

# Negative and Positive Consequences of Cancer Treatment Experienced by Long-term Osteosarcoma Survivors: A Qualitative Study

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**Abstract.** *Background:* Our study aimed to explore how survivors of osteosarcoma of the lower extremity experience physical and psychosocial late effects several years after undergoing arduous treatment. A qualitative, phenomenological and hermeneutic approach was applied. *Materials and Methods:* Osteosarcoma survivors ( $n=8$ ) previously treated at the Norwegian Radium Hospital, Oslo University Hospital, participated in the study. In-depth and semi-structured interviews were conducted. The interviews were analyzed using inductive thematic analysis. *Results:* Three to ten years after diagnosis, the majority of participants had experienced both negative and positive consequences following treatment. Changes in activity and exclusion from participation in different areas were the most challenging consequences. Several of their experiences are similar to those described by people with disabilities. *Conclusion:* It is important to understand osteosarcoma survivors' own experiences in order to assist those who struggle to reorient in life and to construct a new identity for themselves.

Following cancer treatment, survivors may face a wide range of late physical and psychosocial consequences (1). Analysis by the National Health Interview Study in the United States showed that respondents with a history of cancer were more likely to report poor health, as well as to have other chronic conditions, disabilities and limitations to their usual activities (2, 3). Similarly, UK-based studies have found that cancer

survivors report poor general health (4, 5). Other studies note that late effects involve fatigue and cognitive challenges (6-8), fear of recurrence (9), sexuality and reproductive issues (10-14), changes in body image (15-17), and limitations in vocational life (18-21).

Primary bone sarcomas are rare and represent fewer than 0.2% of all new cancer cases (22). Osteosarcoma is the most prevalent entity and occurs mainly among adolescents and young adults (23-25). The current relative five-year survival rate for these age groups is between 60% and 70% (26, 27). Curative treatment for osteosarcoma combines the surgical removal of the primary tumour and extensive adjuvant combination chemotherapy (28). Today, almost 90% of patients with osteosarcoma of the lower extremity can be radically operated on using a limb-sparing procedure; hence amputation of the affected extremity can be avoided (27, 29). One difficulty in limb salvage surgery is that internal prostheses only have a limited lifespan. Prosthetic failure, including fractures (30), and a high rate of reoperation thus remain a challenge (31). Extensive surgery also implies the removal of muscles, which may result in weakness associated with movement and balance problems. In addition, insufficient bending of the knee is common (31, 32). The surgical sequelae, as well as toxic effects of chemotherapy (33), mean that osteosarcoma survivors frequently experience considerable health challenges and disabilities as a long-lasting consequence of treatment. Examples of such consequences include difficulties with practical challenges of everyday life and psychosocial life, as well as exclusion from recreational (34, 35) and professional life (20).

The quality of life (QoL) among long-term survivors of lower-extremity osteosarcoma has previously been investigated. Lower QoL scores in terms of physical functioning have been consistently reported, both compared to other cancer survivors, as well as to individuals without cancer from a matched population (36-39). In contrast, other quantitative studies of patients with osteosarcoma concerning

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education, employment, fertility, and marital status have revealed inconsistent results (40-42). Methodological differences and challenges have been emphasised in two systematic reviews to account for these inconsistent findings. (32, 43). A key dimension in most studies cited above is the comparison of function among patients with lower extremity osteosarcoma following limb salvage procedures or amputations. Qualitative research addressing the individual and long-term psychosocial effects of treatment is sparse. Despite this limited evidence, bone cancer treatment clearly has a profound impact on survivors in many life domains. A qualitative study concerning mothers' views of their children (N=12) with bone sarcomas showed that cancer and its treatment had a great impact on their lives (35). The participants described how functional impairment caused major disruption to their child's everyday life, school, sports activities and social life. One study reported that bone sarcoma treatment had an adverse effect on the work status of patients, with 25% of participants (N=9) describing an 'altered' work status such as prolonged periods of disability or unemployment (20). Parsons *et al.* aimed to characterize the experiences of adolescent and young adult osteosarcoma survivors (N=14) in general, as well as in relation to resuming vocational pursuits (34). They found that participants engaged in three types of 'work': illness work (*i.e.* going through treatment and struggling with consequences), identity work, and vocational work. Importantly, their participants described an active process of 'identity work' – work that is characterised by 'becoming other' through self-reflection and effort (34). A qualitative study of bone cancer survivors' experiences after extensive surgery in the hip/pelvis region demonstrated how treatment and long-term consequences affect bone sarcoma survivors (*i.e.* how late effects have affected their current daily life, as well as influencing their future possibilities and their identity) (44). These studies show that moving from being healthy prior to cancer to being functionally impaired following treatment is a transformation that has an extensive impact on a survivor's identity, daily life, and vocational life.

A cancer survivor is someone who is "living with or beyond cancer" (45). It is, however, not necessarily true that the absence of disease is the same as having good health and a meaningful life (46). The aim of medical treatment should thus not only be survival. Although technical cancer treatments are prerequisites for a cure, they are not sufficient. For those affected by cancer, the actual life they are living, their relationships and their environment are important. From a psychosocial and sociocultural perspective, cancer can be understood as a serious incident in life, as a biographical disruption (47-49), as well as a loss of self (50). Firstly, the patient with cancer is a person for whom illness has suddenly broken into their daily life and altered their life experiences. Not only is their own previous life story disrupted, so is their

identity (47, 48, 51). When affected by a serious disease and its consequences, people often have to reorient their life and construct something of a new narrative using their affected body as the starting point. This might help them to create new meaning in life, both in terms of understanding themselves and the world of their illness, as well as recognising new limitations in their lives (47, 52).

The aim of the current study was to understand the long-term experiences of osteosarcoma survivors from a sociocultural and psychosocial perspective through the use of a qualitative methodology. We, therefore, explored how cancer, its treatment and its long-term consequences affect the daily lives of former osteosarcoma patients. Two particular characteristics of the participants are significant: the extent of the surgery they have undergone and the fact that many of them are young. Specifically, we sought to answer the following research questions: How has cancer changed the lives of osteosarcoma survivors in the long-term? Has cancer changed them as a person and, if so, in what way?

## Materials and Methods

**Study design.** In order to explore how former osteosarcoma patients experience life after cancer, we applied a phenomenological experience-based and hermeneutic interpretation-based perspective on their illness. In phenomenological research, the aim is to investigate individual human experiences (phenomena) as manifested in daily life and in specific situations (49, 53-55). Hermeneutics is concerned with the various dimensions of meaning and how to achieve understanding, in addition to how phenomena have to be interpreted in order to be understood. Here, understanding develops through the entire process based on both the participant's and the researcher's pre-understanding, as well as the historical and cultural context (56, 57). This process influences the generation of the research questions, the interview process and the subsequent analysis (54, 56, 58). Svenaeus, inspired by the philosophy of Heidegger (55), has developed a phenomenological approach to health and illness (48, 49). He claims that serious illness forces people to reorient in life. Patients' ailments are multidimensional phenomena and are, at the same time, meaningful on several levels (49). We attempted to connect the biological concept of disease with patients' experiences, as well as the psychosocial and sociocultural aspects of illness, in order to gain new insight and understanding of a phenomenon.

**Participants and recruitment.** We interviewed former osteosarcoma patients who were treated at Norwegian Radium Hospital, Oslo University Hospital (NRH OUS). Potential respondents were identified from the prospective clinical sarcoma database (Med Insight) at NRH OUS, which treats approximately 80% of osteosarcoma patients in Norway (25). Out of the eight eligible patients contacted, all agreed to participate.

Four men and four women, all aged between 18-50 years, who had been treated for osteosarcoma in the lower extremity participated in the study. All were treated with surgery. Their tumors were removed and their bones reconstructed using an internal

prosthesis or, in one case, an allograft. Seven participants received chemotherapy. All participants were diagnosed between 2002 and 2009 (Table I), and were followed-up at the oncological or orthopaedic surgical outpatient clinic at NRH OUS. None had experienced recurrence of the disease for at least three (and up to a maximum of 10) years following primary diagnosis.

The treating physician at NRH OUS made initial contact with the participants. The first author, LF, then provided further details regarding the project before the participants gave informed consent. The interviews were conducted face-to-face by LF in connection with a routine clinical follow-up appointment that took place at NRH OUS.

**Procedure.** This research was anchored by the fundamentals of the Declaration of Helsinki. Permission to conduct the interviews, as well as to collect and store sensitive data, was obtained from both our Institutional Review Board and the Regional Committee for Medical Research Ethics, REK South East, approval number 2012/918. All information was stored confidentially. The analyses were carried out from anonymised transcripts.

The interviews lasted an average of 54 minutes (range=30-104 minutes), and were audiotaped and then transcribed verbatim by a medical secretary at NRH OUS. Field notes were written following each interview in order to document any observations made by the interviewer. The interview guide referenced the following topics: How the participants had experienced the functional, practical, psychosocial, emotional and vocational consequences of their disease and treatment; and whether their cancer experiences had changed them as a person. The interview guide was designed to allow the participants to chronologically tell their whole cancer story. The guide was, however, only loosely followed, so that the participants were able to significantly influence the depth of the interview. Using this approach, structure and meaning are produced jointly by the participant and the researcher (59). As such, certain interpretations emerged during the interview on both sides. This enabled the confirmation or rejection of the interviewer's perceptions of what the participants expressed (54).

**Data analysis.** The participants' accounts were analyzed by hand by LF and GO using thematic analyses. The analysis took place in stages and followed an inductive strategy (60) within a contextualised framework (54). Firstly, the transcribed interviews were read through to gain an overall impression and to identify preliminary themes. Secondly, the entire dataset was coded in detail, then organised into themes, and concepts were developed. The themes were reflected on in accordance with the study's objectives and were also compared against the available literature and theory highlighting the interaction between cancer and the patient's life experiences.

## Results

In the present study, the participants expressed the consequences and changes in their lives some three to ten years after diagnosis of osteosarcoma. The majority of participants communicated both negative and positive experiences (Table II). Only one of the participants reported that he did not feel there existed any negative consequences

Table I. Demographic and clinical data.

	Number
Gender	
Female	4
Male	4
Age, years	
18-25	3
26-35	1
36-50	4
Diagnosis	
Osteosarcoma	8
Time of diagnosis	
3-5 Years prior to study	3
6-10 Years prior to study	5
Treatment	
Surgery	8
Prosthesis	7
Allograft	1
Chemotherapy	7

of his cancer treatment. In contrast, another participant had nothing positive to say about his current life following cancer treatment. More than half of the participants expressed how their experiences had influenced them positively with regard to how they now look at life and themselves. Below, we address the negative and positive consequences that were identified in our analyses.

Participant quotes are identified by participant ID and gender. For instance, MP1 indicates male participant number 1, while FP3 indicates female participant 3.

**Negative consequences of osteosarcoma treatment. Daily practical challenges.** Seven out of the eight participants expressed negative consequences in terms of impaired function and reduced mobility. They had all undergone limb-sparing surgery with an internal prosthesis in either the femur or tibia, with all involving the knee joint. Most still experienced reduced mobility, lessened strength, stability and flexibility, as well as poor balance. Climbing stairs was challenging for six of them. As FP3 described: "I cannot use the stairs properly. I walk like a small kid, one step at a time." Carrying any load, particularly on the stairs, can be difficult or impossible. MP1 stated the following: "When I go shopping, I have to call my son or daughter to help with carrying." This also applies to lifting and carrying heavy things in the house, especially in connection with housework. More than half of the participants mentioned that they walked with a limp. Some needed to use walking poles when they moved over rough terrain. In Norway, there is snow in wintertime and it is slippery for several months of the year, making it difficult for the participants to move when

Table II. Key findings extracted from the individual patient accounts.

Finding	Participant (gender and number)							
	MP1	MP2	FP3	FP4	FP5	MP6	MP7	FP8
Age, years	>35	<35	>35	<35	>35	<35	<35	>35
<b>Negative consequences</b>								
Impaired functionality in daily life	X		X	X	X	X	X	X
Balance problems	X		X	X	X	X	X	X
Climbing stairs is challenging	X		X		X	X	X	X
Limping	X		X	X	X	X		X
Carrying loads is difficult	X		X		X	X	X	X
Afraid of falling	X		X	X	X	X	X	X
Broken leg/prostheses	X	X						X
Reduced ability to bend the leg/knee	X		X	X	X	X	X	X
Fatigue/tired/less energy			X	X	X	X	X	X
Less sporting activities than before	X		X	X	X	X	X	X
Loss of important hobbies	X		X	X	X	X	X	
Less social				X	X	X	X	X
Limited activities with own children	X		X		X			
Do not expose altered leg	X				X		X	
Negative impact on work/studies						X		X
Concerns about recurrence				X	X			X
Affected fertility		X			X	?	?	X
<b>Positive consequences</b>								
Appreciate every day more		X	X				X	X
More mature, confident, stronger	X		X	X			X	X
New knowledge and experiences	X		X	X	X		X	
More empathetic	X			X	X			X
Changed lifestyle				X				

Participants are identified by participant ID and gender. For instance, MP1 indicates male participant number 1 while FP3 indicates female participant 3.

outdoors. The majority emphasised that icy roads were a challenge and some even had to use crampons. MP6 had to use crutches throughout the winter, although he was not dependent on them during the summer. He and many other participants reported being afraid of falling and breaking their prosthesis. Three of them have already been re-operated on. As MP6 said: "I have no control. If I go out without crutches, I am afraid of falling. The risk of falling is much greater if I were to lose my foothold with the healthy leg. Then I have no chance of standing up on my feet with just the injured leg."

**Lack of energy.** More than half of the participants stated that they struggle with low energy and tiredness. Some were exhausted after work and a few had to spend considerable time simply recovering when they returned home. Two of those with children said that this lack of energy also affected their interaction with their children. FP3 stated: "I notice that this has an impact on the children. It does – especially the oldest one." The two participants, both females, said that they had to leave some of the interaction, activity and homework

to their husbands. FP5 expressed that she devoted most of her energy to working full time: "I have less energy at home and I often leave my husband to take care of the little girl, and among other things help her with homework. I just have to sit on the couch, take up knitting and just be myself."

Only two participants mentioned that the late effects of their cancer treatment had affected their current studies or vocational life. FP8 said she would like to work but struggled with fatigue that limited her job opportunities. Eventually, she hoped to be able to engage in some non-strenuous work. She expressed dissatisfaction with her current situation: "Yes, I feel somewhat ashamed that I stay at home. I should be able to do an easy job."

**Body image concerns.** In addition to functional challenges, extensive surgery also implies changes in body image. As mentioned above, more than half of the participants limped, although they all stated that it did not bother them much. All participants had scars, and one also had discolouration of the skin. Many had a leg that is much thinner than normal because the muscles were removed.

The majority of participants commented on their altered body. Three of the participants reported that they did not like to expose their body because either they thought it did not look neat or else they preferred not to have to focus on their cancer. In particular, FP5 stated that she was not happy with her appearance following cancer treatment. Before treatment, she was slim and fit; she had liked what she saw in the mirror. After, one leg was thinner than the other and looked strange. In addition, she had put on weight: "I was proud of my body before and I think I had a decent body, but I do not think so now." The other five participants were not ashamed of their altered bodies. They undressed as before when they were among others, for example at the beach. In contrast, the three participants mentioned above were reluctant to undress or to wear shorts or a bikini when they were with others. They were especially reluctant to show the injured leg in their hometown where people they knew might recognise them. Interestingly, they felt it was okay to undress abroad where no one knew them. MP7 is a young man who had chosen not to expose his mutilated leg: "There is a vast difference between my two legs. [...] The operated leg is thinner than the other, and there are scars after skin grafting on the other leg." He expressed a desire to travel to warmer places abroad and said that there, where no one would know him, he would wear just swimming trunks. It appears that for some participants it is harder to reveal a deviating body among people they know than among strangers.

*Changes and losses in leisure time and social life.* The majority of participants were active and took part in one or more sports before being diagnosed with cancer. For almost all of the participants, their current activity levels were drastically reduced. Most were no longer able to run, ski, play football or bandy (floor ball), or do aerobics, dance or ride horses. For many of them, their leisure time had completely changed. FP5, a former trainer and athlete, felt her activity opportunities to be considerably limited when compared to what she had been able to do before having cancer: "I had to resign from my trainer job and I miss my classes a lot. Leisure activities that others do, such as mountaineering and skiing, I cannot do any more. I miss that during wintertime." She was, however, still physically active and worked out at a gym. FP5 also mentioned that another woman she knew who was treated before her and had to have her leg amputated, was able to return to her job as a trainer despite her prosthesis. FP5 herself had limb-preserving surgery and so has to be more careful. She would not, however, have preferred to be an amputee: "I see that she is struggling much more than I, anyway."

For most participants, the loss of physical activity is a significant loss. They have lost a meaningful hobby and their identity has changed from being physically active to being partially disabled. MP7 is a young man who lost his main

hobby, football, which previously took up all of his leisure time. However, even though he said he misses football, he stated that his experiences with cancer were not solely negative, commenting that he had more spare time, time that he did not have when playing football. This has changed his focus from football to studies: "My grades really went up following cancer treatment, from average to above average." However, it should be mentioned that several of the participants still exercised in the gym or in controlled forms. It was particularly important for them to keep active, albeit at a different level and scale than before.

All three participants with children also expressed their loss at not being able to participate in some of their children's activities, especially sport and outdoor activities. For FP5, this was a great loss: "My daughter has a disabled mother, and she is often sad when I cannot be with them. I can never be with them."

Not being able to participate in sport and outdoor activities can also affect former cancer patients' social lives. For MP7, the football field was a focal point for social gathering: "Before I had cancer, I had a lot more friends. After I had to quit football, I lost contact with many... The cancer served as a filter, a few friends got through and it is them I still have contact with." Taking part in outdoor activities, such as skiing in the mountains, is also a venue for socialising. FP5 said that her friends did not always recognise that she can no longer participate when they plan their trips together: "No, people forget a little. Planning things that I cannot attend, and those things. [...] I think that it is obvious that people do so because they do not think that I am disabled and cannot manage what they do. And then, I become sad."

*Infertility concerns.* As a result of chemotherapy, three participants knew that their fertility was reduced due to either reduced sperm quality or premature menopause. As MP2 stated: "The only current late effect I have is reduced fertility. Nothing else. [...] Of course, it is very sad. Indeed." In addition, two of the male adolescents still lived with uncertainty regarding whether or not they would be able to have children. They were both worried about how this would influence dating and family planning.

*Existential considerations.* Most of the participants expressed thoughts and concerns about having lost much of what had previously been of vital importance in life due to the late effects of cancer treatment. In terms of a fear of recurrence, half of the participants stated that they did not think much about cancer, and that they were not afraid of relapse. Three mentioned that they sometimes thought about it, but that they did not worry any more. MP7 said that he rarely thought about it: "Only the days I'm down here [for his regular follow up at the hospital], maybe a few days after I return

home. Nevertheless, for my part, I would have managed another year of chemotherapy. That would not have been a problem." On the other hand, one young woman mentioned that she was very afraid of experiencing a recurrence and thus having to receive chemotherapy again. None of the participants considered their previous lifestyle to be a cause of their cancer. They expressed no guilt. Only one mentioned that she had, as a result of her cancer, changed her lifestyle, now being more aware of optimal nutrition. Two reported that they had actually become more relaxed about what they ate because they wanted to enjoy life. The others did not report any changes in lifestyle.

*Still struggling.* The majority of participants did not report any current psychological or emotional struggles, such as feelings of uselessness and low mood. Only one participant, MP6, expressed that cancer had had a significant negative impact on his life. He described how cancer had done nothing positive for him. Sport-related activities were a significant part of his previous daily life and he was no longer able to take part in those activities. He did not have any new hobbies and so had lost much of his social contact: "As I said, my leg does not work. In wintertime, I sit indoors most of the time. I do not do much." Not only had his active and social life been affected, lack of energy and motivation had also negatively affected his education and delayed the progress of his studies.

*Positive changes as a consequence of cancer experience.* Towards the end of the interview, after we had mostly talked about the challenges following cancer treatment, two questions were raised: What was the biggest change in your life after you had cancer? How has cancer impacted or changed you as a person? These were intended to be summary questions whereby participants could reflect on their current situation. Seven out of the eight participants used this opportunity to talk about the positive effects cancer had had on their lives and for them as individuals. More than half of the participants expressed how the disease had influenced them positively with regard to how they now looked at life and themselves. They commented that they had gained valuable knowledge and experience through cancer, although they also expressed an ambivalence towards their positive feelings.

*Changed sense of life and self.* MP2 stated that he had always been an optimistic person and, through much exercise, he had achieved nearly the same level of function as before. He expressed that the main difference between his life before and after treatment was his appreciation of every new day: "Now, I appreciate each day even more. Yes, I do [...]. I say we should be grateful for every day that goes well." He said he thought of all those who had had cancer

and did not survive. He stated being thankful that he is one of those who had coped well. More than half of the participants reported that cancer had made them stronger, more mature, tougher, and more confident. They felt better prepared to face challenges. FP3 expressed how, in several contexts, cancer had had a constructive impact on her life: "I can handle more. Can handle things better than others might do. [...] Learned to speak up a little more, and have become a little tougher. [...] Cancer made me stronger, it has. Now, I am more positive about things, maybe. [...] I have a new perspective on life. Yes." FP4 emphasised that she had changed in a positive manner after all she had experienced. She pointed out, however, that she had had cancer when she was still in her teens. So, in addition to personal growth stemming from the cancer, she had also generally matured.

*Compassion for others.* Another positive effect that half of the participants talked about was how their relationships with others had changed. They expressed having become more patient, more empathetic, more relationally oriented, and more understanding towards others. FP4 emphasised this: "I have been quite altered. [...]. I think I have become less judgmental, I think in a way that each person has their own story. You never know what people have experienced or gone through. If something happens to someone, I put myself in his or her situation. I think about how others experience things." Only one participant mentioned that she might have developed a lower threshold for the complaints of others after what she had been through.

*Ambivalence.* Three participants had a clear focus on the positive aspects of their cancer experience. However, there was a certain ambivalence and some contradiction in this regard. Despite the negative late effects, they went so far as to say they were glad that they had had cancer due to their positive experiences. They also stated, however, they would not have had cancer if they could have had a choice. MP7 is a young man for whom cancer had resulted in both negative changes and positive experiences: "No. If I could go back, I would not have had cancer. However, I do not think I would want to be without the experience— all what I have gone through [...]. Yes, both the scars and injuries are trivialities compared to the positive experience." FP4 is a young woman who has reflected on how cancer has influenced her life. She had an ambivalent attitude toward cancer's impact: "Yes. It has not changed me negatively as a person. I would not have the cancer if I could choose, but in a way, it resulted in me having different views on things, a better outlook on things [...]. It sounds very strange to say, but it is good in a way. I am not sure that I would have been where I am now without cancer. On the other hand, I do not know. It sounds very strange that I am glad I had cancer."

## Discussion

In this study, we addressed how the late effects of extensive treatment of osteosarcoma of the lower extremity, especially surgery, have influenced cancer survivors. Our findings regarding functional impairment are consistent with those of the few other qualitative bone cancer studies (20, 34, 35, 61). It seems that our participants described consequences and challenges that are quite similar to those experienced by people with a functional impairment or disability (62), for example victims of traffic or sporting accidents. On the other hand, many of them also experienced personal growth but were ambivalent about how they really felt regarding these positive cancer experiences. Below, we attempt to go beyond these findings and discuss what the late effects actually mean to osteosarcoma survivors.

In the scientific literature, disability is an umbrella term. On the one hand, functional impairment is referred to as a property of the individual – based on an individual and disease-related understanding. In the social model, however, the focus is not on disability as an individual defect, but more on the interplay between subjective function and ambient requirements, social injustice and changes in the social and physical environment. Disability comprises both the individual's reduced function with limitations in activity on the one hand and limitations in participation in different arenas and life situations on the other (63). In a society that is largely based on being a healthy person, an individual with a functional impairment or disability might be excluded from participating in certain activities or situations (64). Among our participants, we observed changes in activity, changed experience in relation to body image and exclusion from participation in different arenas, similar to the experiences described by people with a disability (62).

Research indicates that physical activity has a positive effect on cancer survivors' well-being and good health (65, 66). Several of the participants in this study stated that they were now excluded from sport and outdoor activities. This may involve more than just inactivity. For some, this will result in fewer social activities than before the cancer struck, as has been noted in other studies (35, 61). In particular, those with children stated that they were also excluded from taking part in their children's activities due to the functional impairment. This was the source of considerable grief. In addition, for a few participants, the negative influence of fatigue and having less energy after work affected their opportunities to participate in their children's home-based activities. The negative consequences of cancer treatment mentioned above imply far more than physical conditions. There are also social and individual aspects which result in both a limitation in activities and exclusion from participation in areas that are important to survivors.

Impaired fertility might, for some, be a late effect of chemotherapy (67). One of our participants knew that he was infertile following chemotherapy, while a few of the youngest men still had to face uncertainty in this regard. The two youngest women in our study had been pregnant following their cancer treatment, although the two oldest went into an early menopause. Not being able to have your own children can, particularly for adolescents, be a major loss. Being young and infertile could have an impact on future relationships (12, 68, 69). Research shows that fertility should thus be discussed before treatment. Having sperm cryopreserved or even ovary tissue frozen before starting treatment (70) should be a high priority, especially for adolescents with cancer.

In addition to the physical limitations following cancer treatment, the participants expressed negative feelings about ugly scars and thin deformed legs interfering with their body image. This is in line with findings from other research (11, 15, 71). Three of the participants described how they, as cancer survivors, hid their altered leg when among other people. Contemporary society seems to place increasing importance on appearance and looking good. This can prove distressing for many people, including those whose appearance is, for any reason, different to the norm. Negative self-perception and problems with social interaction are the most commonly reported challenges in this regard. Such challenges may involve fear of others' reactions, low self-esteem and avoiding certain social contexts (72). An ugly scar or a deformed leg can be a stigma that violates the accepted norm of appearance. According to Goffman, people are assessed and judged based on their outward appearance (73). This may entail stigma and cause embarrassment for both the person themselves and for those they meet. They thus hide in order to prevent awkward situations (73).

The majority of participants did not express much fear of recurrence or any guilt. Likewise, hardly any lifestyle changes were reported. There are no known lifestyle issues predisposing to osteosarcoma (27), which might be the reason for the observations mentioned above. Furthermore, there seems not to be any cultural discourse on having cancer in the leg, and the treatment has a focus on surgery and rehabilitation. This is in contrast to, for example, lung cancer (74, 75). Perhaps the localisation of the cancer in the lower extremity with a focus on mobility renders osteosarcoma survivors different from other cancer survivors. Here, more research is warranted. Despite the difficulties that osteosarcoma survivors have overcome, their work in adapting to the new challenges also positively influences their life and identity. This is consistent with other research (76, 77). In our study, the participants expressed experiences of growth (Table II). In the literature, such positive changes are termed posttraumatic growth (PTG) or benefit finding, and are characterised by a changed

sense of oneself, a changed sense of one's relationship with others, and a changed philosophy of life (78). Among patients with cancer, higher growth, both with increasing time from diagnosis and a younger age (79-81), have been found. Thornton's review of benefit finding in the cancer experience reports that a "substantial proportion" of previous cancer patients link positive changes in their life perspective, relation to others and to self with their cancer and illness experiences (76). In some studies, a majority of adolescent cancer survivors report growth (82), including long-term survivors of osteosarcoma (77). In one study, however, PTG was significantly lower among adolescent and young adult survivors of bone sarcoma than among other cancer survivors (83). Evidence suggests that psychological resilience may be present among adolescent and young adult survivors treated for bone tumours of the lower extremities (84). In our study, we did not find any discrepancy between adolescents and adults. Interestingly, three of the participants stated they were glad of their cancer experience. Nevertheless, they also expressed great ambivalence regarding whether cancer had really been worth the personal growth achieved. Hence, they contradicted themselves in this regard.

We note that one of the participants in our study was still struggling years after his cancer treatment. He expressed regrets and sorrow about having missed out on several things in life. He also reported feeling inferior and negative about himself. As mentioned before, being cured of cancer is not always about returning to one's previous life. For some, it requires that they reorient themselves to a new life with an altered body. This pre-supposes having the energy to establish a new understanding of themselves where the new body and the new conditions are integral parts of the new identity. To accept changes and to reconcile with them seems crucial in achieving resilience and growth (52, 69). The small sample size limits the generalizability of this study, although the majority of eligible osteosarcoma survivors in Norway did take part. However, in qualitative research, we are not seeking representative data, but rather aim to illuminate the phenomena that the participants express from their own point of view. Another limitation is that we only interviewed the participants once. Repeated interviews over a longer period from the time of diagnosis might have provided a more complete picture.

A socio-cultural and psychosocial perspective on health may provide a better understanding of what it means to be a cancer survivor. In particular, it would be useful to capture the experiences of those still struggling many years after treatment, those who are excluded from most of what was important to them before cancer struck, and those who do not find meaning in their daily life and identity as a functionally impaired person. It is important that healthcare providers and relatives develop a comprehensive picture of

what it means to be an osteosarcoma survivor. A more comprehensive view of health (46) and treatments other than the traditional biomedical approach could provide measures that are more oriented towards health protection and rehabilitation. This may benefit osteosarcoma survivors.

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