



Facing the unknown: uncertain fertility in young adult survivors of childhood cancer

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Abstract

Purpose The purpose of this research was to describe the challenges young adult childhood cancer survivors (ACCS) experience of living with an unknown fertility status.

Methods In this qualitative study, we analyzed data from in-depth interviews with 25 ACCS (15 females, 10 males) using content-driven inductive thematic analysis and constant comparative techniques.

Results Three themes provide insight into ACCS' experiences of living with an uncertain fertility status. (1) The marked psychological burden, which included fear, worry, anxiety, and sadness, was ubiquitous. (2) Romantic relationships were negatively affected and entailed fear of disappointing one's partner, the difficulty of discussing fertility issues, and shying away from romantic relationships. (3) Communication challenges with healthcare providers were apparent across the cancer trajectory, made worse by provider knowledge gaps and dismissal of fertility concerns. (4) Well-known gender stereotypes about pregnancy and childrearing were replicated, while the emotional and life impacts that uncertain fertility could exert on males were minimized.

Conclusion This study provides beginning insights into the experiences of young ACCS as they navigate an unknown fertility status and highlights existing gaps in fertility-related healthcare services.

Implications for Cancer Survivors Uncertain fertility has implications for survivors' wellbeing that remain poorly addressed by the existing structure of cancer survivor and fertility services. Assessing and addressing the fertility-related needs of ACCS will continue to be a vital component of patient-centered care in the future, and research to develop comprehensive support, resources, and processes of care are vital to meet this priority need.

Keywords Childhood cancer · Cancer survivorship · Oncology · Late effects · Fertility · Infertility · Qualitative

Introduction

As a result of intensive, combined multimodal therapy, over 80% of children diagnosed with cancer now survive into adulthood [1]. Yet, adult childhood cancer survivors (ACCS) are at risk for multiple late effects (conditions that continue to develop 5 or more years after cancer treatment),

including impaired gonadal function and infertility [2–4]. It is well established that infertility, the inability to achieve a successful pregnancy after a 12-month period of appropriately timed, unprotected intercourse [5], can result from various cancer treatments. Patients who received alkylating agents, cisplatin, and other heavy metal-containing chemotherapy are at risk. Radiation therapy to the hypothalamus/pituitary axis, ovaries, or testes has the potential to significantly increase this risk [6, 7]. In females, ovarian failure or fewer ovarian follicles can result in infertility or premature menopause; in males, spermatogenesis can be damaged both temporarily and permanently, resulting in reduced sperm production and infertility [8, 9]. In a research by Barton and colleagues, survivors were shown to have had an increased risk (relative risk 1.48 [95% CI 1.23–1.78]; $p < 0.0001$) of clinical infertility when compared with their siblings, which was most pronounced during early reproductive ages [6]. In another study, fertility problems were self-reported by survivors four times more often than matched healthy controls [10]. Though

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rates largely depend on the type of treatment, roughly 11–26% of female and 42–66% of male childhood cancer survivors face infertility later in life [11, 12].

Many childhood cancer survivors have expressed a strong desire to have children in the future and are hopeful about the possibility [13–15]. Yet, infertility concerns are common and relate to the ambiguity of risk; the health of future biological children; and for women, concerns about their own health during pregnancy [14–18]. Furthermore, psychosocial distress, sexual dysfunction, relationship challenges, and negative effects on feelings of femininity and masculinity have been documented in the context of infertility risk among childhood cancer survivors [14, 16, 18, 19]. Infertility is perhaps one of the most devastating for ACCS as they approach child-bearing age and form romantic relationships. Despite knowledge of the etiology of treatment-related infertility and growing awareness of the risks among both healthcare providers and ACCS, infertility is not always easily anticipated [20]. Survivors have been reported to both underestimate and overestimate their infertility risk [21]. Uncertainty about fertility status has also been documented, reflecting perhaps what survivors recall (or have forgotten) from conversations with parents and/or healthcare providers at the time of treatment and a lack of fertility-related information or education across the cancer trajectory [13, 15, 17, 22, 23]. Uncertain fertility might further reflect the real ambiguity of risk. Reliance on treatment-related risk factors is not straightforward because different responses to therapy yield variability in survivors' fertility [21]. As such, treatment-indicated risk does not translate directly into adult fertility status [21]. During their adolescent and young adult years, many survivors do not undergo fertility evaluations, because either investigations are not made available or they choose not to pursue such evaluations. Moreover, possibility of pregnancy still remains when infertility is considered likely on the basis of cancer treatments. For example, in the study by Barton, nearly two-thirds of survivors with clinical infertility reported a pregnancy [6]. Further complicating the matter, there is no absolute way to test for future fertility potential in either ACCS or the general population. Ovarian reserve tests only provide indirect estimates and do not predict natural fertility potential in regularly ovulating women [24], and likewise, semen analysis cannot absolutely predict sterility in males [25].

Prior research suggested that discussions about infertility and fertility testing should be tailored to individual survivors and their needs, but there is still much to be learned what these needs are [26]. Considering that many ACCS live in a state of uncertainty regarding their fertility status, often during the formative developmental and relational life stage of young adulthood, understanding survivors' experiences is vital to developing patient-centered resources, supports, and healthcare services. Effective means of counseling survivors about their family planning are vitally important to mitigate

the challenges arising from ongoing uncertain fertility. As such, the purpose of this study was to explore ACCS' experiences and challenges of living with an uncertain fertility status.

Methods

In this qualitative thematic analysis study, we drew on interview data from a previous narrative inquiry study that examined ACCS narratives of health management [27], wherein 15 of 30 participants spontaneously discussed in length their unknown fertility-related experiences and challenges. Recognizing this as a prominent experience for ACCS, we then collected 10 additional interviews to extend our initial analysis specific to uncertain fertility. We also considered the ways in which gender and sex were reflected in the participants' accounts. Both the initial study and the additional interviews to extend the analysis were granted ethics approval through the joint University of British Columbia and BC Cancer Research Ethics Board.

Setting and study participants

This research was conducted in the province of British Columbia, Canada, where a public healthcare system provides universal healthcare inclusive of long-term follow-up for cancer survivors. Fertility services, however, are generally not covered through public healthcare, even for cancer survivors. Using convenience sampling, we recruited ACCS for a larger study examining ACCS narratives of health management, through existing follow-up clinics at both the children's and adult cancer centers, as well as online via specific forums and websites. Study fliers were placed in clinics and also distributed to potential participants by clinicians. We interviewed all ACCS who contacted the research team and then used purposive sampling to recruit participants with diverse characteristics. For the additional interviews, purposive sampling through the follow-up clinic at the adult cancer center was particularly useful in identifying individuals whose fertility status was unknown and inviting these individuals to participate in research specifically about their uncertain fertility. With the recruitment of 10 participants in addition to the initial 15, we were confident that we obtained sufficiently high information power [28], owing to the rather specific aim of the study, participants holding characteristics highly specific to the study, and the quality of the communication during the interviews.

Individuals aged 19–36 years who had survived a childhood cancer were included in this study. Further inclusion criteria mandated that each participant be a resident of BC, have had no cancer treatments within the 2 years prior to the study interview, and understand and communicate in English.

Uncertain fertility as determined by their physician (KG) when they provided individuals with the invitation to participate in the study was an inclusion criteria for the additional participants. This one research team member (KG) was involved in data analysis discussions once data was anonymized but did not have access to the individual demographic, medical, or interview data. Research team members involved in confirming eligibility, enrolling individuals into the study, obtaining informed consent, and conducting data collection, had no prior relationship with study participants. Informed consent was obtained from all individual participants included in the study.

The mean age of participants at the time of interview was 29 years (range 23–36 years), with 10 being males and 14 females. The mean age at time of initial cancer diagnosis was 9 years (range < 1–19 years), with the majority ($n = 17$) diagnosed with a type of leukemia or lymphoma and treated with chemotherapy ($n = 25$) and radiation therapy ($n = 17$), and the most commonly reported late effects being bone, joint, or soft tissue related ($n = 10$), and anxiety and/or depression ($n = 8$). See Table 1 for further self-reported demographic information, disease characteristics, and late effects.

Data collection

One investigator (AFH—an experienced qualitative researcher) conducted the interviews with the initial 15 participants, and a second investigator (KN—a registered nurse and graduate student in nursing and public health, trained in qualitative interviewing) conducted the subsequent 10 interviews. Twenty interviews were conducted in person, with 15 in the participants' home, 5 in a private room of a public library, and 5 conducted over the telephone, all lasting 45 to 120 min. For the initial 15 participants, we used an interview guide to query the medical and psychosocial challenges experienced by survivors and the ways in which they managed their health, and based on the preliminary findings specific to uncertain fertility, we developed an interview guide to explore this further in the additional interviews. See [Electronic Supplementary Material](#) for both of the interview guides. Although we used interview guides, we framed the interview for participants as a conversation and encouraged discussion, rather than questions and answers, as well as the sharing of details about what was most important and relevant to the participant. In both phases of data collection, we refined and adjusted our interview questions as we proceeded as a means of exploring important and emergent findings, that is, as analysis of earlier interviews proceeded, our interview questions became more specific to fill in gaps and explore in greater detail important ideas that arose in preceding interviews. We digitally recorded and transcribed all interviews, which we checked for accuracy and then removed identifying information and labeled with an identifier code. We wrote field notes following each interview

to document interactions with the participant, social and contextual factors that might have influenced the interview, and any important or emerging insights.

Data analysis

We analyzed the ACCS interview data and field notes using inductive, thematic analysis and constant comparative techniques. Two investigators (AFH and KN) read the initial 15 interview and field note transcripts numerous times and together developed a preliminary coding frame through discussion of the important concepts, experiences, and interpretations specific to fertility, which was then applied to the data set using the qualitative data management software program NVivo™, version 10. One of the broad themes surfacing at this phase was of uncertain fertility with the preliminary analysis informing the development of the interview guide for the additional 10 interviews. Once the subsequent 10 interviews were added to the data set, we revisited the preliminary coding frame, revised as per inductively derived concepts and ideas in the more recent data, and compared and contrasted the emerging concepts as well as participant experiences. This revised coding frame was then discussed with the larger research team and applied to all 25 interviews. Throughout analysis, one team member (KN) performed the coding and another (AFH), who had worked extensively with the data set from the initial 15 participants, reviewed the coding in NVivo™ and all of the coding output, with ongoing discussions providing interpretive insights. We then grouped and regrouped the data into a meaningful interpretation until we were confident that we captured the predominant ideas and perspectives evident in the 25 participant interviews. Recognizing sex- and gender-related influences on health, illness management, health-seeking behaviors, and supportive care needs [29, 30], we also identified and analyzed the sex- and gender-related narratives shared by study participants.

Results

Three themes provide insight into ACCS' experiences of living with an uncertain fertility status: (1) ongoing psychological burden, (2) the influence on intimate relationships, and (3) communication challenges with healthcare providers. A fourth theme depicts sex and gender commentaries related to uncertain fertility.

The ongoing psychological burden of living with uncertain fertility

The emotional challenge of living with an uncertain fertility status was a ubiquitous commentary throughout the interviews. The majority of women and men participants described

Table 1 Participant self-reported demographic information, disease characteristics, and late effects

		All <i>n</i> = 25
Demographic characteristics		
Age	20–24	4
	25–29	6
	30–34	10
	35+	5
Gender	Male	10
	Female	15
Place of residency	Greater Vancouver area	19
	Other	6
Marital status	Single	15
	Married	10
Living arrangement	Alone	5
	With roommates	1
	With a partner/spouse	12
	With parents	5
Level of education	Did not complete high school	
	Completed high school	1
	Completed or enrolled in university/college	24
Employment status	Unemployed	2
	Student	3
	Employed part- or full-time	20
Disease characteristics		
Age at first diagnosis	0–4	7
	5–9	6
	10+	12
Type of cancer	Leukemia and lymphoma	17
	Brain tumor	2
	Sarcoma (not including brain)	5
	Other solid tumors	1
Treatments	Radiation therapy	17
	Chemotherapy	25
	Surgery	9
	Bone marrow transplant	3
Late effects and health problems		
Bone, joint, or soft tissue late effects		10
Anxiety or depression		8
Learning difficulties or cognitive impairment		7
Endocrine late effects		6
Second cancer		5
Impaired growth and development		5
Respiratory late effects		5
Hearing impairment		5
Dental late effects		4
Digestive late effects		4
Visual impairment		3
Cardiovascular late effects		3

living with fear, worry, anxiety, and sadness due to the uncertainty they felt regarding their future ability to conceive. These participants feared not being able to have their own biological children, undergoing pregnancy complications, having a child whose health would be negatively affected, and disappointing their partner. For example, a 26-year-old male leukemia survivor stated: “It’s been kind of scary because, I mean, I really want to have kids at some point. So it’s kind of a scary thought knowing that maybe I couldn’t.” Likewise, a 35-year-old female leukemia survivor shared how, “It does worry me. I hope that I can have a healthy baby or even have a baby at all. I worry a lot lately because my biological clock is ticking.” Many participants also described struggling to cope with anxiety and the various ways this anxiety became apparent, including for example, interfering with their sleep or appetite, occupying their thoughts for extended periods or when trying to focus on work, and distracting them from everyday activities. Although some participants were able to manage their anxiety through strategies such as talking with their family members, others reported that their anxiety became increasingly troublesome over time. A 23-year-old female non-Hodgkin lymphoma survivor attributed the rollercoaster of emotions and severe anxiety she experienced to ignoring the possibility of her infertility:

It’s been really hard not knowing [about fertility status]. I think when I first found out [about the risk of infertility] it was just horrible. And I didn’t really deal with it, and I kind of just swept it under the rug, and then I started getting anxiety. Honestly the worst part of cancer I think for me was the recovery... this anxiety and fear, all that stuff that comes after is almost worse... it’s like such a process – such a grieving process.

Along with feelings of fear and anxiety associated with uncertain fertility status, many ACCS conveyed a sense of sadness in anticipation that they might never have children of their own. This anticipatory sadness was exemplified by a 26-year-old male leukemia survivor:

It’s sad in a way if I couldn’t [sire a child]. Also, I don’t know yet like it’s something down the road when I’m ready to have children that I’d want to find out then. Because I know if I find out now I’d be I guess kind of upset about it. So that’s the scariest and saddest part of it.

The participants commonly described how anticipatory sadness compelled their decision to wait until they were ready to have children before seeking fertility testing. For example, a 27-year-old neuroblastoma survivor described experiencing anticipatory sadness while also acknowledging the possibility that she might still be able to conceive:

“It’s hard because I mean you’re crying and upset but then you don’t even really know you are infertile. So you’re in limbo kind of. I hate being upset and being like I can never give you kids. But then I’m like well it could still happen I don’t know.”

The constant “nagging” state of uncertainty resulted in many of the participants describing that they felt caught between conflicting emotions, afraid or reticent to imagine a happy future with children.

Despite the stress of not knowing their fertility status, there were participants who indicated that they preferred to live with uncertainty rather than undergo testing and receive confirmation that they were infertile, as exemplified by a 22-year-old female leukemia survivor: “It’s like I want to know, but I don’t want to know. I kind of like being in this like ‘oh it might still happen’ rather than doing it again and knowing for sure it won’t happen, but it is frustrating not knowing.” In this way, the uncertainty enabled some to maintain hope. The majority of the participants, however, would have preferred to know their fertility status.

The influence of uncertain fertility on intimate relationships

All of the study participants indicated that living with an unknown fertility status negatively impacted their romantic relationships in some capacity. They described the challenges of taking someone else’s needs into account, the fear of disappointing their partner, the difficulty of discussing fertility issues with their loved one, and the potential for their unknown fertility status to lead them to shy away from romantic relationships. A 30-year-old male leukemia survivor elaborated on how his unknown fertility status impacted his relationship:

I mean it, it’s definitely worrisome. It’s, it can linger, you know, somebody who wants to have a family and have kids – it can be a serious hindrance on, I guess, emotional aspects of your relationship with your partner. I mean it’s not something that I would express right away or even have that conversation especially when it’s so unknown.

The above participant hinted at the fact that because of the hindrance his possible infertility imposed on his relationship, he was hesitant to disclose his unknown fertility status early in his relationship. Further depicting these challenges, a 26-year-old female sarcoma survivor described how being in a committed relationship forced her to confront fertility issues, because she felt compelled to consider her partner’s needs:

Well now that I’m in a relationship I think that’s the hardest part. Because it’s not only just about what I

wanted for my life, it’s like what he wants too, and then for fertility it influences your partner and your potential future children that, you know, that you could have – so it’s bigger — it’s a different thing to cope with.

The fear of letting one’s partner down, and the hesitation to disclose one’s unknown fertility status, was apparent in multiple interviews. A few participants elaborated on how stressful it was to tell their partner about their possible infertility, wherein they feared disappointing their partner or engendering conflicts in their relationship.

Other young ACCS shied away from certain partners and potential long-term relationships because of their possible infertility and the difficulty they anticipated in broaching fertility-related discussions. A 35-year-old male sarcoma survivor described how he purposely gravitated toward partners who did not want children. For other survivors, the fear of possible infertility prevented them from engaging in any romantic relationship whatsoever. For example, a 30-year-old brain tumor survivor shared that she had never been in a long-term romantic relationship, attributing her hesitation to her possible infertility. Since her teenage years, she had anticipated challenges in discussing fertility with a future partner, not knowing “how to approach it,” which led her to question whether she should ever have a boyfriend.

Interestingly, one participant, a 32-year-old male sarcoma survivor, described how discussing infertility with his romantic partners could have a positive influence on his relationships because opening up to a partner about fertility facilitated deeper intimacy between them:

Because there’s a thing that’s different about me it, it gave me something pretty serious and intimate to talk with partners about early on in a relationship. So often, before I’d ever have sex with any partner, I’d have this conversation with them that I had cancer when I was a kid and there’s, you know, a few things different about me because of that. So for that – on a few occasions, I think, was, was a nice opener to having serious discussions.

Regardless of how well the young ACCS coped with their experiences, living with an unknown fertility status substantially impacted their romantic relationships.

Communication challenges with healthcare providers about fertility

In the narratives shared by the ACCS in this study, the fertility-related communication challenges with healthcare providers included limited discussions, information, and education across the cancer trajectory, made worse by provider knowledge gaps and dismissal of fertility concerns.

Throughout many interviews, the ACCS were hesitant and vague in describing with whom they discussed their fertility; overall, fertility-related communication was minimal.

Most participants in this study reported that they had received limited education regarding their fertility status during the initial cancer diagnosis and the cancer treatment phase, as well as throughout the follow-up period of survivorship. This lack of fertility education was attributed to their young age at cancer onset, failure or reluctance of healthcare providers to elaborate on concrete details around fertility issues, and insufficient knowledge possessed by some providers. Participants acknowledged that being a child at the time of cancer diagnosis, treatment, or oncology follow-up consultation contributed to the lack of education about their risk for infertility. But participants were frustrated that fertility-related discussions had been directed to their parents, even though they were present. For example, a 22-year-old male leukemia survivor described how the only fertility information he had ever received was not actually directed at him:

I think I was like twelve and my doctor, my oncologist, mentioned to my parents, oh yeah, fertility, oh yeah, probably can't have kids or whatnot. I actually didn't know. I was kind of like, wait, I can't have, you know, I can't do that, what? And then he kind of explained it to me. And I kind of wish it was different, you know, because it wasn't really directed at me. It was like oh, he's too young to kind of understand.

When we asked this participant what he knew about his fertility status now, and what he thought his risks could be, he responded: "I don't know much to be honest. They [healthcare providers] don't really talk about it, honestly, they don't really talk about it that much."

Limited fertility-related education was also common among those treated in their teenage years and those seeking information as young adults. A 31-year-old male leukemia survivor commented on the brevity of such exchanges with healthcare providers:

You know, when I'm like nineteen or twenty and asking like, oh what's the like deal with my fertility? And sometimes I'd kind of get like, oh well, are you married? And I'd say no, they'd say, oh, don't worry about it. They're like, that's something to worry about later.

There were many descriptions of such encounters, which indicated that healthcare providers rarely discussed fertility with teenage patients and were prone to avoid fertility discussions throughout the cancer trajectory. By failing to prioritize fertility topics for their young patients, and postponing conversations until a later date, participants felt that their needs were marginalized by healthcare providers. Over time, the

ACCS began to feel as though their thoughts, feelings, and information needs related to potential infertility were insignificant and they were hesitant to broach the topic.

Participants also perceived healthcare providers as ill-versed on the topic of infertility after cancer; there were few proactive discussions, leaving participants with numerous unanswered questions regarding their individual fertility prognosis. Several participants commented on inadequate interactions with their primary care provider in particular. A 35-year-old female leukemia survivor explained:

I would not be near as comfortable and happy if I'd only had my GP because, yes, he was aware of everything and he was with me since I was eight. However, I was the one who went to him, I was the one who would ask these questions, I, I did that. So, if I only had him I wouldn't, I don't think I would have had any of the questions answered as far as fertility. He would have been one to say, oh no don't worry about it.

Several participants explained that they did not trust their family doctor for health issues related to their history of cancer, including their infertility risk, and, as such, felt that it was their responsibility to initiate these conversations. Yet, they were also hesitant to raise the topic themselves because they felt self-conscious and awkward discussing their fertility. These conversations were particularly challenging for the queer participant in the study in part because "hetero assumptions" predominated and queer identities were not acknowledged. A number of female participants further explained that their healthcare providers had not informed them that they would enter menopause at an early age and therefore should not delay decisions about fertility testing and having children. Poor fertility communication with healthcare providers was not confined to family physicians, however. A 22-year-old male leukemia survivor commented that because his oncologist did not seem well educated about his infertility risk, their conversation felt awkward:

There needs to be more information for sure. Like my doctors, they didn't bring it up. And it was still – like even with the oncologist it seemed like kind of an awkward topic a little bit, you know. And they didn't really know much about it. They kind of – it wasn't really their field.

Even when it was their field, communication with healthcare providers was perceived as insensitive at times. A 23-year-old female brain tumor survivor described how her newly appointed endocrinologist "blurted it [patient infertility] right out" as though the unknown outcome was an "absolute."

Many participants recounted how their educational and informational needs regarding fertility were not met by healthcare providers until they were referred to a specialty clinic. Our findings suggest that participants' perceived lack of fertility-related education was related to the limited knowledge and inability of practitioners to elaborate on the details of fertility resources, fertility testing, and current research on the prognosis and expected outcomes for childhood cancer survivors. Participants alluded to this perception in various ways. A 23-year-old female Hodgkin lymphoma survivor described an experience at the time of diagnosis in which fertility was mentioned briefly as a bullet point on a pamphlet, but further education on the topic was not offered. When asked to elaborate on this experience, she highlighted her lack of clarity and understanding about the physiological impact of cancer treatments on her reproductive organs:

The only conversation I can actually remember is in my long-term follow-up and that's because you're given this thirty page questionnaire. And a lot of them are like, are you concerned about fertility? So I always mark the box yes, I have that concern. And it's always so brief. It's like, okay, so you've had this drug, it's a small dose so you're probably fine – how regular are your periods? I'm like, okay, yeah, they're mostly regular – it's fine, but then it's sort of dismissed. There's no like feelings about it – there's no like what does it mean that I'm infertile? Is that to do with my ovulation? Is that to do with like the actual structure of my uterus like fallopian tubes or ovaries? There's a lot of things that can go wrong, what exactly has this drug done to my body which could potentially make me infertile. I mean I understand why you may not be able to get into that but at the same time I think it's important to know.

A number of participants suggested that not only specific details regarding the physiology and biology of possible infertility were neglected but also the psychological impact was ignored. The participants commonly noted that the psychosocial impact of learning about their fertility was a significant need.

Readiness to have children was a factor for many participants in deciding when to seek information about fertility. Participants provided vague responses that incorporated the idea that waiting until an older age may be preferable when seeking information about their fertility, yet they lacked a clear idea as to when this "later time" might be. It is unclear whether these comments reflected their true preferences or if they were repeating what they had been told by healthcare providers.

Sex and gender commentaries on fertility

Overall, the study participants replicated well-known gender stereotypes about females, pregnancy, and childrearing.

Compared with males, female bodies were considered more complicated and a woman's ability to bear a child was considered more important, with infertility described as a greater psychological blow to women. Both men and women in this study minimized the emotional and life impacts that uncertain fertility could exert on men. They suggested that the anatomy and physiology of the female reproductive system made fertility testing and related treatments more difficult and more invasive than for males. Fertility testing for women was assumed to involve multiple blood tests and ultrasounds, whereas fertility testing for men simply required a sperm sample. Fertility testing interrupted young women's birth control practices and hormone therapy treatment. A 26-year-old female brain tumor survivor stated:

That's a big difference too is that I have to think about birth control, not only because I'm in a relationship, but also because I need the hormones because women's bodies are more – a lot more complicated right? Like if I don't have those hormones then I start going into menopause and then my bones will not be the same and I'll get hot flashes. But a guy who is infertile probably doesn't have to deal with that stuff, like they have a test and they find out. For me it's not easy, it's like I have to sacrifice a lot just to have these tests done.

A 30-year-old male leukemia survivor also touched on the differences between male and female reproductive roles, and how missing the opportunity to go through pregnancy might have more of a psychological impact on a woman, whereas he believed he might find it easier to adopt a child. When asked how he thought men and women might be impacted differently, this participant responded:

I would shy away about taking a guess on how it would affect a female, but I would imagine it would be harder, just because of the, you know, with women having babies it's more, I guess, emotionally different for them to have children so it might affect them psychologically more. One thing I've always thought is that if I can't have children then even though it's obviously different, I mean I would adopt. I guess that might be a little bit harder to do because the females actually carry the baby, and it might be different psychologically that way.

There was an assumption by numerous participants that because females physically carry the child from fertilization to birth, they are also psychologically affected to a greater degree by infertility issues than males. An exception to this sentiment was expressed by a queer participant who commented that, "Just because you have a uterus doesn't mean necessarily that you want to be a mom, you might actually want to be a dad and what does that mean?" This

participant went on to acknowledge the pervasive social expectations for traditional roles, “I don’t know how much room there is for that. There’s a lot of cultural things – its just a challenging issue.”

The participants’ broader ideas about gender roles and gender identity influenced how they perceived men and women to experience potential infertility. Many of these notions referenced broader societal values with long-standing gender stereotypes. One such notion, expressed by multiple participants, invoked the idea that when women miss out on childbearing, they also miss out on the opportunity to engage in an important social bonding experience with other childbearing women. A 30-year-old female leukemia survivor expressed the view that childrearing and parenting were a more significant experience for women:

I’m thirty and my friends are mostly around my age, you’re getting to the point where people are talking about having babies and whatnot. And then sometimes you get friends that don’t know about your situation say something about me being a woman and having a baby. And you’re like, that’s kind of in my face, but they didn’t know... Because for a woman, they are the child bearer – I think it’s probably harder on a woman than a man.

This participant conflated the physical act of pregnancy and childbirth with the emotional life experience of becoming a parent and raising a child. Men were also compelled to uphold the traditional gendered perspective that having a child was integral to a women’s self-worth. A 36-year-old sarcoma survivor described how he assumed it would be far less difficult for an infertile man to simply procure a sperm donor and carry on:

If I was a woman and there were questions about my fertility I think I would be a lot more anxious. And I think that’s because in our society there’s still much more duty placed on the woman to be able to provide a child. So like as a man if I, if I have a partner and we want to have kids a really viable option is to just get a sperm donor. And my partner still gets to go through pregnancy and have a child. But that’s a lot – that’s not the same at all if the gender role is reversed. So I think there would be more of a feeling of inferiority for a woman.

In addition to the presumption of shared social experiences associated with pregnancy for women, the participants also perceived women to share a more intimate bond with a child than do men. Therefore, they inferred that the potential emotional toll of not bearing one’s own child was far greater for women. A 22-year-old leukemia survivor stated: “I think it

might be harder on females just because, I don’t know, I think there’s more intimacy with the child and whatnot. I think it’s hard on both sexes but I think it probably would be a little bit harder on a woman.”

When men communicated their own personal experience without comparing genders, they were more apt to share the emotional significance of fertility in their lives. A 35-year-old leukemia survivor who discovered he was infertile after getting married exclaimed how relieved and happy he was to have banked sperm before his childhood cancer treatment. He and his wife accessed the banked sperm to conceive their own children. His narrative provides insight into the importance of fertility for men:

That’s a big one because obviously I’m impacted by that greatly. And I have thought about that, I thought that how freaking lucky I am that I could put some guys on ice in [year] and, seventeen, eighteen years later go back and get them and now my boy is... whereas if I’d been a woman and had the same treatment and had the same consequences the outcome would be very different in terms of a family.

This participant was exuberant that he had been able to have his own children, but herein again, he felt compelled to conflate emotional and physical aspects of infertility, deferring to the female reproductive system and the inability for women to so easily bank eggs. Interestingly, the emotional loss a man might feel due to the inability to have his own children was barely acknowledged by the women and men in this study. One 34-year-old female leukemia survivor mentioned how her miscarriage impacted her husband saying “we got pregnant and we initially had a miscarriage, which was really hard on him and he wasn’t sure if we should try again.” Her narrative portrayed herself as more resilient and able to tolerate the medical interventions, one of the few diverse gender and fertility narratives we encountered.

Discussion

In this qualitative study, a marked psychological burden that included fear, worry, anxiety, and sadness accompanied ACCS’ experiences of living with an uncertain fertility status. Romantic relationships were negatively affected. Participants described the challenges of taking someone else’s needs into account, the fear of disappointing their partner, the difficulty of discussing fertility issues with their loved one, and the potential for their unknown fertility status to lead them to shy away from romantic relationships. The ACCS also described that fertility-related communication challenges with healthcare providers included limited discussions, information, and education across the cancer trajectory, made worse

by provider knowledge gaps and dismissal of fertility concerns. Lastly, the study participants replicated well-known gender stereotypes about pregnancy and childrearing, while minimizing the emotional and life impact that uncertain fertility could exert on males.

This research is consistent with existing literature documenting the worry, fear, and anxiety associated with the possibility of not being able to have one's own biological children, intimate relationship challenges, and anticipating pregnancy complications [13, 14, 18]. One of the nuanced but novel findings in our study was the describing of psychological distress to be ongoing and even increasing for some as they approached childbearing years, which interfered with daily activities at times. This is likely one of the many factors contributing to the anxiety and distress reported among some childhood cancer survivors that in our study was rarely accounted for clinically.

Our examination of sex- and gender-related narratives offers novel evidence of the ways in which gender norms and expectations are potentially harmful. When asked to comment on how the experience of men and women cancer survivors compare, both men and women participants portrayed men as stoic, with a tendency to downplay the emotional impact of uncertain infertility, and yet the psychological burden was apparent in men's accounts of their experiences. These socially preferred stories reinforce gender stereotypes, particularly hegemonic masculinity wherein men are expected to exhibit a lack of emotions [31]. In their examination of the impact of infertility on hegemonic masculinity, Burton [32] argues that infertility poses a direct threat to the image of the ideal man such that infertile men are viewed as deficient and experience a high level of social stigma. In interviews, men's worry, fear, and anxiety, as well as the preference to not disclose their infertility risk to an intimate partner, are perhaps also reflections of these threats to socially preferred versions of masculinity. The downplaying of the psychological burden of uncertain fertility, coupled with the additional masculine ideal of not seeking help [31], places men at particular risk for poor mental health supports.

Our research complements the evidence describing the ways in which uncertain fertility is seen by ACCS' as introducing intimate relationship challenges [14, 18, 26]. In prior research, difficulties with intimacy are apparent such that some survivors experience sexual dysfunction, are older when they have their first boyfriend or girlfriend and sexual relationship, and are less likely to marry when compared with siblings or national averages [33–40]. Moreover, intimate relationships can be considered within the broader context of social relationships. Childhood cancer survivors report having fewer friends, difficulties forming close friendships, and a lower likelihood of spending leisure time with friends [34, 35, 41]. The ability to engage in social activities and build requisite social skills in childhood and adolescents can impair

social development among ACCS, who subsequently experience lower self-esteem and self-confidence, such that social isolation and social anxiety worsen with age [42, 43]. For those survivors who are perhaps struggling socially, the threat of infertility is yet one more complication that is not easily discussed or disclosed and that impacts romantic relationships. Uniquely evident in our study were the first-hand accounts of how, often throughout their young adult years, ACCS navigated their own unease with such discussions and the fear of disappointing a current or potential partner, which led some to shy away from romantic relationships. As such, the lower marriage and parenthood rates previously reported among childhood cancer survivors [6] should not solely be attributed to physiological treatment effects and medical infertility, but are likely also related to the psychological and social challenges related to survivorship and uncertain fertility [44].

This study adds to existing research describing the unmet infertility-related communication, information, and education needs throughout the cancer trajectory and well into survivorship [22]. Consistent with previous research, the participants reported feeling awkward having fertility-related discussions with healthcare providers, but also, healthcare provider appeared uncomfortable and uncertain about how to talk to ACCS [14, 18]. Not surprisingly, fertility-related discussions do require extra sensitivity and for providers to take the initiative, a preference expressed by survivors themselves [22]. Our research uniquely highlights that healthcare provider discomfort and dismissal of fertility-related concerns contributed to survivors themselves judging such conversations to be too early, with the appropriate time when they are ready to have children. This is especially concerning for female survivors at risk for premature menopause, whose risks for infertility begin at an earlier age. Research suggests that survivors do want to discuss their fertility concerns with providers across their cancer experience, including shortly following treatment completion as well as throughout survivorship [22].

The provision of fertility-related information, education, and even medical care cannot be considered in isolation of the larger question of the most appropriate and feasible model of long-term follow-up for ACCS. The feasibility of either oncology or primary care providers in addressing fertility concerns during routine follow-up visits is challenging [22]. Survivorship care guidelines are not specific about whether and how to routinely screen survivors for infertility, and it remains questionable if, how and with whom fertility should be addressed [7, 45, 46]. As it stands, fertility testing has been reported to be low among ACCS [15], and providers are unsure of how to approach fertility-related needs [47]. We can also assume that healthcare providers, be they primary care providers or oncologists, have knowledge gaps specific to infertility in ACCS, just as many do with late effects in general. The long-term impact of cancer treatment on fertility is an

evolving field, and it is vital that providers who are counseling survivors about fertility risk and fertility options stay updated with current research. Alternately, clear paths of referral to specialists are essential.

Owing to the broad impact of uncertain fertility on the psychosocial health of ACCS, sensitive and empathic guidance, supports, and resources for survivors are clearly needed [23]. Access to nurses, counselors, or psychologists with fertility-related expertise across the cancer continuum, but especially early in the survivorship journey, would perhaps assist survivors in navigating the emotional and practical issues that arise prior to considering fertility and future parenthood. While information is necessary, it cannot replace the benefit of discussions with a trusted healthcare provider, as suggested by cancer survivors' preferences for face-to-face conversations [47]. Considering evidence that nurses perceive limited designated responsibility and opportunity in fertility information provision, others have recommended further exploration of the role of nurses with fertility-related training in leading evidence-based interventions [47]. Further research efforts are also needed that focus on understanding and developing the psychological care, supports, and interventions related to uncertain fertility that are gender-sensitive [48] and move beyond the gender binary and stereotypes. This would entail explicating the ways in which men, women, and non-binary survivors' needs could be better met through tailored communication/messaging, education, information, and modes of support. This need was particularly evident and a novel research implication informed by our study.

In this small qualitative study, we offer a description of ACCS' experiences of uncertain fertility that provides insight, but that ought to be considered along with study limitations. The initial 15 interviews were conducted for a larger study that focused on ACCS experiences of managing their health in general, and although these participants discussed uncertain fertility, it is possible that we would have obtained more diverse and nuanced data had these been focused specifically in uncertain fertility. All study participants were recruited in one Canadian province, and most were receiving some form of cancer-related medical follow-up, and thus, this study does not capture the perspectives of those in other healthcare systems or without access to appropriate healthcare. Furthermore, there was only one participant who self-identified as queer in this study, and so the perspectives of individuals who identify as LGBTQ are not adequately represented in this study. Further research that focuses on the experiences of LGBTQ cancer survivors specifically related to uncertain fertility would be an important contribution and is required to inform the development of gender-inclusive supports, resources, and healthcare services.

Despite growing efforts to increase fertility-preservation methods and access to services at the time of diagnosis and primary cancer treatment, this is not always an option, and

some methods remain experimental. Thus, addressing the fertility-related needs of childhood and adolescent cancer survivors will continue to be a vital component of patient-centered care in the future. Uncertain fertility is common and has implications for survivors' wellbeing, capacity for family planning, and ability to access specific fertility services in a timely and appropriate manner. Further research to develop comprehensive support, resources, and processes of care are vital to meet this priority need for survivors.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

Informed consent Informed consent was obtained from all individual participants included in the study.

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