Eye (2011) 25, 403–413

www.nature.com/eye

REVIEW

# Amblyopia and quality of life: a systematic review

J Carlton and E Kaltenthaler


## Abstract

**Amblyopia is a common condition, which can affect up to 5% of the general population. Health-related quality-of-life (HRQoL) implications of amblyopia and/or its treatment have been explored in the literature.**
**A systematic literature search was undertaken during the period of 7–14 May 2010 to identify the HRQoL implications of amblyopia and/or its treatment. A total of 35 papers were included in the literature review. The HRQoL implications of amblyopia related specifically to amblyopia treatment, rather than to the condition itself.**
**These included impact on family life, social interactions, difficulties in undertaking daily activities, as well as feelings and behaviour. The identified studies adopted a number of methodologies. The study populations included children with the condition, parents of children with amblyopia, and adults who had undertaken amblyopia treatment as a child. Some studies developed their own measures of HRQoL, and others determined HRQoL through proxy measures. The reported findings of the HRQoL implications are of importance when considering the management of cases of amblyopia. The issues identified in the literature review are discussed with respect to how HRQoL is measured (treatment compliance *vs* proxy measures), and whether HRQoL is taken from a child's or a parent's perspective. Changing societal views over glasses and occlusion therapy are also discussed. Further research is required to assess the immediate and long-term effects of amblyopia and/or its treatment on HRQoL using a more standardised approach.**

*Eye* (2011) **25,** 403–413; doi:10.1038/eye.2011.4; published online 28 January 2011

*Keywords:* amblyopia; health-related quality of life (HRQoL); child; psychosocial

## Introduction

The impact of amblyopia on health-related quality of life (HRQoL) has not been adequately explored. Amblyopia is an important condition that can affect up to 5% of the general population.[1] Despite an increasing body of evidence describing the effectiveness of amblyopia treatment, little robust evidence regarding HRQoL implications of the condition and/or its treatment is emerging. Within the allocation of health-care resources, there is increasing demand for evidence regarding not only treatment effectiveness but also the implication of the condition and/or the effect its treatment has on the patient in both the immediate and the long term. The use of patient-reported outcomes, such as HRQoL questionnaires, can be useful in determining the impact a condition has on an individual.

Screening programmes currently exist within the United Kingdom to identify children who have or those who are at risk of developing amblyopia. A recent report examined the clinical importance and cost-effectiveness of pre-school vision screening for children aged up to 5 years.[1] It concluded that the cost-effectiveness of screening for amblyopia is dependent on the long-term utility (or HRQoL) effects of unilateral vision loss. However, the authors noted that the evidence of the impact of amblyopia and/or its treatment on HRQoL was limited. The purpose of this study is to undertake a systematic literature review to examine the HRQoL implications of amblyopia and/or its treatment, and to evaluate the measures identified in the reported studies.

## Materials and methods

### *Literature search*

A systematic literature search was undertaken during the period of 7–14 May 2010. The electronic databases searched are detailed in Appendix 1. Specific search strategies were

Health Economics and Decision Science (HEDS), School of Health and Related Research (ScHARR), University of Sheffield, Sheffield, UK

Correspondence: J Carlton, Health Economics and Decision Science (HEDS), School of Health and Related Research (ScHARR), University of Sheffield, Regent Court, 30 Regent Street, Sheffield S1 4DA, UK
Tel: +44 (0)114 2220799; Fax: +44 (0)114 2724095.
E-mail: j.carlton@sheffield.ac.uk

Received: 20 August 2010
Accepted in revised form: 17 December 2010
Published online: 28 January 2011

used for each database. Search strategies were performed to identify literature pertaining to amblyopia terms, amblyopia treatment terms, children terms, and QoL terms. No date or language restrictions were applied. Details of the literature search terms and database search strategies are shown Appendix 1.

A total of 1876 articles were identified through the database searches. An additional 10 articles were identified through a recent HTA publication[1] and two systematic reviews on amblyopia screening and treatment.[2,3] These articles were not identified because the publication was in a journal that was not included in the search engines used (ie, articles were published in journals not found on Medline). After the removal of duplicates, a total of 632 articles were applicable for this review. Every article identified was checked by one reviewer (JC) and subjected to a pre-determine inclusion/exclusion criteria. Articles were rejected at title if they were not related to the subject area ($n=479$), and rejected at abstract if they were in a non-English publication or not pertinent to the research question ($n=111$). Letters, reviews, and editorials describing other studies reporting HRQoL implications of amblyopia were excluded. Where abstracts were ambiguous, the article was obtained. A further seven articles were rejected at full paper stage. These were found to be review papers, summaries of other studies, or contained no data to inform the research question.

A total of 35 articles were included in the review. The PRISMA flow diagram of study identification is shown in Figure 1. Details of the included papers are shown in Table 1.

### *Data extraction and synthesis*

Data were extracted by one reviewer (JC). Papers were assessed and data extracted using a data extraction form (see Appendix 2). Papers were examined in terms of the instruments used in the study. Newly developed HRQoL instruments identified were assessed in terms of reliability, validity, and responsiveness (see Table 2). Studies were also examined to determine whether the study respondents were children, parents, or adults who had undergone amblyopia treatment as a child. Finally, the HRQoL implications of amblyopia were extracted.

## Results

The majority of the studies reported HRQoL from a parental perspective ($n=22$).[4–7,13–23,28,29,31,32,34,36,37] Some studies reported results from adults who had amblyopia as a child ($n=9$).[8–11,24,26,27,30,33] One study involved questioning both parents and children ($n=1$).[25] Only three studies reported results solely from the child's perspective ($n=3$).[12,35,38]

### *Study methodology: instruments used*

From the 35 papers identified, 5 used an existing measure in their study methodology to determine the impact of amblyopia on HRQoL. Three studies used the Children's Visual Function Questionnaire (CVFQ).[4,7,23] One study used the Self-Perception Profile for Children (SPPC),[12] and one study used the Visual Function Index (VF-14).[8]

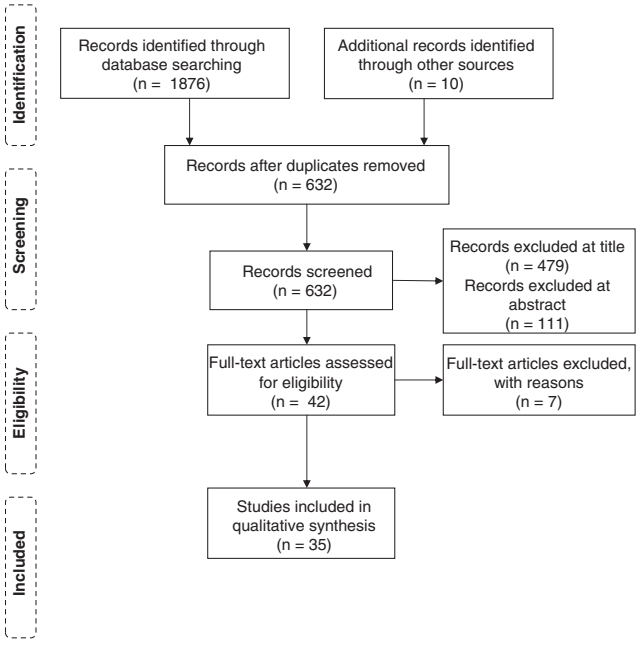


**Figure 1** PRISMA flow diagram of study identification.

**Table 1** Articles included in the review and their source

| *Database* | *Articles identified* |
|---|---|
| HTA | Carlton *et al*[1] |
| CINAHL | Carlton *et al*[1] |
| DARE | Schmucker *et al*[2,3] |
| SCI and SSCI | Carlton *et al*,[1] Schmucker *et al*,[3] Birch *et al*,[4] Holmes *et al*,[5,6] Loudon *et al*,[7] Sabri *et al*,[8] van de Graaf *et al*,[9,10] Vianya-Estopa *et al*,[11] Webber *et al*[12] |
| CRCT | Holmes *et al*,[5,6] Cole *et al*,[13] Göransson *et al*,[14] Hrisos *et al*,[15] Loudon *et al*,[16–19] Newsham,[20] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Roefs *et al*[23] |
| Ovid | Carlton *et al*,[1] Schmucker *et al*,[2,3] Holmes *et al*,[5] van de Graaf *et al*,[9,10,30] Vianya-Estopa *et al*,[11] Webber *et al*,[12] Cole *et al*,[13] Felius *et al*,[24] Koklanis *et al*,[25] Packwood *et al*,[26] Rahi *et al*,[27] Searle *et al*,[28] Searle,[29] Choong *et al*,[31] |
| Carlton *et al*[1] | Newsham,[20,37] Pediatric Eye Disease Investigator Group,[22] Packwood *et al*,[26] Rahi *et al*,[27] Searle *et al*,[28] van de Graaf *et al*,[30] Choong *et al*,[31] Parkes,[32] Chua and Mitchell,[33] Dixon-Woods *et al*,[34] Horwood *et al*,[35] Leach,[36] Williams *et al*[38] |
| Schmucker *et al*[2] | Carlton *et al*,[1] Hrisos *et al*,[15] Horwood *et al*[34] |
| Schmucker *et al*[3] | Pediatric Eye Disease Investigator Group,[22] Packwood *et al*,[26] Chua and Mitchell,[33] Horwood *et al*,[35] Williams *et al*[38] |

Abbreviations: NHS EED, NHS Economic Evaluation Database (0 identified); CMR, Cochrane Methodology Register (0 identified); HTA, Health Technology Assessment database (1 identified); CRCT, Cochrane Register of Controlled Trials (0 identified); DARE, Database of Abstracts of Reviews of Effects (2 identified); CDSR, Cochrane Database of Systematic Reviews (0 identified); CINAHL, Cumulative Index to Nursing and Allied Health Literature (1 identified); SCI and SSCI, Science Citation Index and Social Sciences Citation Index (5 identified).
Ovid, Medline; PsychINFO; AMED (Allied and Complementary Medicine); British Nursing Index and Archive; and Econlit (17 identified).
Scopus; 0 identified.

**Table 2** Assessment of HRQoL measures

| | |
|---|---|
| Reliability | • 'Ability of a measure to reproduce the same value on two separate occasions when there has been no change in health'[39]<br>• Can be over time or between methods of administration[39]<br>• May be considered in terms of internal consistency (the extent to which all items measure the same concept or test–retest reliability (the extent to which the results of the instrument compare if the test is administered to the same subject on more than one occasion when there has been no demonstrable change in health status) |
| Validity | • The extent to which a measure reflects the concept that it is intended to measure<br>• May be considered in terms of content validity ('degree to which the instrument is reflective of aspects important to the patients and disease of interest'), construct validity ('how well a measure correlates with other indicators of similar or related constructs'), concurrent validity ('the extent to which an instrument correlates with other measures of the same or similar construct'), and discriminant validity ('the ability to discriminate between either cases *vs* controls or disease severity groups')[40]<br>• For the purpose of this paper, construct validity will be determined if compared with objective clinical measures such as visual acuity; concurrent validity will be a comparison to an existing vision-specific HRQoL measure<br>• Factor analysis is a method of determining the structure of an instrument in terms of domains or subscales. It can be used to identify redundant or duplicate items. It may also be used to determine the domain structure. Some papers refer to this as a measure of internal validity |
| Responsiveness | • The extent to which the instrument can detect in patients known to have a change in their physical condition |

*The CVFQ*

The CVFQ is a vision-specific instrument designed for use in children up to 7 years of age. Two versions are available for younger (<3 years of age, which contains 34 items) and older children (3–7 years, which contains 39 items). The instrument consists of four dimensions: competence, personality, family impact, and treatment difficulty, and has undergone testing of reliability and validity.[41]

*The SPPC*

Webber *et al*[12] used the SPPC to explore the effect amblyopia has on a child's self-esteem. This measure has been assessed for reliability and validity[42] and has been used in other ophthalmological studies to determine the impact of myopia on a child's well-being.[43,44] The measure consists of 36 items, which form 6 domains (namely scholastic competence, social acceptance, athletic competence, physical appearance, behavioural conduct, and global self-worth). Each domain contains six questions, which are answered on a four-part scale.

*The VF-14*

The VF-14 is a well-recognised measure of vision-related functional status that has been used in many areas

of ophthalmology research, particularly cataract.[45] The measure consists of 14 items relating to activities of daily living, which are answered on a 4-part scale.[45]

*Developed measures*

Five studies were identified that developed their own instruments, and described the psychometric properties of these measures. These include the Amblyopia Treatment Index (ATI),[13] the Amblyopia and Strabismus Questionnaire (A&SQ),[30] the Psychological Impact Questionnaire,[8] the Protection Motivation Theory (PMT) Questionnaire,[28] and the Patching Success Questionnaire (PSQ).[16] These are summarised in Table 3.

The ATI has undergone further testing of validity.[6] The literature search identified that this instrument has been used in subsequent studies to investigate the impact of amblyopia and/or its treatment.[5,21,22] The A&SQ has undergone additional testing of validity and reliability.[9–11] An English version has also been developed and tested.[24]

Sabri *et al*[8] developed a Psychological Impact Questionnaire and administered this in conjunction with the VF-14 to assess the construct validity of their questionnaire. The measure was developed through a literature search, clinician input, and discussion groups with amblyopic subjects. It consists of eight questions, which are answered on a five-part Likert scale.

Searle *et al*[28] produced a questionnaire, based on the application of PMT applied to the results of interviews of parents of amblyopic children.[29] The questionnaire contains seven domains and the questions are asked using a five-point Likert-type scale, although the exact number of questions is not clear from the study methodology.

Loudon *et al*[19] developed a PSQ, a questionnaire based on the PMT. Additional questions were included to incorporate experiences of clinicians treating patients with amblyopia. The PSQ has been used in subsequent studies to explore the impact of amblyopia and/or its treatment.[16–18]

A number of papers ($n=7$) developed their own questionnaires (Table 4).[15,20,26,31,32,36,37] The psychometric properties of these instruments were not disclosed.

**Table 3** Summary of developed HRQoL instruments used in studies

| *Instrument* | *Item pool development* | *Number of questions* | *Likert-type scale used* | *Domains or subscales* | *Mode of administration* | *Psychometrics* |
|---|---|---|---|---|---|---|
| ATI[5,6,13,21,22] | CB, LB | 18 (atropine)<br>19 (patching) | Five-point<br>Five-point | Adverse effects<br>Compliance<br>Social stigma | Parent | IC, CV |
| A&SQ[9,10,11,24,30] | CB | 26 | Five-point | Fear of losing better eye<br>Distance estimation<br>Visual disorientation<br>Diplopia<br>Problems with social contact and cosmetic problems | Self | IC, DV, CV, CCV |
| Protection Motivation Theory Questionnaire[28] | LB | Unclear | Five-point | Protection motivation<br>Severity<br>Vulnerability<br>Response efficacy<br>Distress barrier<br>Stigma barrier<br>Self-efficacy | Parent | IC, CV |
| Psychological Impact Questionnaire[8] | CB, LB, PB | 32 (8 questions asked times in relation to 4 factors; in general daily life, having a weaker eye, wearing glasses, having noticeable strabismus | Five-point | Not categorised | Self | CV, CCV, TRR |
| PSQ[19] | LB, CB | 60 | Five-point | As PMT but additionally:<br>Knowledge disease<br>Knowledge treatment<br>Logistics of treatment | Parent | CV |

Abbreviations: CB, clinician based; CCV, concurrent validity; CV, construct validity; DV, discriminant validity; IC, internal consistency; LB, literature based; PB, patient based; R, responsiveness; TRR, test–retest reliability.

**Table 4** Summary of studies that developed their own questionnaires

| *Study* | *Country of origin* | *Questionnaire development* | *Mode of administration* | *Results compared with any other measure?* |
|---|---|---|---|---|
| Choong *et al*[31] | United Kingdom | CB, PAC | Parents | Perceived Stress Index (PSI) |
| Hrisos *et al*[15] | United Kingdom | CB, LB, PAC | Parents | Revised Rutter Parents Scale for Preschool Children |
| Leach[36] | United Kingdom | CB | Parents | — |
| Newsham[20] | United Kingdom | CB | Parents | — |
| Newsham[37] | United Kingdom | CB | Parents | — |
| Packwood *et al*[26] | United States of America | CB | Self (adults) | — |
| Parkes[32] | United Kingdom | CB | Parents | — |

Abbreviations: CB, clinician based; LB, literature based; PAC, parent of amblyopic child; PB, patient based.

Six studies used qualitative methods to report the HRQoL implications of amblyopia and/or its treatment ($n=6$).[14,25,29,34,35,38] The majority used semi-structured interviews ($n=4$);[14,25,29,34] two studies used a structured interview approach ($n=2$).[35,38] Two studies used proxy methods (such as educational attainment) to report the impact of amblyopia on HRQoL ($n=2$).[27,33]

### *Study methodologies*

The identified studies can be summarised both in terms of their study methodologies (ie, the respondent) and the HRQoL implications identified. The identified studies may be summarised into the following broad categories.

*Questioning parents about the impact of amblyopia treatment on the child's HRQoL*

A total of 22 articles explored the impact of amblyopia treatment on the child's HRQoL from the parental perspective ($n=22$).[4–7,13–23,28,29,31,32,34,36,37]

Some of these articles specifically examined the issue of treatment compliance. Compliance might reflect the presence of QoL implications in amblyopia treatment. However, treatment compliance may also relate to parental non-concordance. Parental choice of treatment modalities and timing of treatment can affect concordance. Parental understanding of the condition was noted to impact treatment compliance.[14,16,17,19,20,28,29,37]

*Questioning children about the impact of amblyopia treatment on their HRQoL*

Four papers examined the impact of amblyopia and/or its treatment on a child's HRQoL from the child's perspective ($n=4$).[12,25,35,38] One used a combination of both parental and child reporting ($n=1$).[25] Three studies used qualitative interviews in their methodology ($n=3$).[25,35,38] It should be noted that two of the identified papers reported findings from the same cohort (part of the ALSPAC (Avon Longitudinal Study of Parents and Children)).

*The impact of amblyopia treatment on adults when they undertook amblyopia treatment as a child*

Nine papers were identified that reported the HRQoL implications of amblyopia and/or its treatment on adults who had undergone amblyopia treatment as a child ($n=9$).[8–11,24,26,27,30,33]

*The impact of amblyopia in later life: the use of proxy measures*

Two papers were identified, which explored the impact of amblyopia on adults using proxy measures of HRQoL ($n=2$).[27,33] The consequences of amblyopia on educational attainment, occupational status, risk of developing long-term vision loss, behaviour, and social functioning were examined. No association was found between amblyopia and educational achievement in one study,[27] whereas the other reported a borderline significant effect of amblyopia on the completion of a university degree qualification.[33] No statistically significant association between amblyopia and occupational classification was found.[27,33] The risk of developing long-term vision loss in the better-seeing eye was reported to be greater in amblyopes.[33] Amblyopia was not found to be associated with significant behavioural problems or bullying.[27]

### *HRQoL implications of amblyopia and/or its treatment*

The HRQoL implications of amblyopia and/or its treatment could be considered to fall into four broad categories: impact on family life, social interactions, undertaking daily activities, as well as feelings and behaviour. These can be examined as to whether they occur as a result of amblyopia itself and/or its treatment (see Table 5).

*Impact on family life*

Amblyopia treatment was reported to impact family life. This resulted in increased stress and anxiety for the parent/guardian facilitating the treatment, and negatively impacted carer–child relationships.[5,6,13,15,21,22,31,34] Other relationships within the family were also affected.[5,6,13,15,21,22]

**Table 5** Summary of quality-of-life implications of amblyopia and/or its treatment identified in the literature search

| *Quality-of-life component* | *Identified by* | *Due to amblyopia* | *Due to amblyopia treatment* |
|---|---|---|---|
| *Family life* | | | |
| Carer–child relationship | Holmes *et al*,[5,6] Cole *et al*,[13] Hrisos *et al*,[15] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Choong *et al*,[31] Dixon-Woods *et al*[34] | X | √ |
| Strained relationships within the family | Holmes *et al*,[5,6] Cole *et al*,[13] Hrisos *et al*,[15] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group[22] | X | √ |
| *Social interactions* | | | |
| Feelings of isolation/ differing from others | Holmes *et al*,[5,6] Sabri *et al*,[8] Cole *et al*,[13] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Koklanis *et al*[25] | X | √ |
| Bullying | Hrisos *et al*,[15] Koklanis *et al*,[25] Packwood *et al*,[26] Dixon-Woods *et al*,[34] Horwood *et al*,[35] Williams *et al*[38] | √ | √ |
| Interaction with peers | Holmes *et al*,[5,6] Sabri *et al*,[8] Felius *et al*,[24] van de Graaf *et al*,[9,10] Vianya-Estopa *et al*,[11] Cole *et al*,[13] Hrisos *et al*,[15] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Koklanis *et al*,[25] van de Graaf *et al*,[30] Williams *et al*[38] | √ | √ |
| *Activities* | | | |
| Impact on activities | Holmes *et al*,[5,6] Sabri *et al*,[8] van de Graaf *et al*,[9,10] Vianya-Estopa *et al*,[11] Cole *et al*,[13] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Felius *et al*,[24] Packwood *et al*,[26] Rahi *et al*,[27] Searle *et al*,[28] Searle,[29] van de Graaf *et al*,[30] Dixon-Woods *et al*[34] | √ | √ |
| Impact on education (immediate and long term) | Holmes *et al*,[5,6] van de Graaf *et al*,[9,10,30] Vianya-Estopa *et al*,[11] Cole *et al*,[13] Hrisos *et al*,[15] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group,[22] Felius *et al*,[24] Packwood *et al*,[26] Rahi *et al*,[27] Dixon-Woods *et al*[34] | √ | √ |
| *Feelings and behaviour* | | | |
| Self-esteem and self-image | Webber *et al*,[12] Newsham,[20] Koklanis *et al*,[25] Packwood *et al*,[26] Searle *et al*,[28] Searle,[29] Choong *et al*,[31] Parkes,[32] Dixon-Woods *et al*,[34] Newsham[37] | √ | √ |
| Depression, frustration, embarrassment | Sabri *et al*,[8] Hrisos *et al*,[15] Koklanis *et al*,[25] Rahi *et al*,[27] Searle *et al*,[28] Searle,[29]Leach[36] | X | X |
| Understanding of amblyopia and its implications | Sabri *et al*,[8] van de Graaf *et al*,[9,10,30] Vianya-Estopa *et al*,[11] Newsham,[20] Felius *et al*,[24] Packwood *et al*,[26] Searle *et al*,[28] Searle,[29] Chua and Mitchell,[33] Newsham[37] | √ | X |
| Sensation of patch/drops/ glasses | Holmes *et al*,[5,6] Cole *et al*,[13] Pediatric Eye Disease Investigator Group Writing Committee,[21] Pediatric Eye Disease Investigator Group[22] | X | √ |

Not mutually exclusive.

Siblings teased or bullied the child who undertook amblyopia treatment. The increased parental attention that treatment is associated with may also be an issue. Compliance with treatment is intrinsically linked to HRQoL. Often the negative aspects of amblyopia treatment are reported, yet treatment may not always be a negative experience. If compliance is good, praise and attention may be given to the child thereby improving the parent–child relationship.

*Social interactions*

Bullying[15,25,26,34,35,38] and interactions with peers[5,6,8–11,13,15,21,22,24,25,30,38] were reported to occur as a result of amblyopia and/or its treatment. Noticeable differences in the change in appearance (by nature of wearing glasses and/or patch) meant that treatment was obvious to others. The age at which emergence of negative opinions towards others has not been adequately explored. Feelings of isolation and noting differences between others were also documented.[5,6,8,13,21,22,25]

*Activities*

One of the frequently reported HRQoL implications of amblyopia was the impact the condition had on career choice and educational attainment.[5,6,9–11,13,15,21,22,24,26,27,30,34] This could be in the immediate (such as if the treatment

was undertaken during school hours) or in the long term (the implication of amblyopia in adulthood). The impact that amblyopia and treatment had on daily living activities was well documented.[5,6,8–11,13,21,22,24,26–30,34]

*Feelings and behaviour*

Feelings of low self-esteem and negative self-image were reported as a result of amblyopia and/or its treatment.[12,20,25,26,28,29,31,32,34,37] Other psychosocial implications included feelings of depression, frustration, and embarrassment.[8,15,25,27–29,36] Literature that explored the understanding of amblyopia and its implications was identified,[8–11,20,24,26,28–30,33,37] with attempts made to understand why compliance to treatment may be poor in some cases. Other studies explored feelings associated with the treatment of amblyopia, specifically the sensation of patch/drops/glasses.[5,6,13,21,22]

## Discussion

The concept of QoL can be considered in terms of four domains: symptoms of the disease and side effects of treatment, physical and functional status, emotional status, and social functioning.[46] It seems that the main HRQoL implications of amblyopia appear to be related to treatment of the condition rather than to the condition itself.

The search strategy used for the literature review incorporated standard terms used in HRQoL studies. However, it is possible that by extending the search strategy to include terms specifically relating to the four domains of QoL, additional studies may have been identified. The functional ability of subjects with amblyopia is an area of increasing interest, with impaired fine motor skills and slower reading speed being reported.[47,48]

Some of the identified studies included subjects who had a diagnosis of strabismus and a diagnosis of amblyopia; and some of the HRQoL instruments used included questions specifically relating to strabismus. Large-angled strabismus has been documented to negatively impact QoL.[49,50] It is possible that the studies identified in the literature review which reported lower HRQoL may actually be detecting HRQoL implications of strabismus rather than those of amblyopia. Only two of the studies identified in the literature review reported HRQoL in subjects with 'straight-eyed' amblyopia (anisometropic or small-angled strabismus measuring $<10$ prism dioptres).[15,26]

### *The adult's* vs *the child's perspective*

Some HRQoL instruments used in the identified studies were derived from consultations with ophthalmic professionals and/or parents of children with amblyopia. Therefore, the items included in the instrument design are deemed to be of importance from an adult's perspective. The included items may be of importance to adults, but not necessarily to the child. For example, a parent may feel that educational attainment and the ability to see well at school is of great importance; however, this view may not be shared by the child. In some of the studies identified, the reported findings are taken from a parental perspective. It is not possible to state that the impact of amblyopia treatment felt by the child is the same as that perceived by adults on how, or what the child should feel or experience. Some of the questions asked included how well the child could see while undertaking treatment. The parent/guardian cannot directly assess this; they can only make a judgement on how they perceive the child is able to see while on treatment. Their judgement could be influenced by how important they judge the activity to be (such as school work or interacting with friends).

Measuring HRQoL in a young age group is challenging; however, this has been achieved in conditions such as childhood cancers, asthma, and dermatitis.[50–54] Some of the difficulties involved include the burden of the task (ie, how difficult it is for the child to complete the questionnaire). This corresponds to the number of items (or questions) included, the scales used to answer the questions (yes/no or Likert-type scales), and the time taken to complete the task. Development of an instrument specifically designed to assess self-reported HRQoL in subjects with amblyopia is currently being undertaken by the author.[55]

Some studies reported HRQoL on adults who had undertaken amblyopia treatment as a child. It is possible that the recollections of adults in terms of amblyopia impacting childhood experiences could be tainted by subsequent events in adulthood. The responses are given from an adult's perspective, despite respondents being asked to recall childhood experiences and events. Recall bias is a recognised challenge in patient-reported outcomes and HRQoL research.[56]

### *Determining QoL by treatment compliance*

Treatment compliance in amblyopia therapy is influenced by both the child and the parent/guardian. Although the child may object to the wearing of glasses or a patch on a personal level, a parent's perspectives can influence the success of such treatment. This may incorporate their own experiences or impressions of patching/wearing glasses, or their understanding of the condition and the importance of treatment. Although these factors have been explored in the literature, to use compliance as a measure of HRQoL is questionable.

Parental understanding of the condition and belief in the prescribed treatment are key components for good treatment compliance. However, parents can be well-informed and positive, yet compliance may still be poor. Another argument against using treatment compliance as a measure of HRQoL is that a child may consent to wearing the patch, but their daily activities and social interactions may still be affected. In this instance, using treatment compliance would not truly represent any HRQoL implications of amblyopia and/or its treatment.

### *Use of proxy measures to determine QoL*

Some of the identified studies used proxy measures to determine the impact of amblyopia and/or treatment on HRQoL. These included educational attainment, occupation, long-term vision loss, and social functioning (as measured by self-reported depression of psychological distress in adult life). Such outcomes are influenced by many factors. The presence of amblyopia cannot be solely used to either explain episodes of psychological distress in adulthood or educational attainment. These studies highlight the importance of making the distinction between HRQoL and functional status or ability. Functional status and health status use measures that determine an individual's ability to perform or carry out an activity. HRQoL incorporates both the ability and an 'evaluation of the subjective experience of being able to complete a given activity'.[57] Some of the identified studies fail to address this issue and report functional status alone.

### *Changing trends in glasses and patches*

The manner by which people who wear glasses are perceived is changing. Glasses are becoming increasingly popular, and the social acceptance of this has much improved. With traditional 'NHS style' glasses being a thing of the past, it could be argued that the reported HRQoL findings from some of the earlier literature may not truly reflect on how things are in modern day practice. Similarly, the choice and style of patches have also changed, with a movement towards coloured patches, and patches that fit over glasses, to improve comfort and appearance. This has started to be explored in recent studies.[23]

It is clear that there are HRQoL implications associated with amblyopia; however, these are related to amblyopia treatment rather than to the condition itself. Despite differing study methodologies, four key components of HRQoL were identified: those of physical ability (undertaking daily tasks), emotional status (feelings and behaviour), social interactions, and impact on family life. Very few of the studies identified assessed HRQoL from the child's perspective. Current recommendations from the Department of Health encourage the participation of children respondents in the assessment of their own health and treatment,[58] and future studies in this area need to address this issue.

The HRQoL measures used in the identified studies failed to report the psychometric properties of the measures themselves (ie, reliability and validity), with the exception of the ATI, A&SQ, and Psychological Impact Questionnaire. Although their reported findings may be of clinical importance, their use in economic evaluations and subsequent policy-making decisions are limited. Further research is required to assess the immediate and long-term utility effects of amblyopia and/or its treatment, using more robust methods of HRQoL assessment.

## Conflict of interest

The authors declare no conflict of interest.

## Acknowledgements

This work is produced by Jill Carlton under the terms of Researcher Development Awards research training fellowship issued by the NIHR. The views expressed in this publication are those of the author and not necessarily those of the NHS, The National Institute for Health Research, or the Department of Health. The author is currently undertaking a PhD to produce a paediatric disease-specific measure of HRQoL in amblyopia.

## References

1 Carlton J, Karnon J, Czoski-Murray C, Smith KJ, Marr J. The clinical effectiveness and cost-effectiveness of screening programmes for amblyopia and strabismus in children up to the age of 4–5 years: a systematic review and economic evaluation. *Health Tech Assess* 2008; **12**(25): iii.
2 Schmucker C, Kleijnen J, Grosselfinger R, Riemsma R, Antes G, Lange S *et al.* Effectiveness of early in comparison to late(r) treatment in children with amblyopia or its risk factors: a systematic review. *Ophthalmic Epidemiol* 2010; **17**(1): 7–17.
3 Schmucker C, Grosselfinger R, Riemsma R, Antes G, Lange S, Lagreze W *et al.* Effectiveness of screening preschool children for amblyopia: a systematic review. *BMC Ophthalmol* 2009; **9**: 3.
4 Birch EE, Cheng CS, Felius J. Validity and reliability of the Children's Visual Function Questionnaire (CVFQ). *J AAPOS* 2007; **11**(5): 473–479.
5 Holmes JM, Beck RW, Kraker RT, Cole SR, Repka MX, Birch EE *et al.* Impact of patching and atropine treatment on the child and family in the amblyopia treatment study. *Arch Ophthalmol* 2003; **121**(11): 1625–1632.
6 Holmes JM, Strauber S, Quinn GE, Cole SR, Felius J, Kulp M. Further validation of the Amblyopia Treatment Index parental questionnaire. *J AAPOS* 2008; **12**: 581–584.

7 Loudon SE, Felius J, Simonsz B, Joosse MV, Fronius M, Simonsz HJ. Quality of life of children treated with occlusion therapy for amblyopia. *Invest Ophthalmol Vis Sci* 2005; **46**: 3596.
8 Sabri K, Knapp CM, Thompson JR, Gottlob I. The VF-14 and psychological impact of amblyopia and strabismus. *Invest Ophthalmol Vis Sci* 2006; **47**(10): 4386–4392.
9 van de Graaf ES, Felius J, van Kempen-du SH, Looman CW, Passchier J, Kelderman H *et al*. Construct validation of the Amblyopia and Strabismus Questionnaire (A&SQ) by factor analysis. *Graefes Arch Clin Exp Ophthalmol* 2009; **247**(9): 1263–1268.
10 van de Graaf ES, van der Sterre GW, van Kempen-du SH, Simonsz B, Looman CW, Simonsz HJ *et al*. Amblyopia and Strabismus Questionnaire (A&SQ): clinical validation in a historic cohort. *Graefes Arch Clin Exp Ophthalmol* 2007; **245**(11): 1589–1595.
11 Vianya-Estopa M, Elliott DB, Barrett BT, Vianya-Estopa M, Elliott DB, Barrett BT. An evaluation of the Amblyopia and Strabismus Questionnaire using Rasch analysis. *Invest Ophthalmol Vis Sci* 2010; **51**(5): 2496–2503.
12 Webber AL, Wood JM, Gole GA, Brown B. Effect of amblyopia on self-esteem in children. *Optom Vis Sci* 2008; **85**(11): 1074–1081.
13 Cole SR, Beck RW, Moke PS, Celano MP, Drews CD, Repka MX *et al*. The Amblyopia Treatment Index. *J AAPOS* 2001; **5**: 250–254.
14 Göransson A, Dahlgren LO, Lennerstrand G. Changes in conceptions of meaning, effects and treatment of amblyopia. A phenomenographic analysis of interview data from parents of amblyopic children. *Patient Educ Couns* 1998; **34**: 213–225.
15 Hrisos S, Clarke MP, Wright CM. The emotional impact of amblyopia treatment in preschool children: randomized controlled trial. *Ophthalmology* 2004; **111**: 1550–1556.
16 Loudon SE, Chaker L, de VS, Fronius M, Looman CWN, Simonsz B *et al*. Electronic Recording of Patching for Amblyopia Group (ERPAG): reasons and a remedy for non-compliance? *Invest Ophthalmol Vis Sci* 2007; **48**: E-abstract 1107.
17 Loudon SE, Verhoef BL, Joosse MV, Fronius M, Awan M, Newsham D *et al*. Electronic Recording of Patching for Amblyopia Study (ERPAS): preliminary results. *Invest Ophthalmol Vis Sci* 2003; **44**: E-abstract 4246.
18 Loudon SE, Simonsz B, Joosse MV, Fronius M, Awan M, Newsham D *et al*. Electronic Recording of Patching for Amblyopia Study: predictors for non-compliance. *Invest Ophthalmol Vis Sci* 2004; **45**: E-abstract 4991.
19 Loudon SE, Passchier J, Chaker L, de VS, Fronius M, Harrad RA *et al*. Psychological causes of non-compliance with electronically monitored occlusion therapy for amblyopia. *Br J Ophthalmol* 2009; **93**: 1499–1503.
20 Newsham D. A randomised controlled trial of written information: the effect on parental non-concordance with occlusion therapy. *Br J Ophthalmol* 2002; **86**: 787–791.
21 Pediatric Eye Disease Investigator Group Writing Committee, Rutstein RP, Quinn GE, Lazar EL, Beck RW, Bonsall DJ *et al*. A randomized trial comparing Bangerter filters and patching for the treatment of moderate amblyopia in children. *Ophthalmology* 2010; **117**(5): 998–1004.
22 Pediatric Eye Disease Investigator Group. A randomized trial of atropine vs patching for treatment of moderate amblyopia in children. *Arch Ophthalmol* 2002; **120**(3): 268–278.
23 Roefs AM, Tjiam AM, Vukovic E, Loudon SE, Felius J, Simonsz HJ. Qol and compliance during occlusion therapy for amblyopia in a prospective, double blind study comparing four brands of occlusion patches. *Invest Ophthalmol Vis Sci* 2008; **49**: E-abstract 2583.
24 Felius J, Beauchamp GR, Stager Sr DR, van de Graaf ES, Simonsz HJ, Felius J *et al*. The Amblyopia and Strabismus Questionnaire: English translation, validation, and subscales. *Am J Ophthalmol* 2007; **143**(2): 305–310.
25 Koklanis K, Abel LA, Aroni R, Koklanis K, Abel LA, Aroni R. Psychosocial impact of amblyopia and its treatment: a multidisciplinary study. *Clin Exp Ophthalmol* 2006; **34**(8): 743–750.
26 Packwood EA, Cruz OA, Rychwalski PJ, Keech RV, Packwood EA, Cruz OA *et al*. The psychosocial effects of amblyopia study. *J AAPOS* 1999; **3**(1): 15–17.
27 Rahi JS, Cumberland PM, Peckham CS. Does amblyopia affect educational, health, and social outcomes? Findings from 1958 British birth cohort. *BMJ* 2006; **332**(7545): 820–825.
28 Searle A, Norman P, Harrad R, Vedhara K, Searle A, Norman P *et al*. Psychosocial and clinical determinants of compliance with occlusion therapy for amblyopic children. *Eye* 2002; **16**(2): 150–155.
29 Searle A. Compliance with eye patching in children and its psychosocial effects: a qualitative application of protection motivation theory. *Psychol Health Med* 2000; **5**(1): 43–54.
30 van de Graaf ES, van der Sterre GW, Polling JR, van KH, Simonsz B, Simonsz HJ *et al*. Amblyopia & Strabismus Questionnaire: design and initial validation. *Strabismus* 2004; **12**(3): 181–193.
31 Choong YF, Lukman H, Martin S, Laws DE, Choong YF, Lukman H *et al*. Childhood amblyopia treatment: psychosocial implications for patients and primary carers. *Eye* 2004; **18**(4): 369–375.
32 Parkes LC. An investigation of the impact of occlusion therapy on children with amblyopia, its effect of their families, and compliance with treatment. *Br Orthopt J* 2001; **58**: 30–37.
33 Chua B, Mitchell P. Consequences of amblyopia on education, occupation, and long term vision loss. *Br J Ophthalmol* 2004; **88**: 1119–1121.
34 Dixon-Woods M, Awan M, Gottlob I. Why is compliance with occlusion therapy for amblyopia so hard? A qualitative study. *Arch Dis Child* 2006; **91**: 491–494.
35 Horwood J, Waylen A, Herrick D, Williams C, Wolke D. Common visual defects and peer victimisation in children. *Invest Ophthalmol Vis Sci* 2005; **46**(4): 1177–1181.
36 Leach C. Compliance with occlusion therapy for strabismic and anisometropic amblyopia: a pilot study. *Binocular Vis Q* 1995; **10**(4): 257–266.
37 Newsham D. Parental non-concordance with occlusion therapy. *Br J Ophthalmol* 2000; **84**: 957–962.
38 Williams C, Horwood J, Northstone K, Herrick D, Waylen A, Wolke D. The timing of patching treatment and a child's wellbeing. *Br J Ophthalmol* 2006; **90**(6): 670–671.
39 Brazier J, Ratcliffe J, Salomon JA, Tsuchiya A. Methods for obtaining health state values: generic preference-based measures of health and the alternatives. In: Brazier J, Ratcliffe J, Salomon JA, Tsuchiya A (eds). *Measuring and Valuing Health Benefits for Economic Evaluation*, 1st edn. Oxford University Press: Oxford, 2007, pp 175–256.
40 Margolis MK, Coyne K, Kennedy-Martin T, Baker T, Schein O, Revicki DA. Vision-specific instruments for the assessment of health-related quality of life and visual functioning. *Pharmacoeconomics* 2002; **20**(12): 791–812.

41 Felius J, Stager Sr DR, Berry PM, Fawcett SL, Stager Jr DR, Salomao SR *et al*. Development of an instrument to assess vision-related quality of life in young children. *Am J Ophthalmol* 2004; **138**(3): 362–372.
42 Marsh HW, Holmes IW. Multidimensional self-concepts: construct validation of responses by children. *Am Ed Res J* 1990; **27**: 89–117.
43 Dias L, Manny RE, Hyman L, Fern K. The relationship between self-esteem of myopic children and ocular and demographic characteristics. *Optom Vis Sci* 2002; **79**: 688–696.
44 Dias L, Hyman L, Manny RE, Fern K. Evaluating the self-esteem of myopic children over a three-year period: the COMET experience. *Optom Vis Sci* 2005; **82**: 338–347.
45 Steinberg EP, Tielsch JM, Schein OD, Javitt JC, Sharkey P, Cassard SD *et al*. The VF-14. An index of functional impairment in patients with cataract. *Arch Ophthalmol* 1994; **112**(5): 630–638.
46 Schumacher M, Olschewski M, Schulgen G. Assessment of quality of life in clinical trials. *Stat Med* 1991; **10**(12): 1915–1930.
47 Webber AL, Wood JM, Gole GA, Brown B. The effect of amblyopia on fine motor skills in children. *Invest Ophthalmol Vis Sci* 2008; **49**(2): 594–603.
48 Stifter E, Burggasser G, Hirmann E, Thaler A, Radner W. Monocular and binocular reading performance in children with microstrabismic amblyopia. *Br J Ophthalmol* 2005; **89**(10): 1324–1329.
49 Satterfield D, Keltner JL, Morrison TL. Psychosocial aspects of strabismus study. *Arch Ophthalmol* 1993; **111**(8): 1100–1105.
50 Jackson S, Harrad RA, Morris M, Rumsey N. The psychosocial benefits of corrective surgery for adults with strabismus. *Br J Ophthalmol* 2006; **90**(7): 883–888.
51 Varni JW, Katz ER, Quiggens DJL, Friedman-Bender A. The Pediatric Cancer Quality of Life Inventory-32 (PCQL-32): I. Reliability and validity. *Cancer* 1998; **82**(6): 1184–1196.
52 Varni JW, Katz ER, Seid M, Quiggens DJ, Friedman-Bender A, Castro CM. The Pediatric Cancer Quality of Life Inventory (PCQL). I. Instrument development, descriptive statistics, and cross-informant variance. *J Behav Med* 1998; **21**(2): 179–204.
53 Christie MJ, French D, Sowden A, West A. Development of child-centred disease-specific questionnaires for living with asthma. *Psychosom Med* 1993; **55**(6): 541–548.
54 Lewis-Jones MS, Finlay AY. The Children's Dermatology Life Quality Index (CDLQI): initial validation and practical use. *Br J Dermatol* 1995; **132**(6): 942–949.
55 Carlton J http://public.ukcrn.org.uk/search/StudyDetail.aspx?StudyID = 6393.
56 Stull DE, Leidy NK, Parasuraman B, Chassany O. Optimal recall periods for patient-reported outcomes: challenges and potential solutions. *Curr Med Res Opin* 2009; **25**(4): 929–942.
57 Clarke SA, Eiser C. The measurement of health-related quality of life (QOL) in paediatric clinical trials: a systematic review. *Health Qual Life Outcomes* 2004; **2**: 66.
58 Department of Health. National Service Framework for Children Young People and Maternity Services: Core Standards. 2007. www.dh.gov.uk/en/Publicationsand statistics/Publications/PublicationsPolicyAndGuidance/Browsable/DH_4094329.

## Appendix 1

The following electronic bibliographic databases were searched.

| | |
|---|---|
| 1. | Scopus |
| 2. | NHS Economic Evaluation Database (NHS EED) |
| 3. | Cochrane Methodology Register |
| 4. | Health Technology Assessment Database |
| 5. | Cochrane Register of Controlled Trials |
| 6. | Database of Abstracts of Reviews of Effects (DARE) |
| 7. | Cochrane Database of Systematic Reviews (CDSR) |
| 8. | Cumulative Index to Nursing and Allied Health Literature (CINAHL) |
| 9. | Science Citation Index (SCI) and Social Sciences Citation Index (SSCI) |
| 10. | Ovid includes Medline; PsychINFO; AMED (Allied and Complementary Medicine); British Nursing Index and Archive; and Econlit |

A specific search strategy was used for each database

| *Database* | *Search strategy* | *Number of articles identified* |
|---|---|---|
| Scopus | 'Amblyopia terms' and 'selected quality-of-life terms' | 0 |
| NHS EED | 'Amblyopia terms' | 7 |
| Cochrane Methodology Register | 'Amblyopia terms' | 3 |
| Health Technology Assessment Database | 'Amblyopia terms' | 5 |
| Cochrane Register of Controlled Trials | 'Amblyopia terms' | 273 |
| Database of Abstracts of Reviews of Effects | 'Amblyopia terms' | 5 |
| Cochrane Database of Systematic Reviews | 'Amblyopia terms' | 15 |
| CINAHL | 'Amblyopia terms' and 'selected quality-of-life terms' | 9 |
| SCI and SSCI | 'Amblyopia terms' and 'selected quality-of-life terms' | 55 |
| Ovid | 'Amblyopia terms' and 'child terms' and 'quality-of-life terms' | 236 |
| | 'Amblyopia treatment terms' and 'quality-of-life terms' and 'amblyopia terms' | 278 |
| | 'Amblyopia terms' and 'quality-of-life terms' | 309 |

Amblyopia terms

| | |
|---|---|
| 1. | Amblyop$ |
| 2. | Lazy eye |
| 3. | 1 or 2 |

$ denotes truncation.

Child terms

| | |
|---|---|
| 1. | child$ or infant$ or kindergarten$ or juvenile$ or preschool$ or pre school$ or pre-school$ or nurser$ or adolesc$ or school$ or infancy$ |

$ denotes truncation.

Amblyopia treatment terms

| | |
|---|---|
| 1. | occlu$ |
| 2. | patch$ |
| 3. | atropine$ |
| 4. | therap$ or treatment$ or manag$ |
| 5. | cosme$ |
| 6. | psychosocial$ |

$ denotes truncation.

Quality-of-life terms

| | |
|---|---|
| 1. | quality of life |
| 2. | life quality |
| 3. | hql |
| 4. | sf 36 or sf36 or sf thirtysix or sf thirty six or short form 36 or short form thirty six or short form thirtysix or shortform 36 |
| 5. | qol |
| 6. | euroqol or euro qol or eq5d or eq 5d |
| 7. | qaly$ |
| 8. | quality adjusted life year$ |
| 9. | hye$ |
| 10. | health$ year$ equivalent$ |
| 11. | health utility$ |
| 12. | hui$ |
| 13. | quality of wellbeing$ |
| 14. | quality of well being |
| 15. | qwb |
| 16. | disability adjusted life$ |
| 17. | daly$ |
| 18. | health status indicators |
| 19. | sf12 or sf 12 or short form 12 or shortform 12 or sf twelve or sftwelve or shortform twelve or short form twelve |
| 20. | disutili$ |
| 21. | willingness to pay |
| 22. | standard gamble$ |
| 23. | SG$ |
| 24. | tto |
| 25. | time trade off |

$ denotes truncation.

Selected quality-of-life terms

| | |
|---|---|
| 1. | quality of life |
| 2. | life quality |
| 3. | hql |
| 4. | qol |
| 5. | quality adjusted life |
| 6. | quality of wellbeing |
| 7. | quality of well being |
| 8. | health related quality of life |
| 9. | hqol |
| 10. | hrqol |
| 11. | hr qol |

$ denotes truncation.

## Appendix 2

Data extraction form.

Details of publication

| | |
|---|---|
| 1. | Author(s) |
| 2. | Title |
| 3. | Source and reference |
| 4. | Country of origin |

Instrument details

| | | |
|---|---|---|
| 1. | Name | |
| 2. | Newly developed: | Yes/no |
| 3. | Questionnaire development | Clinician based/ literature based/ patient based |
| 4. | Domains | |
| 5. | Number of items | |
| 6. | Mode of administration | |
| 7. | Test of validity | Yes/no |
| 8. | (a) Discriminant validity | |
| 9. | (b) Construct validity | |
| 10. | (c) Concurrent validity | |
| 11. | (d) Internal consistency | |
| 12. | (e) Responsiveness | |
| 13. | (a) Test–retest reliability | |

Qualitative study

| | | |
|---|---|---|
| 1. | Type of interview | Structured/semi-structured/open |
| 2. | Conducted by | Clinician/researcher/unknown |
| 3. | Parent present | Yes/no |