BMJ Open

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Journal:	*BMJ Open*
Manuscript ID	bmjopen-2022-062754
Article Type:	Original research
Date Submitted by the Author:	17-Mar-2022
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Keywords:	Glaucoma < OPHTHALMOLOGY, QUALITATIVE RESEARCH, Paediatric ophthalmology < OPHTHALMOLOGY, OPHTHALMOLOGY

Quality of life in children with glaucoma: An interview study

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Word count: 4296

Abstract

Objective: Childhood glaucoma is a chronic vision-threatening condition which poses a substantial threat to an individual's psychosocial well-being. However, there is a paucity of literature investigating the quality-of-life (QoL) in children with glaucoma. The aim of this study was to investigate and report on the QoL issues encountered by children with glaucoma.

Design: This is a qualitative interview study underpinned by interpretive phenomenological analysis. Data were collected through semi-structured interviews. NVivo-12 software (QSR International Pty Ltd, Melbourne, Victoria, Australia) was used to inductively analyse and code data to identify QoL themes.

Participants: Eighteen children with glaucoma, aged 8 to 17 years, were recruited from the Australian and New Zealand Registry of Advanced Glaucoma.

Setting: Interviews were conducted via telephone or videoconferencing.

Results: Median child age was 12.1 years (interquartile range: 9.7–14.5 years) and 33% were female. Seven quality of life themes were identified. Coping strategies, inconveniences and emotional well-being were the most prominent themes. Adaptive coping strategies included resilience throughout clinical examinations and establishing a positive relationship with the ophthalmologist. These minimised inconveniences related to clinic waiting times and pupillary dilatation. External to the clinical setting, children often dissociated from their glaucoma but struggled with glare symptoms and feeling misunderstood by fellow peers. Older children aged 13 to 17 years commonly disengaged from their glaucoma care and expressed an unwillingness to attend ophthalmic appointments. Older children further experienced issues with career options, obtaining a driver's license and family planning under the theme of autonomy.

Conclusions: The psychosocial impact of childhood glaucoma extends beyond the clinical environment and was minimised using coping strategies. Older children may require additional social and ophthalmic support as they transition into adulthood.

Key Words: childhood glaucoma, glaucoma, quality of life, qualitative, interviews

Article Summary

Strengths and limitations of this study

- This study used an appropriate qualitative methodology to develop a novel and in-depth insight into the lived experience of childhood glaucoma from the perspectives of children.
- This study included individuals with varied disease characteristics and thus detailed the lived experience of the disease as a whole.
- Participants were recruited from a national registry and thus may be more willing to participate and may be experiencing a better quality-of-life than nonparticipants.
- Participants were mostly of European ancestry and resided in Australia which may limit the generalisability of the results.

INTRODUCTION

Childhood glaucoma describes a heterogeneous group of rare chronic vision-threatening disorders with onset occurring at any age from birth to less than 18 years of age.[1] It is typically characterised by elevated intraocular pressure (IOP) and irreversible optic neuropathy. Primary childhood glaucoma is caused by isolated abnormal development of the anterior chamber angle and includes primary congenital glaucoma (PCG) and juvenile open-angle glaucoma (JOAG).[1] Secondary childhood glaucoma includes glaucomatous disease that is associated with either other ocular anomalies (e.g., aniridia, Axenfeld-Rieger syndrome), an underlying systemic condition (e.g., Sturge-Weber syndrome) or an acquired ocular condition (e.g., uveitis, trauma).[1] Upon diagnosis, surgical intervention is typical and lifelong monitoring with or without additional surgical interventions and/or adjuvant topical therapies to manage IOP and prevent vision loss is generally required.[2] Additional symptoms can include glare and high myopia, whilst cosmetic concerns can be associated with buphthalmos, occlusion therapy for amblyopia, and spectacle wear.[3]

Children with glaucoma may experience several visual and non-visual challenges as they adapt to living with the condition. Vision-related challenges are captured using vision-related quality of life (VR-QoL) and functional visual ability (FVA) instruments. Previous research has demonstrated that children with glaucoma who have lower best-corrected visual acuity (BCVA) experienced lower VR-QoL,[4-7] and reduced FVA.[5-7] However, none are yet to correlate VR-QoL and FVA with a child’s contrast or glare sensitivity, which are otherwise associated with reduced FVA in adults with glaucoma.[8] Furthermore, these measurements do not provide an understanding of how a child perceives their overall well-being.[9,10] This is instead measured by health-related quality of life (HR-QoL) instruments, which consider the impact of visual and nonvisual-related challenges.[7,10]

There is a paucity of literature investigating HR-QoL in children with glaucoma. Two studies have reported that younger age and lower BCVA are associated with lower HR-QoL.[7,10] However, there has been limited investigation as to why this trend was observed.[7,10] Furthermore, there is no agreement upon which patient-reported outcome measure (PROM) is best utilised to measure HR-QoL in children with glaucoma,[7,10] because a childhood glaucoma-specific PROM does not exist. A qualitative inquiry is therefore required to explore issues that are specifically associated with HR-QoL in children with glaucoma. Findings from this study will inform the development of a childhood glaucoma-specific PROM for future related research and clinical implementation.

METHODS

Participants

Interpretive phenomenological analysis was used to provide an in-depth description of children's lived experience and to identify QoL issues.[11] Children were recruited from a large Australasian disease registry, the Australian and New Zealand Registry of Advanced Glaucoma (ANZRAG),[12] using a non-probability convenience sampling technique. Children were eligible to be interviewed if they currently resided in Australia, were English speaking, had a diagnosis of any subtype of glaucoma as per the Childhood Glaucoma Research Network criteria,[1] and were aged between 8 and <18 years. Children aged ≥8 years are more likely to reliably and independently understand questions relating to QoL than children aged <8 years.[13] Children were excluded if they had coexisting ocular disease unrelated to childhood glaucoma or had a hearing or cognitive impairment or other disability impacting on QoL (e.g., intellectual disability) as informed by their referring specialist or parent/guardian (henceforth abbreviated to parent).

Eligible children, and their parent/s, were posted an invitation to be interviewed and asked to return their interest. If both parties expressed interest, an information pack and consent form were sent. An interview was arranged once written informed consent from one parent and assent from the child were provided. If no response was received within two weeks, parents received a follow-up phone call. Children were deemed non-contactable after at least two unsuccessful attempts.

Children's clinical details were obtained from their most recent medical record and included: glaucoma subtype, age at diagnosis, laterality, BCVA, IOP, number of surgical interventions, and number of topical antiglaucoma medications currently being used. The International Classification of Diseases for Mortality and Morbidity Statistics (11th Revision),[14] was used to categorise BCVA per eye. Because visual field information was not available for every child, BCVA was used as a measure of disease severity. For analysis, children's ages were grouped into 8 to 12 years and 13 to 17 years, as per the Pediatric Quality of Life Inventory Version 4.0 (PedsQL).[15] Glaucoma onset at ≥4 years was considered juvenile.[1] Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee and the study adhered to the tenets of the Declaration of Helsinki.

Interviews

A semi-structured interview guide was developed from a literature review of VR-QoL and HR-QoL PROMs (see Supplementary file 1, which details the semi-structured interview guide used).[15-19] Interviews were conducted in the English language by one of two authors with qualitative research experience (LSWK and BR). LSWK is a clinical and research orthoptist and BR is a health counsellor. No participants were under the clinical care of either interviewer. The child and parent/s were informed that the interviewers were completing a higher research degree. One-on-one semi-structured interviews occurred via telephone or Cisco WebEx videoconferencing (Milpitas, California, USA), subject to the child's preference. Children aged <16 years required a parent chaperone and parents were not to answer questions on their child's behalf. Interviews were audio-recorded and transcribed verbatim. Interviews continued until thematic saturation was achieved (i.e., the point where no new information was gained from subsequent interviews).[20]

Data Analysis

Transcripts were systematically and inductively coded using QSR NVivo 12 (QSR International Pty Ltd, Melbourne, Victoria, Australia) by one author (LSWK). To ensure research credibility, stakeholder coding checks were frequently and separately performed by three authors (BR, MPS and ES).[21] Major QoL themes, and their sub-themes were determined by grouping codes with similar or repetitive patterns of meaning,[22] and were abbreviated to be consistent with previous ophthalmic QoL research.[23,24] Statistical calculations were performed using SPSS version 27.0 for Windows (IBM/SPSS Inc, Chicago, IL, USA). The datasets generated for the current study are not publicly available. This is to protect the confidentiality of research participants.

Patient and Public Involvement

Authors (LSWK, BR and ES) had presented the research aims at a national childhood glaucoma support group meeting prior to conducting the research. Engagement with attendees assisted in the development of the interview guide, and it was agreed that research findings would be disseminated back to the childhood glaucoma community.

RESULTS

Fifty-four eligible children from the ANZRAG were invited to participate and 18 (33%) were interviewed (see Figure S1 in Supplementary file 2, which depicts the recruitment of participants). The proportion of participants and non-participants with bilateral disease was significantly different (11/18, 61% vs 34/36, 94%, respectively, p=0.004) whilst all other demographic and clinical variables were similar (see Table S1 in Supplementary file 3). Reasons for declining to participate were not recorded due to the sensitive nature of the study.

Interviews were conducted between April 2020 and July 2021. The average interview length was 30 ± 14 minutes and the median age of children interviewed was 12.1 years (interquartile range: 9.7–14.5 years). Demographic and clinical characteristics of the children interviewed are detailed in Table 1.

Table 1. Demographic and clinical characteristics of children interviewed

Variable	**n (%)**†
Age at glaucoma diagnosis, years (median [range])	0.5 [0–15]
Time since diagnosis, years (median [IQR])	9.8 [7.3–13.6]
Age at interview	
8–12 years	10 (56)
13–17 years	8 (44)
Gender, female	6 (33)
Laterality of glaucoma, bilateral	11 (61)
Self-reported ancestry, European	16 (89)
Subtype of childhood glaucoma	
Primary congenital glaucoma	12 (67)
Glaucoma associated with non-acquired ocular anomalies	
Aniridia	1 (6)
Axenfeld-Rieger syndrome	1 (6)
Glaucoma associated with non-acquired systemic condition	
Sturge-Weber syndrome	1 (6)
Glaucoma associated with an acquired condition	
Uveitis	2 (11)
Glaucoma following cataract surgery	1 (6)

Number of topical antiglaucoma medications currently using	
0	13 (72)
≥1	5 (28)
Intraocular pressure at last ophthalmic appointment, mmHg (median [range])	18 [14–25]
Time since last ophthalmic appointment, months (median [IQR])	3.8 [2.9–7.4]
Number of surgical interventions per child (median [IQR])	2 [2–4]
Time since last ophthalmic surgical intervention, years (median [IQR])	6.7 [1.6–13.6]
Disease complications	
Corneal disease	1 (6)
Cataract	4 (22)
Molecular diagnosis identified	9 (50)
Autosomal recessive inheritance	2 (11)
Autosomal dominant inheritance	7 (39)

Vision category	**BCVA**	**Better Eye BCVA (n, %)**	**Worse Eye BCVA (n, %)**
No vision impairment	20/20–≥20/40	15 (83)	8 (44)
Mild vision impairment	<20/40–≥20/60	1 (6)	4 (22)
Moderate vision impairment	<20/60–≥20/200	1 (6)	2 (11)
Severe vision impairment or blindness	<20/200–≥20/400	0 (0)	1 (6)
Blindness	<20/400–CF	1 (6)	2 (11)
	HM or LP	0 (0)	1 (6)
	NLP	0 (0)	0 (0)

†: n (%) presented unless otherwise specified

BCVA: Best corrected visual acuity; CF: count fingers; HM: hand movements; IQR: interquartile range; LP: light perception; NLP: no light perception

Seven QoL themes emerged from the data. The total proportion of children experiencing issues per QoL theme and coded segments per theme are shown in Figure 1. Additional sub-themes not presented within the results are provided in a mind map (see Figure S2 in Supplementary file 2).

Theme 1: Coping

All children used coping strategies to manage the impacts of their glaucoma (Figure 1). All children (18/18, 100%) discussed being resilient, which is an adaptive emotion-focused coping strategy (i.e., a strategy which involves regulation or minimisation of negative emotions).[25]

> "*I've grown up with it. I've gotten used to it. I just don't pay much attention to it now.*" (Child aged 13–17 years)

Adaptive problem-focused strategies (i.e., strategies which actively confront the problem),[25] included developing a positive relationship with their ophthalmologist (12/18, 67%), seeking and accepting support from family, friends, or schoolteachers (11/18, 61%) and accepting parents' use of positive reinforcement for appointment attendance (9/18, 50%).

> "*I'm a lot more comfortable with [my ophthalmologist] because he's been doing it with me since basically the first time, I went there... we're friends.*" (Child aged 8–12 years)

Several children (10/18, 56%) discussed adapting to activity limitations secondary to visual abilities or symptoms, such as photophobia. This was observed in children with bilateral (3/3, 100%) or unilateral BCVA <20/60 (3/7, 43%) and children with no BCVA impairment (4/8, 50%). Adapting to visual limitations was improved with the use of electronic devices in the classroom (e.g., laptop computer) whereby text size and contrast could be manipulated. Adapting to photophobia was usually resolved with sunglasses wear. Consequently, 5/18 (28%) children explicitly stated that their glaucoma did not limit them.

> "*A lot of [schooling] stuff is on the computers and not written on the board anymore. So yeah, like I don't really think that I have troubles.*" (Child aged 13–17 years)

Dissociating from one’s glaucoma outside of the clinical setting and ignoring its presence was used by 8/18 (44%) children, most of whom did not have bilaterally impaired BCVA (7/8, 88%). This was considered an adaptive strategy in 4/8 (50%), 3/4 (75%) of whom were aged 8 to 12 years, as these children considered themselves unaffected by their glaucoma. Conversely, it was considered maladaptive in 4/8 (50%) children, irrespective of age or gender, because these children avoided asking for vision-related assistance from teachers or were disinterested in possible disease consequences.

> “*I'm just not interested in my eyes much.*” (Child aged 8–12 years)

Actively leaving medical responsibilities and decision-making to their parent/s was discussed by more children aged 13 to 17 years compared to their younger counterparts (5/8, 63% vs 2/10, 20%, respectively). Gender, antiglaucoma medication use, and BCVA did not appear influential.

> “*I’d let Mom ask the questions… I’m more of a listener. Like a bystander… I’ll get all the information I want out of Mom.*” (Child aged 13–17 years)

Furthermore, 3/4 (75%) children aged ≥16 years discussed strong feelings of wanting to avoid attending their ophthalmic appointments.

> “*I was just yelling and screaming… I really did not want to go [to my appointment].*” (Child aged 13–17 years)

Theme 2: Inconveniences

All children discussed several inconveniences related to their ophthalmic appointments or glaucoma treatment. Clinic waiting time caused boredom for 6/18 (33%) children and 5/18 (28%) discussed negative outcomes related to school absenteeism. These were exacerbated where travelling long distances for ophthalmic review was required. Conversely, 7/18 (39%) reasoned that school absenteeism was a positive experience.

> "*It took us like three hours to get there and to go back... I often had to skip school to go there, and it was often always the fun days*." (Child aged 8–12 years)

Most children (11/18, 61%) discussed the inconvenience of having blurred vision for many hours following pupillary dilatation, whilst 4/18 (22%) considered a visual field test burdensome.

> "*I hate getting drops… everything I see is blurry for six or seven hours… They're still the worst thing that could possibly happen.*" (Child aged 13–17 years)

Spectacle wear was considered inconvenient and uncomfortable by 6/18 (33%) children, particularly during sporting activities. Among children who currently use topical antiglaucoma medication, 2/5 (40%) considered them bothersome.

> "*I don't really like wearing [glasses]... because my nose gets sweaty.*" (Child aged 8–12 years)

Theme 3: Emotional well-being

Negative emotional experiences were discussed by 15/18 (83%) children. Feeling frustrated (13/18, 72%) or anxious (10/18, 56%) were often experienced in the contexts of requiring pupil dilatation or performing certain clinical tests (e.g., visual field test, IOP test).

> "*The sight field test… has like things that blink and it's just like heaps of them, and it's like in a way sort of overwhelming.*" (Child aged 8–12 years)

Several children (7/18, 39%) discussed feeling misunderstood at times by their friends, peers and/or schoolteachers. At times, this led to concealment of their condition.

> "*I like keeping [my glaucoma] a bit of a secret… Because when I try to explain - no one understands and I have to keep explaining, explaining and explaining.*" (Child aged 8–12 years)

Feeling self-conscious of their appearance was expressed by 6/18 (33%) children. Reasons included their eye appearance, wearing spectacles or wearing an eye patch for amblyopia therapy. These were not dependent on BCVA, gender or age with the exception that one child, with bilateral BCVA <20/60, expressed feeling self-conscious whilst using their white cane for mobility.

> "*I hate [all the photos] when I'm younger because of the big, shaded glasses and stuff... I'm not a very photogenic person.*" (Child aged 13–17 years)

Theme 4: Symptoms

The most common symptom experienced by children was blurred vision (13/18, 72%). Of these, 4/13 (31%) had unilateral disease, and 7/13 (54%) had no BCVA impairment. It was usually described in the context of reading the classroom board, reading small texts, and playing sports that involve a small ball (e.g., tennis).

> "*If it's small writing and I'm at the back of the class I can't always get it but if it's like medium like to big writing I can see.*" (Child aged 13–17 years)

Glare (8/18, 44%), sore eyes (4/18, 22%) and reduced peripheral vision (2/18, 11%) were other symptoms experienced, irrespective of any clinical or demographic characteristic.

> “*I hate the sun… It hurts my eyes… I do stay inside most of my life.”* (Child aged 8–12 years)

Meanwhile, reduced contrast sensitivity was experienced by 6/18 (33%) children, all of whom had bilateral disease.

> “*The stronger colours like blue, purple and black I can read but when it goes to like green and all of them other colours like orange I can’t, it’s harder for me to read what it says*.” (Child aged 13–17 years)

Theme 5: Ocular health concerns

Several children (13/18, 72%) discussed ocular health concerns which were often experienced as worry or anxiety. Hypersensitivity of objects touching their eye was experienced most (6/18, 33%), particularly by children with bilateral disease (5/6, 83%).

> “*One time my eye was really sore, and I got kind of worried, and kind of scared, but it turned out it was the ingrown eyelash.”* (Child aged 8–12 years)

Concerns for raised IOP (5/18, 28%) and losing vision (4/18, 22%) were additionally experienced. The former was more typical among children aged between 13 and 17 years (4/5, 80%).

> “*When I go to the like appointment, and I get my pressures checked I get nervous of if I'm going to get like a high pressure*.” (Child aged 13–17 years)

Requiring future surgery (2/18, 11%), forgetting to use their antiglaucoma medication (2/18, 11%) and changing ophthalmologist (1/18, 6%) caused concerns among fewer children.

> "*I don't want any more surgery. I'm done… it's just really scary.*" (Child aged 13–17 years)

Theme 6: Social well-being

Having glaucoma caused social issues for 13/18 (72%) children. Schoolyard bullying was experienced by 5/18 (28%) children irrespective of age. Bullying was attributed to their visual ability, need to wear spectacles, or need for sunglasses in the schoolyard.

> "*There are some kids at our school that have glasses that get bullied... Those kids have tried to bully me and my friends, so we have to defend ourselves.*" (Child aged 8–12 years)

Several children (5/13, 28%), of whom 4/5 (80%) were aged 13 to 17 years, experienced feeling socially isolated by their condition due to its rarity. It was often relieved by a desire to meet another child with glaucoma.

> "*I'm a loner at my school… People are a bit standoffish. I don't think they really know how to approach me.*" (Child aged 13–17 years)

Conversely, 6/18 (33%) children, of whom 4/6 (67%) were aged 8 to 12 years, reasoned that they had good social well-being.

> "*[My friends] all know about [my glaucoma] already… They just treat me the same.*" (Child aged 8–12 years)

Theme 7: Autonomy

Two-thirds (12/18, 67%) of children discussed issues relating to their autonomy. These were typically discussed by children aged 13 to 17 years compared to those aged 8 to 12 years (7/8, 88% vs 5/10, 50%). The main issue related to autonomy raised by younger children was that they wanted to administer their antiglaucoma medication without parental assistance. These children, however, frequently discussed being forgetful of when to use them.

> "*Most of the time I [put in the eye drops] myself and kept on forgetting.*" (Child aged 8–12 years)

All children aged ≥16 years (4/4, 100%) discussed issues becoming responsible for their own glaucoma care. These included actively engaging with the ophthalmologist and attending appointments without their parents, which were often met with feeling nervous or anxious.

> "*There's definitely questions I would like to ask but - I don't know…. I still get nervous asking.'"* (Child aged 13–17 years)

Among children aged 13 to 17 years, 4/8 (50%) wanted to know what caused their glaucoma and the risk involved in passing on their glaucoma to their future children.

> "*I'd definitely be interested to find out where I got it from… [but] if my children [have glaucoma], I guess it should be fine*." (Child aged 13–17 years)

The impact of glaucoma on their future career was discussed by 5/18 (28%) children, all of whom had bilateral or unilateral BCVA <20/60. Four (4/5, 80%) were aged 13 to 17 years.

> "*I can't actually join the Army, because of my lack of vision… It sucks, because now I don't actually have a plan for my life*." (Child aged 13–17 years)

Two children aged 13 to 17 years (2/18, 11%), one of whom had bilateral BCVA <20/60, discussed future issues with obtaining a driver's license whilst 3/18 (17%) children discussed issues with independently navigating environments due to their sight.

> "*I just think about what it'd be like if I could get a [driver's] license, when I'm driving on the road… I don't know if some person would pick on me because of the condition that I have*." (Child aged 13–17 years)

DISCUSSION

To the best of our knowledge, this exploratory interview study is the first qualitative study to explore the lived experience of children with glaucoma. Six of the seven themes identified were consistent with those reported in adults with childhood glaucoma,[24] and adult-onset glaucoma.[26,27] The impact of the condition on a child's autonomy was novel and provided a unique perspective of how childhood glaucoma impacts on the transition from childhood to adulthood. Each theme was relevant to all glaucoma subtypes and thus provided a thorough representation of how a child may live with glaucoma.

There are evidently several glaucoma-related non-visual and non-clinical variables that influence a child's QoL. Most notably, this includes how a child copes with their condition. This is in agreement with a recent study exploring the lived experience of adults with childhood glaucoma which similarly identified that resilience, adaptation and establishing a positive relationship with the ophthalmologist are important coping strategies in childhood glaucoma.[24] Becoming resilient was further identified as a coping strategy in children with cystic fibrosis,[28] spina bifida,[29] and type 1 diabetes.[30] This often assisted in self-management of their condition, as observed in this study whereby children, particularly those aged 8 to 12 years, who expressed a desire to self-manage their antiglaucoma medication.

Conversely, older youths with spina bifida,[29] and children with type 1 diabetes,[31] were more likely to disengage in their care over time, possibly due to having increased medical responsibilities and feeling overwhelmed. The same trend may be occurring in this study whereby children aged ≥16 years discussed issues related to disengagement in clinical care, in conjunction with autonomy-related pressures.

This possible age-related coping trend may be exacerbated by concurrent QoL issues. This study observed an additional trend for older children to experience more issues related to social well-being (social isolation) and ocular health concerns (increasing IOP) compared to their younger counterparts. The former may be particularly due to an increased understanding of glaucoma disease itself. Nonetheless, these collective issues may contribute to a greater psychosocial impact of glaucoma in older children. This observation is opposite to previous childhood glaucoma studies which reported lower psychosocial health in younger children.[7,10] Reasons for this disparity may be due to the non-specificity of the PROM used to measure HR-QoL,[7,10] the child's level of comprehension, or different characteristics in the cohorts studied. Nonetheless, adults with childhood glaucoma in Australia reported feeling misunderstood and were concerned with raised IOP, career, and family planning,[24] which are identical to those raised by older children in this study. Similar age-related observations were noted in children aged 14 to 18 years with cystic fibrosis who reported a greater disease-related impact on body image, emotional state and treatment burden compared to younger children.[32] Adolescents with type 1 diabetes additionally reported issues balancing demands between medical management and non-disease related pressures of being an adolescent.[33] Disease stigmatisation, social isolation, self-image and school absenteeism concerns were otherwise experienced among children of any age with asthma and epilepsy,[34] type 1 diabetes,[30,31] and juvenile idiopathic arthritis.[35]

Thus, the issues identified in children with glaucoma align with the greater childhood chronic disease experience and their impact may be exacerbated when a child approaches adulthood.

Clinicians should be aware of possible issues, particularly experienced during adolescence, as they may cumulatively influence the use of maladaptive coping and lead to medical negligence. This has been reported in adults aged 18 to 40 years with childhood glaucoma,[24] and such coping behaviours could lead to worse visual outcomes. Consequently, adolescents may require additional support to facilitate their transition toward adulthood and medical autonomy. This could involve provision of coping skills training, which aims to increase medical competence and the use of positive coping strategies.[36] This training has been successful for children with type 1 diabetes.[36] Ancillary ophthalmic personnel (e.g., orthoptists) may be best suited to facilitate this and future research could evaluate its effectiveness in children with glaucoma. Parent-to-child transfer of glaucoma self-management may otherwise begin at any age by providing children with an active voice in their care and increasing their knowledge of their glaucoma, as encouraged in other childhood chronic conditions.[37,38] These processes, however, must be tailored to the child's maturity, visual abilities and emotional state, with consideration to potential parental anxiety over relinquishing control of care to their child, as documented in parents of children with glaucoma.[39]

It is important to recognise that the QoL issues identified in this study were experienced by children irrespective of their clinical characteristics (i.e., BCVA and laterality). Previous research has demonstrated that VR-QoL and FVA is negatively associated with BCVA in children with glaucoma, but the impact of laterality remains unclear.[4-7] This may be explained by the results of this study. Firstly, children with bilaterally impaired BCVA used adaptive technology and did not perceive themselves to be limited. The availability and use of such technology may therefore influence how a child responds to VR-QoL and FVA-related questions. Secondly, children reported subjective symptoms including glare and reduced contrast sensitivity. These are yet to be measured as variables that may affect VR-QoL and FVA in children with glaucoma,[4-7] and may explain the variable effect of laterality. Glare is otherwise among the most common symptoms reported by adults with childhood glaucoma and contributed to their nonparticipation in outdoor activities.[24] Lastly, few children in this study subjectively reported that they had reduced BCVA irrespective of objective measurements and laterality. This may further contribute to unexpected or conflicting findings in quantitative association studies. Evidently, the impact of childhood glaucoma on QoL extends beyond a child's clinical characteristics and their subjective experience must be considered in clinical management of the condition.

To guide glaucoma management and enable more accurate investigation of the influence of clinical and demographic variables on QoL, a childhood glaucoma-specific PROM must be developed. Prior research have instead utilised VR-QoL (Impact of Vision Impairment for Children[19])[4-7] and HR-QoL measures (Kidscreen-27 questionnaire,[18] PedsQL[15])[7,10] that do not measure disease-specific QoL issues (e.g., inconveniences, ocular health concerns, symptoms). A childhood glaucoma-specific PROM will substantially improve our understanding of the disease impact and inform clinicians and education providers of QoL issues encountered by children. The results of this study will assist with the identification of items for a childhood glaucoma-specific PROM.

Study limitations include that children were recruited from a national registry and interviewed after receiving parental consent and child assent. Consequently, the child and/or parent may be more willing to participate and may be experiencing a higher QoL than non-respondents and/or their parents. Furthermore, children resided in Australia and the majority were of self-reported European ancestry such that findings may only be extrapolated to cohorts with similar socio-demographics, healthcare and education systems, and access to resources supporting visual functioning. Children with disease onset at age 16 or 17 years were unable to be recruited, likely owing to the narrow time frame between reaching adulthood and time required to conceptualise their diagnosis before agreeing to be interviewed. The experience of someone diagnosed at this age was otherwise captured in our previous study on adults diagnosed with childhood glaucoma.[24] Furthermore, more children interviewed had unilateral disease compared to non-respondents. It is unknown how these characteristics may have influenced results as thematic saturation was reached.

Despite these limitations, this study provided unique insight into the lived experience of childhood glaucoma from the perspective of the child. This rare condition may cause a considerable impact upon a child's physical, emotional, and social well-being which is managed with the use of coping strategies. Overall, it appears that older children experience more QoL issues compared to their younger counterparts. Healthcare professionals and parents should be mindful of this trend, and social and ophthalmic interventions may be required to support a child as they transition into adulthood and achieve medical autonomy. Future research endeavours should evaluate the most appropriate method to facilitate medical autonomy and subsequently ensure that any individual with childhood glaucoma achieves the best possible long-term visual and quality of life outcomes.

FOOTNOTES

Ethics Approval: Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee (HREC/19/WCHN/161) and the study adhered to the tenets of the Declaration of Helsinki. All child participants provided written assent and their parent or guardian provided informed written consent.

Patient consent for publication: Not required.

Acknowledgements: The authors wish to thank the research participants for their time and interest in the project.

Competing interests: None declared.

Funding: This work was supported by the Australian National Health and Medical Research Council (NHMRC) Centres of Research Excellence Grant (GNT1116360), Orthoptics Australia Research Grant (grant number N/A), Flinders Health and Medical Research Institute (FHMRI) Higher Degree Researcher Grant (grant number N/A) and Flinders University Cross-College Research Grant (grant number N/A). JEC was supported by an NHMRC Practitioner Fellowship (GNT1154824) and ES by an Early Career Fellowship from the Hospital Research Foundation (grant number N/A). LSWK was funded by Flinders Health and Medical Research Institute and Flinders Foundation scholarships (grant number N/A). SES was supported by the NHMRC Centre of Research Excellence Grant (GNT1116360). The Centre for Eye Research Australia receives Operational Infrastructure Support from the Victorian Government.

Contributorship statement: LSWK, BR, SES, JEC, MPS and ES were involved in the research conception and design. LSWK and BR were involved in data acquisition and research execution. LSWK and BR, SES, JEC, MPS and ES were involved in data analysis and interpretation. LSWK prepared an initial draft of the manuscript and BR, SES, JEC, MPS and ES edited, revised, and approved the final version of the manuscript.

Data sharing statement: No data are available.

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Figure legends

Figure 1. Quality of life themes identified in children with glaucoma

This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

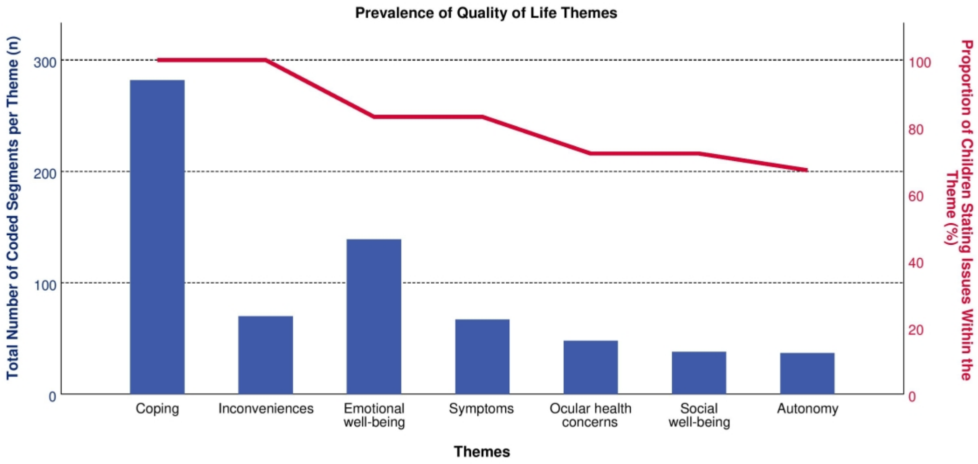

Figure 1. Quality of life themes identified in children with glaucoma. This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

Supplementary File 1. Semi-structured interview guide for children with glaucoma

Period of diagnosis/Emotional

What is it like growing up with glaucoma?

School and Cognitive Functioning

What is school like for you?

Do you think your eyes make some things harder to learn? (like maths, science or geography?)

Do you find it challenging to read your books or papers at school?

How do you find reading the board or laptop in your classroom?

Are you confident to ask for help at school?

How do you feel about the help that you get?

Do you keep up with other children in the classroom or do things take longer for you to finish? Can you tell me why?

Do you feel like your teachers and other children at school understand your eyes?

What do you like to do at playtime or recess? Do you do the same as your friends? Is it because of your eyes?

Can you find your friends easily in the playground? Why not?

Do you think you will finish high school? Why/why not?

Do you think that your glaucoma will determine what you do in the future for work or study?

Are you worried about it?

Relationships

Because of your glaucoma, do you find it easy to make friends?

Do you get along with your brothers and sisters? Do you think that's because of your eyes?

Do you feel that your family and friends understand your eye problem? Why/why not?

Does having glaucoma make you feel different to your friends or brothers or sisters? Why/why not?

Do you feel like other children treat you differently because of your eyes? Can you explain that?

Do you tell your friends about your eyes? Why/why not?

Do your family and friends need to help you do some things? Like what?

Do they give you enough help?

(*For older children if appropriate*) Have you ever been worried if your children will have glaucoma too?

Role Performance and Leisure

Has your eye problem made it hard to do some activities such as sports, playing on the playground, going to the movies, or playing video games?

How does that make you feel?

What do you do when you find something hard to do?

Psychological

Do you worry about your eyes? Why/why not?

Do you ever feel sad or angry about your eyes? What cheers you up?

(*For older children if appropriate*) What worries, or concerns do you have regarding the future?

Treatment/Medical Care

How does going to the eye doctor make you feel? Do you miss out on things because you have to go?

Prompting questions: Do you get nervous before you go? Why? (e.g., reading the vision chart, pressure test, needing eye drops, doing a visual field test, waiting a long time)

Do you feel like your eye doctor helps you? Do you ask questions?

How does putting in eye drops every day make you feel (if applicable)?

Mobility/Autonomy

How do you get to school? Can you catch the bus to school by yourself?

Do you find it hard to cross the road, or go up and down stairs, riding a bike?

Do you feel that you bump into things a lot? When does it happen or what sort of things do you bump into?

Are you worried about driving a car when you’re older?

Low Vision Aids

Do you use any special computers or iPads to make things bigger?

Do you like using them?

Does using them make you feel different?

Figure S1. A flow chart depicting the recruitment of participants

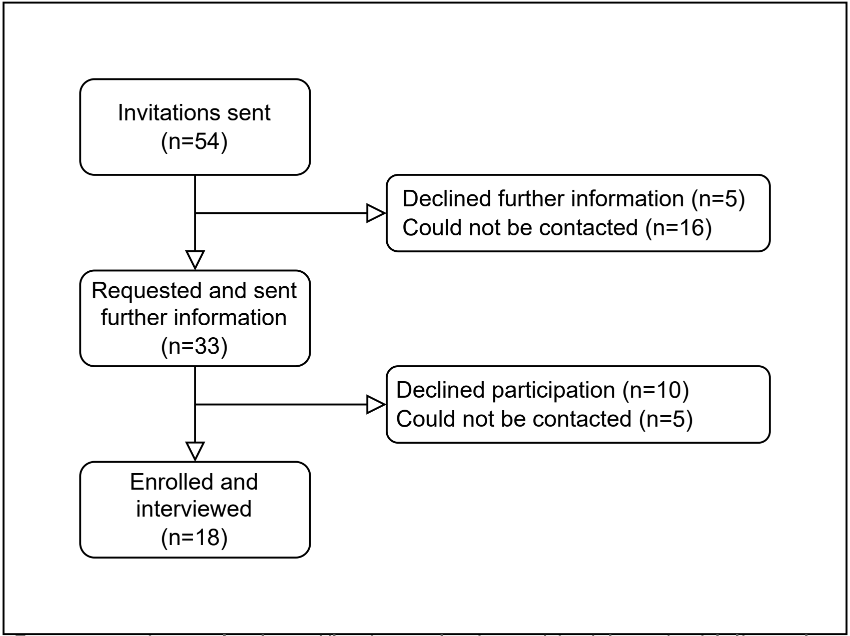

Figure S2. A mind map which illustrates the development of themes and subthemes.

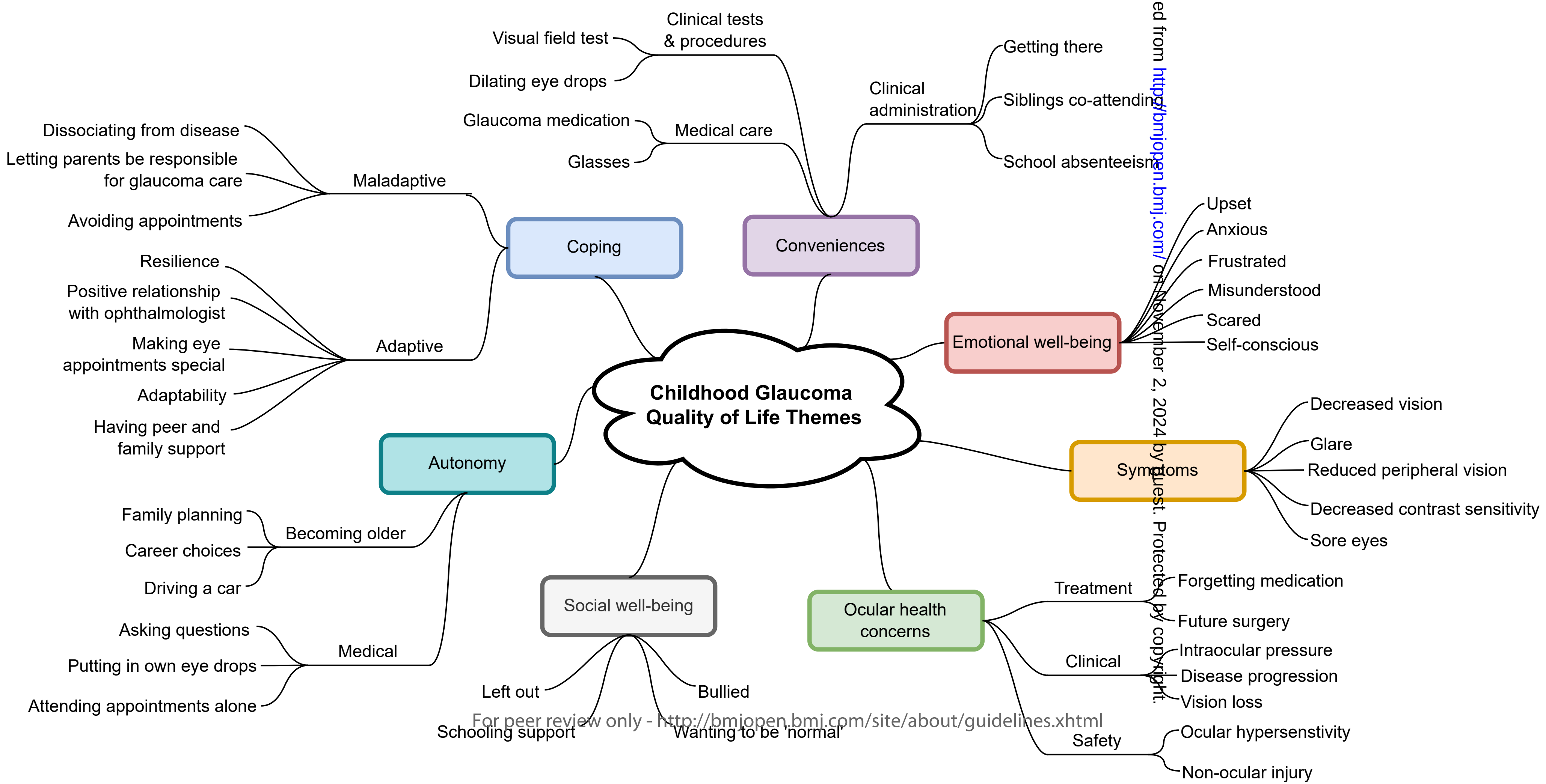

Table S1. Characteristics of children who enrolled and were interviewed compared to individuals who could not be contacted or declined participation.

Characteristic	**Individuals enrolled and interviewed, n (%)†** **(n=18)**	**Could not be contacted or declined participation, (n, %)†** **(n=36)**	**p value**
Current age, years (median [IQR])	12.1 (9.7–14.5)	14.2 (11.4–16.0)	0.14‡
Current age, ≥13 years	8 (44)	21 (58)	0.50§
Years at diagnosis (median [IQR])	0.5 (0.2–4.0)	0.5 (0.0–6.0)	0.89‡
Age at diagnosis, ≥4 years	5 (28)	10 (36)	1.00§
Years since diagnosis (median [IQR])	9.8 (7.3–13.6)	11.2 (8.2–14.0)	0.72‡
Gender, female	6 (33)	17 (47)	0.50§
Laterality of glaucoma, bilateral	11 (61)	34 (94)	**0.004**¶
Self-reported ancestry, European	16 (89)	21 (58)	0.06§
Glaucoma subtype, primary*	12 (67)	28 (78)	0.51¶
BCVA better eye, impaired (<20/60)	3 (17)	6 (17)	1.00¶
Molecular diagnosis identified	9 (50)	9 (25)	0.13§
Family history, first degree	4 (22)	17 (47)	0.12§

†: n (%) presented unless otherwise specified
‡Mann-Whitney U test
§Chi-square test with Yates' correction for continuity
¶Fisher exact test
*Primary glaucoma includes primary congenital glaucoma and juvenile open-angle glaucoma
IQR: interquartile range; BCVA: best-corrected visual acuity

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	
Occupation	3	What was their occupation at the time of the study?	
Gender	4	Was the researcher male or female?	
Experience and training	5	What experience or training did the researcher have?	
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	
Sample size	12	How many participants were in the study?	
Non-participation	13	How many people refused to participate or dropped out? Reasons?	
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	
Field notes	20	Were field notes made during and/or after the inter view or focus group?	
Duration	21	What was the duration of the inter views or focus group?	
Data saturation	22	Was data saturation discussed?	
Transcripts returned	23	Were transcripts returned to participants for comment and/or	

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	
Description of the coding tree	25	Did authors provide a description of the coding tree?	
Derivation of themes	26	Were themes identified in advance or derived from the data?	
Software	27	What software, if applicable, was used to manage the data?	
Participant checking	28	Did participants provide feedback on the findings?	
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	
Data and findings consistent	30	Was there consistency between the data presented and the findings?	
Clarity of major themes	31	Were major themes clearly presented in the findings?	
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

BMJ Open

Quality of life in children with glaucoma: A qualitative interview study in Australia

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2022-062754.R1
Article Type:	Original research
Date Submitted by the Author:	15-Jun-2022
Complete List of Authors:	Knight, Lachlan; Flinders University, Department of Ophthalmology; Women's and Children's Hospital, Department of Ophthalmology Ridge, Bronwyn; Flinders University, Department of Ophthalmology Staffieri, Sandra; The Royal Children's Hospital Melbourne, Department of Ophthalmology; Royal Victorian Eye and Ear Hospital, Centre for Eye Research Australia Craig, Jamie; Flinders University, Department of Ophthalmology Prem Senthil, Mallika; Flinders University, Caring Futures Institute, College of Nursing and Health Sciences Souzeau, Emmanuelle; Flinders University, Department of Ophthalmology
Primary Subject Heading:	Ophthalmology
Secondary Subject Heading:	Paediatrics, Patient-centred medicine, Qualitative research
Keywords:	Glaucoma < OPHTHALMOLOGY, QUALITATIVE RESEARCH, Paediatric ophthalmology < OPHTHALMOLOGY, OPHTHALMOLOGY

Quality of life in children with glaucoma: A qualitative interview study in Australia

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Word count: 4541

Abstract

Objective: Childhood glaucoma is a chronic vision-threatening condition which poses a substantial threat to an individual's psychosocial well-being. There is a paucity of literature investigating the quality-of-life (QoL) in children with glaucoma. The aim of this study was to investigate and report on the QoL issues encountered by children with glaucoma.

Design: This is a qualitative interview study. Data were collected through semi-structured interviews. NVivo-12 software (QSR International Pty Ltd, Melbourne, Australia) was used to analyse and code data to identify QoL themes. The prominence of QoL themes was determined by the number of children who experienced issues within it.

Participants: Eighteen children with glaucoma, aged 8 to 17 years, who resided in Australia were recruited from the Australian and New Zealand Registry of Advanced Glaucoma.

Setting: Interviews were conducted via telephone or videoconferencing between April 2020 and July 2021.

Results: Median child age was 12.1 years (interquartile range: 9.7–14.5 years) and 33% were female. Seven QoL themes were identified: 'Coping', 'inconveniences' and 'emotional well-being' were more prominent themes than 'symptoms', 'ocular health concerns', 'social well-being' and 'autonomy'. Adaptive coping strategies included resilience throughout clinical examinations and establishing positive relationships with ophthalmologists. These minimised inconveniences related to clinic waiting times and pupillary dilatation. External to the clinical setting, children often dissociated from their glaucoma but struggled with glare symptoms and feeling misunderstood by fellow peers. Older children aged 13 to 17 years commonly disengaged from their glaucoma care and expressed an unwillingness to attend ophthalmic appointments. Older children further experienced issues with career options, obtaining a driver's license and family planning under the theme of autonomy.

Conclusions: The psychosocial impact of childhood glaucoma extends beyond the clinical environment and was minimised using coping strategies. Older children may require additional social and ophthalmic support as they transition into adulthood.

Key Words: childhood glaucoma, glaucoma, quality of life, qualitative, interviews

Article Summary

Strengths and limitations of this study

- This study used an appropriate qualitative method to develop a novel and in-depth insight into the QoL issues experienced in childhood glaucoma from the perspectives of children.
- This study included individuals with varied disease characteristics and thus detailed the lived experience of the disease as a whole.
- Participants were recruited from a national registry and thus may be more willing to participate and may be experiencing a better quality-of-life than nonparticipants.
- Participants were mostly of European ancestry and resided in Australia which may limit the generalisability of the results.

INTRODUCTION

Childhood glaucoma describes a heterogeneous group of rare chronic vision-threatening disorders with onset occurring at any age from birth to less than 18 years of age.[1] It is typically characterised by elevated intraocular pressure (IOP) and irreversible optic neuropathy. Primary childhood glaucoma is caused by isolated abnormal development of the anterior chamber angle and includes primary congenital glaucoma (PCG) and juvenile open-angle glaucoma (JOAG).[1] Secondary childhood glaucoma includes glaucomatous disease that is associated with either other ocular anomalies (e.g., aniridia, Axenfeld-Rieger syndrome), an underlying systemic condition (e.g., Sturge-Weber syndrome) or an acquired ocular condition (e.g., uveitis, trauma).[1] Upon diagnosis, surgical intervention is typical and lifelong monitoring with or without additional surgical interventions and/or adjuvant topical therapies to manage IOP and prevent vision loss is generally required.[2] Additional symptoms can include glare and high myopia, and a child may experience cosmetic concerns associated with buphthalmos, occlusion therapy for amblyopia, and spectacle wear.[3]

Children with glaucoma may experience several visual and non-visual challenges as they adapt to living with the condition. However, there is a paucity of literature exploring the impact of these challenges on quality-of-life (QoL). Previous research is limited to quantitative association studies that utilize non-glaucoma specific patient-reported outcome measures (PROMs) that were designed to measure the impact of vision impairment on QoL (called vision-related QoL [VR-QoL])[4-7] or the impact on overall well-being (called health-related QoL [HR-QoL])[7,8]. This is because a childhood glaucoma-specific PROM does not exist. Consequently, the results from these studies may not be providing an accurate account of QoL in children with glaucoma. Nonetheless, several studies have reported that children with glaucoma who have lower best-corrected visual acuity (BCVA) experienced lower VR-QoL.[4-7] Meanwhile, a younger age has been associated with lower VR-QoL and HR-QoL.[7,8] However, there has been limited investigation as to why this trend was observed.[7,8] A qualitative inquiry is therefore required to explore disease-specific issues that are associated with QoL in children with glaucoma. Findings from this study will inform the development of a childhood glaucoma-specific PROM for future related research and clinical implementation.

METHODS

Participants

This study used a post-positivist paradigm to identify QoL issues.[9] Children were recruited from a large Australasian disease registry, the Australian and New Zealand Registry of Advanced Glaucoma (ANZRAG),[10] using a non-probability convenience sampling technique. Children were eligible to be interviewed if they currently resided in Australia, were English speaking, had a diagnosis of any subtype of glaucoma as per the Childhood Glaucoma Research Network criteria,[1] and were aged between 8 and <18 years. Children aged ≥8 years are more likely to reliably and independently understand questions relating to QoL than children aged <8 years.[11] Children were excluded if they had coexisting ocular disease unrelated to childhood glaucoma or had a hearing or cognitive impairment or other disability impacting on QoL (e.g., intellectual disability) as informed by their referring specialist or parent/guardian (henceforth abbreviated to parent).

Eligible children, and their parent/s, were posted an invitation to be interviewed and asked to return their interest. If both parties expressed interest, an information pack and consent form were sent. An interview was arranged once written informed consent from one parent and assent from the child were provided. If no response was received within two weeks, parents received a follow-up phone call. Children were deemed non-contactable after at least two unsuccessful attempts.

Children's clinical details were obtained from their most recent medical record and included: glaucoma subtype, age at diagnosis, laterality, BCVA (logMAR), IOP, number of surgical interventions, and number of topical antiglaucoma medications currently being used. The International Classification of Diseases for Mortality and Morbidity Statistics (11th Revision),[12] was used to categorise BCVA per eye. Because visual field information was not available for every child, BCVA was used as a measure of disease severity. For analysis, children's ages were grouped into 8 to 12 years and 13 to 17 years, as per the Pediatric Quality of Life Inventory Version 4.0 (PedsQL).[13] Glaucoma onset at ≥4 years was considered juvenile.[1] Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee and the study adhered to the tenets of the Declaration of Helsinki.

Interviews

A semi-structured interview guide was developed from a literature review of VR-QoL and HR-QoL PROMs (see Supplementary file 1, which details the semi-structured interview guide used).[13-17] Interviews were conducted in the English language by one of two authors with qualitative research experience (LSWK and BR). LSWK is a clinical and research orthoptist and BR is a health counsellor. No participants were under the clinical care of either interviewer. The child and parent/s were informed that the interviewers were completing a higher research degree. One-on-one semi-structured interviews occurred via telephone or Cisco WebEx videoconferencing (Milpitas, California, USA), subject to the child's preference. Children aged <16 years required a parent chaperone and parents were not to answer questions on their child's behalf. Interviews were audio-recorded and transcribed verbatim. Interviews continued until thematic saturation was achieved (i.e., the point where no new information was gained from subsequent interviews).[18] Thematic saturation occurred after the fourteenth interview. An additional four interviews with participants already recruited to the study were conducted to confirm data saturation. Recruitment ceased thereafter.

Data Analysis

A general inductive approach was used to identify QoL themes [19]. Transcripts were systematically coded using QSR NVivo 12 (QSR International Pty Ltd, Melbourne, Australia) by one author (LSWK) during the study recruitment period. To ensure research credibility, stakeholder coding checks were frequently and separately performed by three authors (BR, MPS and ES).[19] Major QoL themes, and their sub-themes were determined by grouping codes with similar or repetitive patterns of meaning,[20] and were abbreviated to be consistent with our previous ophthalmic QoL research pertaining to QoL issues encountered in adults with childhood glaucoma.[21] The prominence of QoL themes was determined by the number of children who experienced issues within it. Statistical calculations were performed using SPSS version 27.0 for Windows (IBM/SPSS Inc, Chicago, IL, USA). The datasets generated for the current study are not publicly available. This is to protect the confidentiality of research participants.

Patient and Public Involvement

Authors (LSWK, BR and ES) had presented the research aims at a national childhood glaucoma support group meeting prior to conducting the research. Engagement with attendees assisted in the development of the interview guide, and it was agreed that research findings would be disseminated back to the childhood glaucoma community.

RESULTS

Fifty-four eligible children from the ANZRAG were invited to participate and 18 (33%) were interviewed (see Figure S1 in Supplementary file 2, which depicts the recruitment of participants). The proportion of participants and non-participants with bilateral disease was significantly different (11/18, 61% vs 34/36, 94%, respectively, p=0.004) whilst all other demographic and clinical variables were similar (see Table S1 in Supplementary file 3). Reasons for declining to participate were not recorded due to the sensitive nature of the study.

Interviews were conducted between April 2020 and July 2021. The average interview length was 30 ± 14 minutes and the median age of children interviewed was 12.1 years (interquartile range: 9.7–14.5 years). Demographic and clinical characteristics of the children interviewed are detailed in Table 1.

Table 1. Demographic and clinical characteristics of children interviewed

Variable	**n (%)**†
Age at glaucoma diagnosis, years (median [range])	0.5 [0–15]
Time since diagnosis, years (median [IQR])	9.8 [7.3–13.6]
Age at interview	
8–12 years	10 (56)
13–17 years	8 (44)
Gender, female	6 (33)
Laterality of glaucoma, bilateral	11 (61)
Self-reported ancestry, European	16 (89)
Subtype of childhood glaucoma	
Primary congenital glaucoma	12 (67)
Glaucoma associated with non-acquired ocular anomalies	
Aniridia	1 (6)
Axenfeld-Rieger syndrome	1 (6)
Glaucoma associated with non-acquired systemic condition	

Sturge-Weber syndrome	1 (6)
Glaucoma associated with an acquired condition	
Idiopathic uveitis[‡]	2 (11)
Glaucoma following cataract surgery	1 (6)
Number of topical antiglaucoma medications currently using	
0	13 (72)
≥1	5 (28)
Intraocular pressure at last ophthalmic appointment, mmHg (median [range])	18 [14–25]
Time since last ophthalmic appointment, months (median [IQR])	3.8 [2.9–7.4]
Number of surgical interventions per child (median [IQR])	2 [2–4]
Time since last ophthalmic surgical intervention, years (median [IQR])	6.7 [1.6–13.6]
Disease complications	
Corneal disease	1 (6)
Cataract	4 (22)
Molecular diagnosis identified	9 (50)
Autosomal recessive inheritance	2 (11)
Autosomal dominant inheritance	7 (39)

Vision category	**BCVA (logMAR)**	**Better Eye BCVA (n, %)**	**Worse Eye BCVA (n, %)**
No vision impairment	≥0.3	15 (83)	8 (44)
Mild vision impairment	<0.3–≥0.5	1 (6)	4 (22)
Moderate vision impairment	<0.5–≥1.0	1 (6)	2 (11)
Severe vision impairment or blindness	<1.0–≥1.3	0 (0)	1 (6)
Blindness	<1.3–CF	1 (6)	2 (11)
	HM or LP	0 (0)	1 (6)
	NLP	0 (0)	0 (0)

[†]: n (%) presented unless otherwise specified

[‡]: No underlying systemic disease was diagnosed

BCVA: Best corrected visual acuity; CF: count fingers; HM: hand movements; IQR: interquartile range; LP: light perception; NLP: no light perception

Seven QoL themes emerged from the data. The total proportion of children experiencing issues per QoL theme and coded segments per theme are shown in Figure 1. Additional sub-themes not presented within the results are provided in a mind map (see Figure S2 in Supplementary file 2).

Theme 1: Coping

All children used coping strategies to manage the impacts of their glaucoma (Figure 1). All children (18/18, 100%) discussed being resilient, which is an adaptive emotion-focused coping strategy (i.e., a strategy which involves regulation or minimisation of negative emotions).[22]

> "*I've grown up with it. I've gotten used to it. I just don't pay much attention to it now.*" (Child aged 13–17 years)

Adaptive problem-focused strategies (i.e., strategies which actively confront the problem),[22] included developing a positive relationship with their ophthalmologist (12/18, 67%), seeking and accepting support from family, friends, or schoolteachers (11/18, 61%) and accepting parents' use of positive reinforcement for appointment attendance (9/18, 50%).

> "*I'm a lot more comfortable with [my ophthalmologist] because he's been doing it with me since basically the first time I went there... we're friends.*" (Child aged 8–12 years)

Several children (10/18, 56%) discussed adapting to activity limitations secondary to visual abilities or symptoms, such as photophobia. This was observed in children with bilateral (3/3, 100%) or unilateral BCVA <0.5 (3/7, 43%) and children with no BCVA impairment (4/8, 50%). Adapting to visual limitations was improved with the use of electronic devices in the classroom (e.g., laptop computer) whereby text size and contrast could be manipulated. Adapting to photophobia was usually resolved with sunglasses wear. Consequently, 5/18 (28%) children explicitly stated that their glaucoma did not impact their participation in daily activities.

> “*A lot of [schooling] stuff is on the computers and not written on the board anymore. So yeah, like I don’t really think that I have troubles.*” (Child aged 13–17 years)

Dissociating from one’s glaucoma outside of the clinical setting and ignoring its presence was used by 8/18 (44%) children, most of whom did not have bilaterally impaired BCVA (7/8, 88%). This was considered an adaptive strategy in 4/8 (50%), 3/4 (75%) of whom were aged 8 to 12 years, as these children considered themselves unaffected by their glaucoma. Conversely, it was considered maladaptive in 4/8 (50%) children, irrespective of age or gender, because these children avoided asking for vision-related assistance from teachers or were disinterested in possible disease consequences.

> “*I'm just not interested in my eyes much.*” (Child aged 8–12 years)

Actively leaving medical responsibilities and decision-making to their parent/s was discussed by more children aged 13 to 17 years compared to their younger counterparts (5/8, 63% vs 2/10, 20%, respectively). Gender, antiglaucoma medication use, and BCVA did not appear influential.

> “*I’d let Mom ask the questions… I’m more of a listener. Like a bystander… I’ll get all the information I want out of Mom.*” (Child aged 13–17 years)

Furthermore, 3/4 (75%) children aged ≥16 years discussed strong feelings of wanting to avoid attending their ophthalmic appointments.

> “*I was just yelling and screaming… I really did not want to go [to my appointment].*” (Child aged 13–17 years)

Theme 2: Inconveniences

All children discussed several inconveniences related to their ophthalmic appointments or glaucoma treatment. Clinic waiting time caused boredom for 6/18 (33%) children and 5/18 (28%) discussed negative outcomes related to school absenteeism. These were exacerbated where travelling long distances for ophthalmic review was required. Conversely, 7/18 (39%) reasoned that school absenteeism was a positive experience.

> "*It took us like three hours to get there and to go back... I often had to skip school to go there, and it was often always the fun days*." (Child aged 8–12 years)

Most children (11/18, 61%) discussed the inconvenience of having blurred vision for many hours following pupillary dilatation, whilst 4/18 (22%) considered a visual field test burdensome.

> "*I hate getting drops… everything I see is blurry for six or seven hours… They're still the worst thing that could possibly happen.*" (Child aged 13–17 years)

Spectacle wear was considered inconvenient and uncomfortable by 6/18 (33%) children, particularly during sporting activities. Among children who currently use topical antiglaucoma medication, 2/5 (40%) considered them bothersome.

> "*I don't really like wearing [glasses]... because my nose gets sweaty.*" (Child aged 8–12 years)

Theme 3: Emotional well-being

Negative emotional experiences were discussed by 15/18 (83%) children. Feeling frustrated (13/18, 72%) or anxious (10/18, 56%) were often experienced in the contexts of requiring pupil dilatation or performing certain clinical tests (e.g., visual field test, IOP test).

> "*The sight field test… has like things that blink and it's just like heaps of them, and it's like in a way sort of overwhelming.*" (Child aged 8–12 years)

Several children (7/18, 39%) discussed feeling misunderstood at times by their friends, peers and/or schoolteachers. At times, this led to concealment of their condition.

> "*I like keeping [my glaucoma] a bit of a secret… Because when I try to explain - no one understands and I have to keep explaining, explaining and explaining.*" (Child aged 8–12 years)

Feeling self-conscious of their appearance was expressed by 6/18 (33%) children. Reasons included their eye appearance, wearing spectacles or wearing an eye patch for amblyopia therapy. These were not dependent on BCVA, gender or age with the exception that one child, with bilateral BCVA <0.5, expressed feeling self-conscious whilst using their white cane for mobility.

> "*I hate [all the photos] when I'm younger because of the big, shaded glasses and stuff... I'm not a very photogenic person.*" (Child aged 13–17 years)

Theme 4: Symptoms

The most common symptom experienced by children was blurred vision (13/18, 72%). Of these, 4/13 (31%) had unilateral disease, and 7/13 (54%) had no BCVA impairment. It was usually described in the context of reading the classroom board, reading small texts, and playing sports that involve a small ball (e.g., tennis).

> "*If it's small writing and I'm at the back of the class I can't always get it but if it's like medium like to big writing I can see.*" (Child aged 13–17 years)

Glare (8/18, 44%), sore eyes (4/18, 22%) and reduced peripheral vision (2/18, 11%) were other symptoms experienced, irrespective of any clinical or demographic characteristic.

> "*I hate the sun… It hurts my eyes… I do stay inside most of my life.*" (Child aged 8–12 years)

Meanwhile, reduced contrast sensitivity was experienced by 6/18 (33%) children, all of whom had bilateral disease.

> "*The stronger colours like blue, purple and black I can read but when it goes to like green and all of them other colours like orange I can't, it's harder for me to read what it says.*" (Child aged 13–17 years)

Theme 5: Ocular health concerns

Several children (13/18, 72%) discussed ocular health concerns which were often experienced as worry or anxiety. Hypersensitivity of objects touching their eye was experienced most (6/18, 33%), particularly by children with bilateral disease (5/6, 83%).

> "*One time my eye was really sore, and I got kind of worried, and kind of scared, but it turned out it was the ingrown eyelash.*" (Child aged 8–12 years)

Concerns for raised IOP (5/18, 28%) and losing vision (4/18, 22%) were additionally experienced. The former was more typical among children aged between 13 and 17 years (4/5, 80%).

> "*When I go to the like appointment, and I get my pressures checked I get nervous of if I'm going to get like a high pressure*." (Child aged 13–17 years)

Requiring future surgery (2/18, 11%), forgetting to use their antiglaucoma medication (2/18, 11%) and changing ophthalmologist (1/18, 6%) caused concerns among fewer children.

> "*I don't want any more surgery. I'm done… it's just really scary*." (Child aged 13–17 years)

Theme 6: Social well-being

Having glaucoma caused social issues for 13/18 (72%) children. Schoolyard bullying was experienced by 5/18 (28%) children irrespective of age. Bullying was attributed to their visual ability, need to wear spectacles, or need for sunglasses in the schoolyard.

> "*There are some kids at our school that have glasses that get bullied... Those kids have tried to bully me and my friends, so we have to defend ourselves*." (Child aged 8–12 years)

Several children (5/13, 28%), of whom 4/5 (80%) were aged 13 to 17 years, experienced feeling socially isolated by their condition due to its rarity. It was often relieved by a desire to meet another child with glaucoma.

> "*I'm a loner at my school… People are a bit standoffish. I don't think they really know how to approach me*." (Child aged 13–17 years)

Conversely, 6/18 (33%) children, of whom 4/6 (67%) were aged 8 to 12 years, reasoned that they had good social well-being.

> "*[My friends] all know about [my glaucoma] already… They just treat me the same.*" (Child aged 8–12 years)

Theme 7: Autonomy

Two-thirds (12/18, 67%) of children discussed issues relating to their autonomy. These were typically discussed by children aged 13 to 17 years compared to those aged 8 to 12 years (7/8, 88% vs 5/10, 50%). The main issue related to autonomy raised by younger children was that they wanted to administer their antiglaucoma medication without parental assistance. These children, however, frequently discussed being forgetful of when to use them.

> "*Most of the time I [put in the eye drops] myself and kept on forgetting.*" (Child aged 8–12 years)

All children aged ≥16 years (4/4, 100%) discussed issues becoming responsible for their own glaucoma care. These included actively engaging with the ophthalmologist and attending appointments without their parents, which were often met with feeling nervous or anxious.

> "*There's definitely questions I would like to ask but - I don't know…. I still get nervous asking.'"* (Child aged 13–17 years)

Among children aged 13 to 17 years, 4/8 (50%) wanted to know what caused their glaucoma and the risk involved in passing on their glaucoma to their future children.

> "*I'd definitely be interested to find out where I got it from… [but] if my children [have glaucoma], I guess it should be fine.*" (Child aged 13–17 years)

The impact of glaucoma on their future career was discussed by 5/18 (28%) children, all of whom had bilateral or unilateral BCVA <0.5. Four (4/5, 80%) were aged 13 to 17 years.

> "*I can't actually join the Army, because of my lack of vision… It sucks, because now I don't actually have a plan for my life*." (Child aged 13–17 years)

Two children aged 13 to 17 years (2/18, 11%), one of whom had bilateral BCVA <0.5, discussed future issues with obtaining a driver's license whilst 3/18 (17%) children discussed issues with independently navigating environments due to their sight.

> "*I just think about what it'd be like if I could get a [driver's] license, when I'm driving on the road… I don't know if some person would pick on me because of the condition that I have*." (Child aged 13–17 years)

DISCUSSION

To the best of our knowledge, this exploratory interview study is the first qualitative study to explore the QoL issues experienced by children with glaucoma. Six of the seven themes identified were consistent with those reported in adults with childhood glaucoma,[21] and adult-onset glaucoma.[23,24] The impact of the condition on a child's autonomy was novel and provided a unique perspective of how childhood glaucoma impacts on the transition from childhood to adulthood. Each theme was relevant to all glaucoma subtypes and thus provided a thorough representation of how a child may live with glaucoma.

There are evidently several glaucoma-related non-visual and non-clinical variables that influence a child's QoL. Most notably, this includes how a child copes with their condition. This is in agreement with a recent study exploring the lived experience of adults with childhood glaucoma which similarly identified that resilience, adaptation and establishing a positive relationship with the ophthalmologist are important coping strategies in childhood glaucoma.[21] Becoming resilient was further identified as a coping strategy in children with cystic fibrosis,[25] spina bifida,[26] and type 1 diabetes.[27] This often assisted in self-management of their condition, as observed in this study whereby children, particularly those aged 8 to 12 years, expressed a desire to self-manage their antiglaucoma medication. Conversely, older youths with spina bifida,[26] and children with type 1 diabetes,[28] were more likely to disengage in their care over time, possibly due to having increased medical responsibilities and feeling overwhelmed. The same trend may be occurring in this study whereby children aged ≥16 years discussed issues related to disengagement in clinical care.

This possible age-related coping trend regarding disengagement may be underpinned by concurrent QoL issues. In this study, we observed a greater proportion of children aged 13-17 years who described more disruptions to QoL compared to children aged 8-12 years. These disruptions were particularly related to autonomy (becoming responsible for own care, career choices, driving, family planning), social well-being (social isolation) and ocular health concerns (increasing IOP). The latter may be particularly due to an increased understanding of glaucoma disease itself. Subsequently, these collective issues may contribute to a greater psychosocial impact of glaucoma in older children.

This hypothesis is opposite to findings in previous childhood glaucoma studies which reported lower VR-QoL and HR-QoL in younger children compared to their older counterparts.[7,8] These findings were irrespective of BCVA, disease laterality, gender and duration since surgery,[8] and was instead hypothesised to be a result of a child developing a better understanding of their condition and better coping strategies over time.[7,8] This was referred to as the “response shift”.[8] The disparity between our hypothesis and previous findings may be due to the non-specificity of the PROM used to measure QoL,[7,8] the child's level of comprehension, or different characteristics between the cohorts studied. This hypothesis could be explored in future quantitative association studies that utilise a childhood glaucoma-specific PROM. Nonetheless, our hypothesis is consistent with age-related observations reported in children aged 14 to 18 years with cystic fibrosis who reported a greater disease-related impact on body image, emotional state and treatment burden compared to younger children.[29] Adolescents with type 1 diabetes additionally reported issues balancing demands between medical management and non-disease related pressures of being an adolescent.[30] Disease stigmatisation, social isolation, self-image and school absenteeism concerns were otherwise experienced among children of any age with asthma and epilepsy,[31] type 1 diabetes,[27,28] and juvenile idiopathic arthritis.[32] Thus, the issues identified in children with glaucoma align with the greater childhood chronic disease experience and their impact may be exacerbated when a child approaches adulthood.

Clinicians should be aware of possible issues, particularly experienced during adolescence, as they may cumulatively influence the use of maladaptive coping and lead to medical negligence. This has been reported in adults aged 18 to 40 years with childhood glaucoma,[21] and such coping behaviours could lead to worse visual outcomes. Consequently, adolescents may require additional support to facilitate their transition toward adulthood and medical autonomy. This could involve provision of coping skills training, which aims to increase medical competence and the use of positive coping strategies.[33] This training has been successful for children with type 1 diabetes.[33] Ancillary ophthalmic personnel (e.g., orthoptists) may be best suited to facilitate this and future research could evaluate its effectiveness in children with glaucoma. Parent-to-child transfer of glaucoma self-management may otherwise begin at any age by providing children with an active voice in their care and increasing their knowledge of their glaucoma, as encouraged in other childhood chronic conditions.[34,35] These processes, however, must be tailored to the child's maturity, visual abilities and emotional state, with consideration to potential parental anxiety over relinquishing control of care to their child, as documented in parents of children with glaucoma.[36]

It is important to recognise that the QoL issues identified in this study appeared to be experienced by children irrespective of their clinical characteristics (i.e., BCVA and laterality). Previous research has demonstrated that VR-QoL is negatively associated with BCVA in the better-seeing eye in children with glaucoma.[4-7] Despite this, several studies have been unable to establish whether disease laterality is associated with VR-QoL.[4,5,7] Moreover, self-reported HR-QoL has not been found to be associated with disease laterality [7,8]. This suggests that unilateral disease may still impact QoL even if the child has normal BCVA in their better-seeing eye. The results of this study may offer some insight into these contradictory findings. Firstly, children with bilaterally impaired BCVA used adaptive technology and did not consider that their participation in daily activities was impacted. The availability and use of such technology may therefore influence how a child responds to QoL-related questions. Secondly, children reported subjective symptoms including glare and reduced contrast sensitivity. These are yet to be measured as variables that may affect QoL in children with glaucoma.[4-8] Glare is otherwise among the most common symptoms reported by adults with childhood glaucoma and contributed to their nonparticipation in outdoor activities.[21] It is therefore possible that the experience of these symptoms have a greater impact on QoL than disease laterality. Lastly, few children in this study subjectively reported that they had reduced BCVA irrespective of objective measurements and laterality. This may further contribute to unexpected or conflicting findings in quantitative association studies. Evidently, the impact of childhood glaucoma on QoL extends beyond a child's clinical characteristics and their subjective experience must be considered in clinical management of the condition.

To guide glaucoma management and enable more accurate investigation of the influence of clinical and demographic variables on QoL, a childhood glaucoma-specific PROM must be developed. Prior research have instead utilised VR-QoL (Impact of Vision Impairment for Children[17])[4-7] and HR-QoL measures (Kidscreen-27 questionnaire,[16] PedsQL[13])[7,8] that do not measure disease-specific QoL issues such as those identified in this study (e.g., concern for IOP, feeling misunderstood due to disease rarity). A childhood glaucoma-specific PROM will substantially improve our understanding of the disease impact and inform clinicians and education providers of QoL issues encountered by children. The results of this study will assist with the identification of items for a childhood glaucoma-specific PROM.

Study limitations include that children were recruited from a national registry and interviewed after receiving parental consent and child assent. Consequently, the child and/or parent may be more willing to participate and may be experiencing a higher QoL than non-respondents and/or their parents. Furthermore, children resided in Australia and the majority were of self-reported European ancestry. Consequently, the findings of this study may only be relevant to cohorts with similar socio-demographics, healthcare and education systems, and those with similar access to resources supporting visual functioning. Children with disease onset at age 16 or 17 years were unable to be recruited, likely owing to the narrow time frame between reaching adulthood and time required to conceptualise their diagnosis before agreeing to be interviewed. The experience of someone diagnosed at this age was otherwise captured in our previous study on adults diagnosed with childhood glaucoma.[21]

Furthermore, more children interviewed had unilateral disease compared to non-respondents, and most children had no vision impairment in their better eye. It is unknown how these characteristics may have influenced results as thematic saturation was reached. Lastly, the interviews specifically evaluated the impact of glaucoma such that the influence of conditions unique to uveitis, aniridia, Sturge-Weber syndrome and Axenfeld-Rieger syndrome were not included in the analysis. However, it remains possible that the physical manifestations of these conditions have impacted the QoL outcomes of this study.

Despite these limitations, this study provided unique insight into the QoL issues experienced in childhood glaucoma from the perspective of the child. This rare condition may cause a considerable impact upon a child's physical, emotional, and social well-being which is managed with the use of coping strategies. Overall, our findings suggest that older children may experience more QoL issues compared to their younger counterparts and hypothesise that increasing age may be associated with a lower QoL. Healthcare professionals and parents should be mindful of this trend, and social and ophthalmic interventions may be required to support a child as they transition into adulthood and achieve medical autonomy. Future research endeavours should evaluate the most appropriate method to facilitate medical autonomy and subsequently ensure that any individual with childhood glaucoma achieves the best possible long-term visual and quality of life outcomes.

FOOTNOTES

Ethics Approval: Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee (HREC/19/WCHN/161) and the study adhered to the tenets of the Declaration of Helsinki. All child participants provided written assent and their parent or guardian provided informed written consent.

Patient consent for publication: Not required.

Acknowledgements: The authors wish to thank the research participants for their time and interest in the project.

Competing interests: None declared.

Funding: This work was supported by the Australian National Health and Medical Research Council (NHMRC) Centres of Research Excellence Grant (GNT1116360), Orthoptics Australia Research Grant (grant number N/A), Flinders Health and Medical Research Institute (FHMRI) Higher Degree Researcher Grant (grant number N/A) and Flinders University Cross-College Research Grant (grant number N/A). JEC was supported by an NHMRC Practitioner Fellowship (GNT1154824) and ES by an Early Career Fellowship from the Hospital Research Foundation (grant number N/A). LSWK was funded by Flinders Health and Medical Research Institute and Flinders Foundation scholarships (grant number N/A). SES was supported by the NHMRC Centre of Research Excellence Grant (GNT1116360). The Centre for Eye Research Australia receives Operational Infrastructure Support from the Victorian Government.

Contributorship statement: LSWK, BR, SES, JEC, MPS and ES were involved in the research conception and design. LSWK and BR were involved in data acquisition and research execution. LSWK and BR, SES, JEC, MPS and ES were involved in data analysis and interpretation. LSWK prepared an initial draft of the manuscript and BR, SES, JEC, MPS and ES edited, revised, and approved the final version of the manuscript.

Data availability statement: No additional data available.

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Figure legends

Figure 1. Quality of life themes identified in children with glaucoma

This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

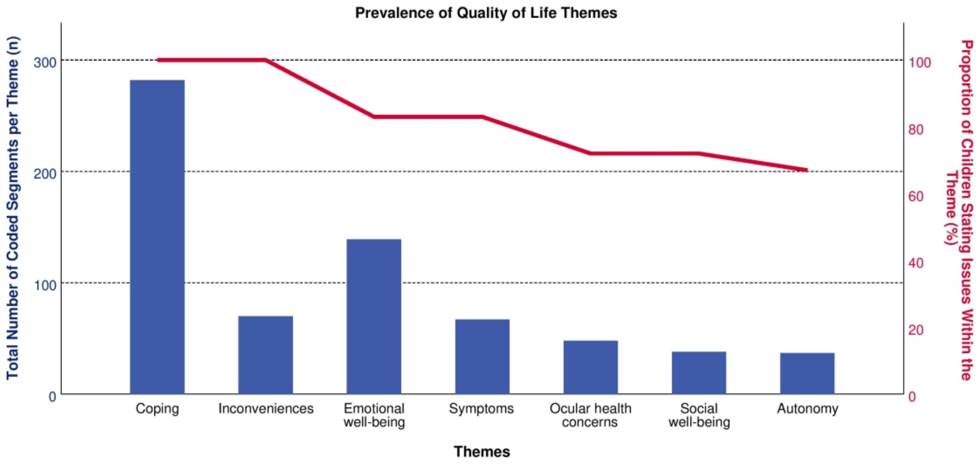

Figure 1. Quality of life themes identified in children with glaucoma. This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

Supplementary File 1. Semi-structured interview guide for children with glaucoma

Period of diagnosis/Emotional

What is it like growing up with glaucoma?

School and Cognitive Functioning

What is school like for you?

Do you think your eyes make some things harder to learn? (like maths, science or geography?)

Do you find it challenging to read your books or papers at school?

How do you find reading the board or laptop in your classroom?

Are you confident to ask for help at school?

How do you feel about the help that you get?

Do you keep up with other children in the classroom or do things take longer for you to finish? Can you tell me why?

Do you feel like your teachers and other children at school understand your eyes?

What do you like to do at playtime or recess? Do you do the same as your friends? Is it because of your eyes?

Can you find your friends easily in the playground? Why not?

Do you think you will finish high school? Why/why not?

Do you think that your glaucoma will determine what you do in the future for work or study?

Are you worried about it?

Relationships

Because of your glaucoma, do you find it easy to make friends?

Do you get along with your brothers and sisters? Do you think that's because of your eyes?

Do you feel that your family and friends understand your eye problem? Why/why not?

Does having glaucoma make you feel different to your friends or brothers or sisters? Why/why not?

Do you feel like other children treat you differently because of your eyes? Can you explain that?

Do you tell your friends about your eyes? Why/why not?

Do your family and friends need to help you do some things? Like what?

Do they give you enough help?

(*For older children if appropriate*) Have you ever been worried if your children will have glaucoma too?

Role Performance and Leisure

Has your eye problem made it hard to do some activities such as sports, playing on the playground, going to the movies, or playing video games?

How does that make you feel?

What do you do when you find something hard to do?

Psychological

Do you worry about your eyes? Why/why not?

Do you ever feel sad or angry about your eyes? What cheers you up?

(*For older children if appropriate*) What worries, or concerns do you have regarding the future?

Treatment/Medical Care

How does going to the eye doctor make you feel? Do you miss out on things because you have to go?

Prompting questions: Do you get nervous before you go? Why? (e.g., reading the vision chart, pressure test, needing eye drops, doing a visual field test, waiting a long time)

Do you feel like your eye doctor helps you? Do you ask questions?

How does putting in eye drops every day make you feel (if applicable)?

Mobility/Autonomy

How do you get to school? Can you catch the bus to school by yourself?

Do you find it hard to cross the road, or go up and down stairs, riding a bike?

Do you feel that you bump into things a lot? When does it happen or what sort of things do you bump into?

Are you worried about driving a car when you’re older?

Low Vision Aids

Do you use any special computers or iPads to make things bigger?

Do you like using them?

Does using them make you feel different?

Figure S1. A flow chart depicting the recruitment of participants

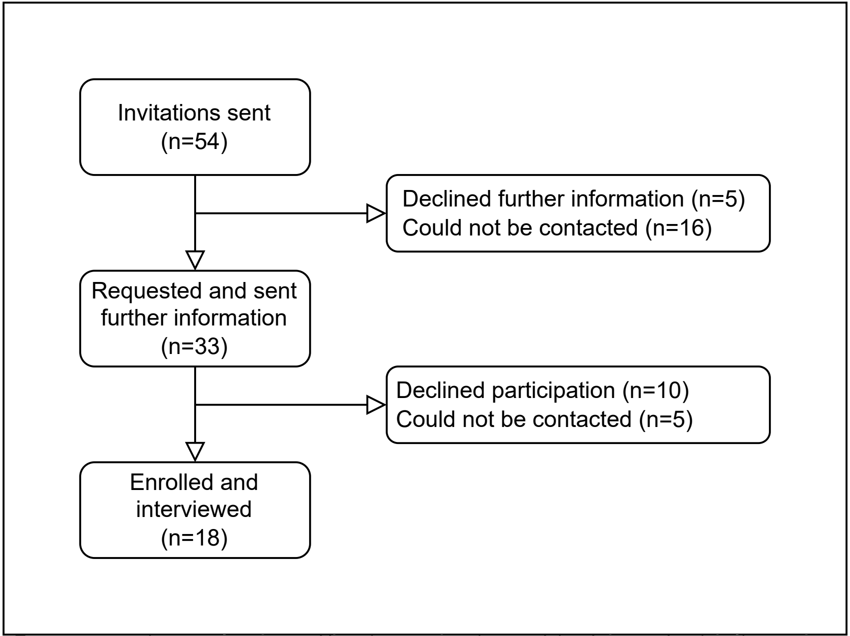

Figure S2. A mind map which illustrates the development of themes and subthemes

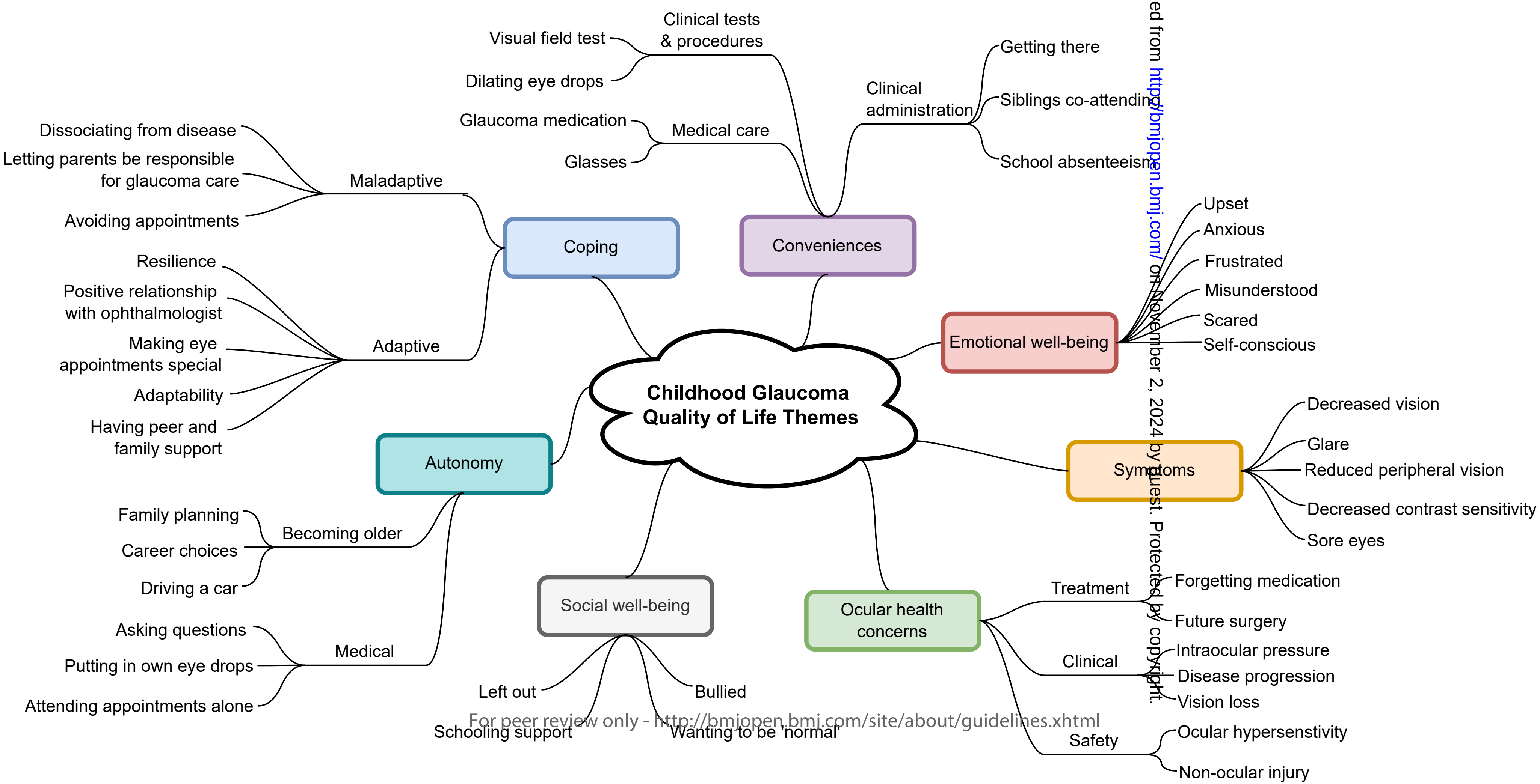

Table S1. Characteristics of children who enrolled and were interviewed compared to individuals who could not be contacted or declined participation.

Characteristic	**Individuals enrolled and interviewed, n (%)†** **(n=18)**	**Could not be contacted or declined participation, (n, %)†** **(n=36)**	**p value**
Current age, years (median [IQR])	12.1 (9.7–14.5)	14.2 (11.4–16.0)	0.14‡
Current age, ≥13 years	8 (44)	21 (58)	0.50§
Years at diagnosis (median [IQR])	0.5 (0.2–4.0)	0.5 (0.0–6.0)	0.89‡
Age at diagnosis, ≥4 years	5 (28)	10 (36)	1.00§
Years since diagnosis (median [IQR])	9.8 (7.3–13.6)	11.2 (8.2–14.0)	0.72‡
Gender, female	6 (33)	17 (47)	0.50§
Laterality of glaucoma, bilateral	11 (61)	34 (94)	**0.004**¶
Self-reported ancestry, European	16 (89)	21 (58)	0.06§
Glaucoma subtype, primary*	12 (67)	28 (78)	0.51¶
BCVA better eye, impaired (<0.5)	3 (17)	6 (17)	1.00¶
Molecular diagnosis identified	9 (50)	9 (25)	0.13§
Family history, first degree	4 (22)	17 (47)	0.12§

†: n (%) presented unless otherwise specified
‡Mann-Whitney U test
§Chi-square test with Yates' correction for continuity
¶Fisher exact test
*Primary glaucoma includes primary congenital glaucoma and juvenile open-angle glaucoma
IQR: interquartile range; BCVA: best-corrected visual acuity

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	
Occupation	3	What was their occupation at the time of the study?	
Gender	4	Was the researcher male or female?	
Experience and training	5	What experience or training did the researcher have?	
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	
Sample size	12	How many participants were in the study?	
Non-participation	13	How many people refused to participate or dropped out? Reasons?	
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	
Field notes	20	Were field notes made during and/or after the inter view or focus group?	
Duration	21	What was the duration of the inter views or focus group?	
Data saturation	22	Was data saturation discussed?	
Transcripts returned	23	Were transcripts returned to participants for comment and/or	

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	
Description of the coding tree	25	Did authors provide a description of the coding tree?	
Derivation of themes	26	Were themes identified in advance or derived from the data?	
Software	27	What software, if applicable, was used to manage the data?	
Participant checking	28	Did participants provide feedback on the findings?	
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	
Data and findings consistent	30	Was there consistency between the data presented and the findings?	
Clarity of major themes	31	Were major themes clearly presented in the findings?	
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

BMJ Open

Quality of life in children with glaucoma: a qualitative interview study in Australia

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2022-062754.R2
Article Type:	Original research
Date Submitted by the Author:	06-Jul-2022
Complete List of Authors:	Knight, Lachlan; Flinders University, Department of Ophthalmology; Women's and Children's Hospital, Department of Ophthalmology Ridge, Bronwyn; Flinders University, Department of Ophthalmology Staffieri, Sandra; The Royal Children's Hospital Melbourne, Department of Ophthalmology; Royal Victorian Eye and Ear Hospital, Centre for Eye Research Australia Craig, Jamie; Flinders University, Department of Ophthalmology Prem Senthil, Mallika; Flinders University, Caring Futures Institute, College of Nursing and Health Sciences Souzeau, Emmanuelle; Flinders University, Department of Ophthalmology
Primary Subject Heading:	Ophthalmology
Secondary Subject Heading:	Paediatrics, Patient-centred medicine, Qualitative research
Keywords:	Glaucoma < OPHTHALMOLOGY, QUALITATIVE RESEARCH, Paediatric ophthalmology < OPHTHALMOLOGY, OPHTHALMOLOGY

Quality of life in children with glaucoma: a qualitative interview study in Australia

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Word count: 4802

Abstract

Objective: Childhood glaucoma is a chronic vision-threatening condition that may significantly impact an individual's psychosocial well-being. There is a paucity of literature investigating the quality-of-life (QoL) in children with glaucoma. The aim of this study was to investigate and report on the QoL issues encountered by children with glaucoma.

Design: This is a qualitative interview study. Data were collected through semi-structured interviews. NVivo-12 software (QSR International Pty Ltd, Melbourne, Australia) was used to analyse and code data to identify QoL themes. The prominence of QoL themes was determined by the number of children who raised issues connected to the corresponding theme.

Setting: Interviews were conducted via telephone or videoconferencing between April 2020 and July 2021.

Participants: Eighteen children with glaucoma, aged 8 to 17 years, who resided in Australia, were recruited from the Australian and New Zealand Registry of Advanced Glaucoma.

Results: Median child age was 12.1 years (interquartile range: 9.7–14.5 years) and 33% were female. Seven QoL themes were identified: 'Coping', 'inconveniences' and 'emotional well-being' were more prominent themes than 'symptoms', 'ocular health concerns', 'social well-being' and 'autonomy'. Adaptive coping strategies included resilience throughout clinical examinations and establishing positive relationships with ophthalmologists. These minimised inconveniences related to clinic waiting times and pupillary dilatation. External to the clinical setting, children often dissociated from their glaucoma but struggled with glare symptoms and feeling misunderstood by fellow peers. Older children aged 13 to 17 years commonly disengaged from their glaucoma care and expressed an unwillingness to attend ophthalmic appointments. Older children further raised issues with career options, obtaining a driver's license and family planning under the theme of autonomy.

Conclusions: The psychosocial impact of childhood glaucoma extends beyond the clinical environment and was minimised using coping strategies. Older children may require additional social and ophthalmic support as they transition into adulthood.

Keywords: childhood glaucoma, glaucoma, quality of life, qualitative, interviews

Strengths and limitations of this study

- This study used an appropriate qualitative method to develop a novel and in-depth insight into the QoL issues experienced in childhood glaucoma from the perspectives of children.
- This study included individuals with varied disease characteristics and thus detailed the lived experience of the disease as a whole.
- Participants were recruited from a national registry and thus may be more willing to participate and may be experiencing a better quality-of-life than nonparticipants.
- Participants were mostly of European ancestry and resided in Australia, which may limit the generalisability of the results.

INTRODUCTION

Childhood glaucoma describes a heterogeneous group of rare chronic vision-threatening disorders with onset occurring at any age from birth to less than 18 years of age.[1] It is typically characterised by elevated intraocular pressure (IOP) and irreversible optic neuropathy. Primary childhood glaucoma is caused by isolated abnormal development of the anterior chamber angle and includes primary congenital glaucoma (PCG) and juvenile open-angle glaucoma (JOAG).[1] Secondary childhood glaucoma includes glaucomatous disease that is associated with either other ocular anomalies (e.g., aniridia, Axenfeld-Rieger syndrome), an underlying systemic condition (e.g., Sturge-Weber syndrome) or an acquired ocular condition (e.g., uveitis, trauma).[1] Upon diagnosis, surgical intervention is typical and lifelong monitoring with or without additional surgical interventions and/or adjuvant topical therapies to manage IOP and prevent vision loss is generally required.[2] Additional symptoms can include glare and high myopia, and a child may experience cosmetic concerns associated with buphthalmos, occlusion therapy for amblyopia, and spectacle wear.[3]

Children with glaucoma may experience several visual and non-visual challenges as they adapt to living with the condition. However, there is a paucity of literature exploring the impact of these challenges on quality-of-life (QoL). Previous research is limited to quantitative association studies that utilize non-glaucoma specific patient-reported outcome measures (PROMs) that were designed to measure the impact of vision impairment on QoL (called vision-related QoL [VR-QoL])[4-7] or the impact on overall well-being (called health-related QoL [HR-QoL])[7,8]. This is because a childhood glaucoma-specific PROM does not exist. Consequently, the results from these studies may not be providing an accurate account of QoL in children with glaucoma. Nonetheless, several studies have reported that children with glaucoma who have lower best-corrected visual acuity (BCVA) experienced lower VR-QoL.[4-7] Meanwhile, a younger age has been associated with lower VR-QoL and HR-QoL.[7,8] However, there has been limited investigation as to why this trend was observed.[7,8] A qualitative inquiry is therefore required to explore disease-specific issues that are associated with QoL in children with glaucoma. Findings from this study will inform the development of a childhood glaucoma-specific PROM for future related research and clinical implementation.

METHODS

Methodology

This study used a post-positivist paradigm to identify QoL issues.[9] This approach was used because we had a theoretical interest in how glaucoma may impact a child's QoL. This was formed by prior literature and our own clinical and research experience. Post-positivism further allows the calculation of the number of children represented within each theme.[9] This was considered useful in enhancing the readability of qualitative findings for positivist researchers and clinicians (e.g., ophthalmologists) who are instrumental in the care of children with glaucoma. Meanwhile, post-positivism acknowledges that the researchers' experiences may influence data collection and interpretation (i.e., researcher objectivity is not entirely possible).[9]

Participants

Children were recruited from a large Australasian disease registry, the Australian and New Zealand Registry of Advanced Glaucoma (ANZRAG),[10] using a non-probability convenience sampling technique. Children were eligible to be interviewed if they currently resided in Australia, were English speaking, had a diagnosis of any subtype of glaucoma as per the Childhood Glaucoma Research Network criteria,[1] and were aged between 8 and

<18 years. Children aged ≥8 years are more likely to reliably and independently understand questions relating to QoL than children aged <8 years.[11] Children were excluded if they had coexisting ocular disease unrelated to childhood glaucoma or had a hearing or cognitive impairment or other disability impacting on QoL (e.g., intellectual disability) as informed by their referring specialist or parent/guardian (henceforth abbreviated to parent).

Eligible children, and their parent/s, were posted an invitation to be interviewed and asked to return their interest. If both parties expressed interest, an information pack and consent form were sent. An interview was arranged once written informed consent from one parent and assent from the child were provided. If no response was received within two weeks, parents received a follow-up phone call. Children were deemed non-contactable after at least two unsuccessful attempts.

Children's clinical details were obtained from their most recent medical record and included: glaucoma subtype, age at diagnosis, laterality, BCVA (logMAR), IOP, number of surgical interventions, and number of topical antiglaucoma medications currently being used. The International Classification of Diseases for Mortality and Morbidity Statistics (11th Revision),[12] was used to categorise BCVA per eye. Because visual field information was not available for every child, BCVA was used as a measure of disease severity. For analysis, children's ages were grouped into 8 to 12 years and 13 to 17 years, as per the Pediatric Quality of Life Inventory Version 4.0 (PedsQL).[13] Glaucoma onset at ≥4 years was considered juvenile.[1] Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee and the study adhered to the tenets of the Declaration of Helsinki.

Interviews

A semi-structured interview guide was developed from a literature review of VR-QoL and HR-QoL PROMs (see Supplementary file 1, which details the semi-structured interview guide used).[13-17] Interviews were conducted in the English language by one of two authors with qualitative research experience (LSWK and BR). LSWK is a clinical and research orthoptist and BR is a health counsellor. No participants were under the clinical care of either interviewer. The child and parent/s were informed that the interviewers were completing a higher research degree. One-on-one semi-structured interviews occurred via telephone or Cisco WebEx videoconferencing (Milpitas, California, USA), subject to the child's preference. Children aged <16 years required a parent chaperone and parents were not to answer questions on their child's behalf. Interviews were audio-recorded and transcribed verbatim. Interview transcripts and overall findings were not returned to children for accuracy or feedback as it was considered burdensome to the child and unethical (i.e., the maturity and comprehension required to understand their contents could not be assured). Instead, at the conclusion of each interview, the child was provided with a verbal summary of their responses for confirmation that they had been interpreted correctly. Interviews continued until thematic saturation was achieved (i.e., the point where no new information was gained from subsequent interviews).[18] Thematic saturation occurred after the fourteenth interview. An additional four interviews with participants already recruited to the study were conducted to confirm data saturation. Recruitment ceased thereafter.

Data analysis

A general inductive approach was used to identify QoL themes [19]. Transcripts were systematically coded using QSR NVivo 12 (QSR International Pty Ltd, Melbourne, Australia) by one author (LSWK) during the study recruitment period. To ensure research credibility, stakeholder coding checks were frequently and separately performed by three authors (BR, MPS and ES).[19] Major QoL themes, and their sub-themes were determined by grouping codes with similar or repetitive patterns of meaning,[20] and were abbreviated to be consistent with our previous ophthalmic QoL research pertaining to QoL issues encountered in adults with childhood glaucoma.[21] The prominence of QoL themes was determined by the number of children who raised issues connected to the corresponding theme. Statistical calculations were performed using SPSS version 27.0 for Windows (IBM/SPSS Inc, Chicago, IL, USA). The datasets generated for the current study are not publicly available. This is to protect the confidentiality of research participants.

Patient and public involvement

Authors (LSWK, BR and ES) presented the research aims at a national childhood glaucoma support group meeting prior to conducting the research. Engagement with attendees assisted in the development of the interview guide, and it was agreed that research findings would be disseminated back to the childhood glaucoma community.

RESULTS

Fifty-four eligible children from the ANZRAG were invited to participate and 18 (33%) were interviewed (see Figure S1 in Supplementary file 2, which depicts the recruitment of participants). The proportion of participants and non-participants with bilateral disease was significantly different (11/18, 61% vs 34/36, 94%, respectively, p=0.004) whilst all other demographic and clinical variables were similar (see Table S1 in Supplementary file 3). Reasons for declining to participate were not recorded due to the sensitive nature of the study.

Interviews were conducted between April 2020 and July 2021. The average interview length was 30 ± 14 minutes and the median age of children interviewed was 12.1 years (interquartile range: 9.7–14.5 years). Demographic and clinical characteristics of the children interviewed are detailed in Table 1.

Table 1. Demographic and clinical characteristics of children interviewed

Variable	**n (%)**†
Age at glaucoma diagnosis, years (median [range])	0.5 [0–15]
Time since diagnosis, years (median [IQR])	9.8 [7.3–13.6]
Age at interview	
8–12 years	10 (56)
13–17 years	8 (44)
Gender, female	6 (33)
Laterality of glaucoma, bilateral	11 (61)
Self-reported ancestry, European	16 (89)
Subtype of childhood glaucoma	
Primary congenital glaucoma	12 (67)
Glaucoma associated with non-acquired ocular anomalies	
Aniridia	1 (6)
Axenfeld-Rieger syndrome	1 (6)
Glaucoma associated with non-acquired systemic condition	
Sturge-Weber syndrome	1 (6)
Glaucoma associated with an acquired condition	
Idiopathic uveitis‡	2 (11)
Glaucoma following cataract surgery	1 (6)
Number of topical antiglaucoma medications currently using	
0	13 (72)
≥1	5 (28)
Intraocular pressure at last ophthalmic appointment, mmHg (median [range])	18 [14–25]
Time since last ophthalmic appointment, months (median [IQR])	3.8 [2.9–7.4]
Number of surgical interventions per child (median [IQR])	2 [2–4]
Time since last ophthalmic surgical intervention, years (median [IQR])	6.7 [1.6–13.6]
Disease complications	
Corneal disease	1 (6)
Cataract	4 (22)
Molecular diagnosis identified	9 (50)

Autosomal recessive inheritance			2 (11)
Autosomal dominant inheritance			7 (39)
Vision category	**BCVA (logMAR)**	**Better Eye BCVA (n, %)**	**Worse Eye BCVA (n, %)**
No vision impairment	≥0.3	15 (83)	8 (44)
Mild vision impairment	<0.3–≥0.5	1 (6)	4 (22)
Moderate vision impairment	<0.5–≥1.0	1 (6)	2 (11)
Severe vision impairment or blindness	<1.0–≥1.3	0 (0)	1 (6)
Blindness	<1.3–CF	1 (6)	2 (11)
	HM or LP	0 (0)	1 (6)
	NLP	0 (0)	0 (0)

†: n (%) presented unless otherwise specified

‡: No underlying systemic disease was diagnosed

BCVA: Best corrected visual acuity; CF: count fingers; HM: hand movements; IQR: interquartile range; LP: light perception; NLP: no light perception

Seven QoL themes emerged from the data. The total proportion of children experiencing issues per QoL theme and coded segments per theme are shown in Figure 1. Additional sub-themes not presented within the results are provided in a mind map (see Figure S2 in Supplementary file 2).

Theme 1: Coping

All children used coping strategies to manage the impacts of their glaucoma (Figure 1). All children (18/18, 100%) discussed being resilient, which is an adaptive emotion-focused coping strategy (i.e., a strategy that involves regulation or minimisation of negative emotions).[22]

> "*I've grown up with it. I've gotten used to it. I just don't pay much attention to it now.*" (Child aged 13–17 years)

Adaptive problem-focused strategies (i.e., strategies that actively confront the problem),[22] included developing a positive relationship with their ophthalmologist (12/18, 67%), seeking and accepting support from family, friends, or schoolteachers (11/18, 61%) and accepting parents' use of positive reinforcement for appointment attendance (9/18, 50%).

> "*I'm a lot more comfortable with [my ophthalmologist] because he's been doing it with me since basically the first time I went there... we're friends*." (Child aged 8–12 years)

Several children (10/18, 56%) discussed adapting to activity limitations secondary to visual abilities or symptoms, such as photophobia. This was observed in children with bilateral (3/3, 100%) or unilateral BCVA <0.5 (3/7, 43%) and children with no BCVA impairment (4/8, 50%). Adapting to visual limitations was improved with the use of electronic devices in the classroom (e.g., laptop computer) whereby text size and contrast could be manipulated. Adapting to photophobia was usually resolved with sunglasses wear. Consequently, 5/18 (28%) children explicitly stated that their glaucoma did not impact their participation in daily activities.

> "*A lot of [schooling] stuff is on the computers and not written on the board anymore. So yeah, like I don't really think that I have troubles.*" (Child aged 13–17 years)

Dissociating from one's glaucoma outside of the clinical setting and ignoring its presence was used by 8/18 (44%) children, most of whom did not have bilaterally impaired BCVA (7/8, 88%). This was considered an adaptive strategy in 4/8 (50%), 3/4 (75%) of whom were aged 8 to 12 years, as these children considered themselves unaffected by their glaucoma. Conversely, it was considered maladaptive in 4/8 (50%) children, irrespective of age or gender, because these children avoided asking for vision-related assistance from teachers or were disinterested in possible disease consequences.

> "*I'm just not interested in my eyes much.*" (Child aged 8–12 years)

Actively leaving medical responsibilities and decision-making to their parent/s was discussed by more children aged 13 to 17 years compared to their younger counterparts (5/8, 63% vs 2/10, 20%, respectively). Gender, antiglaucoma medication use, and BCVA did not appear influential.

> "*I'd let Mom ask the questions… I'm more of a listener. Like a bystander… I'll get all the information I want out of Mom.*" (Child aged 13–17 years)

Furthermore, 3/4 (75%) children aged ≥16 years discussed strong feelings of wanting to avoid attending their ophthalmic appointments.

> "*I was just yelling and screaming… I really did not want to go [to my appointment].*" (Child aged 13–17 years)

Theme 2: Inconveniences

All children discussed several inconveniences related to their ophthalmic appointments or glaucoma treatment. Clinic waiting time caused boredom for 6/18 (33%) children and 5/18 (28%) discussed negative outcomes related to school absenteeism. These were exacerbated where travelling long distances for ophthalmic review was required. Conversely, 7/18 (39%) reasoned that school absenteeism was a positive experience.

> "*It took us like three hours to get there and to go back... I often had to skip school to go there, and it was often always the fun days.*" (Child aged 8–12 years)

Most children (11/18, 61%) discussed the inconvenience of having blurred vision for many hours following pupillary dilatation, whilst 4/18 (22%) considered a visual field test burdensome.

> "*I hate getting drops… everything I see is blurry for six or seven hours… They're still the worst thing that could possibly happen.*" (Child aged 13–17 years)

Spectacle wear was considered inconvenient and uncomfortable by 6/18 (33%) children, particularly during sporting activities. Among children who currently use topical antiglaucoma medication, 2/5 (40%) considered them bothersome.

> "*I don't really like wearing [glasses]... because my nose gets sweaty.*" (Child aged 8–12 years)

Theme 3: Emotional well-being

Negative emotional experiences were discussed by 15/18 (83%) children. Feeling frustrated (13/18, 72%) or anxious (10/18, 56%) were often experienced in the contexts of requiring pupil dilatation or performing certain clinical tests (e.g., visual field test, IOP test).

> "*The sight field test… has like things that blink and it's just like heaps of them, and it's like in a way sort of overwhelming.*" (Child aged 8–12 years)

Several children (7/18, 39%) discussed feeling misunderstood at times by their friends, peers and/or schoolteachers. At times, this led to concealment of their condition.

> "*I like keeping [my glaucoma] a bit of a secret… Because when I try to explain - no one understands and I have to keep explaining, explaining and explaining.*" (Child aged 8–12 years)

Feeling self-conscious of their appearance was expressed by 6/18 (33%) children. Reasons included their eye appearance, wearing spectacles or wearing an eye patch for amblyopia therapy. These were not dependent on BCVA, gender or age with the exception that one child, with bilateral BCVA <0.5, expressed feeling self-conscious whilst using their white cane for mobility.

> "*I hate [all the photos] when I'm younger because of the big, shaded glasses and stuff... I'm not a very photogenic person.*" (Child aged 13–17 years)

Theme 4: Symptoms

The most common symptom raised by children was blurred vision (13/18, 72%). Of these, 4/13 (31%) had unilateral disease, and 7/13 (54%) had no BCVA impairment. It was usually described in the context of reading the classroom board, reading small texts, and playing sports that involve a small ball (e.g., tennis).

> "*If it's small writing and I'm at the back of the class I can't always get it but if it's like medium like to big writing I can see.*" (Child aged 13–17 years)

Glare (8/18, 44%), sore eyes (4/18, 22%) and reduced peripheral vision (2/18, 11%) were other symptoms discussed by children, irrespective of any clinical or demographic characteristic.

> "*I hate the sun… It hurts my eyes… I do stay inside most of my life.*" (Child aged 8–12 years)

Meanwhile, reduced contrast sensitivity was discussed by 6/18 (33%) children, all of whom had bilateral disease.

> "*The stronger colours like blue, purple and black I can read but when it goes to like green and all of them other colours like orange I can't, it's harder for me to read what it says*." (Child aged 13–17 years)

Theme 5: Ocular health concerns

Several children (13/18, 72%) discussed ocular health concerns that were often experienced as worry or anxiety. Hypersensitivity of objects touching their eye was the most common concern raised (6/18, 33%), particularly by children with bilateral disease (5/6, 83%).

> "*One time my eye was really sore, and I got kind of worried, and kind of scared, but it turned out it was the ingrown eyelash."* (Child aged 8–12 years)

Concerns for raised IOP (5/18, 28%) and losing vision (4/18, 22%) were additionally discussed. The former was more typical among children aged between 13 and 17 years (4/5, 80%).

> "*When I go to the like appointment, and I get my pressures checked I get nervous of if I'm going to get like a high pressure*." (Child aged 13–17 years)

Requiring future surgery (2/18, 11%), forgetting to use their antiglaucoma medication (2/18, 11%) and changing ophthalmologist (1/18, 6%) caused concerns among fewer children.

> "*I don't want any more surgery. I'm done… it's just really scary*." (Child aged 13–17 years)

Theme 6: Social well-being

Having glaucoma caused social issues for 13/18 (72%) children. Schoolyard bullying was discussed by 5/18 (28%) children irrespective of age. Bullying was attributed to their visual ability, need to wear spectacles, or need for sunglasses in the schoolyard.

> "*There are some kids at our school that have glasses that get bullied... Those kids have tried to bully me and my friends, so we have to defend ourselves*." (Child aged 8–12 years)

Several children (5/13, 28%), of whom 4/5 (80%) were aged 13 to 17 years, discussed feeling socially isolated by their condition due to its rarity. It was often relieved by a desire to meet another child with glaucoma.

> "*I'm a loner at my school… People are a bit standoffish. I don't think they really know how to approach me*." (Child aged 13–17 years)

Conversely, 6/18 (33%) children, of whom 4/6 (67%) were aged 8 to 12 years, reasoned that they had good social well-being.

> "*[My friends] all know about [my glaucoma] already… They just treat me the same.*" (Child aged 8–12 years)

Theme 7: Autonomy

Two-thirds (12/18, 67%) of children discussed issues relating to their autonomy. These were typically discussed by children aged 13 to 17 years compared to those aged 8 to 12 years (7/8, 88% vs 5/10, 50%). The main issue related to autonomy raised by younger children was that they wanted to administer their antiglaucoma medication without parental assistance. These children, however, frequently discussed being forgetful of when to use them.

> "*Most of the time I [put in the eye drops] myself and kept on forgetting.*" (Child aged 8–12 years)

All children aged ≥16 years (4/4, 100%) discussed issues becoming responsible for their own glaucoma care. These included actively engaging with the ophthalmologist and attending appointments without their parents, which were often met with feeling nervous or anxious.

> "*There's definitely questions I would like to ask but - I don't know…. I still get nervous asking.'"* (Child aged 13–17 years)

Among children aged 13 to 17 years, 4/8 (50%) wanted to know what caused their glaucoma and the risk involved in passing on their glaucoma to their future children.

> "*I'd definitely be interested to find out where I got it from… [but] if my children [have glaucoma], I guess it should be fine*." (Child aged 13–17 years)

The impact of glaucoma on their future career was discussed by 5/18 (28%) children, all of whom had bilateral or unilateral BCVA <0.5. Four (4/5, 80%) were aged 13 to 17 years.

> "*I can't actually join the Army, because of my lack of vision… It sucks, because now I don't actually have a plan for my life*." (Child aged 13–17 years)

Two children aged 13 to 17 years (2/18, 11%), one of whom had bilateral BCVA <0.5, discussed future issues with obtaining a driver's license whilst 3/18 (17%) children discussed issues with independently navigating environments due to their sight.

> "*I just think about what it'd be like if I could get a [driver's] license, when I'm driving on the road… I don't know if some person would pick on me because of the condition that I have*." (Child aged 13–17 years)

DISCUSSION

To the best of our knowledge, this exploratory interview study is the first qualitative study to explore the QoL issues experienced by children with glaucoma. Six of the seven themes identified were consistent with those reported in adults with childhood glaucoma,[21] and adult-onset glaucoma.[23,24] The impact of the condition on a child's autonomy was novel and provided a unique perspective of how childhood glaucoma impacts on the transition from childhood to adulthood. Each theme was relevant to all glaucoma subtypes and thus provided a thorough representation of how a child may live with glaucoma.

There are evidently several glaucoma-related non-visual and non-clinical variables that influence a child's QoL. Most notably, this includes how a child copes with their condition. This is in agreement with a recent study exploring the lived experience of adults with childhood glaucoma, which similarly identified that resilience, adaptation and establishing a positive relationship with the ophthalmologist are important coping strategies in childhood glaucoma.[21] Becoming resilient was further identified as a coping strategy in children with cystic fibrosis,[25] spina bifida,[26] and type 1 diabetes.[27] This often assisted in self-management of their condition, as observed in this study whereby children, particularly those aged 8 to 12 years, expressed a desire to self-manage their antiglaucoma medication. Conversely, older youths with spina bifida,[26] and children with type 1 diabetes,[28] were more likely to disengage in their care over time, possibly due to having increased medical responsibilities and feeling overwhelmed. The same trend may be occurring in this study whereby children aged ≥16 years discussed issues related to disengagement in clinical care.

This possible age-related coping trend regarding disengagement may be underpinned by concurrent QoL issues. In this study, we observed a greater proportion of children aged 13-17 years who described more disruptions to QoL compared to children aged 8-12 years.

These disruptions were particularly related to autonomy (becoming responsible for own care, career choices, driving, family planning), social well-being (social isolation) and ocular health concerns (increasing IOP). The latter may be particularly due to an increased understanding of glaucoma disease itself. Subsequently, these collective issues may contribute to a greater psychosocial impact of glaucoma in older children.

This hypothesis is opposite to findings in previous childhood glaucoma studies that reported lower VR-QoL and HR-QoL in younger children compared to their older counterparts.[7,8] Other characteristics including BCVA, disease laterality, gender and duration since surgery were not found to influence this age-related finding.[8] Consequently, it was hypothesised that an older child may experience better QoL as they may develop a better understanding of their condition and better coping strategies over time.[7,8] This has been referred to as the “response shift”.[8] In contrast, our findings suggest there is an ‘implications shift’ whereby children appeared to be more concerned about limitations their glaucoma may place on their adult life as they enter adolescence. The apparent disparity between findings suggestive of a ‘response shift’ or an ‘implications shift’ may be explained by the studies’ different approaches (i.e., the use of a non-disease specific PROM to measure QoL,[7,8] compared to semi-structured interviews) or the clinical and demographic differences in the cohorts studied, including children’s abilities to respond to QoL-related questions. It would therefore be useful to further investigate the influence of aging on QoL and whether the ‘response shift’ or ‘implications shift’ is more likely to dominate the lived experience. This could be explored in future qualitative studies or quantitative association studies that utilise a childhood glaucoma-specific PROM. Nonetheless, our age-related findings are consistent with observations reported in children aged 14 to 18 years with cystic fibrosis who reported a greater disease-related impact on body image, emotional state and treatment burden compared to younger children.[29] Adolescents with type 1 diabetes additionally reported issues balancing demands between medical management and non-disease related pressures of being an adolescent.[30] Disease stigmatisation, social isolation, self-image and school absenteeism concerns were otherwise experienced among children of any age with asthma and epilepsy,[31] type 1 diabetes,[27,28] and juvenile idiopathic arthritis.[32] Thus, the issues identified in children with glaucoma align with the greater childhood chronic disease experience and their impact may be exacerbated when a child approaches adulthood.

Clinicians should be aware of possible issues, particularly experienced during adolescence, as they may cumulatively influence the use of maladaptive coping and lead to medical negligence. This has been reported in adults aged 18 to 40 years with childhood glaucoma,[21] and such coping behaviours could lead to worse visual outcomes. Consequently, adolescents may require additional support to facilitate their transition toward adulthood and medical autonomy. This could involve provision of coping skills training, which aims to increase medical competence and the use of positive coping strategies.[33] This training has been successful for children with type 1 diabetes.[33] Ancillary ophthalmic personnel (e.g., orthoptists) may be best suited to facilitate this and future research could evaluate its effectiveness in children with glaucoma. Parent-to-child transfer of glaucoma self-management may otherwise begin at any age by providing children with an active voice in their care and increasing their knowledge of their glaucoma, as encouraged in other childhood chronic conditions.[34,35] These processes, however, must be tailored to the child’s maturity, visual abilities and emotional state, with consideration to potential parental anxiety over relinquishing control of care to their child, as documented in parents of children with glaucoma.[36]

It is important to recognise that the QoL issues identified in this study appeared to be raised by children irrespective of their clinical characteristics (i.e., BCVA and laterality). Previous research has demonstrated that VR-QoL is negatively associated with BCVA in the better-seeing eye in children with glaucoma.[4-7] Despite this, several studies have been unable to establish whether disease laterality is associated with VR-QoL.[4,5,7] Moreover, self-reported HR-QoL has not been found to be associated with disease laterality [7,8]. This suggests that unilateral disease may still impact QoL even if the child has normal BCVA in their better-seeing eye. The results of this study may offer some insight into these contradictory findings. Firstly, children with bilaterally impaired BCVA used adaptive technology and did not consider that their participation in daily activities was impacted. The availability and use of such technology may therefore influence how a child responds to QoL-related questions. Secondly, children reported subjective symptoms including glare and reduced contrast sensitivity. These are yet to be measured as variables that may affect QoL in children with glaucoma.[4-8] Glare is otherwise among the most common symptoms reported by adults with childhood glaucoma and contributed to their nonparticipation in outdoor activities.[21] It is therefore possible that the experience of these symptoms have a greater impact on QoL than disease laterality. Lastly, few children in this study subjectively reported that they had reduced BCVA irrespective of objective measurements and laterality. This may further contribute to unexpected or conflicting findings in quantitative association studies. Evidently, the impact of childhood glaucoma on QoL extends beyond a child's clinical characteristics and their subjective experience must be considered in clinical management of the condition.

To guide glaucoma management and enable more accurate investigation of the influence of clinical and demographic variables on QoL, a childhood glaucoma-specific PROM must be developed. Prior research have instead utilised VR-QoL (Impact of Vision Impairment for Children[17])[4-7] and HR-QoL measures (Kidscreen-27 questionnaire,[16] PedsQL[13])[7,8] that do not measure disease-specific QoL issues such as those identified in this study (e.g., concern for IOP, feeling misunderstood due to disease rarity). A childhood glaucoma-specific PROM will substantially improve our understanding of the disease impact and inform clinicians and education providers of QoL issues encountered by children. The results of this study will assist with the identification of items for a childhood glaucoma-specific PROM.

Study limitations include that children were recruited from a national registry and interviewed after receiving parental consent and child assent. Consequently, the child and/or parent may be more willing to participate and may be experiencing a higher QoL than non-respondents and/or their parents. Furthermore, children resided in Australia and the majority were of self-reported European ancestry. Consequently, the findings of this study may only be relevant to cohorts with similar socio-demographics, healthcare and education systems, and those with similar access to resources supporting visual functioning. Children with disease onset at age 16 or 17 years were unable to be recruited, likely owing to the narrow time frame between reaching adulthood and time required to conceptualise their diagnosis before agreeing to be interviewed. The experience of someone diagnosed at this age was otherwise captured in our previous study on adults diagnosed with childhood glaucoma.[21] Furthermore, more children interviewed had unilateral disease compared to non-respondents, and most children had no vision impairment in their better eye. It is unknown how these characteristics may have influenced results as thematic saturation was reached. Lastly, the interviews specifically evaluated the impact of glaucoma such that the influence of conditions unique to uveitis, aniridia, Sturge-Weber syndrome and Axenfeld-Rieger syndrome were not included in the analysis. However, it remains possible that the physical manifestations of these conditions have impacted the QoL outcomes of this study.

Despite these limitations, this study provided unique insight into the QoL issues experienced in childhood glaucoma from the perspective of the child. This rare condition may cause a considerable impact upon a child's physical, emotional, and social well-being, which is managed with the use of coping strategies. Overall, our findings suggest that older children may experience more QoL issues compared to their younger counterparts and hypothesise that increasing age may be associated with a lower QoL. Healthcare professionals and parents should be mindful of this trend, and social and ophthalmic interventions may be required to support a child as they transition into adulthood and achieve medical autonomy. Future research endeavours should evaluate the most appropriate method to facilitate medical autonomy and subsequently ensure that any individual with childhood glaucoma achieves the best possible long-term visual and quality of life outcomes.

FOOTNOTES

Ethics approval: Ethical approval was obtained from the Women's and Children's Health Network Human Research Ethics Committee (HREC/19/WCHN/161) and the study adhered to the tenets of the Declaration of Helsinki. All child participants provided written assent and their parent or guardian provided informed written consent.

Patient consent for publication: Not required.

Acknowledgments: The authors wish to thank the research participants for their time and interest in the project.

Competing interests: None declared.

Funding: This work was supported by the Australian National Health and Medical Research Council (NHMRC) Centres of Research Excellence Grant (GNT1116360), Orthoptics Australia Research Grant (grant number N/A), Flinders Health and Medical Research Institute (FHMRI) Higher Degree Researcher Grant (grant number N/A) and Flinders University Cross-College Research Grant (grant number N/A). JEC was supported by an NHMRC Practitioner Fellowship (GNT1154824) and ES by an Early Career Fellowship from the Hospital Research Foundation (grant number N/A). LSWK was funded by Flinders Health and Medical Research Institute and Flinders Foundation scholarships (grant number N/A). SES was supported by the NHMRC Centre of Research Excellence Grant (GNT1116360). The Centre for Eye Research Australia receives Operational Infrastructure Support from the Victorian Government.

Contributors: LSWK, BR, SES, JEC, MPS and ES were involved in the research conception and design. LSWK and BR were involved in data acquisition and research execution. LSWK and BR, SES, JEC, MPS and ES were involved in data analysis and interpretation. LSWK prepared an initial draft of the manuscript and BR, SES, JEC, MPS and ES edited, revised, and approved the final version of the manuscript.

Data availability statement: No additional data available.

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Figure legends

Figure 1. Quality of life themes identified in children with glaucoma

This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

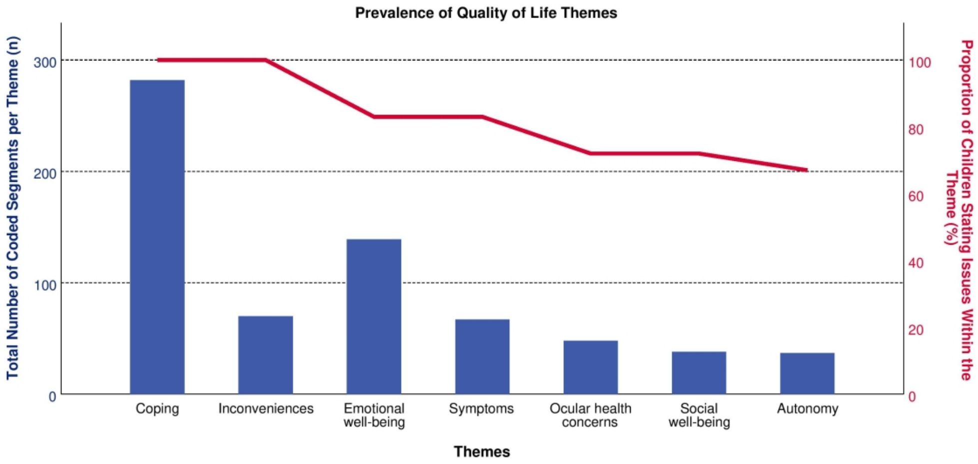

Figure 1. Quality of life themes identified in children with glaucoma. This Dual Y Axis Chart demonstrates the total number of codes per theme (blue bar chart) and the proportion of children who discussed an issue within the theme (red line chart).

Supplementary File 1. Semi-structured interview guide for children with glaucoma

Period of diagnosis/Emotional

What is it like growing up with glaucoma?

School and Cognitive Functioning

What is school like for you?

Do you think your eyes make some things harder to learn? (like maths, science or geography?)

Do you find it challenging to read your books or papers at school?

How do you find reading the board or laptop in your classroom?

Are you confident to ask for help at school?

How do you feel about the help that you get?

Do you keep up with other children in the classroom or do things take longer for you to finish? Can you tell me why?

Do you feel like your teachers and other children at school understand your eyes?

What do you like to do at playtime or recess? Do you do the same as your friends? Is it because of your eyes?

Can you find your friends easily in the playground? Why not?

Do you think you will finish high school? Why/why not?

Do you think that your glaucoma will determine what you do in the future for work or study?

Are you worried about it?

Relationships

Because of your glaucoma, do you find it easy to make friends?

Do you get along with your brothers and sisters? Do you think that's because of your eyes?

Do you feel that your family and friends understand your eye problem? Why/why not?

Does having glaucoma make you feel different to your friends or brothers or sisters? Why/why not?

Do you feel like other children treat you differently because of your eyes? Can you explain that?

Do you tell your friends about your eyes? Why/why not?

Do your family and friends need to help you do some things? Like what?

Do they give you enough help?

(*For older children if appropriate*) Have you ever been worried if your children will have glaucoma too?

Role Performance and Leisure

Has your eye problem made it hard to do some activities such as sports, playing on the playground, going to the movies, or playing video games?

How does that make you feel?

What do you do when you find something hard to do?

Psychological

Do you worry about your eyes? Why/why not?

Do you ever feel sad or angry about your eyes? What cheers you up?

(*For older children if appropriate*) What worries, or concerns do you have regarding the future?

Treatment/Medical Care

How does going to the eye doctor make you feel? Do you miss out on things because you have to go?

Prompting questions: Do you get nervous before you go? Why? (e.g., reading the vision chart, pressure test, needing eye drops, doing a visual field test, waiting a long time)

Do you feel like your eye doctor helps you? Do you ask questions?

How does putting in eye drops every day make you feel (if applicable)?

Mobility/Autonomy

How do you get to school? Can you catch the bus to school by yourself?

Do you find it hard to cross the road, or go up and down stairs, riding a bike?

Do you feel that you bump into things a lot? When does it happen or what sort of things do you bump into?

Are you worried about driving a car when you're older?

Low Vision Aids

Do you use any special computers or iPads to make things bigger?

Do you like using them?

Does using them make you feel different?

Figure S1. A flow chart depicting the recruitment of participants

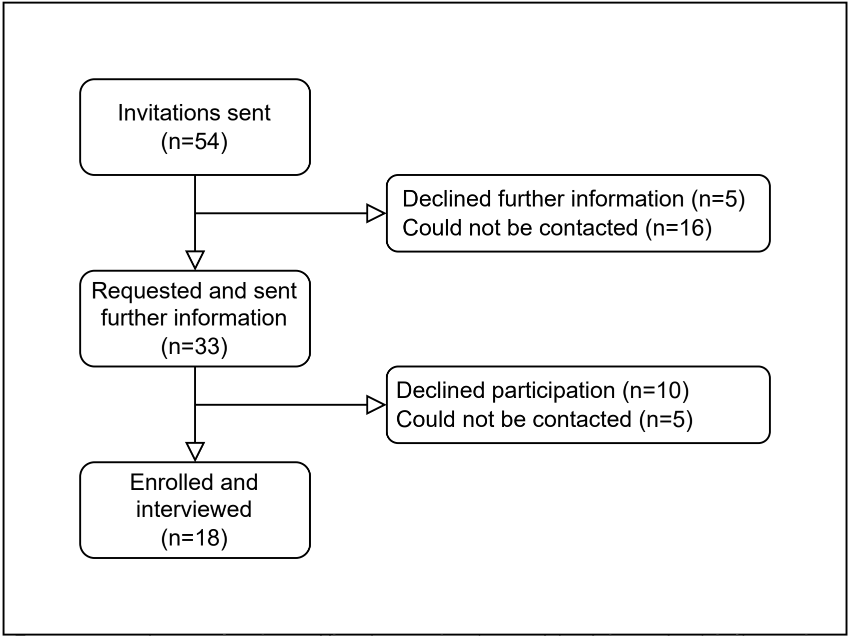

Figure S2. A mind map which illustrates the development of themes and sub-themes.

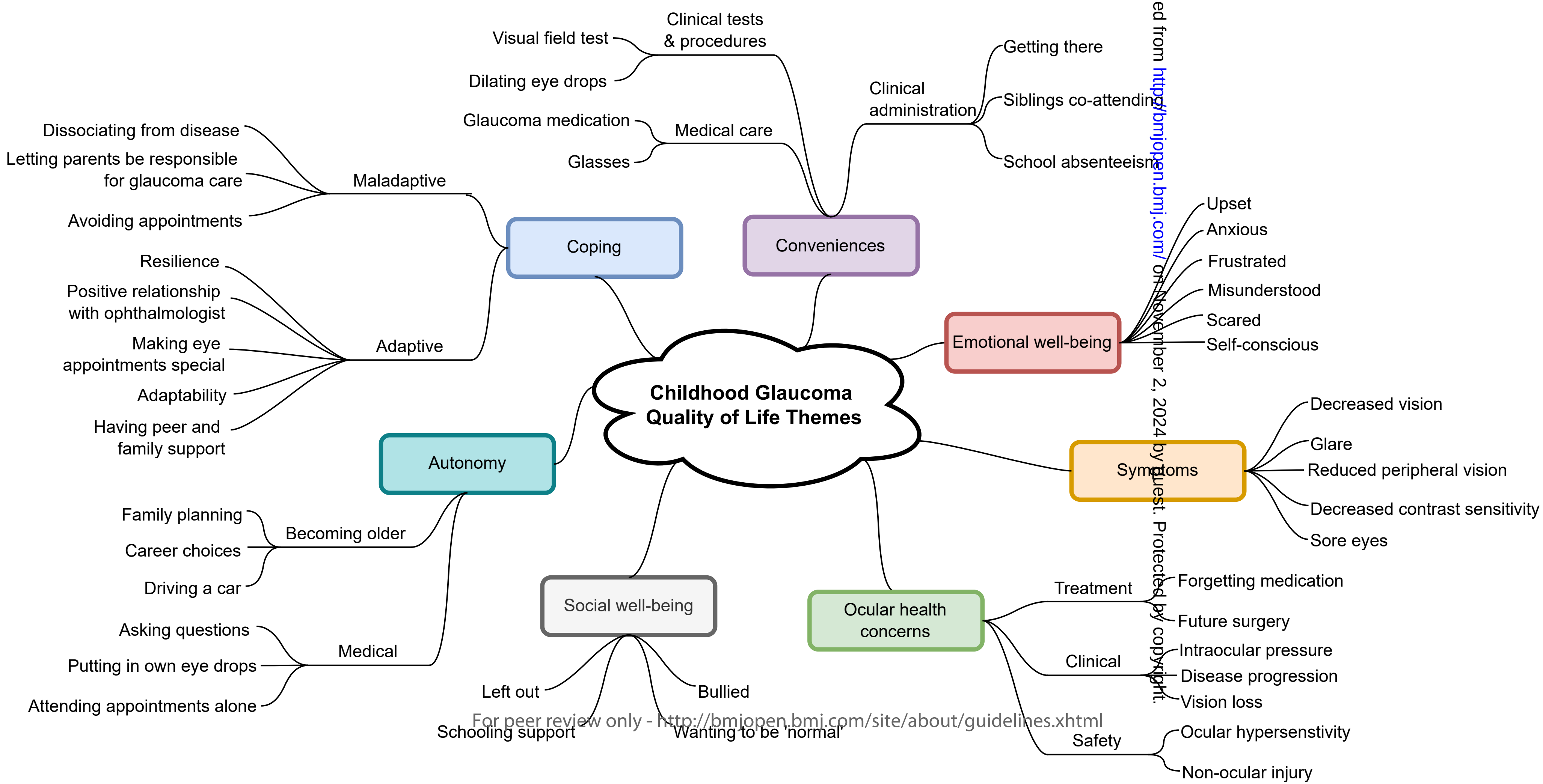

Table S1. Characteristics of children who enrolled and were interviewed compared to individuals who could not be contacted or declined participation.

Characteristic	**Individuals enrolled and interviewed, n (%)† (n=18)**	**Could not be contacted or declined participation, (n, %)† (n=36)**	**p value**
Current age, years (median [IQR])	12.1 (9.7–14.5)	14.2 (11.4–16.0)	0.14‡
Current age, ≥13 years	8 (44)	21 (58)	0.50§
Years at diagnosis (median [IQR])	0.5 (0.2–4.0)	0.5 (0.0–6.0)	0.89‡
Age at diagnosis, ≥4 years	5 (28)	10 (36)	1.00§
Years since diagnosis (median [IQR])	9.8 (7.3–13.6)	11.2 (8.2–14.0)	0.72‡
Gender, female	6 (33)	17 (47)	0.50§
Laterality of glaucoma, bilateral	11 (61)	34 (94)	**0.004**¶
Self-reported ancestry, European	16 (89)	21 (58)	0.06§
Glaucoma subtype, primary*	12 (67)	28 (78)	0.51¶
BCVA better eye, impaired (<0.5)	3 (17)	6 (17)	1.00¶
Molecular diagnosis identified	9 (50)	9 (25)	0.13§
Family history, first degree	4 (22)	17 (47)	0.12§

†: n (%) presented unless otherwise specified
‡Mann-Whitney U test
§Chi-square test with Yates' correction for continuity
¶Fisher exact test
*Primary glaucoma includes primary congenital glaucoma and juvenile open-angle glaucoma
IQR: interquartile range; BCVA: best-corrected visual acuity

COREQ (COnsolidated criteria for REporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	
Occupation	3	What was their occupation at the time of the study?	
Gender	4	Was the researcher male or female?	
Experience and training	5	What experience or training did the researcher have?	
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	
Sample size	12	How many participants were in the study?	
Non-participation	13	How many people refused to participate or dropped out? Reasons?	
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	
Field notes	20	Were field notes made during and/or after the inter view or focus group?	
Duration	21	What was the duration of the inter views or focus group?	
Data saturation	22	Was data saturation discussed?	
Transcripts returned	23	Were transcripts returned to participants for comment and/or	

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	
Description of the coding tree	25	Did authors provide a description of the coding tree?	
Derivation of themes	26	Were themes identified in advance or derived from the data?	
Software	27	What software, if applicable, was used to manage the data?	
Participant checking	28	Did participants provide feedback on the findings?	
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	
Data and findings consistent	30	Was there consistency between the data presented and the findings?	
Clarity of major themes	31	Were major themes clearly presented in the findings?	
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.