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A qualitative study exploring patients experiences of being diagnosed and living with primary bone cancer

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A qualitative study exploring patients experiences of being diagnosed and living with primary bone cancer

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ABSTRACT

Objective: The aim of this study is to explore the experiences of patients with primary bone cancer.

Design: Qualitative study design using semi-structured interviews and focus groups.

Setting: Hospitals across the United Kingdom (UK) and recruitment through UK sarcoma charities and support groups.

Methods: Semi-structured telephone/face-to-face interviews and focus groups with a purposive sample of 26 participants. Data were analysed using Framework Analysis.

Participants: Patients with primary bone cancer aged 13-77 years. The majority were male (69%), white (85%); diagnosed within 4 years (54%); and had lower limb sarcoma (65%). Ten participants had undergone an upper/lower limb amputation (39%).

Results: The health-related quality of life domains of physical, emotional and social well-being and healthcare professionals' role were the overarching themes of analysis. The physical domain anchored patient experiences. The intensity and length of treatment, the severity of side-effects, the level of disability after surgery and the uncertainty of their prognosis had an impact on patient's self-image, confidence, mood and identity, and caused disruption to various aspects of the patients' social life, including their relationships (emotional and sexual) and participation in work/school and leisure activities. Adaptation was influenced by the way patients dealt with stress and adversity, with some finding a new outlook in life, and others struggling with finding their 'new normal'. Family and friends were the main source of support. Healthcare professional's expertise and support was critical. Rehabilitation services had a considerable role in patient's physical and emotional well-being, but inequitable access to these services was apparent.

Conclusions: This study described the impact of primary bone cancer on patients' well-being and adjustment over time with the identification of influencing factors of better/worse experiences. It showed impact was felt after end of treatment and affected patients at different life stages. Holistic models of survivorship care are needed.

Strengths and limitations of this study

- This study is an in-depth exploration of the experiences of 26 patients with primary bone cancer
- The study population varied considerably by age at, and time from, diagnosis, gender and anatomical location
- This study includes the experience of patients who had undergone amputation and other surgical interventions and treatment types
- Self-selected participants might not be representative of the experiences of patients with bone sarcoma, but our findings are consistent with previous studies and add to the research by identifying the role of influencing factors of better/worse experiences

Keywords: patient experience; primary bone cancer; quality of life

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None declared

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Author contributions:

RMT, LAF, MW, LS, RW, JW, CG and JSW were involved in developing the protocol; AM coordinated the running of the study and was responsible for data acquisition; AM, MW, JW and CG contributed to the analysis; AM, MW and CG drafted the manuscript. All authors critically revised and approved the final manuscript.

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No additional data available.

INTRODUCTION

Primary bone cancer (PBC), also referred to as a bone sarcoma, is very rare and makes up approximately 0.2% of all cancers diagnosed in the United Kingdom (UK).[1] In 2015, 551 new patients were diagnosed with PBC in the UK.[2] The commonest subtypes are osteosarcoma and Ewing sarcoma, which have a relatively high incidence in children and young people, and chondrosarcomas, more common in older age groups.[3] There has been no major improvement in 5-year overall survival rates for patients with PBC over the past 25–30 years, with rates static at between 53 and 55%.[4]

To give the potential for cure, PBC requires complete surgical resection of the affected part of the bone. Thus, the patient characteristically experiences a lifelong limitation in physical activity. Less frequently, this can be achieved only by limb amputation, which leads to permanent disability. Given the high physical burden of the disease and its treatment and expectations of survival, poorer patient-reported outcomes[5,6] are recorded in comparison to patients with other cancer types.[7] For example, patients have reported poorer care experiences compared with other cancer types in the National Cancer Patient Experience Survey (NCPES).[8]

When conducting a literature search, it became evident that there were inconsistent findings regarding the impact PBC may have on patients.[5] While research has shown that patients may experience lower levels of physical functioning than population norms or control groups (e.g. other cancer groups or healthy peers), there appears to be a lack of reliable information on the impact on patient's emotional and social well-being.[5] Several studies comparing patients who undergo an amputation or limb sparing surgery have shown no differences in quality of life scores [9,10,11,6,12,13] ; however there are also reports of patients who had an amputation having poorer health-quality of life scores compared with patients who had other types of surgery[14]. The inconsistent results are also shown in studies that report that patients who had amputations had poorer mental health[15,16] but better feelings of self-worth[16] compared with patients who had other type of surgery. Other studies have shown that those who undergo lower limb amputation tended to report more anxiety and depression.[5,17]

In addition to studies comparing patients who undergo an amputation and other types of surgery, there are studies comparing emotional and social well-being of patients with PBC with reference values either of general population data provided with the measure, non-cancer control data collected as part of the study or from patients with other cancer types. Overall the findings reported are also inconsistent.[5] Research with children and young people has shown improvement in emotional functioning over time[7,18], however in studies with older patients Paredes et al.[19] found no improvement in cognitive and emotional well-being from diagnosis to treatment and noted that they were worse than reference norms. No clear pattern can sufficiently explain these inconsistencies, although methodological differences including range of assessment tools used,

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outcome variables measured, as well as diagnosis, anatomical location, treatment, and age groups have to be taken into consideration.[5]

Given the complexity and heterogeneity within the patient population[5,20] more research is needed to explore patients' experiences of PBC. While qualitative research addressing the effects of PBC is sparse, literature has shown the adverse impact of having PBC on patient's emotional and social well-being. Studies focusing on employment[21, 22], and the long-term effects of bone cancer on current daily life, body image, future possibilities and identity[23,24,25] provide some insights into patients' experiences, but they concentrate on specific time points or specific aspects of the patient experience (e.g. body image).

Both qualitative and quantitative studies shed light on the impact of PBC on patients' quality of life; however more research is needed to understand in-depth, how patients of different ages, genders and treatment experiences live with and beyond PBC.[5] The aim of the current study was to explore the experiences of patients with PBC across the UK.

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METHODS

Design and Patient Involvement

This was a qualitative study involving patients with PBC, conducted as part of a larger qualitative study to develop a sarcoma patient-reported outcome measure – the Sarcoma Assessment Measure (SAM).

As the Sarcoma Assessment Measure is about patient experience of being diagnosed and living with sarcoma, patient involvement is central part of the study. Initially, the SAM research team included the Chief Executive of Sarcoma UK (Lindsey Bennister), who represented the patient perspective in the design of the study. Lindsey Bennister will continue to be involved in the study as a patient representative. Maria Onasanya, who was diagnosed with sarcoma over 5 years ago joined the study as a co-investigator and has assisted with data analysis. Both Lindsey and Maria have assisted with data analysis and are co-authors in presentations and research papers. We are also working alongside Sarcoma UK and the Bone Cancer Research Trust to ensure patients are updated about the progress of the study (through study newsletters suitable for non-specialist audience) and they receive copies of all relevant outputs. Healthcare professionals working with patients with sarcoma have been engaged in data discussion, recruitment strategies and dissemination.

Participants and setting

The study was undertaken across the UK recruiting through specialist PBC clinical teams, sarcoma charities and patient support groups. Eligible patients were those with any diagnosis of bone sarcoma, aged over 13 (current age) and were able to communicate in English.

Patients were identified by recruiting hospitals who obtained and emailed the patient's written consent to the researcher, the researcher reaffirmed consent prior to conducting the interview. Patients less than 16 years gave their assent after consent had been received from their legal guardians.

Patients who contacted the research team after seeing information on social media from sarcoma charities were given information about the study and sent a consent form; the interviews proceeded following receipt of consent. Focus groups conducted in patient support groups were only conducted if all members consented to participate; information was given to members by the lead for the support group (either another patient or a clinical nurse specialist) and the research team were invited to the group if patients had given their consent. The study was approved by London - Stanmore Research Ethics Committee (IRAS reference: 217605).

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Data collection

Data were collected through semi-structured individual telephone interview (conducted by AM) and focus groups (led by AM and RT) conducted in sarcoma support group meetings (AM and RT are experienced researchers and were not involved in the patient's care). Young people less than 16 years took part in face-to-face interviews (parents were present during the interview). The interview schedule was developed from the literature and expert opinion (patients and healthcare professionals). This was not prescriptive and was purposefully flexible to enable the researcher to explore new and emerging experiences[26]. This topic guide was shared with participants prior to the interview (Appendix 1), so they knew the topics covered (this allowed participants to decide whether they wanted to talk about the topics covered and gave them more control over the process). With permission, all interviews and focus groups were recorded and transcribed verbatim. Interviews lasted between 26 and 96 minutes and focus groups lasted 2 hours. Field notes were taken during and after the focus groups.

Data analysis

Data analysis was developed in two phases. Transcripts were broadly coded into the three functional domains of quality of life defined by Taylor et al.[27] "subjective, multidimensional and dynamic (…) and includes aspects of physical, psychological and social function" (p1831). Data were analysed line-by-line with the aid of QSR NVivo 11, a specialist software package used to assist with data storage, management and retrieval. The coded data were entered into a framework. Within each overarching domain, data were coded into more detailed themes using in NVivo codes. For example, for the overarching theme of emotional well-being, a subtheme of coping mechanisms was identified. Data exploration of patients' experiences considering age, gender, or if the patient had an amputation, helped uncover commonalities and differences. Initial coding was reviewed by the research team and revised following discussion.

RESULTS

Overall, a purposive sample of 34 of 121 patients participating in the wider Sarcoma Assessment Measure (SAM) study were diagnosed with PBC; 8 patients had craniofacial bone sarcomas and were not included in this analysis as they presented with experiences more akin to those with soft tissue head and neck sarcoma (data available from the overall study which includes patients with soft tissue sarcoma). We report here the findings from the remaining 26 participants (Table 1).

Table 1. Participants' characteristics

		N	%
Gender	Male	18	69
	Female	8	31
Age at study	M=40.5; SD=17.9, range 13-77 years;		
	13-24	4	15
	25-39	11	42
	40-65	6	23
	66+	5	19
Age at diagnosis	M=34.2; SD=18.1, range 8-77 years;		
	8-24	6	23
	25-39	14	54
	40-66+	6	23
Marital status	Married/civil partnership/cohabiting	13	50
	Single	9	35
	Other	4	15
Employment	Employee full-time/part-time job	10	38
	Full-time education at school, college/university	5	19
	Wholly retired from work	5	19
	Other	6	23
Ethnicity	White British/any other white background	22	85
	Other	4	15
Time from diagnosis	0-4 (years)	14	54
	5-20+ (years)	12	46
Treatment status	On treatment	5	19
	Off treatment	21	81
Where in the body was the sarcoma	Lower limb +/- other site	19	73
	Upper limb +/- other site	5	19
	Spine +/- other site	2	8
Treatment Type	Surgery alone	8	31
	Surgery and chemotherapy	12	46
	Surgery, radiotherapy and chemotherapy	4	15
	Surgery and radiotherapy	1	4
	Chemotherapy and radiotherapy	1	4
Amputation	Yes	10	39
	No	16	61

Four overarching themes structured the analysis: the health-related quality of life domains of physical, emotional and social well-being and the role of healthcare professionals. More detailed analysis revealed that the experience of sarcoma in relation to the quality of life domains was particularly influenced by the patient's stage in life (i.e. if they were a teenager, young or older adult), the individual's coping strategies and social support, the nature and extent of adaptation that had to be made as a result of illness (referred to as the adaptive tasks of illness), and where they were in their disease trajectory (e.g. diagnosis, treatment, end of treatment, follow up, or recurrence). These will be described throughout the presentation of results below.

Physical Well-being: *it was literally the worst pain I'd ever experienced*

The physical domain anchored patient experiences and the impact of PBC on emotional and social well-being. The intensity of treatment (surgery, cycles of chemotherapy and/or radiotherapy), the severity of side-effects, the length of treatment or the level of disability after surgery were some of the key features of PBC influencing the experiences described by patients.

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Patients faced specific challenges at different points in their treatment pathway. Starting with diagnosis, many explained the onset of pain as the trigger to seek help. During treatment, the majority of those who had chemotherapy gave vivid descriptions of how debilitating their treatment was and some experienced frequent hospitalisations between chemotherapy cycles. Patients who received radiotherapy talked also about fatigue and burning sensations as negative side effects of treatment. The range and magnitude of side effects described during the interviews reflected the significant impact of treatment on physical well-being, including: mouth ulcers, nausea, fatigue, (chest) infections, constipation, incontinence, diarrhoea, sepsis, tinnitus, hearing loss, anaemia, "*pins and needles in the ends of my fingers and the loss of some feeling in the ends of my fingers*", kidney and heart problems, and foot drop.

Moreover, radiotherapy and surgery were described as causing pain for varied periods of time after the treatment phase had ended. Whether patients had undergone limb sparing or amputation, pain was described as part of their lives and restricted daily activities. Pain management was particularly difficult in the first year after end of treatment. Patients with amputation found the unfamiliar sensations of 'phantom pain' particularly disconcerting.

"There doesn't seem to be a lot of options in dealing with that, especially because, for me, I don't have phantom pains every day, but I'll get it randomly (…) it can be quite disconcerting and painful"

The extensive surgery required had a profound impact on mobility, with lessened strength, instability, loss of flexibility, and poor balance. Patients described use of technical aids to their mobility such as crutches or a cane, use of a wheelchair or mobility scooter. Functional impairment caused major disruption in every aspect of the patients' life such as climbing stairs, lifting, driving, walking: "*going up and down the stairs is a job of work, to be honest. It is quite tiring*". Most patients described the frustration of not being able to be as active as before and this was felt long after end of treatment.

For a few patients, falls were a concern and limited their activities. In those who had experienced falls, the consequences could be severe: "*A little poodle, yapping like anything, I* *suddenly put all my weight, to jump out the way, onto my left leg and I felt it break. It just literally split apart*".

Given the impact on mobility and other everyday tasks, physiotherapy played a major role in the recovery process. It helped patients to become mobile and manage the long-term effects of surgery: "*I found that when we stopped, I got lazy with walking which would then give me more problems with the back and the ribs and the side so I find that I have to go and see a physio every six weeks*".

After lower limb amputation, as well as mobility being severely affected, patients needed to re-learn activities of daily living, including going to the toilet, taking a shower and walking. A well-fitted prosthesis was crucial to successfully gain greater mobility. If this was achieved this positively influenced their overall well-being; however, patients described having issues with their prosthesis "*My prosthesis is where I'm finding it to be quite difficult (…) I have to have a good-fitting prosthesis if I'm going to use it properly, and if my fitting is not correct then I can't really do the things I want to do*".

Even many years after end of treatment, some patients were still struggling with the impact of treatment. For example, for a young adult who was more than 5 years after treatment, the negative impact of loss of memory was still evident: "*the memory thing, that's probably the one that affects me the most (…) it's probably got a little bit worse (…) Whereas before cancer, everyone used to remark about how good my memory was*". The longevity and unpredictability of some of these symptoms was troubling for patients.

Emotional Well-being: *I think emotional has been equally hard and challenging as the physical one*

Patients managed the stages of treatment and the physical and emotional consequences of PBC differently, but there were some particular features of their experience at the stages of diagnosis, treatment decision-making and end of treatment, as described below. As mentioned previously, patients' emotional well-being was significantly affected by the way in which they experienced physical symptoms and disabilities. An overriding theme was also the impact of PBC on body image.

Diagnosis and treatment: *Does this get worse for each cycle because I'm already not coping*

At diagnosis, patients described fear, shock, panic, and feeling overwhelmed, because of this understanding and taking in the information shared at this point was difficult. It helped if they were given the opportunity to ask questions later or had someone with them at hospital appointments.

During treatment, the physical impact (as described in the previous theme) was difficult to cope with and had an impact on emotional well-being: "*It was at that stage where I thought, 'This is only going to get progressively worse.' I can remember asking at the clinic 'Does this get worse for each cycle because I'm already not coping*?'".

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All patients described how emotionally burdensome chemotherapy was due to physical impact it had on them, and gave examples of being angry and frustrated. Such feelings also had an impact on interactions with others: "*Sometimes, it [chemotherapy] makes me more responsive, a little bit angry after the chemo (…) It feels like it wasn't in my control*"

The devastating news of having to have an amputation and the lack of other options available were distressing: "*I couldn't really-, based on the journals that I was reading, and based on the information that was available at the time, amputation just seemed like such a life-altering decision*". Patients managed the decision to have an amputation by focusing on how it would increase their chances of survival ('*I'm losing my leg, but I'm not losing my life*"), how it would remove the cancer from their body. Several were helped by being able to talk with patients who had had an amputation as this had made them see that it was still possible to have a "*normal*" life afterwards. One patient had also found it helpful to talk to someone who had undergone limb sparing surgery: "*having to live a life where you are constantly having to take pain-killers, is not really a good life. Although she would do it again, it's not something she would recommend for somebody who is young*".

End of Treatment: *trying to get used to being on the other side of it*

After treatment ended, the focus and structure of care which was in place while they were going through treatment disappeared and patients found this transition challenging: "*It's difficult, sort of, coming to terms with everything (…) trying to get used to being on the other side of it and not having to deal with any of the chemo or anything*". It was during this transition period that most reflected on what they had gone through and some did not know how to cope with their thoughts and feelings such as the uncertainty associated with the prognosis which was a source of distress for patients: "*I think it was post-traumatic (…) every little thing that went wrong, I thought it had come back (…) so then I was constantly going to the doctors all the time (…) I wasn't sleeping very well*".

The regular appointments and scans that were part of follow-up care were also a source of anxiety, a constant reminder of the uncertainty and potential for recurrence, with some patients finding the period before appointments extremely stressful: "*my anxiety levels peak, you know, just before my three-monthly check-up, and then once I've had my check-up, I, kind of, come right back down (…) next three-month, the cycle starts again (...) I would certainly say the first twelve months were awful*". Anxiety and fear of recurrence was the emotional aspect of having a PBC that was shared by all patients as being difficult: "*you're left in a state of uncertainty, because obviously no-one can give you a definitive five year outlook. You're constantly having re-scans and everything. You've been through a very big thing and it's not really over. It might be, but you don't know, so that level of uncertainty is tricky at first to sort of get to grips with*"

Throughout participants' descriptions of their experiences there was a continuous negotiation between the "old" self and the changes imposed by their diagnosis and treatment, and

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the recovery process took time: "I *was very impatient and just wanted to hurry up all the process of getting, sort of, back to normality, but it was a very slow process (…) you don't get back to where you were before*". Time from diagnosis influenced patient's emotional experiences. Descriptions of feeling devastated, of losing their future, their identity were mainly present in the first two years from being diagnosed. While some patients were able to resume or continue what they were able to do before the diagnosis others found the impact on their lives completely disruptive: "*I had an amputation to deal with, so my identity from what I assumed I looked like had just been completely devastated really, because I didn't look like the way I'd grown to-, I didn't look like what I'd always looked like. I looked like something else, or someone else*".

For patients who had an amputation, the pressure of trying to get back to 'normal', and to be seen as 'normal', was more salient than in patients who did not have an amputation. There was a conscious knowledge that 'normal' was something different after the surgery and it had an impact on how they saw themselves and how others saw them. Being dependent on others was hard, undressing in public spaces took time for them to get used to. In addition to the influence of type of surgery on the patient's experience, patient's stage of life also influenced the emotional experiences. Young adults (i.e. patients aged up to 40 years old) who were transitioning to a new job, building romantic relationships, wanting to start a family, described their identify, expectations for the future in terms of employment, romantic relationships being changed and away from their peers. For example, this young adult reflected on the emotional impact of infertility caused by treatment: *"That made me feel really isolated from people (…) I've always wanted a family, so that has been probably the biggest thing that I've had to overcome. I'm still not really over it (…) it's still very painful".*

The experience of PBC was described as difficult and traumatic. While some patients described having low mood sometimes, others described having depression and made reference to suicidal ideation at some of their lowest points: "*I got put on the antidepressants. (…) I still suffer from very bad depression now (…) emotionally, I wasn't expecting to be that demanding*". Other emotions shared by a few patients included feeling guilty at having survived, and feeling particularly upset due to the impact of the illness on their family.

Besides trying to focus on positive aspects of life (with some patients stating how lucky they were to still be alive), there were examples of other strategies patients used to cope with their diagnosis, treatment and life after end of treatment including: setting up goals which gave a sense of control over what was happening to them; problem-solving, focusing on getting all the information about their diagnosis and treatment; focusing on their own children while going through treatment and in their rehabilitation; focusing on getting better "*Most of the time I was just like, 'I've got to deal with it, and I know I can get better*"'. Additionally, around half those interviewed acknowledged a positive change in their outlook in life "*I just became more relaxed about life, and probably appreciated small things more than a lot of people*".

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Changes to Body Image: *how much your body image affects your confidence*

Throughout the interviews, there were numerous examples of the way in which PBC treatment had affected patients' physical appearance, and as a result, their confidence and identity. As one said: *"I used to be a big gym goer, had a good shape on me, nice hair (…) I lost my shape completely, ended up having to just eat crap just to stay alive, lost my hair, lost my eyebrows. I felt absolutely crap"*.

While some changes were specific to the treatment phase, others were not, and the acceptance of the altered body was an ongoing challenge for patients. It was also something that patients found difficult to discuss with loved ones. Some needed professional support to deal with the impact of body changes on their self-confidence and self-image, long after treatment had ended: *"I suppose I didn't realise how much your body image affects your confidence. You know, that was something else that I would speak to the counsellor about, that perhaps I couldn't speak about to my partner at home".*

Adaptation or acceptance of their altered body was also influenced by other's perceptions. For example, questions about why someone was limping brought to the forefront the changed body "*The hardest thing I have is when someone says, 'Oh, you're limping. What have you done at the weekend?*". Many patients preferred to avoid going out because they did not want to confront other people's reactions to their altered body: "*I still can't get out, or not feel good about it. Due to amputation, I don't want my wife to take me in the wheelchair somewhere here or there so, I have to wait until chemo is over and I might get a little bit better walking with my prosthetic and get a normal body*". Others remarked on how the growth of Paralympics had a positive impact on awareness of people with prostheses by the public.

Social Well-being: *Everyone was very supportive. Everyone came around*

As described above, the physical and emotional impact of PBC and the effect of treatment on patients' body image had a profound impact on social activity and well-being. The diagnosis, duration of treatment, frequent hospitalisations, and restrictions imposed during and after end of treatment caused disruption to various aspects of the patients' social life, including their relationships (emotional and sexual), work/school and leisure activities.

Relationships: *I've got very frustrated about things; she's dealt with that and supported me*

PBC impacted on patients' relationships with family and friends. Overall, they stated that the diagnosis had brought them closer to loved ones, but for a few it meant losing contact with friends who did not support them through their experience. Patients also described a greater appreciation of the time with their loved ones after diagnosis. However, there was also an acknowledgment that sometimes patients had felt overwhelmed with treatment or its side-effects and overreacted or lost control emotionally and that there were implications on their relationships with those closer to them.

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"*She's very positive with me, I must admit. When there have been times when I've got very frustrated about things, she's dealt with that and supported me*".

Family and friends had an important role supporting patients through their treatment and recovery. The support received was both practical and emotional and had a particularly salient role for different age groups. For adolescents, parents had a central role in communicating with healthcare professionals and providing emotional and practical support. Many teenagers reported receiving messages of support though social media while they were in hospital, which made a positive difference. For adults, partners or family were mentioned as being important. Extended family support was crucial, with grandparents and sometimes friends helping parents (patient and partner) with childcare during the treatment phase: "*You know, my partner still had to go to work, and so my mum looked after my children for me, and I'm quite fortunate*".

Being at home came with its own challenges and managing day to day tasks with restricted mobility was difficult for both patients and their partners, particularly when the main carer had additional caring responsibilities: "*my wife and kids, it's affected them a lot more (…) I don't get anybody else to look after me. It's only my wife who has to do it so, there's a lot of little things which is time, energy consuming and extra load on their work as well*".

Conversely, patients who had no partner or close family, or those with elderly parents who were not able to support them, had to rely on professional carers. When their mobility was compromised this led to social isolation: *"This severely restricts me. I only really leave the house to go to [family house] (…) I think I've only gone out at night, sort of, socially, since this happened [more than 2 years ago] probably about eight times"*

After the end of treatment, young adults in particular, described the interaction with new friends, romantic partners, and co-workers being challenged by the decision to disclose their diagnosis: *"I feel it's very isolating because for me it was such a personal experience that I find it hard to share with people".* Friends were important in making patients feel 'normal' again, by not treating them differently, by showing up and continuing to do leisure activities: "*I was still on crutches, and I think I turned round to him when he dropped me home and just said, 'Thank you for this, it just made me feel normal*'" or even by using humour as described here: **"***My friends still act the same way around me. So far I have been called Long John Silver, Hopalong Cassidy, Hoppy, Ironside. Basically, they're all cracking jokes about it, and that I do like*".

Most patients also described the benefits of being in contact with other patients, either face-to-face or online. This helped them to feel normal ("*if you hear they're struggling about certain things and you can relate to them, you start realising it's normal*"), share experiences and find out practical tips (e.g. travel insurance) on how to deal with specific issues linked with their diagnosis and treatment.

Changes in fertility, sexuality & intimacy: I think that's sometimes a bit of an awkward subject

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There were varied experiences of the impact of PBC on fertility, sexuality and intimacy. For those without partners, establishing intimate relationships was difficult, due to their altered body image and side-effects of treatment: "*My CNS said, 'You can't let this define you,' but I don't see how you cannot because it affects every little tiny aspect of your place in life. As we go, I just accept no woman would really have any interest in me*". This was mainly a concern shared by participants in their twenties and thirties. For these participants the reality of infertility and the need to consider alternative methods of conceiving a child was a significant issue: *"when you're dating, when is an acceptable time to tell a girl that you're infertile? (…) I think, like, IVF, to me, obviously I've had lots of needles pushed into me and bits and pieces [but that is not the ideal way of conceiving for a woman]".*

For patients with partners, the experience of PBC had brought them closer. If there was an impact of PBC on sexuality it was limited to the treatment phase: "*Unfortunately, that can't happen at the moment. One of the side-effects of one of the pills I'm on is actually erectile dysfunction*". A few had to adapt their sexual practices to accommodate their changed bodies. Older patients (patients aged 61 or older) adopted other forms of intimacy e.g. by cuddling and supporting each other, with some acknowledging that "*I mean, our sexuality had gone some good time ago to be honest, so it's not really affected that but we're still a close couple*". However, even older patients described the impact of having an amputation on their sexual life long after the end of treatment. "*I think that's more of a mind thing from my point of view, because obviously in bed, I don't wear my prosthesis (…) we don't really discuss it*". A common experience for all participants irrespective of age was the lack of discussion of these issues with their clinical teams.

Changes to school/university/work: physically I can't keep it up for a long period of time

For teenagers and young adults being in hospital disrupted school/university life: *"I haven't been to school for three months now"*. Young people's descriptions of returning to school showed this to be an important sign of regaining normality. The way in which teachers handled changes in the young people's physical abilities, the phased return to school/university and support available was critical.

For adults, the financial impact of the diagnosis was dependant on the type of employment contract the patient had, the size of the company, availability of health insurance and personal savings or family financial support available *"that cleared off my mortgage and also meant that I didn't have to go back to work full-time (…) That took away a lot of stress and worry that I potentially could have had"* . Some patients had their jobs secured for the time they were going through treatment or they were already retired, but other patients opted for early retirement or had to reduce their working hours due to fatigue or physical impairment "*I do work five days a week in our busy periods but I've noticed that physically I can't keep it up for a long period of time*". Most patients had a phased return to work due to fatigue and mobility restrictions, as well as the emotional strain of having PBC. However, the options were limited for patients who owned their business or worked as freelancers.

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Even when patients were able to keep their jobs, young adults felt insecure because of lack of experience in the workplace and the uncertainty of being new in a job: *"I was really concerned with obviously starting a new job, the implications of this illness when I'd just started. I didn't know regarding sick pay and things like that, and whether they might just get rid of me"*. For some patients the physical limitations/restrictions imposed by treatment/surgery led to a career change or redefinition of career aspirations: "*I had to decide to do something else, something which I could do, something where I could improve and where my [physical] situation wouldn't affect my career so badly*".

These experiences were as a result of a combination of financial support available and the possibility to either return to work or find an alternative carer pathway. However, there were some cases where the consequences of treatment were so pervasive that patients were not able to work, had no access to family support or savings, and had to access benefits, this had a negative impact on their financial well-being: "*The effect it's had on me is I can't afford to go out anywhere. I can't afford to buy myself anything (…) people don't understand how tight things can be financially*".

<u>*Changes to participation in leisure activities: everything really changed. I used to play rugby*</u>

The impact of surgery and treatment on mobility affected patient's ability to participate in sports and social activities with peers and family (e.g. patients described being excluded of playing with their children). The interlinked impact of physical and social well-being resulted in a negative effect on emotional well-being: "*I was just really upset, because I want to go ice skating. I'm not allowed to go ice skating and it's just like, everybody else can do it, and I can't, and I'm just-, it's just, sort of, I was quite upset about it, because, I don't know, why did it happen to me?*"

Managing pain whilst trying to take part in the activities they enjoyed was a challenge for patients. For example, a patient who had limb sparing surgery described how he endured the pain and restrictions imposed by it: "*I do go to [activity], but I do experience extreme pain after"*. Similarly, managing fatigue and lack of energy were experienced long after the initial treatment phase for most patients. Patients identified triggers and managed their day to accommodate it, but the reality was that fatigue had a profound effect on their participation in social activities, work and school performance, as well as affecting emotional well-being: *"I'm careful about not pushing myself too much, but also it's quite unpredictable really. I know that if I do a lot then I will pay for it (…) I just allow myself to cancel plans and go to bed"*.

The Role of Healthcare Professionals: *They were experts in what they were doing*

The expertise of clinical teams was valued by participants, especially if their route to diagnosis had been long and complex, which was the case for most patients. The specialist skills and experience of clinical teams restored patients' trust in the healthcare profession, as this had often been eroded by the time patients got to diagnosis. Many patients described how the healthcare team worked collaboratively to best support them.

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All participants referred to being given information by the specialist team, asking questions and feeling supported. However, the timing and delivery of information was not always optimal, with some patients receiving information over the phone and then having to wait for the appointment with the specialist team to have their questions answered or concerns clarified. This was a period of distress for patients, with some looking for information online, which led to more distress. In contrast, when patients were guided towards specific online resources after meeting with their specialist clinical team, they were very positive about this.

The clinical nurse specialist (CNS) was a key point of contact for all patients, s/he provided practical and emotional support, information about diagnosis and treatment and support available (e.g. support groups; professional psychological support). CNS were also described as being approachable and available: "*So, if I needed help, she'd be there to help me (…) So, [CNS] was really a stable point for me to go to for any sort of information or support*".

The transition from specialist centres back to receiving care in local hospitals was at times problematic, as patients felt local care teams were not knowledgeable: "*Awful. Nobody knew what to do with me (…) They weren't equipped, at all, to deal with me. Nobody would use my Hickman line*". Where physiotherapy needs were not met by local services, some patients had to access services privately instead **"***So, I suffer a dropped foot as a result (…) I was just given some exercises to do on a sheet (…) So, I've since been seeing a private physio. They've really helped and worked wonders for my leg*." The importance of timely and expert physiotherapy support was consistently emphasised as crucial to recovery. For example, one patient said "*our local social services immediately came around. My wheelchair was already here when I came home from hospital, but they immediately built a ramp, so I could get out of the house on my wheelchair (…) They were really good and I immediately started to feel so much better*".

<u>Psychological Support: *the psychological impact of trying to recover is probably as bad*</u>

The physical consequences of PBC were so pervasive they affected patients' identity, how they saw themselves and what they could expect from their body, a body with different abilities and restrictions: *"I've just learnt to be really patient and forgiving of myself. I suppose just coming to terms with the fact that my body and my mind have changed and that's part of it. It's not just being ill and having the treatment, it's everything that comes afterwards too".* Patients dealt with the impact of PBC in different ways, for some professional support was needed.

Patients needed and accessed professional psychological support at different points in their timeline. For some it was while they were receiving treatment, for others it was a couple of months, sometimes years, after the end of treatment before they looked for professional help. The barriers to accessing support included not wanting to look weak or finding it difficult to admit that after treatment they were not feeling happy. Conversely, being advised by other patients to seek help was often a catalyst: "*Get counselling as quickly as you can, because it really does help (…) because the psychological impact of trying to recover is probably as bad, in a different way, as the*

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physical side of treatment". Having a good relationship with the General Practitioner (GP) also facilitated the process of telling a health professional they needed psychological support.

Access to psychological support was not consistent across patient's experiences. A few patients described not feeling well and not discussing this with their clinical team. At least half of the patients described feeling low mood, anxiety, mild depression for a period of time before sharing this with professionals at primary or secondary care which then triggered signposting to support or receiving information about where they could access support *("but they always ask how you're doing emotionally, as well. It was just at that point where I said, 'I'm actually not doing very well.' So, at the [hospital] they said, 'Pop along there,' and they've got details of, sort of, the surrounding areas, offices, etc., and they put us in touch with the local hospice in our area, who then helped us")*. The type of professional psychological support received varied from seeing a psychologist while in hospital, to referral by their GP ("*I'm on the waiting list for a mindfulness course, which my GP has referred me on*"), to receiving counselling over the phone or through visits at home. In a few cases, patients who had received psychological support as part of their hospital/primary care, felt they needed more support and had to look for support through charities. For teenagers and young adults who were treated in specialist centres for young people the support from youth support coordinators helped to facilitate interactions with other young people, which lessened the boredom and sadness of being in hospital. Psychological support was not needed by all patients and some relied on family and friends support or had their own coping mechanisms.

DISCUSSION

The aim of this study was to explore the experiences of patients with PBC. These cancers are characterised by: commonly being associated with severe pain as a presenting symptom, more often affecting younger people; the need for very intensive and toxic chemotherapy regimens; surgical treatments which cause life-long morbidity; and unsatisfactory cure rates.

While the altered body and level of impairment had a significant impact on patient's physical, emotional and social well-being, it was shown that support and expertise of professionals involved in patients' care, the perceived emotional and practical support received/available and patients own way of adapting to the changes caused by PBC influenced the recovery process. In addition, patients' experience was influenced by where they were in their disease trajectory (e.g. diagnosis, treatment, end of treatment, follow up, or recurrence) and their developmental stage in life (i.e. if the patient was a teenager, a young or older adult). The disease trajectory helped explain that there were certain points in their pathway that were particularly challenging for patients, such as the end of treatment when patients felt the support structures available during treatment were absent and they had more time to process what they had been through. Another important aspect was the stage where the patients were in their lives, if they were teenagers going through the challenges of school and establishing friendships or young adults establishing new romantic relationships,

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entering a new job or starting a family, or adults with children and mortgages, or older adults close to retirement or retired. These different challenges were intertwined with their experience of PBC and affected the support needed and how they managed their own experience. For example, impaired fertility was an issue for (young) adults, who reported not being able to have children as both a major loss and significant impediment to romantic relationships. Alongside these potential differences in experience, there were also commonalities as described below.

The main findings showed that physical impairment had a significant impact on a patient's identity, with patients trying to get back to 'normal' or finding a 'new normal', which meant changing or adapting their daily activities, leisure time and school/work, this extended long after the initial treatment phase. This is in line with findings from other qualitative research [21,28,14,22,24]. In addition to the physical limitations following PBC treatment, self-perception and others perceptions of the altered body interfered with patient's body image, which had an impact on confidence in social interactions (e.g. not wanting to go out) and intimate relationships (e.g. feeling less attractive), findings which are consistent with previous research[23,29,30]. This study showed that emotional well-being changed along the different phases of PBC trajectory with the first and second years after diagnosis particularly physically and emotionally challenging for patients[5]. After the end of treatment, patients reported fear of recurrence and this was constantly brought to the forefront of their minds due to regular follow up appointments and scans (mainly patients within 5 years of diagnosis). These findings are contrary to Fauske's et al.[23] findings where the majority of the eight patients with osteosarcoma in the lower extremity (who were aged 18-50 and had been diagnosed for 3 to 10 years) who took part in the study did not report fear of recurrence. These contrasting results might be due to the focus of the interviews in Fauske's et al.[23] study – how has cancer changed your life and how it has changed you as a person – and our goal to cover patients experience from the point of diagnosis onwards.

Moreover, patients in our study reported low mood, depression, anxiety and even suicidal ideation, with a few patients reporting more severe cases of depression or anxiety. This is consistent with previous research that has shown that although the majority of participants did report non-clinical anxious and depressive symptoms a significant minority did experience clinically relevant distress symptoms long after their treatment ended[19]. One last element in understanding patient's emotional well-being, shared by all, was the intensity and duration of chemotherapy and radiotherapy and how difficult it was for them physically and emotionally. Although exposure to intensive chemotherapy has been identified as adding to the risk for depression and somatic distress among both leukaemia and lymphoma survivors[31], we do not know the impact/risk of treatments on patients with PBC. Conversely, there were also reports of positive growth in our study. This was, for some, more straightforward due to their personality and/or timely professional and informal support available. But this might also reflect Bekkering's et al.[32] suggestion that similar psychological status of patients with sarcoma and healthy peers found in their study was due to

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positive emotions felt after completion of treatment and survival, in what was termed the “response shift”.

It was in the absence of certain elements of support that more negative experiences were revealed. It was shown that family and friends gave emotional and practical support, but those who did not have this support were at risk for worse emotional and social well-being. While the majority of patients in this study reported adapting to the changes in mobility, there were some who had either their mobility severely compromised or were physically constrained by side effects of treatment such as severe fatigue. These were also patients who were at higher risk for worse emotional and social well-being. Consistent with a review of survivorship after extremity sarcoma[5], this study showed that physiotherapy and psychological support were crucial to the recovery process; however, neither was reliably provided at the most needed times for all patients.

Our study had a number of limitations. A self-selected group of patients with a wide age range (13-77 years old) and at different points in their timeline (from newly diagnosed to patients who were more than 10 years from diagnosis) took part in the study. While this heterogeneity of the sample might be representative of the heterogeneity of this group of patients; some patients were reporting their experience of being newly diagnosed while others were reflecting on their experience of more than 10 years ago. It should be noted that patients described their experiences throughout the different phases and while newly diagnosed patients were able to give more detailed descriptions of that phase, this information was complemented by patients who had some perspective over their thoughts and feelings during that phase and what followed. Another limitation refers to the sample in terms of age, considering PBC peaks during adolescence/young adulthood, younger patients aged 13-24 years (current age) accounted for 15% of participants in this study. While saturation was reached on the issues raised by those who participated, a larger sample would have given us greater certainty that we could identify all the key issues across all the age groups. Nevertheless, this is a qualitative study and the aim was not generalisability, but the in-depth and credible analysis of patients’ experiences of PBC across different age groups, gender, time from diagnosis, treatment types and anatomical sites. Moreover, our study makes an important contribution to the evidence, most notably the influencing factors of patient experiences across time.

PBC has a negative impact on physical, emotional and social well-being. The extent of this impact might be felt long after treatment has ended or be specific to the end of treatment phase. Longitudinal studies are needed to better understand how patients recover, adapt and adjust to the changes imposed by diagnosis and treatment, their needs at different points in the timeline, and the impact of the support received. In addition, prospective studies, whether qualitative or quantitative, must include an exploration of changes in sexuality, fears of recurrence and impact on social and work activities, in order to underpin more holistic models of survivorship care. In clinical practice, the assessment of support in different phases of the disease trajectory is important for the identification of patients who may be at a higher risk for poor adjustment, and therefore, for targeting screening

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measures and interventions to those patients most likely to benefit. Clinical interventions must take into consideration the timing and types of support provided. In particular for this population, it was shown again the need to ensure patients have access to physiotherapy and psychology services and the relevance of support to return to work or school/university. Lastly, as improvements in survival seem elusive at the moment then improving the experiences and quality of life for this patient group is paramount. While qualitative research has detailed the disruption and changes in patient's experiences as a consequence of PBC, quantitative research has not consistently shown the impact of PBC on patient's quality of life; hence research is needed to improve patient experience and quality of life measures.

Patients with PBC experiences were influenced by the intensity of treatment and side effects that had an impact long after the treatment phase had ended. Mobility constraints, pain and fatigue influenced their participation in employment/education and leisure activities. The physical changes had also an impact on body image and their identity. Physical recovery was critical for their overall well-being and management of getting 'back to normal'. This study gave insight into factors which might influence adjustment over time, for better or for worse, showing that these affect patients at different life stages in which illness can disrupt important developmental milestones. These factors need to be taken into consideration to better understand and improve patients with PBC experiences.

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Appendix 1 - Interview schedule

Welcome and introduction; Study aim; Participation & confidentiality	Thank you for agreeing to take part in this study. I want to remind you that what we talk about will be kept confidential. Nothing you say will be shared with your healthcare team. Only a report summary of the interviews will be given out. Your name will not be on the written record of your interview and the audio recording will be destroyed when the study is complete. Can I check that you are still happy to take part and for me to record the interview? If at any time you want to stop talking just let me know and I will stop. The reason I want to talk to you is to find out what it is like being diagnosed, treated and living with a sarcoma diagnosis. There may be things that have been challenging but also things that have been okay. I want to know about the things that are most important to you. Do you have any questions before we start?
Diagnosis, treatment timeline and experiences	Do you want to tell me about the time you found out you had sarcoma? • *What type of sarcoma do you have [location]?* • *What was your treatment(s) experience?* • *What was your care experience [place of care and delivery of care]?* • *Where are you now in the journey [treatment end, relapse etc.]?* • *If treatment ended, how long ago?*
Physical health	How satisfied are you with your health? (If not, then explore why not) How important is was physical health to you? How do you ensure you are living a healthy life? Is your physical health impaired due to their sarcoma [amputation, general weakness, pain, fatigue]? • *Do you need anyone to help you with activities of daily living (washing, dressing, going to the toilet)?* • *Establish this is due to sarcoma and not present pre-diagnosis* • *How do you feel about this?* • *Do you have any late-effects (if some time out of treatment) – impact on general well-being*
Emotional health	How have you been feeling in the last 3 months? • *Do you feel sad [prompts – feel low, anxious, depressed]? How often?* • *If they do, explore what triggers this, what they do about it, how often it happens* • *Have they been able to access any professional help (if they wanted it and ease of accessing it)* How do you feel about your body image and appearance?
Social well-being	Are you employed/in education? (Explore how this has been since diagnosis) • Have there been any challenges? • Have new opportunities been available that weren't pre-diagnosis? How has your relationship with other people been since your diagnosis? • *Explore impact on family, friends and changes in relationships*. • *For patients over 16, explore sexual identify/sex life* How has your diagnosis affected your financial well-being? Do you have opportunities for leisure activities?
Final comments and wrap up	Any other information you feel is important for me to know, but that I didn't ask about? Thank you for sharing your experiences with us.

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COREQ

Domain 1: Research team and reflexivity

Personal Characteristics

1. Interviewer/facilitator - Page 6

2. Credentials – Page 1

3. Occupation – Page 1

4. Gender – Page 6/Page 1

5. Experience and training - Page 1/Page 6

Relationship with participants

6. Relationship established - Page 6

7. Participant knowledge of the interviewer - Page 6

8. Interviewer characteristics – X

Domain 2: study design

Theoretical framework

9. Methodological orientation and Theory - Page 6

Participant selection

10. Sampling – Page 8

11. Method of approach - Page 6

12. Sample size – Page 7

13. Non-participation – X

Setting

14. Setting of data collection- Page 6

15. Presence of non-participants - Page 6

16. Description of sample - Page 7

Data collection

17. Interview guide – Appendix 1

For peer review only

18. Repeat interviews – Page 6

19. Audio/visual recording - Page 6

20. Field notes - Page 6

21. Duration - Page 6

22. Data saturation – Page 19

23. Transcripts returned - X

Domain 3: analysis and findings

Data analysis

24. Number of data coders - Page 6

25. Description of the coding tree - Page 6

26. Derivation of themes - Page 6, 7 and 8

27. Software - Page 6

28. Participant checking – X

Reporting

29. Quotations presented – Page 8 to 17

30. Data and findings consistent – Page 8 to 17

31. Clarity of major themes – Page 8 to 17

32. Clarity of minor themes – Page 8 to 17

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A qualitative study exploring patients experiences of being diagnosed and living with primary bone cancer in the UK

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A qualitative study exploring patients experiences of being diagnosed and living with primary bone cancer in the UK

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ABSTRACT

Objective: The aim of this study is to explore the experiences of patients with primary bone cancer.

Design: Qualitative study design using semi-structured interviews and focus groups.

Setting: Hospitals across the United Kingdom (UK) and recruitment through UK sarcoma charities and support groups.

Methods: Semi-structured telephone/face-to-face interviews and focus groups with a purposive sample of 26 participants. Data were analysed using Framework Analysis.

Participants: Patients (n=26) with primary bone cancer aged 13-77 years. The majority were male (69%), white (85%); diagnosed within 4 years (54%); and had lower limb sarcoma (65%). Ten participants had undergone an upper/lower limb amputation (39%).

Results: The health-related quality of life domains of physical, emotional and social well-being and healthcare professionals' role were the overarching themes of analysis. The physical domain anchored patient experiences. The intensity and length of treatment, the severity of side-effects, the level of disability after surgery and the uncertainty of their prognosis had an impact on patient's self-image, confidence, mood and identity, and caused disruption to various aspects of the patients' social life, including their relationships (emotional and sexual) and participation in work/school and leisure activities. Adaptation was influenced by the way patients dealt with stress and adversity, with some finding a new outlook in life, and others struggling with finding their 'new normal'. Family and friends were the main source of support. Healthcare professional's expertise and support was critical. Rehabilitation services had a considerable role in patient's physical and emotional well-being, but inequitable access to these services was apparent.

Conclusions: This study described the impact of primary bone cancer on patients' well-being and adjustment over time with the identification of influencing factors of better/worse experiences. It showed impact was felt after end of treatment and affected patients at different life stages. Holistic models of survivorship care are needed.

Strengths and limitations of this study

- This study is an in-depth exploration of the experiences of 26 patients with primary bone cancer
- The study population varied considerably by age at, and time from, diagnosis, gender and anatomical location
- This study includes the experience of patients who had undergone amputation and other surgical interventions and treatment types
- Self-selected participants might not be representative of the experiences of patients with bone sarcoma, but our findings are consistent with previous studies and add to the research by identifying the role of influencing factors of better/worse experiences

Keywords: patient experience; primary bone cancer; quality of life

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

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Data Availability:

No additional data available.

INTRODUCTION

Primary bone cancer (PBC), also referred to as a bone sarcoma, is very rare and makes up approximately 0.2% of all cancers diagnosed in the United Kingdom (UK).[1] In 2015, 551 new patients were diagnosed with PBC in the UK.[2] The commonest subtypes are osteosarcoma and Ewing sarcoma, which have a relatively high incidence in children and young people, and chondrosarcomas, more common in older age groups.[3] There has been no major improvement in 5-year overall survival rates for patients with PBC over the past 25–30 years, with rates static at between 53 and 55%.[4]

To give the potential for cure, PBC requires complete surgical resection of the affected part of the bone. Thus, the patient characteristically experiences a lifelong limitation in physical activity. Less frequently, this can be achieved only by limb amputation, which leads to permanent disability. Given the high physical burden of the disease and its treatment and expectations of survival, poorer patient-reported outcomes[5,6] are recorded in comparison to patients with other cancer types.[7] For example, patients have reported poorer care experiences compared with other cancer types in the National Cancer Patient Experience Survey (NCPES).[8]

When conducting a literature search, it became evident that there were inconsistent findings regarding the impact PBC may have on patients.[5] While research has shown that patients may experience lower levels of physical functioning than population norms or control groups (e.g. other cancer groups or healthy peers), there appears to be a lack of reliable information on the impact on patient's emotional and social well-being.[5] Several studies comparing patients who undergo an amputation or limb sparing surgery have shown no differences in quality of life scores [9,10,11,6,12,13] ; however there are also reports of patients who had an amputation having poorer health-quality of life scores compared with patients who had other types of surgery[14]. The inconsistent results are also shown in studies that report that patients who had amputations had poorer mental health[15,16] but better feelings of self-worth[16] compared with patients who had other type of surgery. Other studies have shown that those who undergo lower limb amputation tended to report more anxiety and depression.[5,17]

In addition to studies comparing patients who undergo an amputation and other types of surgery, there are studies comparing emotional and social well-being of patients with PBC with reference values either of general population data provided with the measure, non-cancer control data collected as part of the study or from patients with other cancer types. Overall the findings reported are also inconsistent.[5] Research with children and young people has shown improvement in emotional functioning over time[7,18], however in studies with older patients Paredes et al.[19] found no improvement in cognitive and emotional well-being from diagnosis to treatment and noted that they were worse than reference norms. No clear pattern can sufficiently explain these inconsistencies, although methodological differences including range of assessment tools used,

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outcome variables measured, as well as diagnosis, anatomical location, treatment, and age groups have to be taken into consideration.[5]

Given the complexity and heterogeneity within the patient population[5,20] more research is needed to explore patients' experiences of PBC. The few qualitative studies conducted have shown the adverse impact of having PBC on patient's emotional and social well-being; however these studies focused on specific time points or specific aspects of the patient experience such as employment[21, 22], and the long-term effects of bone cancer on current daily life, body image, future possibilities and identity[23,24,25].

Both qualitative and quantitative studies shed light on the impact of PBC on patients' quality of life; however more research is needed to understand in-depth, how patients of different ages, genders and treatment experiences live with and beyond PBC.[5] Qualitative research provides richness, depth, nuance, context, multi-dimensionality and complexity of patients' lived experiences and is thus well suited to explore these patients' experiences[26]. The aim of the current study was to explore the experiences of patients with PBC across the UK.

METHODS

Design

This was a qualitative study involving patients with PBC, conducted as part of a larger qualitative study to develop a sarcoma patient-reported outcome measure – the Sarcoma Assessment Measure (SAM).

Patient Involvement

The Sarcoma Assessment Measure is about patient experience of being diagnosed and living with sarcoma, thus patient involvement is a central part of the study. Initially, the SAM research team included the Chief Executive of Sarcoma UK (Lindsey Bennister), who represented the patient perspective in the design of the study. Lindsey Bennister will continue to be involved in the study as a patient representative. Maria Onasanya, who was diagnosed with sarcoma over 5 years ago joined the study as a co-investigator and has assisted with data analysis. Both Lindsey and Maria have assisted with data analysis and are co-authors in presentations and research papers. We are also working alongside Sarcoma UK and the Bone Cancer Research Trust to ensure patients are updated about the progress of the study (through study newsletters suitable for non-specialist audience) and they receive copies of all relevant outputs. Healthcare professionals working with patients with sarcoma have been engaged in data discussion, recruitment strategies and dissemination.

Participants and setting

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The study was undertaken across the UK recruiting through specialist PBC clinical teams, sarcoma charities and patient support groups. Eligible patients were those with any diagnosis of bone sarcoma, aged over 13 (current age) and were able to communicate in English.

Patients were identified by recruiting hospitals who obtained and emailed the patient's written consent to the researcher, the researcher reaffirmed consent prior to conducting the interview. Patients less than 16 years gave their assent after consent had been received from their legal guardians.

Patients who contacted the research team after seeing information on social media from sarcoma charities were given information about the study and sent a consent form; the interviews proceeded following receipt of consent. Focus groups conducted in patient support groups were only conducted if all members consented to participate; information was given to members by the lead for the support group (either another patient or a clinical nurse specialist) and the research team were invited to the group if patients had given their consent. The study was approved by London - Stanmore Research Ethics Committee (IRAS reference: 217605).

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Data collection

Data were collected through semi-structured individual telephone interview (conducted by AM) and focus groups (led by AM and RT) conducted in sarcoma support group meetings (AM and RT are experienced researchers and were not involved in the patient's care). Young people less than 16 years took part in face-to-face interviews (parents were present during the interview). The interview schedule was developed from the literature[27] and expert opinion (patients and healthcare professionals) and included questions about diagnosis, treatment timeline and experiences; physical and emotional health; and social well-being. This was not prescriptive and was purposefully flexible to enable the researcher to explore new and emerging experiences[28]. This topic guide was shared with participants prior to the interview (Appendix 1), so they knew the topics covered (this allowed participants to decide whether they wanted to talk about the topics covered and gave them more control over the process). With permission, all interviews and focus groups were recorded and transcribed verbatim. Interviews lasted between 26 and 96 minutes and focus groups lasted 2 hours. Field notes were taken during and after the focus groups.

Data analysis

Data analysis was developed in two phases. Transcripts were broadly coded into the three functional domains of quality of life defined by Taylor et al.[29] "subjective, multidimensional and dynamic (…) and includes aspects of physical, psychological and social function" (p1831). A preliminary framework was developed by AM including these three overarching domains which then evolved during analysis of the initial transcripts. Data were analysed line-by-line with the aid of QSR NVivo 11, a specialist software package used to assist with data storage, management and retrieval. The coded data were entered into a framework that included the overarching themes of quality of life and the themes that emerged from the data (the fourth overarching theme – healthcare professional's role emerge from the data). Within each overarching domain, data were coded into more detailed themes using in NVivo codes. For example, for the overarching theme of emotional well-being, a subtheme of coping mechanisms was identified. Data exploration of patients' experiences considering age, gender, or if the patient had an amputation, helped uncover commonalities and differences. Initial coding was reviewed by the research team and revised following discussion.

RESULTS

Overall, a purposive sample of 34 of 121 patients participating in the wider Sarcoma Assessment Measure (SAM) study were diagnosed with PBC; 8 patients had craniofacial bone sarcomas and were not included in this analysis as they presented with experiences more akin to those with soft tissue head and neck sarcoma (data available from the overall study which includes patients with soft tissue sarcoma). We report here the findings from the remaining 26 participants (Table 1).

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Table 1. Participants' characteristics

		N	%
Gender	Male	18	69
	Female	8	31
Age at study	M=40.5; SD=17.9, range 13-77 years		
	13-24	4	15
	25-39	11	42
	40-65	6	23
	66+	5	19
Age at diagnosis	M=34.2; SD=18.1, range 8-77 years		
	8-24	6	23
	25-39	14	54
	40-66+	6	23
Marital status	Married/civil partnership/cohabiting	13	50
	Single	9	35
	Other	4	15
Employment	Employee full-time/part-time job	10	38
	Full-time education at school, college/university	5	19
	Wholly retired from work	5	19
	Other	6	23
Ethnicity	White British/any other white background	22	85
	Other	4	15
Time from diagnosis	0-4 (years)	14	54
	5-20+ (years)	12	46
Treatment status	On treatment	5	19
	Off treatment	21	81
Where in the body was the sarcoma	Lower limb +/- other site	19	73
	Upper limb +/- other site	5	19

	Spine +/- other site	2	8
Treatment Type	Surgery alone	8	31
	Surgery and chemotherapy	12	46
	Surgery, radiotherapy and chemotherapy	4	15
	Surgery and radiotherapy	1	4
	Chemotherapy and radiotherapy	1	4
Amputation	Yes	10	39
	No	16	61

Four overarching themes structured the analysis: the health-related quality of life domains of physical, emotional and social well-being and the role of healthcare professionals. More detailed analysis revealed that the experience of sarcoma in relation to the quality of life domains was particularly influenced by the patient's stage in life (i.e. if they were a teenager, young or older adult), the individual's coping strategies and social support, the nature and extent of adaptation that had to be made as a result of illness (referred to as the adaptive tasks of illness), and where they were in their disease trajectory (e.g. diagnosis, treatment, end of treatment, follow up, or recurrence). These will be described throughout the presentation of results below.

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Physical Well-being: *it was literally the worst pain I'd ever experienced*

The physical domain anchored patient experiences and the impact of PBC on emotional and social well-being. The intensity of treatment (surgery, cycles of chemotherapy and/or radiotherapy), the severity of side-effects, the length of treatment or the level of disability after surgery were some of the key features of PBC influencing the experiences described by patients. Patients faced specific challenges at different points in their treatment pathway and these are presented in Table 2.

The extensive surgery required had a profound impact on mobility, with lessened strength, instability, loss of flexibility, and poor balance. Patients described use of technical aids to their mobility such as crutches or a cane, use of a wheelchair or mobility scooter. Functional impairment caused major disruption in every aspect of the patients' life such as climbing stairs, lifting, driving, walking: "*going up and down the stairs is a job of work, to be honest. It is quite tiring*". Most patients described the frustration of not being able to be as active as before and this was felt long after end of treatment.

After lower limb amputation, as well as mobility being severely affected, patients needed to re-learn activities of daily living, including going to the toilet, taking a shower and walking. A well-fitted prosthesis was crucial to successfully gain greater mobility. If this was achieved this positively influenced their overall well-being; however, patients described having issues with their prosthesis which had a negative impact on their lives.

Given the impact on mobility and other everyday tasks, physiotherapy played a major role in the recovery process. It helped patients to become mobile and manage the long-term effects of surgery: "*I found that when we stopped, I got lazy with walking which would then give me more* *problems with the back and the ribs and the side so I find that I have to go and see a physio every six weeks*".

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Table 2. PBC impact on physical well-being at different points in the treatment pathway

Time Point	Impact on physical well-being
At diagnosis	Onset of pain as the trigger to seek help for many patients
During treatment	
• Radiotherapy	Fatigue and burning sensations as negative side-effects of treatment
• Chemotherapy	The majority of patients gave vivid descriptions of how debilitating their treatment was and some experienced frequent hospitalisations between chemotherapy cycles Debilitating physical effects and frequent hospitalisations
• Side-effects	Mouth ulcers, nausea, fatigue, (chest) infections, constipation, incontinence, diarrhoea, sepsis, tinnitus, hearing loss, anaemia, "*pins and needles in the ends of my fingers and the loss of some feeling in the ends of my fingers*", kidney and heart problems, and foot drop
After treatment	
• Surgery	Pain Impact on mobility: lessened strength, instability, loss of flexibility, and poor balance - For a few patients, falls were a concern and limited their activities as consequences could be severe: "*A little poodle, yapping like anything, I suddenly put all my weight, to jump out the way, onto my left leg and I felt it break. It just literally split apart*"
• Amputation and limb sparing surgery	Pain and restricted daily activities (irrespective of type of surgery). Pain management was particularly difficult in the first year after end of treatment
• Amputation	Unfamiliar sensations of 'phantom pain' were particularly disconcerting *"There doesn't seem to be a lot of options in dealing with that, especially because, for me, I don't have phantom pains every day, but I'll get it randomly (…) it can be quite disconcerting and painful"*
• Long term effects	Even many years after end of treatment, some patients were still struggling with the impact of treatment - "*The memory thing, that's probably the one that affects me the most (…) it's probably got a little bit worse (…) Whereas before cancer, everyone used to remark about how good my memory was*" (young adult, 5 years after treatment). The longevity and unpredictability of some of these symptoms was troubling for patients

Emotional Well-being: *I think emotional has been equally hard and challenging as the physical one*

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Overall, the experience of PBC was described as difficult and traumatic. Patients managed the stages of treatment and the physical and emotional consequences of PBC differently (i.e. coping strategies) and some had to have professional support. While some patients described having low mood sometimes, others described having depression and made reference to suicidal ideation at some of their lowest points: "*I still suffer from very bad depression now (…) emotionally, I wasn't expecting to be that demanding*". Anxiety and fear of recurrence was the emotional aspect of having a PBC that was shared by all patients as being difficult: "*you're left in a state of uncertainty, because obviously no-one can give you a definitive five year outlook. You're constantly having re-scans and everything. You've been through a very big thing and it's not really over. It might be, but you don't know, so that level of uncertainty is tricky at first to sort of get to grips with".* In Table 3 the particular features of patients' experience at the stages of diagnosis, treatment decision-making and end of treatment (e.g. end of treatment was a transition point when support was needed) are presented, and Table 4 has the coping strategies and how patients accessed professional support across the disease trajectory. Moreover, as mentioned previously, patients' emotional well-being was significantly affected by the way in which they experienced physical symptoms and disabilities. We will describe below how their experience of PBC affected their identity and body image.

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Table 3. PBC impact on emotional well-being at different points in the treatment pathway

Time Point	**Impact on emotional well-being**
At diagnosis	Fear, shock, panic, and feeling overwhelmed resulting in an inability to take in information It helped if they were given the opportunity to ask questions later or had someone with them at hospital appointments
During treatment	The physical impact was difficult to cope with and had an impact on emotional well-being: "*I can remember asking at the clinic 'Does this get worse for each cycle because I'm already not coping?*'"
- Chemotherapy	Chemotherapy was emotionally burdensome due to the physical impact it had on them
- Amputation	The devastating news of having to have an amputation and the lack of other options available were distressing "*I couldn't really-, based on the journals that I was reading, and based on the information that was available at the time, amputation just seemed like such a life-altering decision*"
End of Treatment	The transition to being off treatment was challenging: the focus and structure of care while they were going through treatment disappeared: "*It's difficult, sort of, coming to terms with everything (…) trying to get used to being on the other side of it and not having to deal with any of the chemo or anything*" Reflection during this transition period resulted in some patients not knowing how to cope with their thoughts and feelings such as the uncertainty associated with PBC: "*I think it was post-traumatic (…) every little thing that went wrong, I thought it had come back (…) so then I was constantly going to the doctors all the time (…) I wasn't sleeping very well*"
Follow-up	**Fear of recurrence** The regular appointments and scans (part of follow-up care) were a source of anxiety, a constant reminder of the uncertainty and potential for recurrence - the period before appointments was extremely stressful: "*my anxiety levels peak, you know, just before my three-monthly check-up, and then once I've had my check-up, I, kind of, come right back down (…) next three-month, the cycle starts again (...) I would certainly say the first twelve months were awful*" **Time from diagnosis influenced patient's emotional experiences** - Descriptions of feeling devastated, of losing their future, their identity were mainly present in the first two years from being diagnosed **Negative Emotions** - Guilt at having survived - Feeling upset due to the impact of the illness on their family

Table 4. How patients managed the impact of PBC impact on emotional well-being

Coping mechanisms	Professional support
Strategies used by patients to manage the impact of PBC: - Focus on positive aspects of life (with some patients stating how lucky they were to still be alive) - Setting up goals which gave a sense of control over what was happening to them - Problem-solving, focusing on getting all the information about their diagnosis and treatment - Focusing on their own children while going through treatment and in their rehabilitation - Focusing on getting better "*I've got to deal with it, and I know I can get better*" - Around half those interviewed acknowledged a positive change in their outlook in life "*I just became more relaxed about life, and probably appreciated small things more than a lot of people*" **How patients managed the decision to have an amputation**: - Focus on how it would increase their chances of survival: '*I'm losing my leg, but I'm not losing my life*" - Focus on how it would remove the cancer from their body - Talk with patients who had amputation to that it was still possible to have a "*normal*" life afterwards - Talk to someone who had undergone limb sparing surgery: "*having to live a life where you are constantly having to take pain-killers, is not really a good life. Although she would do it again, it's not something she would recommend for somebody who is young*"	**Timing of access to support:** no one specific time; some during treatment, for others many years from treatment ending **Access to professional support varied on the type of professional seen and how patients got access to this support:** - Not discussing how they were feeling with their clinical team - Feeling low mood, anxiety, mild depression for a period of time before sharing with professionals who signposted to support - Seeing a psychologist while in hospital - Referral by their GP: "*I'm on the waiting list for a mindfulness course, which my GP has referred me on*" - Receiving counselling over the phone - Home visits by professionals - Psychological support provided in hospital/primary care not adequate so additional support sought from charities - Youth support coordinators (only available for teenagers and young adults (TYA) treated in specialist TYA centres **Barriers to support:** - Patients not wanting to look weak - Patients finding it difficult to admit that after treatment they were not feeling happy **Facilitators of support:** - Being advised by other patients to seek help was often a catalyst- Having a good relationship with the General Practitioner (GP) facilitated patient's disclosure of needing support

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The physical consequences of PBC were so pervasive they affected patients' identity, how they saw themselves and what they could expect from their body, a body with different abilities and restrictions: *"I suppose just coming to terms with the fact that my body and my mind have changed and that's part of it. It's not just being ill and having the treatment, it's everything that comes afterwards too".* Throughout participants' descriptions of their experiences there was a continuous negotiation between the "old" self and the changes imposed by their diagnosis and treatment, and the recovery process took time. There were also numerous examples of the way in which PBC treatment had affected patients' physical appearance, and as a result, their confidence and identity. While some changes were specific to the treatment phase, others were not, and the acceptance of the altered body was an ongoing challenge for patients *"I didn't realise how much your body image affects your confidence. You know, that was something else that I would speak to the counsellor about that perhaps I couldn't speak about to my partner".*

For patients who had an amputation, the pressure of trying to get back to 'normal', and to be seen as 'normal', was more salient than in patients who did not have an amputation. While some patients were able to resume or continue what they were able to do before the diagnosis others found the impact on their lives completely disruptive: "*I had an amputation to deal with, so my identity from what I assumed I looked like had just been completely devastated really (…) I looked like something else, or someone else*". There was a conscious knowledge that 'normal' was something different after the surgery and it had an impact on how they saw themselves and how others saw them. Being dependent on others was hard, undressing in public spaces took time for them to get used to "*Due to amputation, I don't want my wife to take me in the wheelchair somewhere here or there so, I have to wait until chemo is over and I might get a little bit better walking with my prosthetic and get a normal body*". Others remarked on how the growth of Paralympics had a positive impact on awareness of people with prostheses by the public.

In addition to the influence of type of surgery on the patient's experience, patient's stage of life also influenced the emotional experiences. In particular, young adults (i.e. patients aged up to 40 years old) who were transitioning to a new job, building romantic relationships, wanting to start a family, described their identify, expectations for the future in terms of employment, romantic relationships being changed and away from their peers. For example, this young adult reflected on the emotional impact of infertility caused by treatment: *"That made me feel really isolated from people (…) I've always wanted a family, so that has been probably the biggest thing that I've had to overcome".*

Social Well-being: *Everyone was very supportive. Everyone came around*

As described above, the physical and emotional impact of PBC and the effect of treatment on patients' body image had a profound impact on social activity and well-being. The diagnosis,

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duration of treatment, frequent hospitalisations, and restrictions imposed during and after end of treatment caused disruption to various aspects of the patients' social life, including their relationships (emotional and sexual), work/school and leisure activities. The impact on these sub domains of social well-being and how these experiences varied according to patient's age or stage in life and disease related factors are shown in more detail in Appendix 2. A common element of patients' experiences was feeling closer to their family, but the caring responsibilities the patient had varied according to whether they were teenagers, young or older adults (Appendix 2). Similarly it is shown how the impact on romantic and sexual relationships and fertility concerns were mainly concerns of young adults (under 40), but the lack of discussion about sexuality and intimacy with their clinical teams was common irrespective of participant's age.

The impact of surgery and treatment on mobility and its side-effects affected patient's ability to participate in school/work, sports and social activities with peers and family (e.g. patients described being excluded of playing with their children). The interlinked impact of physical and social well-being resulted in a negative effect on emotional well-being: "*I was just really upset, because I want to go ice skating. I'm not allowed to go ice skating and it's just like, everybody else can do it, and I can't, and I'm just-, it's just, sort of, I was quite upset about it, because, I don't know, why did it happen to me?*"

The analysis of the impact of PBC on different domains of social well-being also revealed that relationships had a central role in supporting patients in the management of the impact of PBC as described below.

Relationships: *I've got very frustrated about things; she's dealt with that and supported me*

Family and friends had an important role supporting patients through their treatment and recovery. The support received was both practical and emotional and had a particularly salient role for different age groups. For teenagers, parents had a central role in communicating with healthcare professionals and providing emotional and practical support. Many teenagers reported receiving messages of support though social media while they were in hospital, which made a positive difference. For adults, partners or family were mentioned as being important. Extended family support was crucial, with grandparents and sometimes friends helping parents (patient and partner) with childcare during the treatment phase: "*You know, my partner still had to go to work, and so my mum looked after my children for me, and I'm quite fortunate*".

Conversely, patients who had no partner or close family, or those with elderly parents who were not able to support them, had to rely on professional carers. When their mobility was compromised this led to social isolation: *"This severely restricts me. I only really leave the house to go to [family house] (…) I think I've only gone out at night, sort of, socially, since this happened [more than 2 years ago] probably about eight times"*

Friends were important in making patients feel 'normal' again, by not treating them differently, by showing up and continuing to do leisure activities: "*I was still on crutches (…)and just*

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said, 'Thank you for this, it just made me feel normal'" or even by using humour as described here: **"***My friends still act the same way around me. So far I have been called Long John Silver, Hopalong Cassidy, Hoppy, Ironside. Basically, they're all cracking jokes about it, and that I do like*".

Most patients also described the benefits of being in contact with other patients, either face-to-face or online. This helped them to feel normal ("*if you hear they're struggling about certain things and you can relate to them, you start realising it's normal*"), share experiences and find out practical tips (e.g. travel insurance) on how to deal with specific issues linked with their diagnosis and treatment.

The Role of Healthcare Professionals: *They were experts in what they were doing*

The expertise of clinical teams was valued by participants, especially if their route to diagnosis had been long and complex, which was the case for most patients. The specialist skills and experience of clinical teams restored patients' trust in the healthcare profession, as this had often been eroded by the time patients got to diagnosis. Many patients described how the healthcare team worked collaboratively to best support them.

All participants referred to being given information by the specialist team, asking questions and feeling supported. However, the timing and delivery of information was not always optimal, with some patients receiving information over the phone and then having to wait for the appointment with the specialist team to have their questions answered or concerns clarified. This was a period of distress for patients, with some looking for information online, which led to more distress. In contrast, when patients were guided towards specific online resources after meeting with their specialist clinical team, they were very positive about this.

The clinical nurse specialist (CNS) was a key point of contact for all patients, s/he provided practical and emotional support, information about diagnosis and treatment and support available (e.g. support groups; professional psychological support). CNS were also described as being approachable and available: "*So, [CNS] was really a stable point for me to go to for any sort of information or support*".

The transition from specialist centres back to receiving care in local hospitals was at times problematic, as patients felt local care teams were not knowledgeable: "*Awful. Nobody knew what to do with me (…)Nobody would use my Hickman line*". Where physiotherapy needs were not met by local services, some patients had to access services privately instead **"***So, I suffer a dropped foot as a result (…) I was just given some exercises to do on a sheet (…) So, I've since been seeing a private physio. They've really helped and worked wonders for my leg.*" The importance of timely and expert physiotherapy support was consistently emphasised as crucial to recovery. For example, one patient said "*My wheelchair was already here when I came home from hospital, but they immediately built a ramp, so I could get out of the house on my wheelchair (…) They were really good and I immediately started to feel so much better*".

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DISCUSSION

The aim of this study was to explore the experiences of patients with PBC. These cancers are characterised by: commonly being associated with severe pain as a presenting symptom, more often affecting younger people; the need for very intensive and toxic chemotherapy regimens; surgical treatments which cause life-long morbidity; and unsatisfactory cure rates.

While the altered body and level of impairment had a significant impact on patient's physical, emotional and social well-being, it was shown that support and expertise of professionals involved in patients' care, the perceived emotional and practical support received/available and patients own way of adapting to the changes caused by PBC influenced the recovery process. In addition, patients' experience was influenced by where they were in their disease trajectory (e.g. diagnosis, treatment, end of treatment, follow up, or recurrence) and their developmental stage in life (i.e. if the patient was a teenager, a young or older adult). The disease trajectory helped explain that there were certain points in their pathway that were particularly challenging for patients (e.g. end of treatment). Another important aspect was the stage where the patients were in their lives, if they were teenagers going through the challenges of school and establishing friendships or young adults establishing new romantic relationships, entering a new job or starting a family, or adults with children and mortgages, or older adults close to retirement or retired. These different challenges were intertwined with their experience of PBC and affected the support needed and how they managed their own experience.

Our study adds to the growing body of evidence supporting the need to adopt an age-specific approach to the management of patients with sarcoma as their support needs might be different. Research as shown, for example, that a cancer diagnosis during teenage and young adulthood has an acute and unique impact on this critical and complex stage of life development, disrupting physical health, social and educational goals as well as psychological well-being.[30] This is further supported in a national survey of patients' experiences in the UK which showed differences according to age – teenagers and young adults were significantly more likely to report most side-effects and post-treatment concerns than older patients; elderly patients were more satisfied with the information and emotional support provided than younger patients.[31]

The main findings showed that physical impairment had a significant impact on a patient's identity, with patients trying to get back to 'normal' or finding a 'new normal', which meant changing or adapting their daily activities, leisure time and school/work, this extended long after the initial treatment phase. This is in line with findings from other qualitative research [21,32,14,22,24]. In addition to the physical limitations following PBC treatment, self-perception and other's perceptions of the altered body interfered with patient's body image, which had an impact on confidence in social interactions (e.g. not wanting to go out) and intimate relationships (e.g. feeling less attractive), findings which are consistent with previous research[23,33,34]. This study showed that emotional well-being changed along the different phases of PBC trajectory with the first and second years after

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diagnosis particularly physically and emotionally challenging for patients[5]. After the end of treatment, patients reported fear of recurrence and this was constantly brought to the forefront of their minds due to regular follow up appointments and scans (mainly patients within 5 years of diagnosis). These findings are contrary to Fauske's et al.[23] findings where the majority of the eight patients with osteosarcoma in the lower extremity (who were aged 18-50 and had been diagnosed for 3 to 10 years) who took part in the study did not report fear of recurrence. These contrasting results might be due to the focus of the interviews in Fauske's et al.[23] study – how has cancer changed your life and how it has changed you as a person – and our goal to cover patients experience from the point of diagnosis onwards.

Moreover, patients in our study reported low mood, depression, anxiety and even suicidal ideation, with a few patients reporting more severe cases of depression or anxiety. This is consistent with previous research that has shown that although the majority of participants reported non-clinical anxious and depressive symptoms a significant minority experienced clinically relevant distress symptoms long after their treatment ended[19]. One last element in understanding patient's emotional well-being, shared by all, was the intensity and duration of chemotherapy and radiotherapy and how difficult it was for them physically and emotionally. Although exposure to intensive chemotherapy has been identified as adding to the risk for depression and somatic distress among both leukaemia and lymphoma survivors[35], we do not know the impact/risk of treatments on patients with PBC. Conversely, there were also reports of positive growth in our study. This was, for some, more straightforward due to their personality and/or timely professional and informal support available. But this might also reflect Bekkering's et al.[36] suggestion that similar psychological status of patients with sarcoma and healthy peers found in their study was due to positive emotions felt after completion of treatment and survival, in what was termed the "response shift".

It was in the absence of certain elements of support that more negative experiences were revealed. It was shown that family and friends gave emotional and practical support, but those who did not have this support were at risk for worse emotional and social well-being. While the majority of patients in this study reported adapting to the changes in mobility, there were some who had either their mobility severely compromised or were physically constrained by side effects of treatment such as severe fatigue. These were also patients who were at higher risk for worse emotional and social well-being. Consistent with a review of survivorship after extremity sarcoma[5], this study showed that physiotherapy and psychological support were crucial to the recovery process; however, neither was reliably provided at the most needed times for all patients.

Our study had a number of limitations. A self-selected group of patients with a wide age range (13-77 years old) and at different points in their timeline (from newly diagnosed to patients who were more than 10 years from diagnosis) took part in the study. While this heterogeneity of the sample might be representative of the heterogeneity of this group of patients; some patients were

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reporting their experience of being newly diagnosed while others were reflecting on their experience of more than 10 years ago. It should be noted that patients described their experiences throughout the different phases and while newly diagnosed patients were able to give more detailed descriptions of that phase, this information was complemented by patients who had some perspective over their thoughts and feelings during that phase and what followed. Another limitation refers to the sample in terms of age, considering PBC peaks during adolescence/young adulthood, younger patients aged 13-24 years (current age) accounted for 15% of participants in this study. While saturation was reached on the issues raised by those who participated, a larger sample would have given us greater certainty that we could identify all the key issues across all the age groups. Nevertheless, this is a qualitative study and the aim was not generalisability, but the in-depth and credible analysis of patients' experiences of PBC across different age groups, gender, time from diagnosis, treatment types and anatomical sites. Moreover, our study makes an important contribution to the evidence, most notably the influencing factors of patient experiences across time.

PBC has a negative impact on physical, emotional and social well-being. The extent of this impact might be felt long after treatment has ended or be specific to the end of treatment phase. Longitudinal studies are needed to better understand how patients recover, adapt and adjust to the changes imposed by diagnosis and treatment, their needs at different points in the timeline, and the impact of the support received. In addition, prospective studies, whether qualitative or quantitative, must include an exploration of changes in sexuality, fears of recurrence and impact on social and work activities, in order to underpin more holistic models of survivorship care. In clinical practice, the assessment of support in different phases of the disease trajectory is important for the identification of patients who may be at a higher risk for poor adjustment, and therefore, for targeting screening measures and interventions to those patients most likely to benefit. Clinical interventions must take into consideration the timing and types of support provided. In particular for this population, it was shown again the need to ensure patients have access to physiotherapy and psychology services and the relevance of support to return to work or school/university. Lastly, as improvements in survival seem elusive at the moment then improving the experiences and quality of life for this patient group is paramount. While qualitative research has detailed the disruption and changes in patient's experiences as a consequence of PBC, quantitative research has not consistently shown the impact of PBC on patient's quality of life; hence research is needed to improve patient experience and quality of life measures.

Patients with PBC experiences were influenced by the intensity of treatment and side effects that had an impact long after the treatment phase had ended. Mobility constraints, pain and fatigue influenced their participation in employment/education and leisure activities. The physical changes had also an impact on body image and their identity. Physical recovery was critical for their overall well-being and management of getting 'back to normal'. This study gave insight into factors which might influence adjustment over time, for better or for worse, showing that these affect patients at

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different life stages in which illness can disrupt important developmental milestones. These factors need to be taken into consideration to better understand and improve patients with PBC experiences.

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Appendix 1 - Interview schedule

Welcome and introduction; Study aim; Participation & confidentiality	Thank you for agreeing to take part in this study. I want to remind you that what we talk about will be kept confidential. Nothing you say will be shared with your healthcare team. Only a report summary of the interviews will be given out. Your name will not be on the written record of your interview and the audio recording will be destroyed when the study is complete. Can I check that you are still happy to take part and for me to record the interview? If at any time you want to stop talking just let me know and I will stop. The reason I want to talk to you is to find out what it is like being diagnosed, treated and living with a sarcoma diagnosis. There may be things that have been challenging but also things that have been okay. I want to know about the things that are most important to you. Do you have any questions before we start?
Diagnosis, treatment timeline and experiences	Do you want to tell me about the time you found out you had sarcoma? • *What type of sarcoma do you have [location]?* • *What was your treatment(s) experience?* • *What was your care experience [place of care and delivery of care]?* • *Where are you now in the journey [treatment end, relapse etc.]?* • *If treatment ended, how long ago?*
Physical health	How satisfied are you with your health? (If not, then explore why not) How important is was physical health to you? How do you ensure you are living a healthy life? Is your physical health impaired due to their sarcoma [amputation, general weakness, pain, fatigue]? • *Do you need anyone to help you with activities of daily living (washing, dressing, going to the toilet)?* • *Establish this is due to sarcoma and not present pre-diagnosis* • *How do you feel about this?* • *Do you have any late-effects (if some time out of treatment) – impact on general well-being*
Emotional health	How have you been feeling in the last 3 months? • *Do you feel sad [prompts – feel low, anxious, depressed]? How often?* • *If they do, explore what triggers this, what they do about it, how often it happens* • *Have they been able to access any professional help (if they wanted it and ease of accessing it)* How do you feel about your body image and appearance?
Social well-being	Are you employed/in education? (Explore how this has been since diagnosis) • Have there been any challenges? • Have new opportunities been available that weren't pre-diagnosis? How has your relationship with other people been since your diagnosis? • *Explore impact on family, friends and changes in relationships.* • *For patients over 16, explore sexual identify/sex life* How has your diagnosis affected your financial well-being? Do you have opportunities for leisure activities?
Final comments and wrap up	Any other information you feel is important for me to know, but that I didn't ask about? Thank you for sharing your experiences with us.

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Appendix 2. PBC impact on social well-being

Social Well-Being Domain	Impact
Relationships	**Impact on patients' relationships with family and friends** Diagnosis brought them closer to loved ones, but loss of contact with friends who did not support them through their experience. Greater appreciation of the time with loved ones after diagnosis **Impact of feeling overwhelmed by treatment** Resulted in implications on relationships with those closer to them **Additional caring responsibilities for partners** Challenges in managing day to day tasks with restricted mobility was difficult for both patients and their partners, particularly when the main carer had additional caring responsibilities: "*my wife and kids, it's affected them a lot more (…) I don't get anybody else to look after me. It's only my wife who has to do it so, there's a lot of little things which is time, energy consuming and extra load on their work as well*". **Challenge of disclosing diagnosis to others** At the end of treatment interaction with new friends, romantic partners, and co-workers was challenged by the decision to disclose their diagnosis (particularly in young adults): *"I feel it's very isolating because for me it was such a personal experience that I find it hard to share with people"* **Sexuality, fertility and intimacy** <u>Patients without partners (mainly patients aged 20-30 years old)</u> - Difficulty in establishing intimate relationships: altered body image and side-effects of treatment: "*I just accept no woman would really have any interest in me*" - Infertility and the need to consider alternative methods of conception: *"when you're dating, when is an acceptable time to tell a girl that you're infertile?".* <u>Patients with partners</u> - Brought couples closer together - If there was an impact of PBC on sexuality it was limited to the treatment phase: "*Unfortunately, that can't happen at the moment. One of the side-effects of one of the pills I'm on is actually erectile dysfunction*" - Adaptation in sexual practices to accommodate changed bodies <u>Older patients (patients aged 61 or older)</u> - Adopted other forms of intimacy e.g. by cuddling and supporting each other

	- “*I mean, our sexuality had gone some good time ago to be honest, so it’s not really affected that but we’re still a close couple*” - Impact of having an amputation on sex life long after the end of treatment. “*I think that's more of a mind thing from my point of view, because obviously in bed, I don't wear my prosthesis (…) we don't really discuss it*”
School/University/ Work	**Teenagers and young adults** Being in hospital disrupted school/university life: *“I haven’t been to school for three months now”*. Returning to school showed this to be an important sign of regaining normality but depended on the way teachers handled changes in physical abilities. Phased return to school/university and support available was critical. Feelings of insecurity because of lack of experience in the workplace and the uncertainty of being new in a job: *“I was really concerned with obviously starting a new job, the implications of this illness when I’d just started. I didn’t know regarding sick pay and things like that, and whether they might just get rid of me”* **Adults** Some patients had their jobs secured for the time they were going through treatment or they were already retired Some patients opted for early retirement or had to reduce their working hours due to fatigue or physical impairment Most patients had a phased return to work due to fatigue and mobility restrictions, as well as the emotional strain of having PBC, but the options were limited for patients who owned their business or worked as freelancers For some patients the physical limitations/restrictions imposed by treatment/surgery led to a career change or redefinition of career aspirations: “*I had to decide to do something else, something which I could do, something where I could improve and where my [physical] situation wouldn’t affect my career so badly*” **Financial impact** The experiences were a result of a combination of financial support available and the possibility to either return to work or find an alternative carer pathway - type of employment contract, size of the company, availability of health insurance and personal savings or family financial support available had an impact on financial wellbeing - When the consequences of treatment were so pervasive, patients were not able to work, had no access to family support or savings, and had to access benefits, this had a negative impact on their financial well-being: “*The effect it’s had on me is I can’t afford to go out anywhere. I can’t afford to buy myself anything (…) people don’t understand how tight things can be financially*”.

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Participation in leisure activities	**Impact of pain** Managing pain whilst trying to take part in the activities they enjoyed was a challenge - e.g., following limb sparing surgery a patient described how he endured the pain and restrictions imposed by it: “*I do go to [activity], but I do experience extreme pain after”* **Impact of fatigue** Fatigue and lack of energy were experienced long after the initial treatment phase Patients identified triggers and managed their day to accommodate it, but the reality was that fatigue had a profound effect on their participation in social activities, work and school performance, as well as affecting emotional well-being: *“I’m careful about not pushing myself too much, but also it’s quite unpredictable really. I know that if I do a lot then I will pay for it (…) I just allow myself to cancel plans and go to bed”*.

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COREQ

Domain 1: Research team and reflexivity

Personal Characteristics

1. Interviewer/facilitator - Page 6

2. Credentials – Page 1

3. Occupation – Page 1

4. Gender – Page 6/Page 1

5. Experience and training - Page 1/Page 6

Relationship with participants

6. Relationship established - Page 6

7. Participant knowledge of the interviewer - Page 6

8. Interviewer characteristics – X

Domain 2: study design

Theoretical framework

9. Methodological orientation and Theory - Page 6

Participant selection

10. Sampling – Page 8

11. Method of approach - Page 6

12. Sample size – Page 7

13. Non-participation – X

Setting

14. Setting of data collection- Page 6

15. Presence of non-participants - Page 6

16. Description of sample - Page 7

Data collection

17. Interview guide – Appendix 1

For peer review only

18. Repeat interviews – Page 6

19. Audio/visual recording - Page 6

20. Field notes - Page 6

21. Duration - Page 6

22. Data saturation – Page 19

23. Transcripts returned - X

Domain 3: analysis and findings

Data analysis

24. Number of data coders - Page 6

25. Description of the coding tree - Page 6

26. Derivation of themes - Page 6, 7 and 8

27. Software - Page 6

28. Participant checking – X

Reporting

29. Quotations presented – Page 8 to 17

30. Data and findings consistent – Page 8 to 17

31. Clarity of major themes – Page 8 to 17

32. Clarity of minor themes – Page 8 to 17

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