

# BMJ Open Psychological adaptation and recovery in youth with sarcoma: a qualitative study with practical implications for clinical care and research

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**To cite:** Kosir U, Bowes L, Taylor RM, *et al.* Psychological adaptation and recovery in youth with sarcoma: a qualitative study with practical implications for clinical care and research. *BMJ Open* 2020;**10**:e038799. doi:10.1136/bmjopen-2020-038799

► Prepublication history and supplemental material for this paper is available online. To view these files, please visit the journal online (<http://dx.doi.org/10.1136/bmjopen-2020-038799>).

Received 24 March 2020  
Revised 11 October 2020  
Accepted 16 October 2020



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## ABSTRACT

**Objectives** This study explored factors that play a role in psychological adaptation and recovery of young people with sarcoma.

**Design** Qualitative study.

**Setting** National Health Service hospitals in the UK.

**Methods** Using purposive sampling, participants were recruited for semistructured interviews over the telephone or face to face in order to answer questions about how cancer impacted various domains of their life. Data were analysed using a framework approach.

**Results** Thirty participants, aged 15–39 years with primary sarcoma diagnosis provided in-depth accounts of their experience. Emerging themes from the interviews were grouped into two overarching themes that relate to one's adaptation to illness: individual level and environmental level. The qualitative nature of our study sheds light on meaningful connections between various factors and their role in one's psychological adaptation to sarcoma. We devised a visual matrix to illustrate how risk and protective factors in adaptation vary between and within individuals.

**Conclusions** This study demonstrates that young people with sarcoma report an array of both positive and negative factors related to their illness experience. The route to recovery is a multifactorial process and a one-size-fits-all approach to psychosocial care proves inadequate. We propose that moving beyond the latent constructs of resilience and psychopathology towards a dynamic model of psychological adaptation and recovery in this population can result in optimisation of care. We offer some recommendations for professionals working with young people with sarcoma in clinic and research.

## INTRODUCTION

Sarcomas are a rare form of cancer but nevertheless represent an important health issue and often afflict young people.<sup>1,2</sup> Adolescents and young adults (AYAs) present a particularly vulnerable group of patients due to their developmental period marked by physical, social and emotional maturation,<sup>3</sup> all of which may be disrupted by a cancer diagnosis.<sup>4,5</sup> Consequently, AYAs often experience

## Strengths and limitations of this study

- Framework analysis allows a structured and organised approach for theme-based and case-based analyses, which produces a visual matrix of themes by participants.
- The qualitative nature of our study sheds light on meaningful connections between various factors and their role in recovery and adaptation to life with and after sarcoma.
- Participants in this study are a heterogeneous group and were self-selected, as such, the factors identified may be limited and not generalisable to other adolescents and young adults (AYA) populations.
- Psychopathology and resilience in AYA with sarcoma may result from vastly different pathways, therefore, understanding one's unique trajectory may help us in offering a more personalised approach to psychosocial care.

different challenges and needs compared with younger or older patients facing cancer.<sup>6,7</sup>

Over the past two decades the improvements in treatments for sarcomas resulted in higher survival rates, however, these remain low compared with other tumours, hovering around 50%–60%.<sup>8,9</sup> Patients with sarcoma are often subjected to an array of treatments; radiation and surgical procedures are the first line of treatment for soft tissue sarcoma,<sup>10</sup> while osteosarcomas respond best to a multimodal approach including multi-agent chemotherapy.<sup>11</sup> Due to these treatments many individuals do not reach full recovery; up to 50% report lifelong disability and significantly compromised health-related quality of life.<sup>12</sup> Qualitative work with adults shows that sarcoma diagnosis and treatment can be burdensome and psychologically taxing,<sup>13</sup> however, a recent review of the literature revealed that the impact of sarcoma diagnosis and treatment on psychosocial well-being is not yet well understood.<sup>14</sup>



Psychological adaptation and recovery in cancer are complex phenomena with biological, individual and societal factors contributing varying degrees of risk for poor outcomes. Although distress is a normal response to life-threatening illness, a subset of young patients report psychopathology on reaching remission.<sup>15–17</sup> The causes of this may be many and multifaceted; treatment and medications may directly interfere with the endocrinology,<sup>18</sup> disrupt education or career plans,<sup>19 20</sup> compromise fertility,<sup>21 22</sup> or lead to a lifelong disability.<sup>6</sup> On the other hand, studies have found that despite the challenges, many AYAs with cancer become resilient or experience post-traumatic growth.<sup>23–25</sup>

In the recent years, national as well as international initiatives have recognised the importance of psychosocial impact of cancer diagnosis in youth and are calling for research initiatives that investigate psychopathology and promote psychological well-being in young patients.<sup>26 27</sup> However, the field lacks a common definition of resilience<sup>28</sup> and still relies on latent models of assessing mental health (ie, having depression leads to fatigue and/or loss of interest).<sup>29</sup>

We propose a different way of understanding psychological adaptation and recovery in cancer and conceptualise the two as interlinked and dynamic processes that encompass individuals' mental, physical and social function along the illness trajectory and into survivorship. Resilience and mental health are malleable constructs contained within, brought about by complex interactions between different factors. While successful adaptation and recovery (ie, maximised resilience factors and minimised risk factors for psychopathology beyond physical functioning) is a shared goal in increasingly patient-centred healthcare systems, we need to account for the fact that the configuration of these constructs will vary between people and may result from different pathways.

As the majority of patients with sarcoma experience life-long disability<sup>30</sup> and the number of AYA sarcoma survivors keeps increasing, it is important to develop a novel model of understanding psychological adaptation and recovery in sarcoma and describe the factors that are important to individuals. This will allow us to better address the psychological needs of young patients with sarcoma.

Thus, this study had three aims: (1) To describe factors that impact psychological adaptation and recovery in sarcoma in AYA and demonstrate variability between individuals, (2) To contribute evidence towards a dynamic understanding of psychological adaptation and recovery in AYA with sarcoma, and (3) To provide recommendations for clinical care and research.

To best address these aims, we relied on qualitative methods that allow for an in-depth exploration of patient-reported outcomes, their personal circumstances and response styles.<sup>31</sup>

## MATERIALS AND METHODS

### Participants and recruitment

Individuals who responded to the study advertisement online (via social media) or in the recruiting UK hospitals were invited to participate. Eligible participants were those with any

diagnosis of sarcoma, aged over 13 years (current age) and able to communicate in English. Patients less than 16 years of age gave their assent after consent had been received from their legal guardians; the interviews proceeded following receipt of consent. Purposive sampling<sup>32</sup> was guided by a sampling framework (see online supplemental material for details) to recruit participants across the cancer trajectory, with experience of multiple treatment modalities and aimed at approximately representing a distribution of patients across demographic (eg, gender, age) and illness (eg, type of sarcoma) characteristics. The final sample size was determined when data saturation was reached, and no new themes emerged from the individual semistructured interviews.

The analysis presented in this paper represents a subgroup of the total sample (n=121) who were aged 15–39 years, corresponding to the National Institute of Health definition of AYAs, which has been identified as a unique subgroup of cancer patients and survivors.<sup>33</sup>

### Design and procedure

This study is a part of a larger study of health-related quality of life in patients with sarcoma across the UK, aiming to develop a patient-reported outcome measure specific for patients with sarcoma. Data were collected in this phase from February 2017 to August 2017.

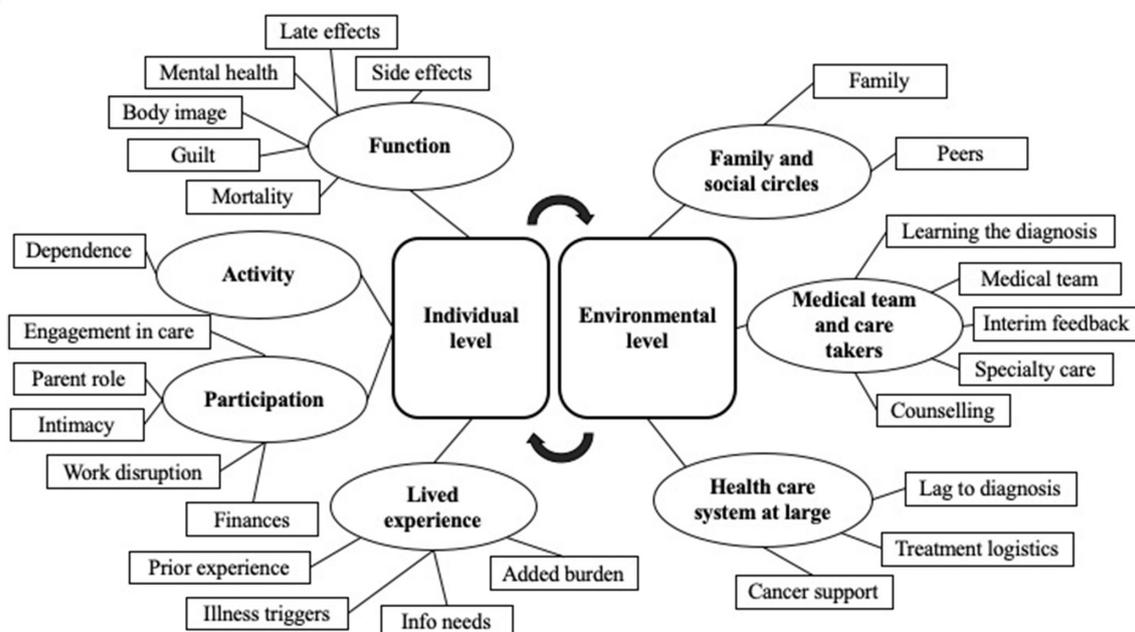
Two modes of interviews were offered, in person, or over the phone. In-person interviews were held at home and at the National Health Service (NHS) premises and phone interviews were conducted with individuals far from the study site. People aged under 16 years were interviewed face to face and had their parents present at their request.

The interview guide was based on the existing theoretical models of health-related quality of life<sup>34 35</sup> and refined with an advisory group that included patient advocates, sarcoma experts, members of the sarcoma unit, as well as the research team. This guide, previously published elsewhere,<sup>36</sup> was not prescriptive and was purposefully flexible to enable the researcher to explore new and emerging experiences.<sup>37</sup> The topic guide was shared with participants prior to the interview, so they knew the topics covered; this allowed participants to have more control over the process. The participants were aware of the study aims, and the role of the researchers. No other information about the research team was shared with the participants.

Researchers with graduate-level training in qualitative methods were responsible for conducting and moderating the semistructured individual interviews. Individual interviews lasted on average 45 min, were audio recorded and transcribed verbatim. Names, locations or other identifiable information were omitted from the transcripts for confidentiality. This manuscript followed the COnsolidated criteria for REporting Qualitative research (COREQ) guidelines<sup>38</sup> for reporting of qualitative research (online supplemental material).

### Data analysis

This study is based on in-depth data analysis of 30 transcripts of the semistructured interviews.



**Figure 1** The conceptual framework on which the themes were mapped.

The transcripts were analysed using a framework approach.<sup>39</sup> Framework analysis allows a structured and organised approach for theme-based and case-based analyses, which produces a visual matrix of themes by participants. The approach is appropriate for semistructured interviews that discuss similar topics, and results in sensible data reduction, which allows for multidisciplinary and collaborative approaches to analysis.<sup>40</sup>

Researchers (UK and AM) read the transcripts repeatedly to familiarise themselves with the data, and then independently coded them. The first phase of the analysis followed a deductive approach by devising codes and key concepts, which were mapped onto the first conceptual framework of psychological adaptation and recovery developed by UK (PhD candidate). The conceptualisation of some themes (ie, function, activity, participation) was informed by the WHO International Classification of Functioning, Disability and Health framework, which describes recovery as a process of improvement in the psychological, physical and social domain.<sup>41</sup> The second phase of the analyses relied on an inductive approach, adding themes from the initial coding in order to complement the framework and consolidate the findings.<sup>42</sup> This resulted in a framework (figure 1) on which data were mapped. Themes were then compared within and between individuals with the help of visual matrices (figures 2 and 3). The themes were described systematically, and the findings discussed among the team and embedded in the broader literature.

The first author (UK) kept a reflexive journal to explore the concepts and repetitive themes. In order to account for participants' and researchers' cultural context and understanding of the interviews,

meanings from data were produced jointly within the team discussions. Due to the nature of this analysis the exact inter-rater reliability was not determined.<sup>43</sup> Interpretations on both sides enabled confirmation or rejection of researchers' perceptions of data.<sup>44</sup> The goal of this work was not to address the questions of prevalence and extent, therefore quasi-statistical phrases such as 'some', 'many' or 'several' are used to not make the findings quantifiable and consequently misleading.<sup>45</sup> Please see figure 4 for the detailed steps of the analysis.

### Patient and public involvement

Patients and advocates for sarcoma were involved in the design of the overarching research study on health-related quality of life in sarcoma and included as co-applicants on the grant. They were invited to help with the recruitment of participants and dissemination strategy, as well as in the analysis, interpretation of the results and, when appropriate, acknowledged in the publication.

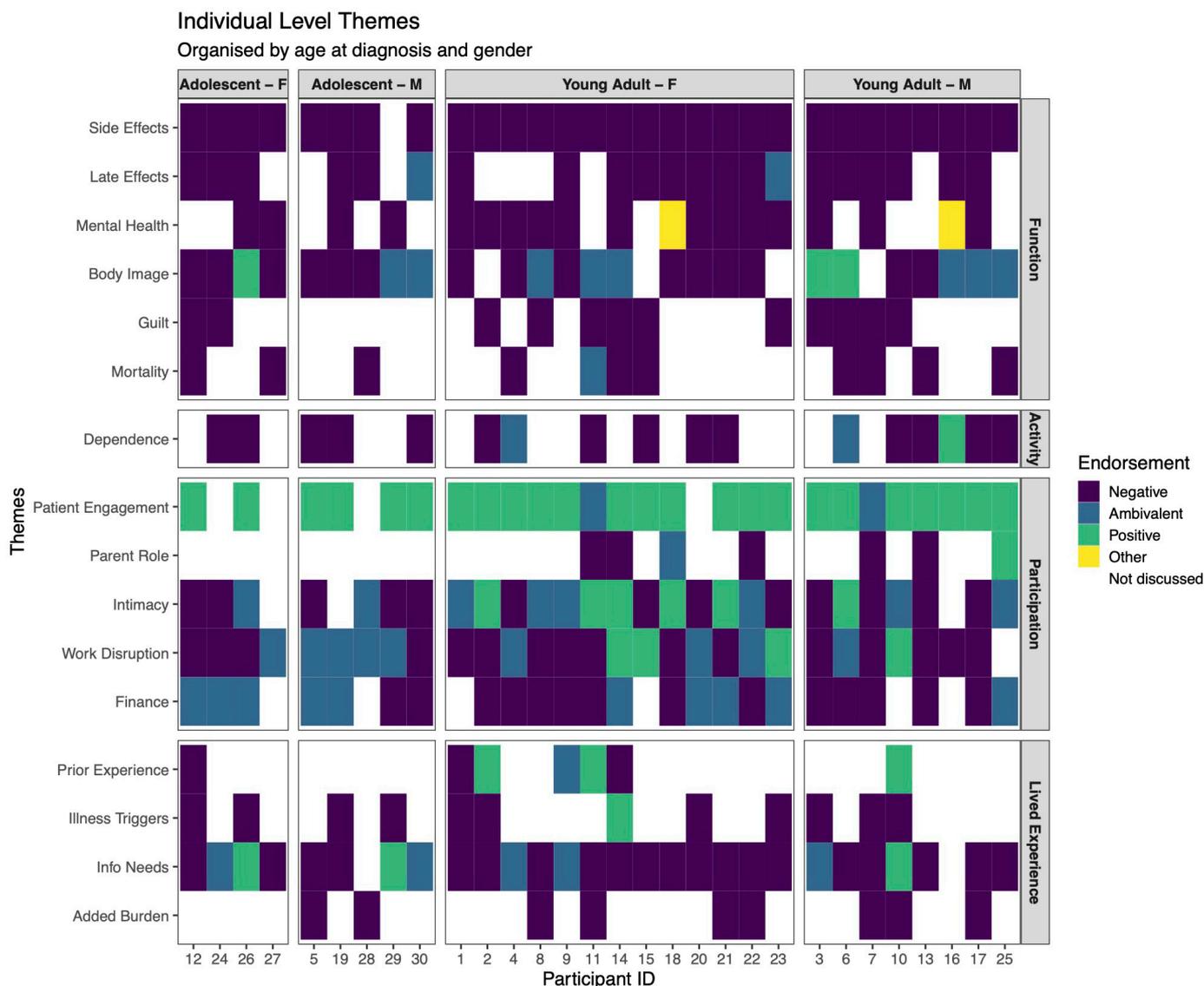
## RESULTS

### Participants

A total of 175 patients expressed interest in participating in the study, of whom 121 provided valid consent and were interviewed or joined a focus group. This study includes the 30 young people who were aged 15–39 years who participated in interviews. Descriptive demographic and medical information of our sample can be found in table 1.

### Factors that play a role in psychological adaptation and recovery in sarcoma

Our analyses revealed 7 themes and 26 subthemes that can be viewed as factors, which were grouped



**Figure 2** Visual matrix of individual-level themes, grouped by age at diagnosis and gender. Note: Adolescents are individuals who were diagnosed at 24 years or under, and young adults are those who were diagnosed above age 24 years. F, female; M, male.

into two overarching themes: individual-level factors and environmental-level factors. When describing these factors, individuals' accounts were rated as either describing a given factor in a negative, positive or ambivalent light. These factors form the basis for our conceptualisation of psychological adaptation and recovery (see figure 1).

Illustrative quotes from participants are provided within the text and fully anonymised for confidentiality reasons. The remaining quotes are included in table 2, which is organised to follow the themes' framework.

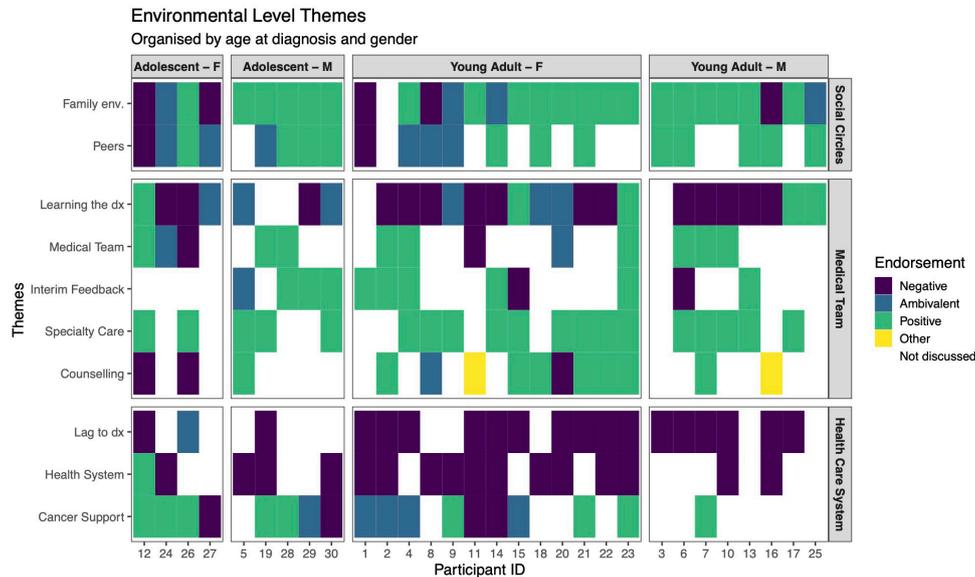
### Individual-level factors

Individual-level factors encompass those that were individually bound and sometimes independent from external circumstances. These were not necessarily mutually exclusive with environmental-level factors, and sometimes overlapped on the continuum between individual

and societal. Figure 2 demonstrates the discussed themes by all participating individuals.

### Function

Function refers to an individual's physical or bodily functioning including psychological well-being and mental health.<sup>41</sup> Treatment side effects were endorsed entirely as negative, impacting participants' quality of life, and though they varied in magnitude and number, participants commonly reported trouble eating, feeling nauseous, and experiencing fatigue or pain. Side effects limited individuals' participation in activities, however, they tended to subside after the treatment ended. For some participants symptoms persisted over time. Among the late effects, issues and worries related to fertility were the most commonly endorsed and discussed equally between genders.



**Figure 3** Visual matrix of environmental-level themes, grouped by age at diagnosis and gender. Note: Adolescents are individuals who were diagnosed at 24 years or under, and young adults are those who were diagnosed above age 24 years. F, female; M, male.

Mental health concerns and acute episodes of psychopathology were reported by 17 participants. Two participants reported that their mental health was directly influenced by their physical function and resolved with physical recovery (see ‘Other’ in figure 2). Several participants reported thoughts about their mortality, which provoked anxiety and negative emotions, and the majority emphasised that the mental health problems and psychopathology started after the phase of active treatment.

I think the main thing that I struggled with was the bit afterwards when some people were ‘You’re alright now. It’s finished, you’re okay.’ I found that hard to talk to some people about, because they saw that actually the physical side of it is over, but actually the mental side isn’t.

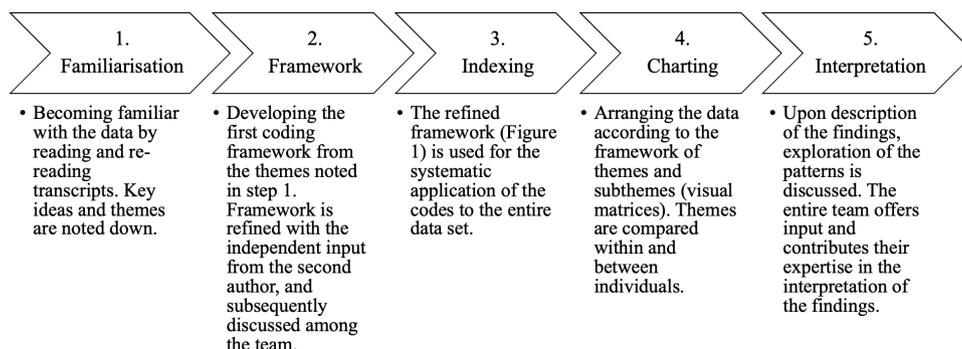
Twelve participants also endorsed feelings of guilt, which tended to persist beyond active treatment. Young people expressed feeling guilty in relation to failing to fulfil their roles as partners or parents, unable to carry out their daily tasks or having to rely on their parents for financial or logistical support. Some participants

experienced guilt in their relationship to other cancer peers who had a poorer prognosis, although others expressed feeling ‘lucky’.

Body image concern was also prevalent; 15 participants reported negative feelings associated with it, and 3 individuals talked about their body image improving with illness due to weight loss or improved self-care and healthier lifestyle brought on by their illness (see table 2).

### Activity

Activity is the ability to carry out a task or an action.<sup>41</sup> The most commonly described factor was loss of independence by 17 participants, which often resulted from physical limitations associated with treatment procedures. Among those who reported feeling dependent, two individuals did not perceive it as problematic because of the temporary nature of dependence and adjustments made to cope. One individual reported remaining relatively independent, which resulted in a sense of accomplishment, and resultantly positive attitude.



**Figure 4** The steps included in framework analysis.

**Table 1** Participant demographic and medical information

	N %	M (SD) range
<b>Gender</b>		
Male	14 (47%)	
Female	16 (53%)	
<b>Age at study, years</b>		
15–24	5 (17%)	30.3 (5.5)(15 – 39)
25–39	25 (83%)	
<b>Age at diagnosis, years</b>		
11–24	11 (37%)	27.5 (6.4)(11 – 39)
25–39	19 (63%)	
<b>Treatment status</b>		
On treatment	10 (33%)	
Off treatment	20 (67%)	
<3 years	13 (65%)	
3–6 years	6 (30 %)	
>6 years	1 (5%)	
<b>Marital Status</b>		
Single	16 (53%)	
Married/cohabiting	12 (40%)	
Divorced	2 (7%)	
<b>Employment</b>		
Full-time student	4 (13%)	
Part-time employment	4 (13%)	
Full-time employment	12 (40%)	
Other	10 (34%)	
<b>Ethnicity</b>		
White	23 (77%)	
Other	7 (23%)	
<b>Treatment type</b>		
Surgery alone	6 (20%)	
Chemotherapy alone	1 (3%)	
Surgery and chemotherapy	10 (33%)	
Surgery, radiotherapy and chemotherapy	6 (20%)	
Surgery and radiotherapy	3 (10%)	
Chemotherapy and radiotherapy	3 (10%)	
Other	1 (3%)	
<b>Sarcoma type</b>		
Soft tissue sarcoma	13 (43%)	
Bone sarcoma	17 (57%)	
<b>Sarcoma site</b>		
Upper limb	4 (13%)	
Lower limb	11 (37%)	
Other*	15 (50%)	
<b>Amputation</b>		
Yes	4 (13%)	
No	23 (77%)	
N/A	3 (10%)	

\*Other site: head and neck, pelvis, spine, finger, breast, abdominal origin. N/A, not applicable.

## Participation

Participation involves active engagement in one's life and daily activities such as education or work, parenting, engagement in one's care, and romantic relationships and intimacy.<sup>41</sup> Disruption in education or work was reported by 29 participants. Sixteen reported a negative impact and felt like they fell behind and had to change their educational and vocational plans, which was 'hard to accept'. Nine participants felt ambivalent about their vocational disruption, most often explaining that it was a 'small price to pay', or the disruption was temporary. Four participants, all of them young adults, endorsed disruption as a positive thing, allowing them to rethink their priorities and pursue more fulfilling endeavours (see table 2).

Financial strain resulting from work disruption was discussed by 24 participants, and mostly felt by those with limited support from their family or partners. Young adults in particular reported hidden costs of sarcoma, which included travelling for treatment, parking or family lodging at a hotel. A few participants reported limited impact because their employers continued to provide them with a salary, or they were financially secure.

Of 30 participants, 7 were parents. Five of them reported that their parenting role was compromised by illness, and they expressed worry about their children. One individual reported that being a parent was his most important role and endorsed it as a positive factor, explaining that it provided him with a sense of purpose during treatment and recovery. Twenty-seven participants also spoke about romantic relationships and intimacy, which were commonly impacted by the diagnosis and treatment. Several talked about romantic relationships in a negative way and suggested that their relationships fell apart, describing illness and associated distress as the main reasons. Eight individuals felt ambivalent about the impact of the sarcoma on their relationship, while six reported that their illness strengthened the appreciation of their romantic partner and made the relationship stronger as they learnt ways to remain intimate despite the limitations imposed by illness.

A widely endorsed positive factor was individuals' participation in one's own care. Twenty-four young people reported being proactive about their care and sought ways to improve their health. Shared decision making and patient engagement were seen as empowering.

## Lived experience

Lived experience describes other factors that are pertinent in one's participation and recovery. A common theme discussed by 28 participants was the need for information. Young people were not always satisfied with the amount of information they were provided. In a few cases the information felt overwhelming, and several reported inadequate information at the time of diagnosis (see table 2).

Twelve participants spoke about illness triggers or 'reminders', which had a negative impact on all but one

**Table 2** Supporting quotes from participants about individual-level factors

Individual		
Function—an individual's physical or bodily functioning including psychological well-being and mental health	Side effects	"I got the ulcers in my mouth a few times after my methotrexate, and the ulcers were quite bad. I'd probably go, like, 3 days without eating or drinking just because my mouth was so painful."
	Late effects	"I found out a couple of months ago that the chemo has made me totally infertile, and so that has probably been the thing that I've struggle with the most out of everything."
	Mental health	"I was on a lot of diazepam to try and help me sleep and just really get through those first few days(after diagnosis). I think I just cried probably for the first few weeks because I didn't know what was going to happen. Nobody seemed to be giving me any indication as to what the next steps were..." "I got put on the antidepressants. That was, kind of, the worst thing, how it affects you mentally. I just couldn't even get out of bed...(..)Depression was that bad. That's one of the worst side effects."
	Body image	Negative: "Losing my hair didn't really bother me that much at the beginning. Towards the end it kind of did bother me. I got really tired of just looking like a cancer patient." Positive: "I think my physical condition, in terms of body, like, I've lost a lot of weight compared with, I think I'm 15 kg down pre-cancer, which I love and I take very much pride in the fact that I'm a lot fitter than I was..."
	Guilt	"The hardest thing is when you see how much pain and upset you caused other people(...). So that's what I really struggle with today all the time. It's horrible that you've ruined someone's life for a year."
	Mortality	"You've got your kids and your family, and you've got to work, because you need the money. In your head, you're thinking, 'I'm dying. I'm literally dying as I'm working."
Activity—ability to carry out a task or an action	Dependence	Negative: "I couldn't do anything. So, during this time, my husband had to wash my feet, change my underwear and dress me.(..)I didn't have a shower for 4 months, just stand-up washes because I didn't want to get the dressings wet, and my husband had to wash my hair and do all the housework." Positive: "I had family around, but in terms of people helping me get dressed and everything, I didn't need that."
Participation—active engagement in one's life and daily activities such as education or work, parenting, engagement in one's care, and romantic relationships and intimacy	Patient engagement in care	"I think it's still important to have an element of control and be involved in your decisions.(..)I've always had the final say, but I think it's always important to feel as though you're part of your treatment. So you know what's going on, and you're not, kind of, just letting it happen to you."
	Parent role	Negative: "It was really hard from things like-, so I had to stop breastfeeding." Positive:(interviewer): "Being able to see your daughter grow up as being something that has helped you throughout this period..."(participant): "Most definitely, yes."
	Romantic relationship and intimacy	Negative: "(Partner) was essentially my carer throughout the whole treatment, because obviously she was there... and our relationship unfortunately fell apart after. We were made aware of this. They did say, 'Look, after(cancer), people can change." Positive: "Sex life, basically that was non-existent for 9 months,(..)we'd have to spend quality time doing something else together, instead of duvet action, so we might watch a DVD or a TV programme, or play cards, and just make it a special quality time, doing something together."
	School and work disruption	"I had a really good relationship with my manager, so it wasn't hard to tell my manager about it. They were very supportive, you know, I obviously had to go for chemo almost immediately, so I had to leave work straight away, and I told them. They were very good, they offered to continue paying me until I come back, and stuff. They kept in touch with me, so that was really good." "I still did all my exams, and so I've had to work harder than anyone to make it up, and I feel really short changed that I'm only graduating with an ordinary degree, and I worry that it's going to hold me back."
	Financial strain	Negative: "I mean obviously when I got ill I was supporting myself via my student loans and my grant, and my part-time job, and that all ended obviously when I had to interrupt my studies, so they stopped paying me." Ambivalent: "Paying for taxis is not exactly cheap, so my friends help. My brothers help, obviously. They drive. My parents help and my colleagues help as well, so I wouldn't say it's dire if you know what I mean? It's manageable."

Continued

Table 2 Continued

Individual	
Lived experience—describes other factors that are pertinent in one's participation and recovery	<p>Prior experience</p> <p>Negative: "Because my mum died of cancer, and having cancer now really kind of reopened a lot of my grief over that, because I just felt like I was reliving her history."</p> <p>Positive: "I have a genetic predisposition to cancer, and so it's not my first cancer. In fact it's not my first sarcoma. I know it might sound weird to say but to be honest since of (other medical condition) diagnosis I've kind of been waiting for osteosarcoma. Yes, I was just kind of-, well, kind of prepared."</p>
Illness triggers	<p>Negative: "Again, you get what I call shadowy pain where the tumour was, so I'll get a sore shoulder or something and that, sort of, leans on your mind because it takes you back to the exact same symptom that gave away the fact that you had cancer originally."</p> <p>Positive: "Now my scar is extremely neat and tidy, even though it's large. I think it reminds me that I've fought something and won, at the moment, and I'm still here."</p>
Information needs	<p>"(Interviewer) When you were with that first appointment where you received the diagnosis, she (Dr.) mentioned reconstructive surgery, at that time did you understand why she was talking about that? To be honest, I didn't. (Interviewer): She just gave you the information? Yes. She didn't really mention what it would entail. She just said, you know, they do great reconstructive surgery, so I'm, like, 'Oh, okay. It sounds like I'm going to have to have surgery,' but she didn't go into detail about what the treatment would be."</p> <p>"Obviously all the information's bombarding, coming into you, so you can't process it all."</p>
Added burden	<p>"I had moved to new jobs at the time. I was going through a break-up in a relationship at the time also which added to the problems, you see."</p> <p>"I mean, my mum used to be my best friend. Just after I got into remission, she had her cancer back again, and she passed."</p>

person. These triggers, ranging from bodily sensations to visits to the hospital, reminded participants of illness and caused feelings of distress and anxiety. One participant reported the illness reminder—a large scar—as a positive sign, giving her a sense of pride and strength.

Another aspect that proved important in one's understanding and adaptation was prior experience. Prior experience defines one's experience with cancer, be it their own or familial, or significant prior experience with the medical care (eg, major surgery, unrelated to cancer). Six participants discussed prior experience, two endorsed it as negative because they have lost a person close to them to cancer. They worried about their own prognosis and felt like they could not openly talk about it with their family because of the negative connotations. On the other hand, three participants reported their prior experience as a positive factor that allowed them to better manage their expectations, navigate the medical system and advocate for themselves.

Finally, nine participants talked about the added burden resulting from external factors not related to their illness, for example, breaking a limb and family bereavement. This burden was an additional stressor and was exclusively negative.

### Environmental-level factors

Environmental-level factors refer to aspects of individuals' social and environmental context within which they found themselves during their illness experience. They range from more intimate circles of friends and family to one's medical team. Figure 4 demonstrates the discussed themes by all participants.

### Family and social circles

Twenty participants endorsed family as a positive factor. Many reported their relationship within the family improving and resulting in a more cohesive environment due to support and greater appreciation of one another. Some individuals could only draw partial support from their family, and for a few, family was not a source of support but rather an impediment (see table 3).

Twelve participants spoke about peers and close friends in a positive light. Close friends provided additional support for the individual, which was different from familial support and provided them with a sense of normalcy. However, several participants reported that they also lost friends and felt abandoned due to illness.

### Medical team

Participants often spoke about their medical teams and healthcare professionals. The initial impact of diagnosis often stemmed from the time they learnt their diagnosis. Learning the diagnosis was a negative experience for 15 individuals and almost entirely resulted in feelings of shock and distress. Seven participants learnt the news over the phone, which did not allow them to ask questions. Some felt that the bad news was attenuated by the fact that they were not alone at the time of diagnosis, and some found great relief in getting an answer for their previously unexplained symptoms.

Nine participants spoke about their positive relationship with the medical team. Those who reported feelings of trust and being able to openly express their concerns reported greater levels of satisfaction with their care and less distress associated with treatment. Additionally,

**Table 3** Supporting quotes from participants about environmental-level factors

Environmental		
Family and social circles	Family environment	Negative: <i>"I'm just not very close to them, so it was quite strange to have bring them to something and they don't talk to each other. They said that I needed to bring them both, and at the time it was almost extra stressful having to bring them."</i> Positive: <i>"I would say, out of everything, it's brought us closer. The impact has been far more positive than negative, because I think everyone just values life a bit more now."</i>
	Peers and friends	Negative: <i>"I feel a slight disconnect with my friends, and it's hard because you can't just kind of slot back in.... It's just a sense of feeling so disconnected because my problems are just different and it's not relatable.(...)It's hard for my friends to see me sick, and it's hard for them to see me really suffering."</i> Positive: <i>"Quite a few of my mates had all arranged a big charity bike ride for me and they'd arranged it at this time because that was when it was going to fit in with my surgery, you see, and before I started my chemo again."</i>
Medical team and caretakers	Learning the diagnosis	Negative: <i>"I actually got told over the phone. I found it just really shocking.... Yes, to have it through a phone call, it was a bit, I don't know, strange to be told that through a phone call, really."</i> Ambivalent: <i>"It was a bit conflicting because obviously it's not something that you expect to hear, and it's not something you want to hear, but at the same time it was kind of nice to know that I wasn't crazy."</i>
	Relationship with the medical team	<i>"I do have quite a good relationship with Dr.(...)I trust that he'll tell me what he thinks is the best thing for me to do."</i>
	Interim feedback	<i>"There's actually been a small reduction in size in my largest mass and everything else has stayed perfectly stable. There's nothing else in my body so really, really pleased. My consultant's very, very pleased with how everything's working so it's very, very positive."</i>
	Specialty sarcoma and/or AYA care	<i>"I hear about people who don't have a clinical nurse specialist and they don't understand how important it is until you have one. You realise she does so much for us. She makes it easy. She knows when you don't understand something. She'll stay behind and explain things. When I had surgery, she travelled to the different hospitals, she came to see us. She looks after your family, friends, everything. Everyone knows her, but it's the fact that she takes time to explain. She listens to you."</i>
	Counselling or psychotherapy	Negative: <i>"One of the hospices arranged for some therapy, but I didn't really get on with it very well, so I only went a couple of times... I just didn't really like it, I found it made me feel more worried than less worried."</i> Positive: <i>"I did have some professional support at the very beginning. I had four or five sessions with a counsellor. Again, that was all done privately because I wasn't offered anything on the NHS,(...). It was really useful to go and talk to somebody and just open up and explain everything to them. She gave me lots of advice and tips on how to deal with the days when I feel down."</i>
Healthcare system at large	Prolonged route to diagnosis	<i>"So, I went to my GP about 1000 times, and they said I had growing pains and prescribed me diclofenac, which I took for a long time. Then because I couldn't sleep because the pain was so bad, and I was trying to do my exams, my mum decided to send me privately."</i> <i>"Every time I went to the GP, they, kind of, still said, 'Oh, look, you're just doing a lot of overtime. You're just standing on your legs a lot. It's just, you've probably got a shin splint."</i>
	Health system and treatment logistics	<i>"Travel can be quite hectic.(...)Yes, the thing is on the travel days it is about muscling through it, you know."</i>
	Cancer support groups	Negative: <i>"I did look on some sarcoma websites, but I found them quite depressing. A lot of people were sort of-, some people were not as successful as I'd been and I felt that I couldn't relate to them because they were in a different situation to me, so I found that those forums didn't really help because they were people offloading because they'd been dealt a bad card."</i> Positive: <i>"Yes, these support groups are fantastic because you can communicate with people the same age as you at any time.(...)They may have a very different cancer and be at a very different stage in their treatment or their journey but they understand because they're the same and they have the same feelings and the same thoughts and the same worries. Those groups are priceless."</i>

AYA, adolescents and young adults; GP, general practitioner.

interim feedback on disease and treatment status was desired. Several individuals emphasised that receiving positive news during treatment was encouraging. Two participants reported feeling a negative impact due to the lack of positive news.

In addition to the medical team, the specialist nurses or in some cases AYA coordinators were endorsed as the most helpful resource by 19 participants. Specialist nurses were able to spend more time with participants and answer questions related to their illness and its impact.

My MacMillan nurse, she's phenomenal. If I'm ever feeling a bit low or anything, when I was on chemo or anything, she was there. She was fantastic. Absolutely fantastic.

Fourteen participants spoke about psychotherapy and counselling. Some received it as part of their oncological care, while others sought it externally. For eight, counselling was a positive experience, teaching them adaptive coping mechanisms. However, four participants found



counselling services inadequate in addressing their needs as sarcoma patients, which resulted in frustration. Two individuals explicitly asked for and wanted psychological support but were unable to obtain any (see figure 3, 'Other').

### Healthcare system at large

Two-thirds of our participants reported a prolonged route to diagnosis. In all but one case this was a negative factor. Participants reported several months, in one case 2 years, before their symptoms were recognised and treated seriously. Several resorted to private care in order to speed up the diagnostic process. Two individuals reported an initial misdiagnosis.

Sixteen individuals also discussed difficulties in navigating the healthcare system. Some individuals had to travel long distances for their treatment, and two reported that their care was not age-appropriate. Managing the logistics of care was burdensome.

Participants were often encouraged to join cancer support groups, face-to-face or online, some of which were specific to sarcoma. Nine participants endorsed them as positive, but young adult men rarely discussed the topic or engaged with the services. Among cancer peers, participants felt supported, understood and less isolated due to their illness. However, half of the participants felt as though the groups were not tailored to their needs, found them too depressing due to generally poor prognosis, or hard to relate to due to different outlooks or age differences.

## DISCUSSION

This qualitative study aimed to describe both positive and negative factors along the illness trajectory in young people with sarcoma. By relying on qualitative methodology, we demonstrated the extent of between-individual variability in mental, physical and social function along the illness trajectory, and contribute evidence towards a dynamic approach to understanding of psychological adaptation and recovery. Practical recommendations for clinical care and future research are discussed below.

Thirty AYA participants from across the UK provided in-depth subjective accounts about their experience with sarcoma. The present study contributes to the existing body of knowledge with a thorough investigation and description of commonly reported factors and demonstrates that when faced with cancer, individuals stem from vastly different backgrounds and life experience. Consequently, different individual-level or environmental-level factors will contribute to one's psychological adaptation and recovery. As psychosocial well-being remains an unmet need and among the most important topics for AYAs with cancer,<sup>26</sup> we have to develop a better understanding of how different subgroups of patients, though afflicted with the same diagnosis, respond differently based on their individual biopsychosocial profile.

We propose that successful psychological adaptation and recovery may not be considered as latent entities, but rather as dynamic systems composed of factors or symptoms. As such, the concept extends on the static models of psychopathology (ie, meeting the symptom threshold for a mental disorder) and resilience (ie, absence of psychopathology given adversity),<sup>46</sup> and accounts for the fact that one might at once experience psychosocial growth, as well as symptoms of psychopathology.

Moving beyond the latent constructs of resilience and psychopathology in youth with sarcoma may be particularly important because unlike in the general population where mental health disorders are thought to occur via the activation of symptoms through adverse life events,<sup>47 48</sup> in patients with chronic conditions (ie, cancer) the pathway to psychopathology may be different.<sup>49 50</sup> The illness may be perceived as both an adverse life event, as well as a chronic health condition; we suggest that psychological adaptation and recovery in AYA with sarcoma will follow unique pathways and factors or symptom presentation. For example, our results imply that education and work are salient aspects of AYAs' identities and often disrupted by illness. So, we might have two individuals experiencing anxiety (ie, not achieving full recovery) who are of the same age, gender, same type of sarcoma and with a comparable treatment plan. While one's feelings of anxiety will revolve around the thoughts of mortality and follow-up consultations (*follow-up consultation* → *thoughts of mortality* → *anxiety*), another might experience anxiety due to occupational disruption and financial strain (*work disruption* → *financial strain* → *anxiety*). Similarly, we show that certain concepts such as body image, which most commonly carry negative connotations,<sup>51 52</sup> can in fact act as resilience factors if appraised as positive (*healthier lifestyle* → *improved body image after cancer* → *improved self-image*). This further strengthens our claim that individual circumstances vary in important ways and should be accounted for in clinical practice. The high level of individual variability also suggests that the use of person-centred statistical approaches (eg, growth mixture models)<sup>53</sup> would be informative in further study of this heterogeneous population.

It would be erroneous to expect young people exposed to cancer to show uniform outcomes in facing their illness. Most of the existing literature in AYA cancer survivorship addressing psychological well-being relies on the conceptualisations of psychopathology and resilience from healthy populations, which have not been adapted to young people with cancer. Therefore, an understanding of psychological adaptation and recovery as dynamic systems is needed to take into account the interplay between many resilience and risk factors on an intraindividual, as well as interindividual level. Our study offers a first step in this conceptualisation and provides invaluable insights for future work in this field.

**Table 4** A summary of recommendations for improvements of care and future research

Individual level	System-wide level	Future research
<ul style="list-style-type: none"> <li>▶ Better side effect management</li> <li>▶ Education and information about late effects (management and prevention)</li> <li>▶ Health literacy and promotion of patient engagement in care</li> <li>▶ Consultation for partners and peers</li> <li>▶ Psychoeducation (promoting adoption of adaptive coping techniques)</li> <li>▶ Early physical rehabilitation (<i>prehabilitation</i>)</li> </ul>	<ul style="list-style-type: none"> <li>▶ Structured support service for return to school or work</li> <li>▶ Streamlined schemes for financial aid for those in need</li> <li>▶ Age-appropriate services for adolescents and young adults</li> <li>▶ Assuring for quality care at smaller clinics</li> <li>▶ Better organised and multidisciplinary approach to diagnosis and treatment</li> <li>▶ Information on how to navigate services</li> <li>▶ Family planning and fertility preservation services</li> <li>▶ Signposting to support for mental health when active treatment ends</li> </ul>	<ul style="list-style-type: none"> <li>▶ Mixed-methods approach with qualitative and quantitative analyses will provide a more holistic interplay of various factors</li> <li>▶ Longitudinal data collection</li> <li>▶ Moving away from assessing psychopathology and resilience as latent constructs, and instead look at individual factors and symptoms</li> </ul>

### Limitations and future directions

Certain limitations are important to note in the present study. Our sample was heterogeneous, including individuals with all types of sarcoma, from a broad age range, and is limited to the UK, which might limit generalisability of the findings, particularly in the domain of the health-care system. Additionally, participants were a self-selected group and mostly provided retrospective accounts, which may have been impacted by recall bias. Though asked about their illness and sarcoma history, this information was self-reported. Lastly, this study relied on an existing data set, which might explain why some concepts were not explored in-depth by all the participants, or in greater detail. The themes mapped onto the framework were not mutually exclusive and often spanned across domains. Despite its limitations, our study is the largest in-depth exploration of the impact of sarcoma on AYA, which adds important insights for future research and targeted therapeutic approaches for this vulnerable population of patients.

### Conclusion and clinical implication

Sarcoma impacts young people on psychological, physical and social levels. Understanding the subjective experience and individual circumstances is at the centre of personalised approach to healthcare. Our findings suggest that young people with sarcoma require holistic guidance and care beyond active treatment. A one-size-fits-all approach is inadequate and healthcare professionals working with this population should actively engage with their patients from the time of diagnosis. Considering individual context will aid in identifying those in need of help in a specific domain (eg, return to work). While certain factors cannot be prevented (ie, infertility), promoting a cohesive atmosphere between the patients and medical professionals can result in better management of an individual and optimisation of service and resources utilisation. Furthermore, across the UK, nationally based services for AYAs mostly end at the age of 24 years.<sup>54</sup> In our

study we included individuals up to 39 years of age and demonstrated that young individuals who are ineligible for the NHS AYA services also present with needs that may not be adequately addressed in adult cancer units. We recommend that special attention is paid to young people outside the dedicated AYA services. A summary of recommendations for improvements of care and future research directions can be found in [table 4](#).

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**Acknowledgements** The authors thank all the participants who took part in this study and all the Principal Investigators at the Trusts opened for recruitment: Fiona Cowie; Helen Hatcher; Amit Kumar; Dan Stark; Craig Gerrand; Robert Ashford; Claire McKenzie; Sarah Massey; Julie Woodford; Jonathan Gregory; Robin Young; Mariam Jafri; Paula Wilson; Robert Ashford; Nicola Keay and Paul Cool. The authors also thank all the professionals involved in patient recruitment.

**Contributors** RMT, MO, RW and CG were involved in developing the protocol; AM coordinated the running of the study and was responsible for data acquisition; UK, AM, LB, CG, RW and RMT contributed to the analysis; UK and AM drafted the manuscript. All authors critically revised and approved the final manuscript.

**Funding** This work was supported by Sarcoma UK (grant number SUK102.2016LG) and Bone Cancer Research Trust (grant number BCRT/49/16). RMT is a National Institute for Health Research (NIHR) Senior Nurse Research Leader; RMT acknowledges support from University College London Hospitals BRC. UK would also like to acknowledge the financial support from Economic and Social Research Council (ESRC) as well as Ad Futura (Slovenia).

**Disclaimer** The views are of the authors and do not necessarily reflect those of Sarcoma UK or Bone Cancer Research Trust, or the NIHR/NHS. None of the funders were involved in conducting this review or drafting this manuscript.

**Competing interests** None declared.

**Patient consent for publication** Not required.

**Ethics approval** The Study was approved by the London Stanmore National Health Service (NHS) Research Ethics Committee (reference: 16/LO/2152; IRAS reference: 217605).

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data availability statement** No data are available.

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