

Title:

Experiences of living with cancer for adolescents and young adults and their families:
a narrative review and synthesis

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ABSTRACT

Introduction: Adolescence is a critical life stage marked by significant physical, psychological, and social change. Cancer diagnosis during adolescence profoundly alters this experience for adolescents and young adults (AYA) and their families with impacts that continue throughout life. It is important to understand these experiences to ensure delivery of appropriate and high quality supportive care. This narrative review critically appraised and synthesised qualitative literature that explored the experiences of AYAs and their family living with cancer.

Methods: Narrative review and synthesis of qualitative research of AYAs' and their family's experiences of cancer. MEDLINE, CINAHL and PsycINFO were searched between February 2000 and September 2019 using search terms including "adolescent", "young people", "young adult", "cancer", "family", and "qualitative". Literature was appraised and synthesised using Popay et al.'s (1) framework.

Results: 3,016 articles were retrieved (Medline n=1298, CINAHL n=1632, PsycINFO n=86). Of these, 151 duplicates were removed. 2,865 papers were screened with 121 abstracts considered for eligibility for inclusion. 18 papers met the inclusion criteria. Three inter-related themes were identified: (1) being diagnosed with cancer; (2) uncertainty - holding on to life; (3) gaps in care delivery.

Discussion and Recommendations: Few studies discuss the impact of cancer on the families of AYA living with cancer. Future research should explore this experience. By doing so the relational impact of cancer will be better understood as the basis of supportive family-centred care.

PROSPERO Registration: CRD42017084148

Keywords: adolescent, young adult, cancer, family, qualitative.

INTRODUCTION

Adolescence is a life stage marked by rapid and significant physical change (2, 3), along with development in cognitive, psychological and social behaviours (4). It can be a time of physical, emotional and social turmoil, and of growing self-awareness (5). Understanding this evolving sense of awareness and individuality is essential if health and social care professionals are to provide care in appropriate and flexible ways that recognises specific needs (6). For adolescents and young adults (AYA), the family is an important part of these care relationships, especially as they may still be living at home (5,6). It is therefore vital that understandings of AYAs' experiences of cancer include the way in which a cancer diagnosis impacts individuals within the family, such as parents/carers or significant others, and the relationships between family members. Evidence around the interrelationships and interconnections between AYAs and their families is currently limited within cancer research. An earlier study by Taylor et al. (7) highlighted the findings of a meta-synthesis of the literature from 1988 to 2010 and proposed a conceptual model of teenager and young adult experiences of cancer. Interestingly 'social support' as a mediator is included in the model but has not included the family as being important within the AYA cancer journey, or the dynamic interrelationships between AYAs, their families and caregivers.

The aim of this narrative review was to synthesise existing literature about the experiences of AYAs living with cancer and their family. The review question was: "What are the experiences of living with cancer for AYAs and their family?"

METHODS

We conducted a narrative review and synthesis of qualitative research papers. Narrative review is a systematic and established approach to gather and synthesise qualitative evidence to inform practice (8).

Search Strategy

Literature published between February 2000 and September 2019 was searched using three databases: MEDLINE, Cumulative Index of Nursing & Allied Health Literature (CINAHL), and, as the review question was not solely related to nursing, PsycINFO. As AYAs with cancer are cared for by several health and social care professionals it is essential that three different healthcare databases were included to capture the various disciplines involved.

There is a wide variation in categorisation and definitions used to describe AYAs, stemming from wider debate around the use and appropriateness of the term adolescent in comparison to others, such as teenagers, young people, young adults (9) and complexity around the extension of adolescence in developmental terms into young adulthood (10, 11, 12). However, in this review the term 'adolescent and young adult' (AYA) was applied broadly to capture international variation in terminology and is in line with the World Health Organisation (WHO) definition of young people as those aged between 12 and 39 years (9).

The search strategy used the following main terms to capture the largest number of relevant articles:

- disease ('neoplasm'; 'cancer');
- age ('adolescent'; 'teenager'; 'young adult');
- relationships ('family');
- study design ('qualitative'); and
- focus ('experience').

Medical Subject Heading (MeSH) terms were used as appropriate. Boolean operators (e.g., 'AND' and 'OR') and wildcards (e.g., *) were used to ensure maximum inclusion. In addition to database searches, experts who have written extensively within the field of AYA cancer care (e.g., Professor Faith Gibson and Dr Rachel Taylor, from the UK, and Professor Zack Zebrack from the USA) were consulted to identify additional papers that may have been missed. The following inclusion/exclusion criteria were applied.

Inclusion

- Peer reviewed primary research studies
- Teenager and/or young adult, or wildcard variations of other terms such as adolescent, young people, adolescent and young adult (AYA) or teenager and young adult (TYA) in the title
- Family

- Age range 12 to 39 years
- Papers written in English
- Published between February 2000 to September 2019.

Exclusion

- This review did not include children aged under 12 years or young adults over 39 years
- Non-malignant disease
- Young people with severe cognitive impairment
- Survived cancer > 5 years
- Quantitative studies, editorials, policy, dissertations, commentaries.

Quality assessment

Qualitative studies were assessed using the guidance framework provided by Popay et al. (1). This provides a structure for narrative reviews and increased transparency and rigour of the process. While there may be risks with over interpretation of study data, the framework supported the transparency and reproducibility of narrative review.

Rigour

Guba & Lincoln's (13) framework was used to assess the quality criteria for each of the individual papers identified in the literature review to ensure their trustworthiness. These criteria were credibility, transferability, dependability and confirmability. To ensure the credibility of the review, the researchers evaluated available frameworks best suited to a narrative review. The credibility of each of the papers was dependent on independent analysis, ethical approval, and whether the findings were clear. In order to ensure transferability, constant checking was completed through a rigorous review of the papers. The transferability was completed by ensuring that the findings were current and that the reported results were trustworthy and credible. Dependability and confirmability were continuous, and this was carried out through a constant update of the literature and being reflexive at all stages of the process (14).

Findings

Figure 1 shows the PRISMA flowchart for the review. 3,016 articles were retrieved (MEDLINE n=1298, CINAHL n=1632, PsycINFO n=86). Of these, 151 duplicates were removed. 2,865 papers were screened with 121 articles assessed for eligibility and 18 papers were included and synthesised in the review.

[Insert Figure 1 PRISMA here]

Study characteristics

An overview of the study characteristics is shown in Table 1. The review included four studies from Canada, three from the USA and UK, two from Sweden, Finland, and Jordan, and one from Taiwan and New Zealand. A range of qualitative designs were applied, four used phenomenology with two specifically using Interpretive Phenomenological Analysis (IPA). Two used survey design with one using a cross-sectional survey design, two longitudinal exploratory design, one ethnography, and one narrative and one descriptive. Two papers identified no design with three qualitative exploratory methods and one action research. Only the ethnography paper included the family in their data collection, although three mentioned the family as being important within the data analysis. The age range of the participants varied, but the majority were in the 12 to 20 age range.

[Insert Table 1]

Synthesis of findings

Using an inductive approach, thematic synthesis was carried out to allow for a systematic approach to the organisation, analysis and synthesis of the literature. The narrative framework by Popay et al (1) supported the synthesis and organisation of the information extracted from each of the papers. From each of the papers, the aim was to construct meaning from the narrative and illuminate understanding. First, one of the authors (WMcl) read each paper in its entirety, taking notes and key messages from each one. Second, once each paper had been fully critiqued, coding was used to highlight key emergent themes. Third, the key themes and sub-themes were mapped and tabulated to support synthesis. These were discussed with the co-authors (CGB, ZC, RGK).

Themes reported in the identified studies are presented in Table 2. Following synthesis, three overarching themes and seven sub-themes were identified. This paper reports these themes supported by the relevant literature and includes illustrative quotations, from the study participants.

[Insert Table 2]

Theme 1 represents the beginning of the journey, being diagnosed with cancer. Theme 2 denotes the uncertainty following the diagnosis and holding on to life. Theme 3 highlights the gaps in the care delivery and identifies parts of the cancer journey that were missing, for example, 'specialist services'. Themes and subthemes are presented as a conceptual visualisation in Figure 2 and discussed in turn.

[insert Figure 2].

Being diagnosed with cancer

While cancer is relatively uncommon in adolescents compared to adults, AYAs reported that receiving a cancer diagnosis was the most challenging part of their cancer journey (15, 16). This experience was compounded by the need to wait for an appointment or start their treatment. A delay in diagnosis from the first symptoms was evident in two of the studies (16). Delay was considered just as crucial to the cancer journey experience as the impact of the diagnosis. Gibson et al's (16) narrative study involved 24 AYAs between the ages of 16 to 24 in England who told their stories about physical pain and alterations to their bodies. These young people did not connect these symptoms to anything being wrong, nor did they see the need to visit their General Practitioner (GP). For example, as one young person commented: "*I thought I had just twisted [my leg] at a party*" (16, p.2587). However, this study reported that the experience of cancer can be worsened where there is diagnostic delay by the young person, their family or healthcare professional, which was similar to the other studies identified in this theme (16).

Five studies highlighted that AYAs who are experiencing a diagnosis of cancer need their family and friends to support them through the cancer journey (17, 18, 19, 20,

21). Within these studies there was an emphasis on maintaining normal family routines, while recognising the impact that cancer had on family and peer relationships. Some relied on close family and friends to help them feel safe. One participant reported, for example, the importance of *'being there to hold my hand'* (21, p127) and another *"the only reason I have come through this far is because of them [family and friends]"* (21,125).

Uncertainty – holding on to life

The experience of cancer can often occur at a time when AYAs are in the process of developing their early adult life plans, transitioning from being a child to teenager and from teenager to young adult (22, 23, 24). Seven studies discussed how a cancer diagnosis can bring uncertainty and was often associated with hope, fear, and loss of self (25, 26, 17, 18, 27, 28, 29). During this period, AYAs often found themselves trying to hold on to the life they had imagined. AYAs experienced fear of the unknown which was mainly due to feeling afraid of having cancer and not being able to cope (25, 26,18). Nevertheless, AYA were not prepared for the treatment or the side effects. Some began to realise that they may even die sooner than they had previously expected and this preoccupied their thoughts. As one young participant expressed *"the idea of death became dominant in my life, cancer is a killer"* (25, p 4). These feelings were all-consuming and the fear of being alone and uncertainty about their future and became the main focus of their experience.

Eight studies highlighted how AYAs experience a sense of loss of self as their bodies dramatically changed from the chemotherapy, radiotherapy and surgery (25, 26, 17, 18, 20, 21, 29, 30). Within the cancer experience, they often lost sight of themselves, as physically their bodies had changed and they no longer recognised the person they had become. As one participant explained, *"I am a girl, and I like my hair, without my hair, my friends won't see me"* (26; p37, 38).

Gaps in the care delivery

The delivery of high quality evidence-based care, by healthcare professionals who are cancer specialists, was a fundamental requirement of the cancer experience (29,30). Ten studies described the need for information and support, leading up to and

following a diagnosis of cancer (31, 22, 16, 32,17,18, 19, 33, 29, 30). For example, participants reported that healthcare professionals' attitudes and the way they communicated were often inappropriate. Young people commented on how the Consultant Oncologists were quite stern – *“it's like talking to a piece of cement”* (19, p1756) – and did not address them directly *“I wish people talked to me rather than my parents”* (31, p725).

Studies also reported the tendency to view the ideal treatment and care experience as arising out of specialist services that are age-appropriate (34, 35). This specialist care was not, however, always readily available for all AYAs with cancer in the reviewed studies due to different healthcare systems and resources. As highlighted in one study, by a participant who was 20 years old, *“there are only old peopleso why was I there?”* and one participant who was 16 years old *“now we shall do some painting, and only the small children came out of the room, the teenagers stayed in their rooms”* (33, p578). This study also found that these specific needs of having an age-appropriate environment and psychosocial support varied over time and throughout the cancer journey and were unique to each person. It was clear from Olsson's (33) study and other reviewed studies (31, 22, 16, 32,17,18, 19, 29, 30) that these needs were not being met. AYAs wanted the hospital environment to be different and equipped for them and to be cared for within an area where there were people of their own age.

DISCUSSION AND RECOMMENDATIONS

Our review found 18 qualitative research papers focusing on AYAs' experience of cancer published between February 2000 and September 2019. We found that there is limited research that involved both AYAs with cancer and their families; only one of the 18 papers included the family (18). This is a significant omission and underscores the need for further research focussed on the relational experience of cancer within families. This is important for three reasons linked to each of the three overarching themes identified.

First, our review found that the experience of receiving a cancer diagnosis was an important marker of how AYAs experienced cancer from that point onwards (15,16, 17, 18, 19, 20, 21). Yet, a key finding of this review was that the word 'cancer' was

rarely used within the healthcare setting at diagnosis, and diagnosis itself is frequently delayed. It was unclear from the reviewed literature why the word 'cancer' was not used by healthcare professionals. Indeed, not using this terminology could contribute to AYAs perceiving that their diagnosis was delayed because it was only later in the clinical pathway that the term cancer was used. Delays in diagnosis can be detrimental to the overall disease process and early intervention is essential (16) as delay can have adverse impacts on AYA experiences and outcomes (16, 21). Our review suggests that improvements are required to the timing and content of conversations about cancer diagnosis. Specifically, when, where, by whom, and how a cancer diagnosis is shared with AYAs and their families requires further investigation, especially to explore family members' involvement in these conversations as well as their own experiences of delayed diagnosis.

Second, our review highlighted that AYAs experience of living with cancer is characterised by uncertainty about their future. However, alongside this uncertainty, there was hope for recovery and life in the future. The fear of having cancer, and the reality that death may be pending did feature, but quickly subsided as AYAs moved along the cancer journey. In addition, AYAs often felt loss of self as they faced losing their hair or identify from receiving the cancer treatment (25, 26, 17,18, 27, 28, 29). The complex ways in which emotions, such as fear and hope, emerge and intertwine after cancer diagnosis is an area that requires further attention. Specifically, the entanglements of emotion experienced by family members and how these amplify or attenuate AYAs own emotional responses to cancer should be explored. This could ensure that professionals support the uncertainty AYAs and their families experience together.

Third, the appropriateness of care environments was a key aspect of AYAs' experiences of cancer. Age-appropriate settings and specialist services were often viewed as ideal, but not always available in specific contexts. Identifying service preferences and gaps with AYAs and their families is an important priority for future research and practice development (31, 22, 16, 32,17,18, 19, 33, 29, 30). Specifically, the impact that different care environments have on family involvement in care and availability of psychosocial support for both AYAs and their family are areas that require attention.

CONCLUSIONS

Limited evidence exists that explores the experiences of AYAs living with cancer that includes the experiences of their families. This omission potentially inhibits optimal service design and psychosocial support for both AYAs and families following cancer diagnosis. Increasing understanding of AYAs' and their families' experiences of communication during diagnosis, the emotional impacts of living with cancer, and the appropriateness of care environments are key to enable practice improvement.

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doi:10.1371/journal.pmed1000097