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'It's not meant to be for life, but it carries on': a qualitative investigation into the psychosocial needs of young retinoblastoma survivors

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ABSTRACT

Objective and design Retinoblastoma (Rb) is a rare childhood eye cancer, with 45% of individuals impacted by heritable disease and the remainder impacted non-heritably. The condition can leave survivors with life-long psychological and social challenges. This qualitative study examined the psychosocial needs of teenagers and young adults living beyond Rb.

Setting A qualitative, exploratory study was conducted using focus groups with teenagers and interviews with young adults. Participants were recruited via the Childhood Eye Cancer Trust and the two national Rb treatment centres in the UK. Reflexive thematic analysis was used to analyse data using exploratory and inductive methods.

Participants 32 young survivors of Rb (10 heritable, 21 non-heritable, 1 unknown; 23 unilateral, 9 bilateral) aged between 13 and 29 years (12 male, 20 female).

Results Data were rich and spanned the life course: three key themes were generated, containing eight subthemes. Theme 1 describes participants' experiences of childhood and trauma, including survivor guilt, memories from treatment and impact on personality. Theme 2 focuses on the challenges of adolescence, including the psychological impact of Rb, the impact on identity, and the sense of normality and adaptation to late effects. The third theme considered adulthood and the development of acceptance, a state of being widely considered unachievable during childhood, as well as the 'work' needed to feel supported, including seeking out information, peer support and therapeutic strategies.

Conclusions This study provides in-depth insight into the experiences of life beyond Rb. Findings highlight the need for specific psychosocial interventions informed by codesign.

STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ This is one of the first qualitative studies to seek an in-depth understanding of the range of psychosocial experiences of retinoblastoma (Rb) survivors, who are now teenagers and young adults.
- ⇒ It includes a relatively large sample (given the small population impacted by Rb), with heritable, non-heritable, unilateral or bilateral diagnoses.
- ⇒ It uses reflexive thematic analysis, which is flexible enough to accommodate different data collection methods (interviews and focus groups), to suit varied ages.
- ⇒ The majority of participants were recruited through a single charitable organisation, which may have a minor impact on the transferability of findings.

INTRODUCTION

Retinoblastoma (Rb) is a rare childhood cancer affecting the eye(s), which is diagnosed in 40–50 children in the UK a year. Around 45% are heritable,[1 2] the remainder occurring spontaneously. Although highly curable (~80% survival 10 years postdiagnosis),[3] Rb can greatly impact psychological well-being long after treatment ends.[4] For many, vision is impaired, and they may experience eye loss or facial changes.[5 6] Others may show no outward indication of their experience, but experience internal lifelong anxiety about the development of second cancers and the possibility of their own children developing Rb.[7]

Most instances of Rb are diagnosed before age 5, during a time of rapid development, when attachments with caregivers, cognitive and linguistic abilities, and emotional regulation are evolving in line with the environment.[8] As Rb impacts very young children, treatment is mostly complete before the individual is able to process the experience with meaningful language; their developmental stage hindering the ability to verbalise their experiences. This can distort memories and complicate psychosocial outcomes as those affected become teenagers and young adults.[9]

Unprocessed trauma is generally considered an integrated mind-body response, and the adaptive information processing (AIP) model describes the dysfunctional way the human brain stores such experiences,[10] potentially resulting in psychological distress, maladaptive thoughts or behaviours, and even physical pain. This has been verified in children experiencing medical trauma, with procedures found to be linked to a high degree of threat and frightening associations with pain, along with an immature understanding of events, including confusing feelings of betrayal towards their parents, whom they may associate with their treatment.[11] Consequently, parents often experience strong psychological reactions to their child's diagnosis,[12] including feelings of powerlessness, guilt and trauma.[13]

Despite the challenging circumstances associated with Rb, there is currently no known specific, evidence-based psychological support offered to young people in the UK, as they transition to adulthood and begin to navigate life beyond Rb. Existing research has highlighted the need for psychosocial support, yet what is required is under-researched and largely unavailable.[4 12 14 15] To address this, the current study aimed to explore, understand and describe the psychosocial needs of young people living beyond Rb, to inform future psychoeducation interventions.

METHODS

This study is part of a broader portfolio of work to understand the psychosocial needs of Rb survivors and develop a novel psychoeducation resource. Methods and results used the Consolidated criteria for Reporting Qualitative research to ensure appropriate empirical reporting.[16]

Study design

A qualitative study was conducted, with in-depth, semi-structured focus groups for teenage participants (aged 13–19 years) and individual interviews for young adults (aged 20–29). The rationale for the different approaches was based on evidence highlighting the distinct psychosocial needs of the two groups, in terms of identity, autonomy, emotional and sexual intimacy, cognitive ability, family planning, financial responsibility, education, and employment.[17]

Setting

All participants were treated in the UK at one of the two national Rb treatment centres: The Royal London Hospital shared with Great Ormond Street Hospital and Birmingham Children's Hospital. The third organisation involved was the Childhood Eye Cancer Trust (CHECT), a UK charity dedicated to supporting people impacted by Rb.

Sample

Sampling was purposive to include participants diagnosed with any form of Rb at any stage of childhood, receiving any type of treatment. Information power was used to guide the sample size.[18] Data quality and participant diversity were monitored during data collection to determine the final sample size.

Patient and public involvement

Patient and public involvement (PPI) involvement was reported according to GRIPP2 guidance on effectively involving experts by experience in health research[19] and conducted according to National Institute for Health Research guidelines.[20] Continuous involvement of CHECT members maintained the personal experience perspective throughout, including identifying the need for the study, piloting measures and commenting on initial findings.

Recruitment

Recruitment commenced on 9 June 2022 and ceased on 15 January 2023 and used online posters circulated via social media, as well as physical posters in hospital clinic waiting rooms.

Interested individuals contacted researchers via email and if eligible (see figure 1) were invited to take part. All participants completed brief questionnaires about their demographic and clinical characteristics (see online supplemental materials). Data collection techniques are discussed separately, for each method.

Data collection

Written consent was obtained from all participants, including for the use of direct quotations. Topic guides were prepared and used to structure interviews and focus groups (box 1) and were informed by the PPI group associated with the study (described in box 1). Using a semistructured approach, questions were open-ended to allow participants to discuss topics that felt meaningful to them. Interviews and focus groups were audio recorded, transcribed verbatim, anonymised and rechecked.

Teenage focus groups

Focus groups were conducted online via video call, between August 2022 and January 2023. Each was facilitated by the lead researcher (NO'D) and a second researcher (BP) to ensure consistency and reduce bias. Alongside conversational content, group dynamics and reflexive notes were kept in a field diary (NO'D).

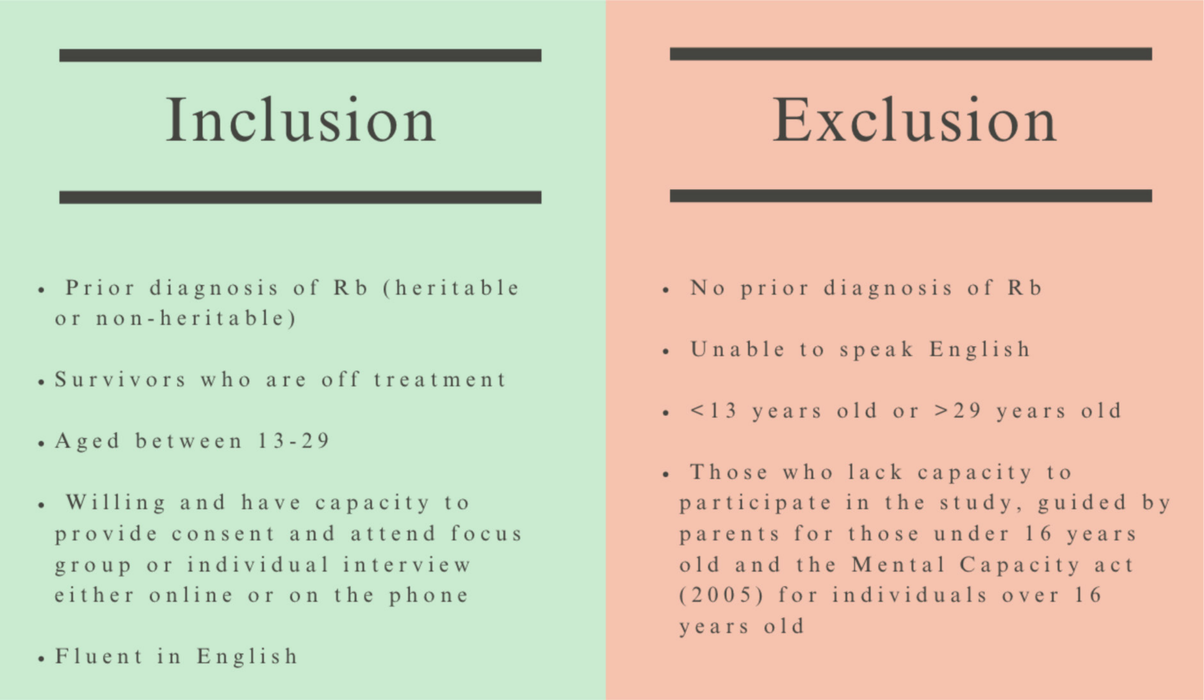
Young adult interviews

One-to-one, in-depth, semi-structured interviews were conducted by video call, between June 2022 and December 2022 (NO'D). To enable choice and maximise accessibility, participants were offered the opportunity to undertake these interviews over the phone. In this event, all participants opted for video call interviews.

All participants were debriefed at the end and contacted 2 days later to check-in on well-being and provide signposting to support services if required. Participants received a £20 retail voucher in recognition of their participation.

Analysis

Data were analysed using reflexive thematic analysis (RTA)[21] and coded in NVivo.[22] To become familiar with the data, NO'D read all transcripts and DH read 10%.



Inclusion

- Prior diagnosis of Rb (heritable or non-heritable)
- Survivors who are off treatment
- Aged between 13-29
- Willing and have capacity to provide consent and attend focus group or individual interview either online or on the phone
- Fluent in English

Exclusion

- No prior diagnosis of Rb
- Unable to speak English
- <13 years old or >29 years old
- Those who lack capacity to participate in the study, guided by parents for those under 16 years old and the Mental Capacity act (2005) for individuals over 16 years old

Figure 1 Inclusion and exclusion criteria. Rb, retinoblastoma.

Preliminary notes were made and quotations were collaborated across scripts to form initial codes. Through discussion (NO'D and DH), new codes were developed iteratively, eventually becoming themes and subthemes. Additional researchers (see the 'Acknowledgements' section) independently reviewed two transcripts each, before the team came together to discuss, develop, and refine codes, themes, and subthemes.

RESULTS

Overall, 32 individuals took part. Four focus groups were conducted with 15 teenagers, each lasting 2 hours, and 17 young adults were interviewed, for between 22 and 65 min (mean=43.3). Demographic details of participants are shown in table 1 (see online supplemental materials).

Box 1 Topic guide for interview and focus group discussion

- ⇒ Would you mind telling me a little bit about yourself and your experience with Rb?
- ⇒ What do you understand about Rb?
- ⇒ Can you tell me what you remember about your treatment?
- ⇒ Do you, or have you ever, have any worries about your mental health?
- ⇒ Has Rb impacted how you feel about yourself? (Is this the same as when you were a teenager? If no, how is this different?)
- ⇒ Has Rb impacted how you live your life? (Is this the same as when you were a teenager? If no, how is this different?)
- ⇒ Has there ever been a time when you felt you would like some support for how your mental health, or how you are thinking or feeling?
- ⇒ If yes, did you receive any? (Particularly during adolescence) Can you tell me about that? Was it helpful?
- ⇒ If you have not accessed any psychological support, but think it would have been helpful, can you tell me what you think this should look like?

Summary of themes

Using RTA,[21] three key themes were identified from data analysis, and eight subthemes, as stated in figure 2.

Themes and subthemes are discussed in detail below, with verbatim quotations reported by participant number (P) and self-reported Rb type (B=bilateral, U=unilateral, H=heritable, NH=non-heritable, eg, P1:U-NH). Occasionally, the self-reported Rb type could not be biologically correct (ie, some people with bilateral disease said this was not heritable Rb, which is not possible), and such instances are marked with an asterix, for example, P2:B-NH*.

Theme 1: childhood: 'the legacy of trauma' (P27:B-H)

A key issue noted within most participant narratives is that of experiencing lifelong trauma while living beyond Rb. Variations in this legacy appear to be influenced by several factors, including age at diagnosis, the genetic nature of the diagnosis, wider family history, the severity of visual and facial changes, and late effects of treatment. The theme also encapsulates 'family experiences and survivor guilt', which considers the role of parents and siblings in the trauma experience. It also deliberates 'memories from treatment', and how these link to the individuals' emotional response and understanding of the world and others around them. Lastly, the combination of these trauma experiences and 'the long-lasting impact on personality' are considered.

Table 1 Demographic characteristics of participants

Participants N (%)		Adolescents (13–19)	Young adult (20–29)
Total	32 (100)	15 (47)	17 (53)
Type of retinoblastoma			
Heritable	10 (31)	5 (33)	5 (29)
Non-heritable	21 (66)	10 (66)	11 (65)
Unknown	1 (3)	0 (0)	1 (6)
Eye(s) impacted			
Unilateral	23 (72)	10 (66)	13 (76)
Bilateral	9 (28)	5 (33)	4 (24)
Median age in years (range)	20 (13–29)	15 (13–19)	25 (20–29)
Age at diagnosis			
<6 months	8 (25)	3 (20)	5 (29.4)
7–11 months	5 (15.6)	2 (13.3)	3 (17.6)
1 year to 1 year 11 months	5 (15.6)	2 (13.3)	3 (17.6)
2 years to 2 years 11 months	8 (25)	5 (33.3)	3 (17.6)
3 years to 3 years 11 months	4 (12.5)	1 (6.6)	3 (17.6)
4 years to 4 years 11 months	1 (3.1)	1 (6.6)	0 (0)
>5 years	1 (3.1)	1 (6.6)	0 (0)
Treating hospital			
Birmingham Children’s	13 (41)	8 (53.3)	5 (29.4)
Royal London/Great Ormond Street	19 (59)	7 (46.6)	12 (70.6)
Treatment (in isolation or combination)			
Enucleation	24 (75)	10 (66.6)	14 (82.3)
Cryotherapy	7 (22)	4 (26.6)	3 (17.6)
Systemic chemotherapy	17 (53)	6 (40)	11 (64.7)
Intra-arterial chemotherapy	6 (19)	1 (6.6)	5 (29.4)
Laser therapy	5 (16)	4 (26.6)	1 (5.8)
Radioactive plaque	7 (22)	0 (0)	7 (41.2)
Gender			
Male	12 (38)	5 (33)	7 (41)
Female	20 (63)	10 (67)	10 (59)
Religion			
None	19 (59.4)	11 (73.3)	8 (47)
Christian	10 (31.2)	2 (13.3)	8 (47)
Other (unspecified)	3 (9.4)	2 (13.3)	1 (6)
Ethnicity			
White British	24 (75)	11 (73.3)	13 (76.5)
White Irish	1 (3)	0 (0)	1 (6)
Other white	2 (6)	1 (6.6)	1 (6)
Indian	1 (3)	1 (6.6)	0 (0)
Mixed/multiple	3 (9)	1 (6.6)	2 (11.8)
Hispanic	1 (3)	0 (0)	1 (6)
Relationship status*			
Single	22 (69)	15 (100)	7 (41.2)
In a relationship	4 (13)	0 (0)	4 (23.5)

Continued

Table 1 Continued

Participants N (%)		Adolescents (13–19)	Young adult (20–29)
Cohabiting	6 (19)	0 (0)	6 (35.3)
Highest level of educational			
Pre-GCSE	9 (28.1)	9 (60)	0 (0)
GCSE	4 (12.5)	4 (26.6)	0 (0)
BTEC	2 (6)	0 (0)	2 (11.8)
A-level or equivalent	3 (16)	2 (13.3)	1 (6.6)
Degree or equivalent	14 (43.75)	0 (0)	14 (82.3)
Employment status*			
Student	18 (56)	14 (93.3)	4 (23.5)
Employed full time	13 (41)	1 (6.6)	12 (70.6)
Unemployed	1 (3)	0 (0)	1 (6)

*At time of study.
A-level, advance level; BTEC, Business and Technology Education Council; GCSE, General Certificate of Secondary Education.

Family experiences and survivor guilt

The concept that Rb *'started with you' (P17:U-H)* was often brought up, in the context of survivor guilt: *"I started this gene. So, my mother and father didn't have it. My brother doesn't have it…I'm the 'lucky' one who started with this gene." (P17:U-H)*

Many acknowledged empathy for their parents and the difficult experiences they endured at this time, and the decisions they had had to make:

> my parents told me at a certain age and described the pressure there had been … the stress that had been there. My mum had told me that she'd kind of gone through this really, really kind of distressing time (P23:U-H)

The wider impact was feeling unable to talk to parents about their diagnosis and feelings as teenagers and young adults. *"My Mum doesn't really like reliving it.… She said that the few weeks between me getting diagnosed and my eye being removed, it's just a blur for her.… she doesn't really remember any of it, 'cos she was just going through the motions and stuff, so I don't really ask her about it." (P19:U-NH)*

Memories from treatment

Due to the age of Rb diagnosis, many participants spoke about others' expectations, and sometimes the reality of not having fully formed memories of diagnosis and treatment themselves. This fed into the narrative of guilt and difficulties discussing what had happened with parents, because of perception that their parents '*had it worse' (P24:U-NH)* as they had adult recollection of events.

> [parents'] memory is vivid and it is that specific that they can remember every blood transfusion, every

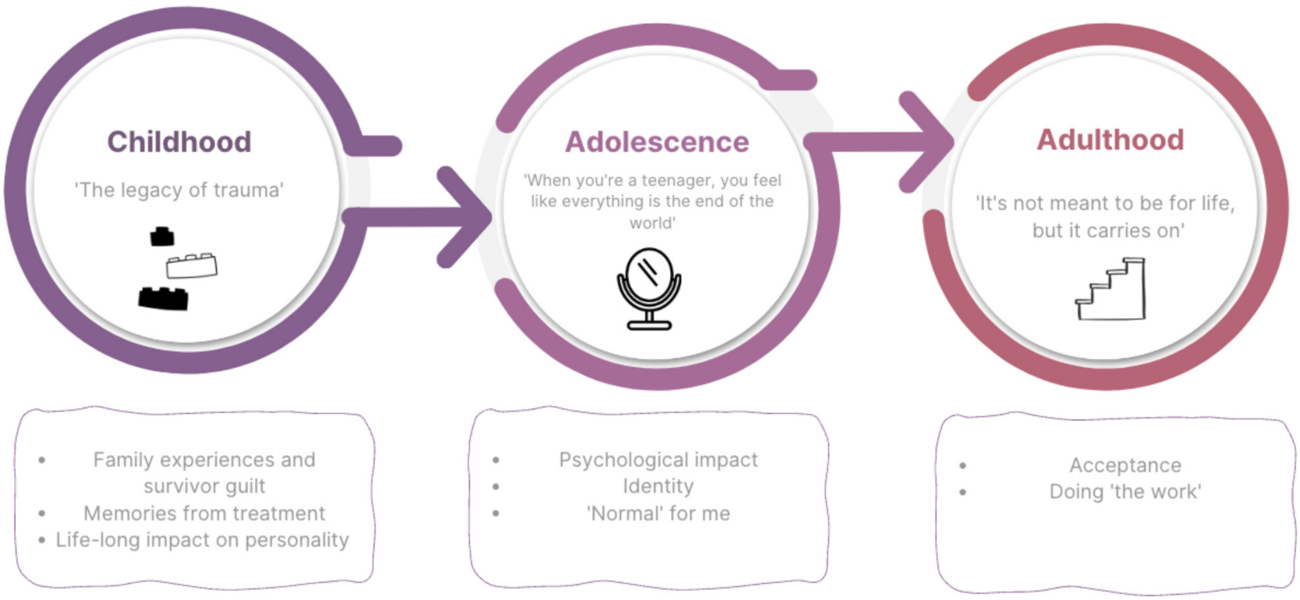


Figure 2 Diagrammatic overview of themes and subthemes.

> chemo…everything that went wrong, what day it was, what month it was, where they were at the time. So, I always just felt very guilty speaking about how I felt with them, because in my opinion, they'd had it worse than I had. (P24:U-NH)

Various memories about treatment were discussed, countering the myth that these are forgotten when this occurs at a young age. These ranged from distressing and traumatic to neutral or even fond recollections, particularly of the '*fun' (P25:U-NH*) and '*kindness' (P13:U-H*) clinical staff instilled into inherently traumatic procedures, such as enucleation. One individual remembered particularly distinctively behaviour post-treatment, when people got close to her face.

> I kind of remember kicking and screaming at the doctors… I just remember absolutely throwing a fit because of it. (P5:U-NH)

Although many participants received treatment while very young; key memories could be vividly recalled during adolescence and young adulthood. For some, these remained dormant until triggered, often by external sensory experiences, primarily focused on distinctive smells, which could elicit strong emotional reactions. This was often combined with attending routine health appointments, as may be expected.

> certain smells set me off as well, like when you go to the dentist and you've got that anaesthetist kind of smell (P24:U-NH)

For others, this was more shocking, with memories seeming to come from nowhere, with some describing feeling *'overwhelming sadness' (P7:U-H*) reminiscent of clinical trauma responses.

Long-lasting impact on personality

Many felt that their experiences of Rb had shaped them as a person. *"If I didn't have Rb when I was young, I would have been a totally different person." (P17:U-H*)

For some, this focused on feeling '*special' (P23:U-H*), prominent during younger childhood, before this tapered off as they got older. For a few, this positive sense of self lasted throughout their lifetime, often due to consistent reassurance from their parents about being themselves:

> My mum just drilled it into me that, Do you know what, you're unique, just go along with it (P5:U-NH)

The idea that it is easier to '*be me' (P18:U-U**) when young, while it is '*worse later on' (P25:U-NH*) reflects typical childhood experiences and the turbulence of adolescence, including transition to secondary school, hormonal changes, complex social dynamics, seeking independence and a focus on appearance.

> up until the age of year six, I was fine, completely fine. In fact, I used to talk about it [Rb] all the time and bring it [prosthetic] into school—you know, bring my spare eyes into school and sort of show off a bit about it…it gets worse later on (P25:U-NH)

Despite the best efforts of parents to instil a sense of pride and individuality, for some the '*realisation' (P18:U-U**) that you are not '*like everyone else*' (*P18:U-U**) still leads to a sense of insecurity, suggesting that the onus of support should not be on families, but on a wider network.

Some thought that trauma had '*made me stronger' (P23:U-H*), suggesting that personality can be positively shaped from negative formative experiences. This was sometimes linked to ambition and a desire to achieve, with goal setting that may not otherwise have been aspired to.

> If I didn't have Rb when I was young… I would have different goals and aspirations in life. (P17:U-H)

Theme 2: adolescence: 'when you're a teenager, you feel like everything is the end of the world' (P28:U-NH)

Adolescence was widely considered the most difficult period and when support skills are required, with survivors of Rb managing late effects alongside the usual challenges faced by peers: *"I feel really lonely and I feel really weird" (P32:B-NH**)

Psychological, social and behavioural impact

Adolescence was considered the most difficult life stage across participants, due to them often being in '*sustained periods of stress mode' (P23:U-H),* managing all the '*normal*' challenges of adolescence, alongside being a childhood cancer survivor. Many felt this was when they were most aware of how they looked, and experienced their peers as more critical, thus adding to the sense of feeling '*abnormal' (P16:U-NH*).

For some, these feelings led to changed behaviour, with many overcompensating for their eye(s) by trying to improve other areas of their image.

> I think there was a time where I was more worried about my appearance and whatnot and I was trying to compensate by getting in the gym and making myself look better (P18:U-U*)

Ironically, adolescence was said to be the time when help was most needed, but also most difficult to acquire. This appeared particularly problematic for males, who seemed more isolated, perhaps reflecting wider cultural norms and expectations *"(I) just grit my teeth together and get through it" (P29:U-NH),* and stigma for expressing emotions, which may be perceived as weak:

> I feel like there's definitely a stigma attached to men nowadays, because there's like this whole thing of "Oh, you've got to be a man, you've got to be tough…a strong, tough, alpha male, I guess." (P29:U-NH).

For a few this led to maladaptive coping strategies, such as drug use and joining cult internet forums. This did not differ by heritable and non-heritable Rb status, with every male involved in this research describing it as the first time they had felt able to speak about the impact of Rb on how they think and feel.

> I do think I could have got help if I actually asked for it. I think my problem was that I probably just suffered a bit in silence. (P28:U-NH)

Developing a culture of open discussion around mental health may therefore be particularly crucial for pre-adolescent boys.

> I found a lot of… relatability, online. And there were all these other angry young men who I thought, "Oh, these guys are the same as me," which they weren't… people who've had bad experiences, when they feel inadequate, they'll eventually fall into this like circle or this group of less than pleasant people online… I was sort of like teetering on the edge of it. (P29:U-NH)

As these individuals reached older adolescence, they often behaved contrary to expected norms, with many taking more responsibility for living a healthy lifestyle compared with their peers. Such behaviour change and self-care can be positive but needs to be balanced to prevent the anxiety and constant body surveillance described by some.

> One of the hangover effects of Rb is that I felt this sense of responsibility to look after myself, so I always felt quite strongly against things like smoking or excessive alcohol consumption, starting about 17 [years old] I suppose (P23:U-NH)

High levels of anxiety were prominent across the sample, beginning in adolescence and often extending into adulthood. These were commonly linked to three areas: sex and relationships, having your own children and developing second cancers.

> It was really panic inducing… I just thought I look ugly… you know when you're growing up, you're reading like Roald Dahl and watching pirate films, and it's all kind of this really ugly thing [one eye] that is attributed to people that are weird or mean, and so I think I didn't yet have the kind of separation from that. (P25:U-NH)

> I am less inclined to have casual flings, to be quite honest, because there's always the potential for that being an unfair burden for anybody to carry, basically, unless you're in a committed relationship (P27:B-H)

> The side effects of like, catching cancer again is quite scary (P12:U-NH)

Identity

For many young adults looking back to adolescence, there was a recognition that they had numerous questions about themselves, and how Rb impacted their sense of self.

> it's just a time where you don't feel, you don't even know who you are, you're trying to figure out, you know, what kind of clothes am I supposed to wear, what's my style, to what do I want to do. Like, who am I? (P32:B-NH*)

Many kept coming back to the idea that life was unfair, in terms of their Rb diagnosis and experience of childhood cancer, but also the impact this had had on their perceptions of self, others and the world around them.

> I always did ask why did it happen—this is a very rare disease, like why did it have to happen to me. I kind of still ask that now, like why me (P29:U-NH)

Dealing with identity as a teenager was complicated by the nature of Rb, with many doubting the legitimacy of their diagnosis, particularly after hearing it described as a '*good cancer*' due to its high survival rates. This was particularly prominent among people not treated with enucleation, who had no visible '*signs*' of anything having been '*wrong*', leading to them making downward comparisons to others.

> it would almost be a bit of a, I guess a joke that I'd say, "Oh, you know, I struggled as well," sitting around with all these people that aren't as fortunate as I was. (P24:U-NH)

Such comparisons were also noted as barriers to accessing support, particularly from generic cancer support groups with children who had cancers that were perceived '*worse*' than Rb.

> It was kind of upsetting to me to be around other kids that also had cancer and had it worse than I did. And I think I was quite aware of that even when I was younger, that there were people that had it worse, and I felt kind of sad about that, so I didn't engage with that [support] as much. (P25:U-NH)

'Normal' for me

As Rb largely affects very young children, many individuals could not remember a life any different to what they knew now. This was the same across individuals with and without prosthetics, as well as those whose treatment had resulted in facial changes.

> It's not really a big part of my life, to be honest, 'cos I was so young when I lost it [eye], I don't think about it much on a daily basis. It's just something that I kind of exist with (P25:U-NH)

It was often other people and their comments that reminded people of their differences; behaviour that was experienced throughout childhood into young adulthood, from peers, friends and strangers.

> I came home one day and I was like, "Oh, what's a Cyclops?" And they [parents] were like, "Oh, it's a mythical creature with one eye." And I was like, "Oh, that's what this boy calls me at school." (P30:U-NH)

Some individuals choose to embrace and make a feature of their difference through wearing a coloured or uniquely designed prosthetic, which could fuel hateful comments and encourage self-blame.

> I still get horrible comments … people tend to go, "Oh well, you're asking for it by wearing something like a coloured prosthetic" (P19:U-NH)

Theme 3: adulthood: 'it's not meant to be for life, but it carries on' (P5:U-NH)

Adulthood was both a time for reflection and projection, dependent on the life stage of the participant at the time of interview.

Acceptance

Many young adults developed the ability to accept themselves and their identity, which involved acknowledging their experience of Rb, without making it the only thing about them.

> Don't compare yourself to anybody… You need to make your reality your possibility… figure out, write it down on a piece of paper what are you able to do… I started to accept, not appreciate, not be grateful, because I'm not grateful for what I've been through, I'm grateful for the strength that I developed to get out of it (P32:B-NH*)

Acknowledging that the impact of Rb does not go away seemed to come with time, with one individual clearly highlighting the potential lifelong impact of their childhood cancer.

> It's not meant to be life but it does carry on throughout your years (P5:U-NH)

The ability to validate experience and provide self-reassurance was common, and for some this involved efforts to '*make friends*' with their Rb and accept how it had shaped their identity and life, rather than fighting it and denying its' existence.

Doing 'the work'

'The work' (P32:B-NH)* is an overarching term used by a participant and developed within this study to refer to the different coping strategies used by participants. A key feature of this was the need to seek accurate information, which was prominent for developing personal understanding, and for preventing childhood beliefs and anxieties being carried into adulthood.

> I'd internalised that, that there's these tumours that could suddenly…go off like a volcano (P27:B-H)

Contrasting with the diagnostic and treatment periods when it was parents who were given information, participants focused on the need for '*information for me, not my parents' (P4:U-H)*. Many however, said it could be hard to know what information was needed, and who to ask for for advice:

> it's small stuff like that that concerns me personally. Like basically, to sum it up in a word, how can I live my life without relying on other people to take my hand and—and gently guide my way through it, you know. (P1:U-NH)

It was apparent across the sample that a sense of being proactive and having someone independent to talk to was important for preventing later difficulties.

> I didn't really understand it growing up. I think you need to be told early, then it prepares for the future if that makes sense, rather than just seek out the information when like you get curious or you need it. (P8:U-U*)

Many young adult participants stated that '*the work*' included accessing and integrating therapy into their lives, which was said to bring substantial benefits, and may be useful in earlier life alongside peer support.

> It sort of feels like there could have been a few things nipped in the bud just by talking… psychological support in the sense of therapy, talking about my experience, talking about how I view the world; talking about how the impact of it on my family has impacted me. (P31:U-NH)

Others expressed a desire to have someone to talk to who was independent of doctors and parents but had not accessed this.

> Maybe having someone to talk to growing up—'cos, you know, you have down days and you have up days, and if you're having a down day, maybe just talking to someone who isn't your parents might be helpful, who really, really gets it. (P31:U-H)

This was summarised by one adult who stated you '*just need someone to steer you in the right direction' (P23:U-H)*.

DISCUSSION

This study explored experiences of living beyond Rb, with a focus on childhood (family experiences and survivor guilt, memories from treatment, lifelong impact on personality), adolescence (psychological impact, identity, 'normal' for me') and adulthood (acceptance and doing 'the work').

For most, Rb was said to cause long-term distress, heightened feelings of anxiety and lowered self-esteem. Despite this, many found that this experience enabled them to develop resilience and enhanced coping strategies that may not otherwise have developed. We identified several themes and subthemes that were common across Rb types, providing a unique contribution to the growing evidence on psychosocial experiences among young survivors of Rb.

A key theme identified in this study was the lifelong trauma impacting the survivor and their family following Rb diagnosis. This is common among childhood cancer generally[23–26] and is well understood in more frequently diagnosed cancer types. The experiences of family members, particularly parents, is better understood than that of affected individuals.[27–29] This may be because most Rb diagnoses are made in early childhood, so it is parents or carers who take on information and make key decisions. Many participants also shared complex thoughts about 'putting' their parents through difficult experiences, reporting emotional difficulties, stress and anxiety as they adapted to the diagnosis and authorised medics to treat their child, which in some cases involved enucleation and use of a prosthetic eye.[27 29] Many participants acknowledged empathy for their parents and the treatment decisions they had to make, as well as the wider impact on their own behaviour, and their inability to talk about how they feel. This finding is reflected in wider childhood cancer research[30] and emphasises the need for young people to access information and support independently from parents and family members, and at an age and developmentally appropriate time.

'Memories from treatment' was a key finding and was linked to the individuals' emotional response and understanding of the world and others around them, as indicated by the verbatim quotations. Although reported in previous childhood cancer survivor literature,[31 32] this has not been noted in Rb before or cancers affecting very young children, who may be preverbal, yet still have sensory, salient recollections of this period. This finding can be described by the AIP model, which states that early life trauma can have lifelong impact.[33 34] For individuals who experience cancer as teenagers or young adults, it has been noted that cancer diagnosed earlier in life disrupts key developmental stages and shapes cognitive processes.[35] This fits with broader research on trauma experiences and the impact this can have on personality development throughout the lifespan, both in terms of post-traumatic growth and influencing positive coping mechanisms like resilience, and more negatively with risks for later mental health difficulties.[36–39]

Correlating with existing childhood cancer survivorship literature, some participants thought that their traumatic experiences had 'made them stronger'. Such positivity is common with respect to cancer diagnoses and negative formative experiences,[40–42] which has been termed 'post-traumatic growth'.[43 44] This can be particularly relevant to children diagnosed at a very young age, who often adapt well to their situation, as they do not recall ever living differently; a position shared by many in the current study.

Adolescence was widely considered the most difficult phase, with a particular need for support and coping mechanisms. We highlighted the psychological impact of Rb at this time, including feelings of unfairness and powerlessness in the face of restrictions (perceived and actual), and demonstrating how survivors from a rare cancer may struggle to identify others with whom they can relate, express themselves and share anxieties and difficulties. Previous literature has referred to this as 'negotiating the psychosocial challenges from two different worlds—'the healthy' and 'the ill'.[45]

The ability to share feelings and be understood is vital for self-esteem, which if absent may result in loneliness. This contradicts work suggesting survivors of Rb report lower levels of depressive, anxious and somatic symptoms than non-Rb childhood cancer survivors.[14] However, these findings may not reflect survivors' true feelings, but instead, reluctance to disclose complex emotional responses and distress through fear of judgement. Existing research[4] has found that adult Rb survivors have 20% more psychological difficulties, such as anxiety and depression, than a healthy comparison group. These findings are echoed by Morse *et al*,[46] who found that although survivors of Rb report typical emotional health and quality of life, they are more anxious than a comparison group of peers who have not experienced cancer.

Regarding the concept of Rb being a 'good cancer', this sentiment has been shared by other survivors who are told they should be 'grateful' as they have a 'good' cancer that they are unlikely to die from or may live 'with' for many years.[47–50] These studies highlight how some cancer experiences can be downplayed, potentially leading to downward comparisons that create a barrier to help-seeking and emotional expression, meaning individuals do not feel they are 'genuine' survivors or 'entitled' to support.

Uncertainty was expressed about many aspects of adult life, notably sex and relationships, having children and developing a second cancer. Such feelings are well documented in cancer survivors of all ages, often leading to over-appraisal of the likelihood of something 'bad' happening again.[51–55] This can be amplified when access to information and social support is limited, and ambiguous healthcare systems are encountered.[56] As may be expected of adolescence, identity was a key theme, with individuals having many questions about themselves, and how Rb had impacted their sense of self. Rb can differ to other childhood cancers as it can leave a physical reminder, commonly via enucleation and use of a prosthetic eye. Existing literature, however, mainly focuses on temporary appearance changes during treatment, for example, loss of hair from chemotherapy,[57 58] or weight gain from steroids[59–61]; or permanent changes that can be hidden, for example, implant surgery or prothesis use in breast cancer survivors.[62 63] Other work addresses permanent non-cancer-related illnesses or accidents, for example, burns,[64 65] cleft palate,[66 67] scarring[68–70]; or inherited conditions like neurofibromatosis,[71–73]which may have different psychosocial impacts to cancer. All these conditions can have a deeply distressing psychosocial impact,[58 74 75] although Rb, particularly when heritable[15] or treated with enucleation[76] is distinct, and must be supported as such.

Adulthood was a time for reflection and projection, dependent on life stage at the time of interview, with Rb acceptance universally considered unachievable while still young. 'Doing the work' encapsulates the need to answer questions, and the role of strategies such as psychotherapy and peer support to improve well-being. In comparison to adolescence, many young adults had developed the ability to accept themselves and their identity, acknowledging the experience of Rb without making it the only thing about them. This corroborates wider cancer survivorship literature, which emphasises the psychosocial challenges unique to adolescent cancer survivors who must manage 'normal' life while working through questions about identity,[77] interpersonal relationships,[78] education,[79] and cancer-specific anxieties.[80 81] These appear to ease as individuals transition to young adulthood, particularly if receiving psychological support with a specific acceptance component.[82–89] Concurring with the current study, these findings highlight how structured psychological therapy may be useful earlier in life alongside peer support, another factor considered useful to survivors of Rb.

Strengths and limitations

To our knowledge, this is one of the first qualitative studies seeking to understand the psychosocial experiences of survivors of Rb, regardless of their genetic status or the unilateral or bilateral impact. It presents a novel exploration of the interplay between Rb, self-perception, interpersonal dynamics, and wider societal and healthcare contexts. Furthermore, it provides insight into the psychological complexity of rare childhood cancer survivorship during the transition from adolescence to young adulthood and beyond. Despite the rarity of Rb, a relatively large sample was recruited, mostly via CHECT's social media platforms, reflecting a desire to 'give back' to the charity and community; this may, however, have risked self-selection bias. Interviews were said to offer a cathartic experience from which participants could view and process their journey. As is increasingly common in qualitative health research,[90 91] we experienced a problem with 'bot' or fraud participants, disrupting our recruitment timescale.[92] All recruitment material was designed for people from 13 years old to promote inclusivity, with video information for visually impaired candidates, or those who preferred to hear/see the research team discuss the study aims and processes. The methodology used, RTA, is a flexible approach which allowed the opportunity to combine different methods of data collection through conducting both interviews and focus groups. At the outset of this research we did not intend to analyse the data as one, but it made sense to do so due to the heterogeneity of themes across adolescent and young adult participants. As an iterative process, this approach allowed us to continually revisit and refine the themes over a long period, holding discussions with the wider research team to delve into the nuances of participant meaning.

Implications for practice and policy

We have clearly demonstrated the profound psychological and emotional impact of Rb on survivors and highlighted the need for integrated, specific psychosocial support, delivered within long-term follow-up care. Regarding policy, our findings align with the National Health Service Long-Term Plan, which aims to provide patients with more support options, particularly for psychological needs and preventative care.[93] It also fits with healthcare changes post-COVID-19, whereby non-hospital-based follow-up in community settings, with digital and remote support options is emphasised.

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Ethics approval This study involves human participants and data governance and ethical guidelines were followed, with approvals from the Health Research Authority (HRA) on 27 May 2022 (Research Ethics Committee reference: 22/NI/0082). Participants gave informed consent to participate in the study before taking part.

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