionals	30
Glaucoma	1235	78	182 PPI 25 Professionals	30
Inherited retinal diseases	280	63	27 PPI 25 Professionals	30
Neuro-ophthalmology	125	43	15 PPI 21 Professionals	30
Ocular cancer	26	19	Not required	19

Ocular inflammatory diseases	472	66	27 PPI 21 Professionals	30
Refractive error and ocular motility	188	70	21 PPI 23 Professionals	31
Retinal vascular diseases	205	56	15 PPI 12 Professionals	30
Vitreoretinal and ocular trauma	265	59	21 PPI 8 Professionals	30

PPI: Inclusive of patients, carers, relatives, organisation representatives.

Professionals: Inclusive of eye health professions.

Table 2 Final workshop participants

Category	Total number of workshop participants	Number of patients, relatives, carers, patient groups and organisations	Number of eye health professionals
Age-related Macular Degeneration	17	9	8
Cataract	11	5	6
Childhood-Onset Disorders	16	7	9
Corneal and External Diseases	12	5	7
Glaucoma	17	9	8
Inherited Retinal Diseases	19	11	8
Neuro-ophthalmology	10	6	4
Ocular Cancer	10	6	4
Ocular Inflammatory Diseases	10	5	5
Refractive Error and Ocular Motility	12	5	7
Retinal Vascular Diseases	11	3	8
Vitreoretinal and Ocular Trauma	10	7	3
TOTAL	**155**	**78**	**77**

Table 3 **Top ten lists per category**

	Age-related macular degeneration	***Cataract***	***Childhood-onset disorders***	***Corneal and external diseases***	***Glaucoma***	***Inherited retinal diseases***
1	Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?	How can cataracts be prevented from developing?	How can cerebral visual impairment be identified, prevented and treated in children?	Can new therapies such as gene or stem cell treatments be developed for corneal diseases?	What are the most effective treatments for glaucoma and how can treatment be improved?	Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?
2	What is the cause of AMD?	Can the return of cloudy or blurred vision after cataract surgery known as posterior capsule opacity (PCO) or secondary cataract be prevented?	How can treatment for visual pathway damage associated with pre-term birth be developed?	What is the most effective management for dry eye and can new strategies be developed?	How can loss of vision be restored for people with glaucoma?	How can sight loss be prevented in an individual with inherited retinal disease?
3	How can AMD be prevented?	How can cataract progression be slowed down?	How do we improve screening and surveillance from the ante-natal period through to childhood to ensure early diagnosis of impaired vision and eye conditions?	Can treatments to save eye sight from microbial keratitis be improved?	How can glaucoma be stopped from progressing?	Is a genetic (molecular) diagnosis possible for all inherited retinal diseases?
4	Are there ways of restoring sight loss for people with AMD?	What alternatives to treat cataracts other than cataract surgery are being developed?	Can the treatment of amblyopia be improved to produce better short and long term outcomes than are possible with current treatments?	How can the rejection of corneal transplants be prevented?	What can be done to improve early diagnosis of sight-threatening glaucoma?	What factors affect the progression of sight loss in inherited retinal diseases?
5	Can the development of AMD be predicted?	What is the cause of cataract?	How can cataract be prevented in children?	Can the outcomes of corneal transplantation be improved?	What causes glaucoma?	What causes sight loss in inherited retinal diseases?
6	What is the most effective way to detect and monitor the progression of early AMD?	How can cataract surgery outcomes be improved?	What are the causes of coloboma and microphthalmia/anophthalmia and how can they be prevented?	What causes keratoconus to progress and can progression be prevented?	What is the most effective way of monitoring the progression of glaucoma?	What is the most effective way to support patients with inherited retinal disease?
7	What factors influence the progression of AMD?	How safe and effective is laser assisted cataract surgery?	Can vision be corrected in later life for people with amblyopia?	Can non-surgical therapy be developed for Fuchs' corneal dystrophy?	How can glaucoma patients with a higher risk to progress rapidly be detected?	Can the diagnosis of inherited retinal diseases be refined so that individuals can be given a clearer idea about their specific condition and how it is likely to progress?
8	Can a non-invasive therapy be developed for wet AMD?	Should accommodative lenses be developed for cataract surgery?	How can retinoblastoma be identified, prevented and treated in children?	Can corneal infections be prevented in high-risk individuals such as contact lens wearers?	Why is glaucoma more aggressive in people of certain ethnic groups, such as those of West African origin?	What is the relationship between sight loss and mental health for people with inherited retinal diseases?
9	Can dietary factors, nutritional supplements, complementary therapies or lifestyle changes prevent or slow the progression of AMD?	What is the best measure of visual disability due to cataract?	Can better treatments for glaucoma in children be developed?	What is the cause of keratoconus and can it be prevented?	How can glaucoma be prevented?	Would having a treatment for an inherited retinal disease preclude a patient from having another treatment?
10	What are the best enablement strategies for people with AMD?	Can retinal detachment be prevented after cataract surgery?	Can a treatment be developed to improve vision for people with albinism?	What is the most effective management of ocular complications associated with Stevens Johnson Syndrome?	Is there a link between treatment adherence and glaucoma progression and how can adherence be improved?	With regard to inherited retinal diseases what is the role of pre-natal and pre-implantation diagnosis in helping parents make informed choices?
11		What are the outcomes for cataract surgery among people with different levels of cognitive		Can severe ocular surface disease in children, such as blepharokeratoconjunctivitis and		

		impairment (all causes excluding dementia, stroke, neurological conditions, head injuries)?		vernal keratoconjunctivitis be managed better?		
	Neuro-ophthalmology	***Ocular cancer***	***Ocular inflammatory diseases***	***Refractive error and ocular motility***	***Retinal vascular diseases***	***Vitreoretinal and ocular trauma***
1	What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What can be done to help ocular cancer sufferers?	What are the most effective treatments for ocular and orbital inflammatory diseases?	What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?	What are the best methods to prevent retinopathy of prematurity?	How can surgical techniques be improved to save sight for eyes damaged by injury?
2	What are the most effective treatments and rehabilitation for optic neuropathies, e.g. Leber's hereditary optic neuropathy and anterior ischaemic optic neuropathy?	Can gene-based targeted therapies for ocular cancers be developed?	What causes thyroid eye disease?	What is the cause of both congenital and acquired nystagmus?	How can sight loss from diabetic retinal changes be prevented and reduced?	How can the risk of losing sight for people with retinal detachment be reduced?
3	Can vision loss due to optic nerve diseases such as giant cell arteritis, Leber's hereditary optic neuropathy, optic neuritis and optic atrophy, be restored, for example through gene therapy and stem cell treatment?	How can immunotherapy be used to fight metastatic ocular melanoma?	Can the severity of ocular and orbital inflammatory disease in an individual be predicted?	How can the development of binocular vision in young children with squint and amblyopia be promoted, and would the same approach work in older individuals without inducing intractable diplopia?	What are the predictive factors for the progression to sight threatening diabetic eye disease?	How can better interventions be developed that are effective in treating vitreous opacities/eye floaters?
4	What rehabilitation or treatment methods are most effective for vision loss following brain damage due to stroke, brain injury, cerebral vision impairment, tumours and dementias?	What are the most effective detection and screening methods for follow up to detect metastasis of ocular melanoma?	Is it possible to prevent further occurrences of retinal damage caused by toxoplasmosis?	Would correction of refractive error have a positive impact on early life learning and development?	Is there a way to improve screening of premature babies for retinopathy of prematurity?	What causes retinal detachment and can it be prevented?
5	What is the most effective way to assess vision in patients with neurological visual impairment i.e. stroke, dementia and cerebral/cortical visual impairment?	How can follow-up for ocular complications be managed in patients with ocular melanoma?	What causes birdshot retinopathy?	Does early diagnosis of refractive error improve long-term prognosis and promote faster, more effective treatment?	Can an effective long lasting treatment for diabetic macular oedema, both ischaemic and non-ischaemic, be developed?	Can more effective diagnostic tools be developed for assessing the vitreous and eye floaters?
6	Can the early stages of optic neuropathy be detected?	What is the best management of metastatic choroidal melanoma?	Why does disease burn out in patients with ocular and orbital inflammatory diseases?	What is the effect of congenital nystagmus on visual and emotional development?	Can a retinal vein occlusion be predicted and prevented?	Can a functioning prosthetic eye be developed to replace an eye damaged by injury?
7	How can optic neuropathies be prevented, for example anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What activates choroidal melanoma metastasis in the liver after the primary melanoma has been treated?	Can early detection methods be developed for ocular and orbital inflammatory diseases?	What is the most effective treatment for exotropia and when should it be delivered?	Can new non-invasive treatments be developed to slow down the progression of diabetic retinopathy?	How can epiretinal membrane/fibrosis be prevented or treated?
8	Can treatments be developed for visual field and ocular	Can adjuvant therapies be developed to treat ocular	What medications best prevent the development of eye disease in	How can the functional effects of surgical treatment for squint best	What are the barriers that prevent diabetic patients	Can stem cells be used to regrow an eye or part of an eye?

	motility manifestations following stroke?	melanoma?	Behcets?	be assessed?	having regular eye checks?	
9	How can electronic devices improve or restore vision for people with optic neuropathies?	What are the causes of ocular cancer and how can they be prevented?	What causes scleritis?	Could the accurate testing of refractive error be made less dependent on a subjective response i.e. the person's own response?	What rehabilitation programmes are best for the management of distorted vision from retinal diseases?	What causes posterior vitreous detachment/vitreous syneresis?
10	Can an alternative or new treatment be developed that will treat the sight loss caused by giant cell arteritis?	What is the most effective treatment for primary ocular melanoma?	Can diet or lifestyle changes prevent uveitis from developing?	How can myopia be prevented?	What is the efficacy and safety of anti-VEGF agents in the treatment of retinopathy of prematurity?	Are there methods to prevent and improve the treatment of macular holes?

Figure legends

Figure 1: Flowchart showing the steps of the process from Stage 1 when establishing the PSP through to Stage 5 at the final prioritisation.

Figure 2: Background of respondents showing that questions were largely received from people who have sight loss or an eye condition, but also including eye health professions, organisations, parents, family and carers.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP): Overview and results of the research prioritisation survey process.

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Data sharing statement: All original data are held by Fight for Sight. Extra data is available from: http://www.library.nhs.uk/duets/SearchResults.aspx?catID=14501 The additional unpublished data includes research questions not included in the final top ten lists of research priorities.

Abstract

Objectives: The Sight Loss and Vision Priority Setting Partnership aimed to identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians. These priorities can be used to inform funding bodies' decisions and enhance the case for additional research funding.

Design: Prospective survey with support from the James Lind Alliance.

Setting: UK wide NHS and non-NHS.

Participants: Patients, carers and eye health professionals. Academic researchers were excluded solely from the prioritisation process. The survey was disseminated by patient groups, professional bodies, at conferences and through the media, and was available for completion online, by phone, by post and by alternative formats (Braille and audio).

Outcome measure: People were asked to submit the questions about prevention, diagnosis and treatment of sight loss and eye conditions that they most wanted to see answered by research. Returned survey questions were reviewed by a data assessment group. Priorities were established across eye disease categories at final workshops.

Results: 2,220 people responded generating 4,461 submissions. Sixty-five percent of respondents had sight loss and/or an eye condition. Following initial data analysis, 686 submissions remained which were circulated for interim prioritisation (excluding cataract and ocular cancer questions) to 446 patients/carers and 218 professionals. The remaining 346 questions were discussed at final prioritisation workshops to reach agreement of top questions per category.

Conclusions: The exercise engaged a diverse community of stakeholders generating a wide range of conditions and research questions. Top priority questions were established across 12 eye disease categories.

Keywords: Sight loss; Vision; Research; Priorities; Partnership; James Lind Alliance

Article summary

Strengths and limitations of this study:

- Wide ranging and comprehensive coverage
- Substantial response
- The hardest to reach and those with the least opportunity to be heard may indeed have not been heard
- Any such groups now feeling excluded should have an opportunity to redress this.

Introduction

In the UK it is estimated that almost 2 million people are affected by sight loss and this number is expected to double by 2050[1]. Currently 50% of sight loss in the UK is avoidable but there are also many unavoidable sight loss conditions. Whether it is to address childhood eye conditions or those affecting adults, research is needed to inform us about prevention, to develop new techniques for early diagnosis and to develop new and more effective treatments for many eye conditions.

The Sight Loss and Vision Priority Setting Partnership (SLV-PSP) was developed from earlier work by the Eye Research Group (ERG) of VISION 2020 UK whose mission is the elimination of avoidable sight loss and the amelioration of the effects of sight loss when it cannot be avoided[2]. The UK Vision Strategy sets out ways of addressing avoidable sight loss in the UK. There is much that can be done to improve the nation's eye health, and to eliminate avoidable sight loss. There are however gaps in the evidence base regarding eye health interventions and the best way to deliver services. Research is needed to support the UK Vision Strategy. In key areas research is urgently required to enable the Vision Strategy to be implemented in an evidence-based way that ensures efficient and effective development.

Despite on-going research in the UK and other countries, there are still many questions about the prevention, diagnosis and treatment of sight loss and eye conditions that remain unanswered. Given that resources for research are limited, it

is important for research funders to understand how patients, carers and eye health professionals prioritise these unanswered questions so that future research can be consolidated and targeted accordingly[3].

The purpose of this project was to undertake a comprehensive, UK-wide, survey of patients, carers and clinicians to identify research questions and priorities to inform decisions of funding bodies and enhance the case for additional research funding.

The priority setting process has been well established by the James Lind Alliance (JLA) (http://www.lindalliance.org/) which has supported partnerships on a range of topics since 2004. This partnership initiated by Fight for Sight, the UK's leading eye research charity was established with support, financial and in kind, from the College of Optometrists, the Royal College of Ophthalmologists, the NIHR Moorfields Biomedical Research Centre, the RNIB, UK Vision Strategy and the Cochrane Eyes and Vision Group. A representative from the JLA convened meetings of the steering committee and provided independent chairmanship for this and the priority setting workshops. Their extensive experience in this process ensured no single voice exerted undue influence over the prioritisation process and that the views of patients, their carers and clinicians were paramount. The views of researchers with no clinical involvement with patients and views of commercial organisations were not included in the prioritisation.

Methods and materials

The detailed methods for this prioritisation process have been described in detail elsewhere[4]. In brief, the process comprised five stages (figure 1). Our study did not require ethical approval or consent from participants. James Lind Alliance priority setting partnerships do not require ethical approval. Dissemination of the survey was via open communications through professional bodies, charities and related organisations. The survey was not undertaken through Higher Education Institutes or through NHS organisations and does not recruit NHS patients. The survey contained clear information on the aims of the priority setting partnership, how the process works and how data will be used. In addition, submission of questions was anonymised. For the workshops in which priorities were discussed and agreed,

participants choose to voluntarily attend and consent was not required for this. We followed the ethical guidance for participation, information and evaluation from the James Lind Alliance guidebook (http://www.jlaguidebook.org/jla-guidebook.asp?val=56).

Establishing the SLV-PSP

A steering committee and data assessment group comprising the authors of this article oversaw the process. Each member was responsible for contributing to and managing a part of the process and was selected for their expertise and association with eye research. The steering committee also included patient representatives and eye health professionals. In April 2012 an initial stakeholder meeting was held to engage the groups and organisations with member bases and community influence. This was to ensure that the initial survey would be disseminated and completed by as many patients, relatives, carers and eye health professionals as possible in the UK.

Main survey

The SLV-PSP survey was launched on 1 May 2012 and was open until 31 July 2012. The aim of the survey was to identify patients', carers' and eye health professionals' unanswered questions about sight loss and eye conditions. The survey's primary question was "What question(s) about the prevention, diagnosis and treatment of sight loss and eye conditions would you like to see answered by research?"

Data analysis

Following closure of the survey, all submissions were examined. Out-of-scope submissions were removed including those not related to the topic and uncertainties better suited to social research. In-scope uncertainties were allocated into disease-specific groups and re-worded in PICO format (Population, Intervention, Comparison, Outcome). Searches were then undertaken to ascertain whether or not each uncertainty could be answered by an up-to-date systematic review. All unanswered uncertainties were then allocated to one of 12 eye disease categories, with duplicates removed and similar questions combined. Checks were also made to identify any on-going trials which might address the uncertainty. The 12 categories

were formed following discussions by the steering group on the most logical and pragmatic way to organise the data within the time and resources available.

Interim prioritisation

In order to start reducing the number of uncertainties, an interim prioritisation exercise was conducted over email and by post. Patients, carers and eye health professionals were invited to examine the long lists and then choose and rank 10 of the uncertainties.

Final prioritisation

The remaining uncertainties were ranked by patients, carers, relatives, organisation representatives and eye health professionals in one-day workshops facilitated by the JLA, using Nominal Group Technique – a mix of discussion and ranking. For each category, the top 10/11 questions were agreed.

Results

Main survey

In response to the survey, 2220 people generated 4461 submissions. Of these respondents, 17% identified themselves as healthcare professionals including primarily ophthalmologists, optometrists, orthoptists, ophthalmic nurses, opticians and people working in social care and rehabilitation (figure 2). Over 60% were people with sight loss or an eye condition. The average age of survey participants was 65.7 years old (range:16 months (proxy completion of survey by adult carer)to 105 years). Just under two thirds (62%) of respondents were female. The geographical split was: England 89%; Scotland 6%; Wales 4%; Northern Ireland 1%.

Data analysis

Following data analysis to remove duplicate/answered/out of scope uncertainties, 686 uncertainties remained. These were divided into twelve eye disease categories. Table 1 shows each category with the initial number of submissions received after the survey responses were submitted, the number of uncertainties sent to interim

prioritisation, the number of participants at interim prioritisation and the number of uncertainties considered at the final prioritisation workshops.

Interim prioritisation

Respondents from the initial survey, organisations and eye healthcare professionals with expertise in the eye diseases in ten categories were contacted to provide interim priority rankings. The smaller number of questions asked in the categories relating to cataract and ocular cancer meant that an interim prioritisation exercise was not required for either category. A large response was received for the interim exercise, with input from 446 patients, carers and relatives plus 218 eye health professionals. Uncertainties accumulated scores based on rank and frequency, resulting in short lists of around 30 uncertainties per category which were taken to final workshops.

Final prioritisation

In April and May 2013, 12 final prioritisation workshops were held: one for each eye disease category. Balanced numbers of patients/carers/relatives and eye health professionals participated. In total, 155 participants attended across all 12 workshops: 78 patients and 77 clinicians (table 2). The topics debated at each workshop comprised between 19-31 questions per category. Overall 153 questions about sight loss and vision were considered resulting in lists of 10/11priorities for each of the 12 categories (table 3). The questions addressed the broad topics of aetiology, prevention, identification and interventions with the number 1 questions as follows:

Age-related macular degeneration

1. Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?

Cataract

1. How can cataracts be prevented from developing?

Childhood-onset disorders

1. How can cerebral visual impairment be identified, prevented and treated in children?

Corneal and external diseases

1. Can new therapies such as gene or stem cell treatments be developed for corneal diseases?

Glaucoma

1. What are the most effective treatments for glaucoma and how can treatment be improved?

Inherited retinal diseases

1. Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?

Neuro-ophthalmology

1. What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?

Ocular cancer

1. What can be done to help ocular cancer sufferers?

Ocular inflammatory diseases

1. What are the most effective treatments for ocular and orbital inflammatory diseases?

Refractive error and ocular motility

1. What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?

Retinal vascular diseases

1. What are the best methods to prevent retinopathy of prematurity?

Vitreoretinal and ocular trauma

1. How can surgical techniques be improved to save sight for eyes damaged by injury?

Discussion

About 50% of sight loss in the UK is currently avoidable[1]. Recognising this and the UK's pre-eminent position in eye research, the Vision 2020UK ERG considered that

any UK Research Agenda must look at addressing unavoidable sight loss for it to be credible. The challenge was to produce a coherently constructed and constituted prioritised research agenda whose methods were clear and for which there had been inclusive and widespread consultation, where everyone with an interest had been offered the opportunity to contribute and be heard. As a result of this priority setting partnership we have established top ten lists of research questions for a range of eye conditions.

There are a number of strengths to this study. The SLV-PSP is unique because it sought the combined views of patients, carers and eye health professionals to identify uncertainties about the prevention, diagnosis and treatment of sight loss and eye conditions and prioritise them for research to address[5]. It is rare that those with direct experience of conditions are able to influence the research agenda[3,5,6]. The views of patients, carers and professionals were given equal merit. All submitted questions were evaluated independently and equally. Duplicate questions and out of scope questions were removed. We did not encounter particular misunderstandings between lay persons and professionals or insufficient knowledge of the public. Open discussions occurred during the face-to-face workshops with good communication and facilitation to encourage respectful listening in accordance with James Lind Alliance guidelines. Questions could only be pooled if this was agreed by patients, carers and professionals. Where there was no agreement, the questions remained separate. Thus, any differing perspectives of priorities between participants were acknowledged. We did not aim to compare and contrast questions from patients, carers and professionals but to represent and act on all.

This SLV-PSP provided an extensive set of unanswered questions prioritised by patients, carers and eye health professionals across twelve categories of eye conditions. These questions addressed a broad range of eye conditions and considered issues of aetiology, prevention, screening, assessment and management. Importantly, the public were as likely to propose questions in relation to aetiology, assessment and management just as professionals were as likely to raise questions regarding impact of sight loss.

In addition to the strengths of our study, we identified a number of limitations. We were unable to calculate a response rate for the survey because of the nature of its

design and implementation. We did not request the views of 'pure' researchers (i.e. scientists with no current clinical practice) as these individuals are intentionally excluded from the priority setting process by the James Lind Alliance. This step is a key feature of the James Lind Alliance methodology in which the remit is to provide an opportunity for patients, carers and clinicians to influence the research agenda. We acknowledge this is a different approach but do not consider their exclusion as a flaw in this process as we include clinical researchers who did take part alongside clinicians and the public.

These questions may now be used to encourage researchers to investigate what is most important to these groups. We do not know how our research questions compare to the prioritisation of research areas by scientists, government agencies or other organisation research funders. We are unaware of any systematic data collating such data.

Various organisations in the sector have set out priorities for eye health and eye research in the past, for example Vision2020UK[7]. In addition, organisations representing the interests of patients/carers and eye health professionals took part in the process, both in promoting the survey and being directly involved in priority setting. Future work to review the SLV-PSP projects priorities with these organisations could be helpful in developing an understanding of how these new, patient and clinician led priorities can inform the sectors approach to commissioning research and focusing resources. Organisations in the sector are already working to review their organisational priorities with the SLV priorities, and have begun to invite researchers seeking funding to consider how their proposed research relates to the SLV priorities.

Similar to other PSP processes, we have provided information on our research priorities openly to national funding organisations and it is envisaged that research funders will be able to use the list to inform commissioned calls for research and identify which research applications to response mode funding opportunities can answer questions that these groups have agreed are a priority. Furthermore, any questions or uncertainties not prioritised in this process were submitted to and are currently available on the UK Database of Uncertainties about the Effects of Treatments (UK DUETs). Thus individuals looking for uncertainties for their research

can access such information direct from UK DUETs. This sharing of information contributes to the quality assurance process of avoiding waste in research.

The SLV-PSP will also help to increase awareness of why research into sight loss and vision is necessary and important. It will be used to campaign for major funders to invest in sight loss and eye conditions, all of which are placing increased emphasis on researchers demonstrating how they have consulted and involved the public and patients in the process of developing their research.

These remain significant goals. For a sector with around 700 organisations to arrive at any kind of consensus for research priority areas, a process that was genuinely consultative, open and engaging to the individuals whose interests these organisations represent as well as at an organisational level was recognised as being critical. For a prioritisation exercise to be useful to the sector it needed to make sense to funders and statutory bodies with responsibilities and interests in these areas as well as to researchers. It was recognised that a prioritisation of research areas produced by a small group within the sector would not be credible and would never engage the support required for it to achieve the goals listed above.

Conclusions

Following a systematic process of national consultation and widespread survey of patients, carers and clinicians, 2220 individuals generated 4461 questions. Through a process of data analysis, interim prioritisation and final workshops, a top ten or eleven research questions have been identified for twelve categories of eye conditions. This is the first time, to our knowledge, that an exercise like this has been carried out anywhere in the world for sight loss and vision. Not only is this the most wide ranging and ambitious James Lind Alliance priority setting partnership, it also engaged a diversity of participants and enabled them to reach consensus together. For the first time, we have a clear idea of what the consumers of eye research – the patients and the people who care for and treat them – believe research money should be spent on. It has provided a focus for research in sight loss and vision and it is intended that these priorities are used to inform funders, researchers, clinicians and the public.

Figure legends

Figure 1: Flowchart showing the steps of the process from Stage 1 when establishing the PSP through to Stage 5 at the final prioritisation.

Figure 2: Background of respondents showing that questions were largely received from people who have sight loss or an eye condition, but also including eye health professions, organisations, parents, family and carers.

References

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6. Caron-Flinterman J, et al. (2006) Patients prioritiesconcerning health research: the case of asthma andCOPD research in the Netherlands. Health Expectations,8, pp253–263.
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Table 1 Categories of eye condition

	Initial number of survey questions	Number of questions at interim prioritisation	Number of participants at interim prioritisation	Number of questions at final prioritisation
Age-related macular degeneration	763	43	101 PPI 25 Professionals	29
Cataract	191	27	Not required	27
Childhood-onset disorders	125	69	12 PPI 20 Professionals	30
Corneal and external diseases	292	93	25 PPI 38 Professionals	30
Glaucoma	1235	78	182 PPI 25 Professionals	30
Inherited retinal diseases	280	63	27 PPI 25 Professionals	30
Neuro-ophthalmology	125	43	15 PPI 21 Professionals	30
Ocular cancer	26	19	Not required	19
Ocular inflammatory diseases	472	66	27 PPI 21 Professionals	30

Refractive error and ocular motility	188	70	21 PPI 23 Professionals	31
Retinal vascular diseases	205	56	15 PPI 12 Professionals	30
Vitreoretinal and ocular trauma	265	59	21 PPI 8 Professionals	30

PPI: Inclusive of patients, carers, relatives, organisation representatives.

Professionals: Inclusive of eye health professions.

Table 2 **Final workshop participants**

Category	Total number of workshop participants	Number of patients, relatives, carers, patient groups and organisations	Number of eye health professionals
Age-related Macular Degeneration	17	9	8
Cataract	11	5	6
Childhood-Onset Disorders	16	7	9
Corneal and External Diseases	12	5	7
Glaucoma	17	9	8
Inherited Retinal Diseases	19	11	8
Neuro-ophthalmology	10	6	4
Ocular Cancer	10	6	4
Ocular Inflammatory Diseases	10	5	5
Refractive Error and Ocular Motility	12	5	7
Retinal Vascular Diseases	11	3	8
Vitreoretinal and Ocular Trauma	10	7	3
TOTAL	**155**	**78**	**77**

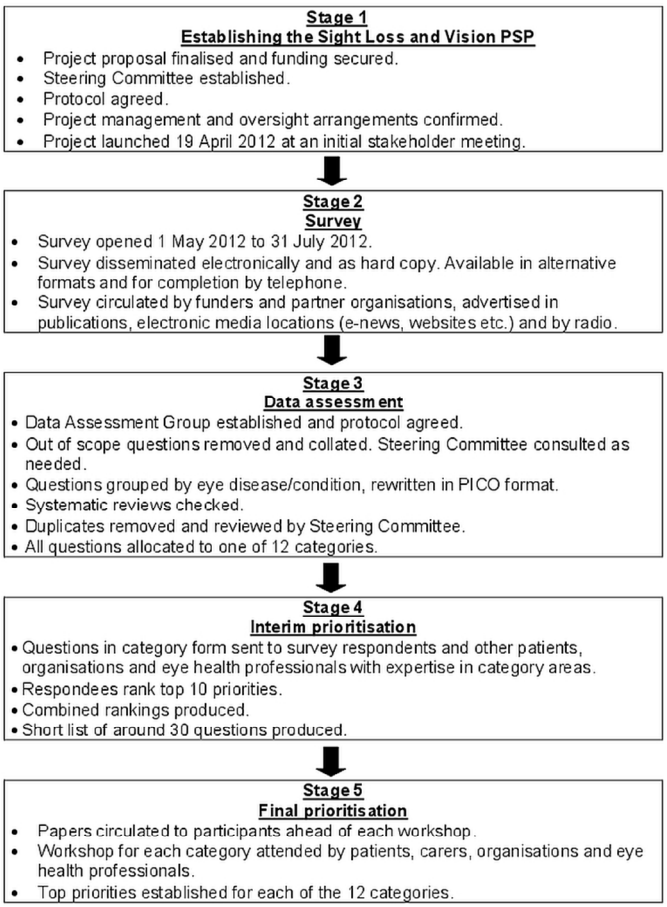
Table 3 Top ten lists per category

	Age-related macular degeneration	*Cataract*	*Childhood-onset disorders*	*Corneal and external diseases*	*Glaucoma*	*Inherited retinal diseases*
1	Can a treatment to stop dry AMD progressing and/or developing into the wet form be devised?	How can cataracts be prevented from developing?	How can cerebral visual impairment be identified, prevented and treated in children?	Can new therapies such as gene or stem cell treatments be developed for corneal diseases?	What are the most effective treatments for glaucoma and how can treatment be improved?	Can a treatment to slow down progression or reverse sight loss in inherited retinal diseases be developed?
2	What is the cause of AMD?	Can the return of cloudy or blurred vision after cataract surgery known as posterior capsule opacity (PCO) or secondary cataract be prevented?	How can treatment for visual pathway damage associated with pre-term birth be developed?	What is the most effective management for dry eye and can new strategies be developed?	How can loss of vision be restored for people with glaucoma?	How can sight loss be prevented in an individual with inherited retinal disease?
3	How can AMD be prevented?	How can cataract progression be slowed down?	How do we improve screening and surveillance from the ante-natal period through to childhood to ensure early diagnosis of impaired vision and eye conditions?	Can treatments to save eye sight from microbial keratitis be improved?	How can glaucoma be stopped from progressing?	Is a genetic (molecular) diagnosis possible for all inherited retinal diseases?
4	Are there ways of restoring sight loss for people with AMD?	What alternatives to treat cataracts other than cataract surgery are being developed?	Can the treatment of amblyopia be improved to produce better short and long term outcomes than are possible with current treatments?	How can the rejection of corneal transplants be prevented?	What can be done to improve early diagnosis of sight-threatening glaucoma?	What factors affect the progression of sight loss in inherited retinal diseases?
5	Can the development of AMD be predicted?	What is the cause of cataract?	How can cataract be prevented in children?	Can the outcomes of corneal transplantation be improved?	What causes glaucoma?	What causes sight loss in inherited retinal diseases?
6	What is the most effective way to detect and monitor the progression of early AMD?	How can cataract surgery outcomes be improved?	What are the causes of coloboma and microphthalmia/anophthalmia and how can they be prevented?	What causes keratoconus to progress and can progression be prevented?	What is the most effective way of monitoring the progression of glaucoma?	What is the most effective way to support patients with inherited retinal disease?
7	What factors influence the progression of AMD?	How safe and effective is laser assisted cataract surgery?	Can vision be corrected in later life for people with amblyopia?	Can non-surgical therapy be developed for Fuchs' corneal dystrophy?	How can glaucoma patients with a higher risk to progress rapidly be detected?	Can the diagnosis of inherited retinal diseases be refined so that individuals can be given a clearer idea about their specific condition and how it is likely to progress?
8	Can a non-invasive therapy be developed for wet AMD?	Should accommodative lenses be developed for cataract surgery?	How can retinoblastoma be identified, prevented and treated in children?	Can corneal infections be prevented in high-risk individuals such as contact lens wearers?	Why is glaucoma more aggressive in people of certain ethnic groups, such as those of West African origin?	What is the relationship between sight loss and mental health for people with inherited retinal diseases?
9	Can dietary factors, nutritional supplements, complementary therapies or lifestyle changes prevent or slow the progression of AMD?	What is the best measure of visual disability due to cataract?	Can better treatments for glaucoma in children be developed?	What is the cause of keratoconus and can it be prevented?	How can glaucoma be prevented?	Would having a treatment for an inherited retinal disease preclude a patient from having another treatment?
10	What are the best enablement strategies for people with AMD?	Can retinal detachment be prevented after cataract surgery?	Can a treatment be developed to improve vision for people with albinism?	What is the most effective management of ocular complications associated with Stevens Johnson Syndrome?	Is there a link between treatment adherence and glaucoma progression and how can adherence be improved?	With regard to inherited retinal diseases what is the role of pre-natal and pre-implantation diagnosis in helping parents make informed choices?
11		What are the outcomes for cataract surgery among people with different levels of cognitive		Can severe ocular surface disease in children, such as blepharokeratoconjunctivitis and		

		impairment (all causes excluding dementia, stroke, neurological conditions, head injuries)?		vernal keratoconjunctivitis be managed better?		
	Neuro-ophthalmology	***Ocular cancer***	***Ocular inflammatory diseases***	***Refractive error and ocular motility***	***Retinal vascular diseases***	***Vitreoretinal and ocular trauma***
1	What is the underlying cause of optic nerve damage in optic neuropathies, such as anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What can be done to help ocular cancer sufferers?	What are the most effective treatments for ocular and orbital inflammatory diseases?	What factors influence the development of refractive error (myopia, astigmatism, presbyopia and long-sightedness)?	What are the best methods to prevent retinopathy of prematurity?	How can surgical techniques be improved to save sight for eyes damaged by injury?
2	What are the most effective treatments and rehabilitation for optic neuropathies, e.g. Leber's hereditary optic neuropathy and anterior ischaemic optic neuropathy?	Can gene-based targeted therapies for ocular cancers be developed?	What causes thyroid eye disease?	What is the cause of both congenital and acquired nystagmus?	How can sight loss from diabetic retinal changes be prevented and reduced?	How can the risk of losing sight for people with retinal detachment be reduced?
3	Can vision loss due to optic nerve diseases such as giant cell arteritis, Leber's hereditary optic neuropathy, optic neuritis and optic atrophy, be restored, for example through gene therapy and stem cell treatment?	How can immunotherapy be used to fight metastatic ocular melanoma?	Can the severity of ocular and orbital inflammatory disease in an individual be predicted?	How can the development of binocular vision in young children with squint and amblyopia be promoted, and would the same approach work in older individuals without inducing intractable diplopia?	What are the predictive factors for the progression to sight threatening diabetic eye disease?	How can better interventions be developed that are effective in treating vitreous opacities/eye floaters?
4	What rehabilitation or treatment methods are most effective for vision loss following brain damage due to stroke, brain injury, cerebral vision impairment, tumours and dementias?	What are the most effective detection and screening methods for follow up to detect metastasis of ocular melanoma?	Is it possible to prevent further occurrences of retinal damage caused by toxoplasmosis?	Would correction of refractive error have a positive impact on early life learning and development?	Is there a way to improve screening of premature babies for retinopathy of prematurity?	What causes retinal detachment and can it be prevented?
5	What is the most effective way to assess vision in patients with neurological visual impairment i.e. stroke, dementia and cerebral/cortical visual impairment?	How can follow-up for ocular complications be managed in patients with ocular melanoma?	What causes birdshot retinopathy?	Does early diagnosis of refractive error improve long-term prognosis and promote faster, more effective treatment?	Can an effective long lasting treatment for diabetic macular oedema, both ischaemic and non-ischaemic, be developed?	Can more effective diagnostic tools be developed for assessing the vitreous and eye floaters?
6	Can the early stages of optic neuropathy be detected?	What is the best management of metastatic choroidal melanoma?	Why does disease burn out in patients with ocular and orbital inflammatory diseases?	What is the effect of congenital nystagmus on visual and emotional development?	Can a retinal vein occlusion be predicted and prevented?	Can a functioning prosthetic eye be developed to replace an eye damaged by injury?
7	How can optic neuropathies be prevented, for example anterior ischaemic optic neuropathy, Leber's hereditary optic neuropathy, optic neuritis and other optic neuropathies?	What activates choroidal melanoma metastasis in the liver after the primary melanoma has been treated?	Can early detection methods be developed for ocular and orbital inflammatory diseases?	What is the most effective treatment for exotropia and when should it be delivered?	Can new non-invasive treatments be developed to slow down the progression of diabetic retinopathy?	How can epiretinal membrane/fibrosis be prevented or treated?
8	Can treatments be developed for visual field and ocular	Can adjuvant therapies be developed to treat ocular	What medications best prevent the development of eye disease in	How can the functional effects of surgical treatment for squint best	What are the barriers that prevent diabetic patients	Can stem cells be used to regrow an eye or part of an eye?

	motility manifestations following stroke?	melanoma?	Behcets?	be assessed?	having regular eye checks?	
9	How can electronic devices improve or restore vision for people with optic neuropathies?	What are the causes of ocular cancer and how can they be prevented?	What causes scleritis?	Could the accurate testing of refractive error be made less dependent on a subjective response i.e. the person's own response?	What rehabilitation programmes are best for the management of distorted vision from retinal diseases?	What causes posterior vitreous detachment/vitreous syneresis?
10	Can an alternative or new treatment be developed that will treat the sight loss caused by giant cell arteritis?	What is the most effective treatment for primary ocular melanoma?	Can diet or lifestyle changes prevent uveitis from developing?	How can myopia be prevented?	What is the efficacy and safety of anti-VEGF agents in the treatment of retinopathy of prematurity?	Are there methods to prevent and improve the treatment of macular holes?

Figure 1 Flowchart of SLVPSP process



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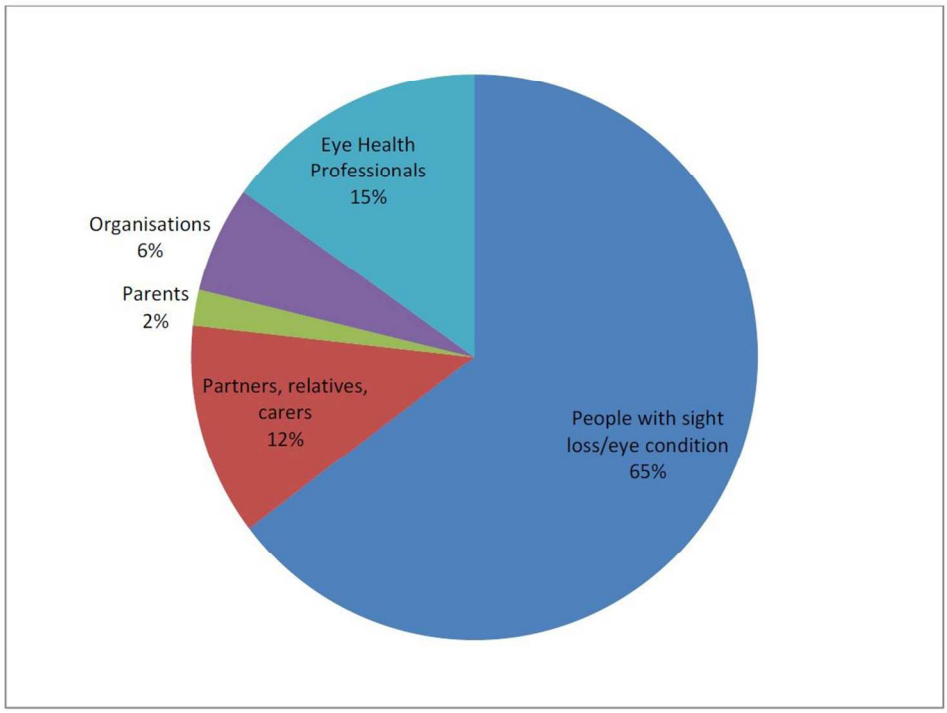

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