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Willingness of patients with sarcoma to participate in cancer surveillance research: a cross-sectional patient survey

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ABSTRACT

Objectives To determine the proportion of patients with extremity sarcoma who would be willing to participate in a clinical trial in which they would be randomised to one of four different postoperative sarcoma surveillance regimens. Additionally, we assessed patients' perspectives on the burden of cancer care, factors that influence comfort with randomisation and the importance of cancer research.

Design Prospective, cross-sectional patient survey.

Setting Outpatient sarcoma clinics in Canada, the USA and Spain between May 2017 and April 2020. Survey data were entered into a study-specific database.

Participants Patients with extremity sarcoma who had completed definitive treatment from seven clinics across Canada, the USA and Spain.

Main outcome measures The proportion of patients with extremity sarcoma who would be willing to participate in a randomised controlled trial (RCT) that evaluates varying postoperative cancer surveillance regimens.

Results One hundred thirty complete surveys were obtained. Respondents reported a wide range of burdens related to clinical care and surveillance. The majority of patients (85.5%) responded that they would agree to participate in a cancer surveillance RCT if eligible. The most common reason to participate was that they wanted to help future patients. Those that would decline to participate most commonly reported that participating in research would be too much of a burden for them at a time when they are already feeling overwhelmed. However, most patients agreed that cancer research will help doctors better understand and treat cancer.

Conclusions These results demonstrate that most participants would be willing to participate in an RCT that evaluates varying postoperative cancer surveillance regimens. Participants' motivation for trial participation included altruistic reasons to help future patients and deterrents to trial participation included the overwhelming burden of a cancer diagnosis. These results will help inform the development of patient-centred RCT protocols in sarcoma surveillance research.

Level of evidence V.

Strengths and limitations of this study

- The primary objective of this study was to investigate the proportion of patients with extremity sarcoma who would be willing to participate in a clinical trial in which they would be randomised to one of four different postoperative cancer surveillance regimens.
- The results of this study have been used to directly inform the definitive phase of the Surveillance AFter Extremity Tumor SurgerY (SAFETY) trial.
- Patient engagement in the preliminary trial development is expected to improve the trial's relevance, increase transparency and, ultimately, accelerate the adoption of findings into practice.
- Patients who agreed to participate in the survey study may be more likely to participate in research in general, thus possibly introducing selection bias.
- This may have resulted in an overestimation of the acceptance rate of the SAFETY study and interest in clinical research; however, our response rate of 92% may have somewhat mitigated these concerns.

INTRODUCTION

Sarcomas are a rare and heterogenous group of cancers with distinct biology that represent <1% of all malignancies.[1–6] Following treatment for a sarcoma, patients remain at risk for the development of local and systemic disease recurrence, which necessitates careful postoperative surveillance. Almost 50% of all patients with sarcoma will develop a local or distant recurrence; however, the risk of recurrence is greatest in the first few years, with 68% occurring by 2 years and 90% by 5 years.[7–9] Metastasis to the lung is the most frequent single location of disease recurrence in patients with sarcoma, occurring in approximately one-half of all patients.[9–12] Earlier detection of less advanced and resectable disease relapse may prolong patient survival; however, once advanced metastases are detected, the median length of survival is 12–15 months.[9]

As such, routine follow-up following the completion of sarcoma treatment is standard

practice, and generally entails regular visits to sarcoma outpatient clinics in the first 5–10 years after surgery. These visits typically include a clinical history, a physical examination and imaging of the lungs. Regular, intensive surveillance is more likely to identify recurrent disease earlier than would less intensive surveillance. This may provide reassurance to patients and clinicians as if the interval screening is negative, the patient is considered at that time to be disease-free.

However, the adverse effects of intensive surveillance practices on patients are also noteworthy. Intensive surveillance can threaten the financial security of patients, due in part to the direct costs, including travel, accommodation, personal care and homemaking, and indirect costs, including lost wages for patients and their caregivers, incurred as a result of follow-up appointments.[13] As a result, patients' health and quality of life can be dramatically impacted should they decide to forego further treatment or alter their lifestyles in order to alleviate financial difficulties.[13–15] Furthermore, intensive surveillance investigations can also induce anxiety, and earlier knowledge of disease recurrence may adversely impact patients' psychosocial well-being for those whose mortality risk cannot be significantly reduced by further medical interventions.[16] In fact, the first recommendation put forward by *Choosing Wisely Canada* for oncology is not to 'order tests to detect recurrent cancer in asymptomatic patients if there is not a realistic expectation that early detection of recurrence can improve survival or quality of life'.[17]

A randomised controlled trial (RCT) would be the ideal approach to determine the optimal postoperative surveillance strategy that balances potential gains in survival, costs and quality of life. Given the rarity of sarcoma, possible patient anxiety related to both less-intensive and more-intensive sarcoma surveillance and the fact that clinical trial recruitment is often slower than anticipated, such an RCT will require extensive international collaboration and patient willingness to be randomly allocated to varying surveillance regimens. Patients' perceptions of surveillance and of participation in a surveillance RCT are required in order to develop a study protocol that is patient-centred, compelling and feasible, and is capable of answering this high priority clinical question in a reasonable timeframe.[18 19] In this study, we conducted a patient survey to investigate the proportion of patients with extremity sarcoma that would be willing to participate in a clinical trial in which they would be randomised to one of four different postoperative sarcoma surveillance regimens. We also assessed the burden of cancer care on patients, the factors that influence patient comfort with being randomised to different surveillance protocols and we explored patients' views on the importance of cancer research.

METHODS

We conducted a cross-sectional multicentre survey between May 2017 and April 2020 at seven sarcoma outpatient clinics in Canada (three sites), the USA (three sites) and Spain (one site).

Participants

Clinical sites

The clinical sites within our international orthopaedic oncology research network were carefully screened for the following criteria: (1) sufficiently high sarcoma volume defined as ≥20 participants per year; (2) adequate research personnel and infrastructure to manage the study and (3) an interest in participating in the Surveillance AFter Extremity Tumor surgerY (SAFETY) trial. The clinical sites that met the eligibility criteria were invited to participate in this cross-sectional study.

Patients

In order to be eligible for participation, patients must have: (1) been at least 18 years of age; (2) been able to read, understand and write in English, French or Spanish; (3) have recently completed treatment of an extremity sarcoma and (4) provided consent to participate.

Questionnaire objectives

Given that patients' willingness to participate in cancer surveillance research is the ultimate determinant of overall study feasibility, the primary objective of this questionnaire was to determine whether patients with extremity sarcoma would be willing to participate in the SAFETY trial.[20] The SAFETY trial, initiated in early 2020, is a 2×2 factorial design RCT in which patients with sarcoma are randomised to one of four different surveillance regiments. The primary objective of the SAFETY trial is to determine the effect of surveillance intensity on long-term survival in the soft-tissue sarcoma population. The current cross-sectional survey served as background work for the trial's development.

The secondary objectives of this cross-sectional patient survey included: (1) assessment of the burden of cancer care on patients; (2) assessment of factors that influence patient comfort with being randomised to different surveillance protocols and (3) the exploration of patients' views on the importance of cancer research.

Questionnaire development

Item generation

We developed a unique patient questionnaire for the purpose of this study. The development of this questionnaire was informed by a review of the current literature on patient surveillance and in consultation with experts in orthopaedic oncology, research methodology and patient recruitment. We utilised a 'sampling-to-redundancy' approach in which we solicited feedback from new orthopaedic oncologists and research methodologists until no new items for the questionnaire emerged.

Pretesting and validity assessments

The questionnaire was reviewed by nine additional experts, who were either orthopaedic oncologists or health research methodologists. These experts

evaluated whether the questionnaire as a whole appeared to adequately address the question of whether patients with extremity sarcoma would participate in cancer surveillance research (face validity) and whether the individual questions adequately addressed the objectives of the current study (content validity). These nine experts also assessed the questionnaire's comprehensiveness and flow, as well as identified any redundant, irrelevant or poorly worded questions.

Survey description

The final survey comprised 58 questions using Likert scales, multiple choice and brief open-ended questions. The following sections were included: (a) demographics, including medical history and income; (b) cancer history, including the number of treatment visits thus far required; (c) perceptions of cancer research; (d) financial burden of cancer care; (e) logistical burden of cancer care and (f) the SAFETY trial, including perceptions of cancer surveillance, the trial design and willingness to participate in such a trial and reasons for accepting or declining to participate. The survey is provided in online supplemental appendix 1.

All questions were straightforward and used clear and layman terminology to enhance the validity of the results. The survey length was kept to a minimum in an effort to maximise the response rate and to limit barriers that could have affected its proper completion.

Sample size

Convenience sampling of consecutive patients was used at the seven participating sites. One hundred thirty patients completed the patient survey, which represents a robust sample in the study of rare diseases.[21]

Survey administration and data collection

Initially, we approached all patients with extremity sarcoma in person that had consented for sarcoma surgery. However, after consulting with the SAFETY trial's Steering Committee members on the study's protocol in May 2018, we determined that patients would be approached, consented and randomised into the SAFETY trial after definitive treatment for their extremity sarcoma, as it was deemed a less stressful time for patients to make an informed decision, as well as a time point closer to the initiation of surveillance. After this decision was made, we began approaching all recent postoperative patients with extremity sarcoma for participation in this survey study, either at a postoperative clinical appointment or via telephone. After obtaining informed consent, the site study coordinator provided each participant with a paper copy of the questionnaire to complete in a private location. Participants were allowed to leave a question blank if they found it uncomfortable to answer. On completion, the participant returned the questionnaire to the site study coordinator who verified that all questions had been answered. Completed questionnaires were then entered into a study-specific database using the REDCap electronic data capture software system.

Statistical analysis

Descriptive analyses, including frequency counts and percentages, were calculated for all collected data. Continuous data were presented as means and SD.

Role of the funding source

The funding source had no role in the design or conduct of the study; the collection, management, analysis or interpretation of the data or the preparation, review or approval of the manuscript. None of the authors has been paid to write this article. The study team had full access to all of the study data and takes responsibility for the integrity of the data and the accuracy of the data.

Patient and public involvement

Although this study evaluates the patients' perspectives on participating in clinical trials and cancer research, patients were not involved in the design, conduct or reporting or dissemination of this research. However, the results of this study will help inform the development of patient-centred clinical trial protocols in sarcoma surveillance research.

RESULTS

Characteristics of respondents

A total of 142 patients were approached to complete the survey and 130 agreed (response rate 92%). To the best of our knowledge, no patients were missed during the recruitment period. Participant demographic and cancer history data are shown in Table 1. The mean participant age was 56.4 years (SD 16.9 years) and 60.8% of participants were male. The majority of patient respondents were white (82.3%) and country of residence was reported as Canada in 40.8%, the USA in 52.3% and Spain in 6.9% of respondents. Most respondents were married or in a common law relationship (70.5%). There was a broad range of educational levels reported with a high school diploma as the most common response (31.3%), and a wide range of household incomes were reported. The most common anatomic location for the sarcoma was the lower extremity (66.7%), and participants reported receiving multidisciplinary treatment including chemotherapy (21.9%) and radiotherapy (68.4%). Travel times to the clinic ranged evenly across the spectrum from <30 min to >2 hours. Most participants reported travelling to medical appointments by personal vehicle (75%) by themselves (46.9%) or with a spouse (41.4%). Seventy-five per cent of patient respondents reported not having previously been involved in a clinical research study.

Burden of cancer care

Respondent details for burden of cancer care are shown in table 2. The majority of participants reported at least some form of financial burden related to their cancer care and surveillance. These included transportation

Table 1 Participant demographics

Characteristic	N=130
Age (years), mean (SD)	56.4 (16.9)
Gender, n (%)	
Male	79 (60.8)
Female	51 (39.2)
Ethnicity, n (%)	
White/Caucasian	107 (82.3)
Black	3 (2.3)
Native	1 (0.8)
Asian	4 (3.1)
Hispanic	9 (6.9)
Other (specify)	5 (3.8)
Country, n (%)	
Canada	53 (40.8)
USA	68 (52.3)
Spain	9 (6.9)
Marital status, n (%)	
Single	20 (15.5)
Separated	0 (0)
Divorced	11 (8.5)
Common law	8 (6.2)
Married	83 (64.3)
Widowed	7 (5.4)
Highest level of education, n (%)	
Did not complete high school	11 (8.6)
High school diploma	40 (31.3)
College/Trade diploma	31 (24.2)
Undergraduate degree	18 (14.1)
Masters degree	11 (8.6)
Doctorate degree	3 (2.3)
Professional degree	7 (5.5)
Annual household income, n (%)*	
<$20 000	12 (9.8)
$20 000–$39 999	25 (20.3)
$40 000–$59 999	21 (17.1)
$60 000–$79 999	13 (10.6)
$80 000–$99 999	15 (12.2)
$100 000 +	37 (30.1)
Cancer type, n (%)	
Chondrosarcoma	5 (3.9)
Ewing's sarcoma	1 (0.8)
Fibrosarcoma	8 (6.3)
Fibrous histiocytoma	2 (1.6)
Leiomyosarcoma	4 (3.1)
Liposarcoma	16 (12.6)
Osteosarcoma	8 (6.3)
Rhabdomyosarcoma	4 (3.1)
Synovial sarcoma	11 (8.7)
Other	49 (38.6)
Location of tumour, n (%)	
Upper extremity	29 (22.5)
Lower extremity	95 (73.6)
Other	5 (3.9)
Pelvis	2 (1.6)
Trunk	3 (2.3)
Cancer treatment modalities, n (%)	
Chemotherapy	25 (21.9)
Radiation therapy	78 (68.4)
Physiotherapy	4 (3.5)
Other	46 (40.4)
Travel time to sarcoma clinic, n (%)	
<30 min	24 (18.6)
30–59 min	38 (29.5)
60–89 min	19 (14.7)
90–119 min	23 (17.8)
120 min +	25 (19.4)
Primary mode of transportation to sarcoma clinic, n (%)	
Public transit	8 (6.5)
Personal vehicle	93 (75.0)
Taxi	3 (2.4)
Bicycle	0 (0)
Foot	1 (0.8)
Hospital transportation	2 (1.6)
Relative's/Friend's vehicle	13 (10.5)
Other (specify)	4 (3.2)
Primary caregiver, n (%)	
Self	60 (46.9)
Spouse/Partner	53 (41.4)
Parent	8 (6.3)
Sibling	1 (0.8)
Child	5 (3.9)
Grandchild	0 (0)
Friend	1 (0.8)
Other (specify)	0 (0)
Previous participation in research study, n (%)	
No	98 (75.4)
Yes	32 (24.6)
1	22 (71.0)
2	8 (25.8)
3	1 (3.2)
>3	0 (0)

*Participants reporting household income in Euros (€) were converted to $C and placed in the respective group at the time of manuscript preparation. Reported household income values include both $C and US$ as currency was not collected from participants when responding to this question.

Table 2 Burden of cancer care

Burden	n=130
Financial burdens	
Transportation and travel expenses, n (%)	
No	16 (12.3)
Yes	114 (87.7)
Accommodation and meal expenses, n (%)	
No	30 (23.4)
Yes	98 (76.6)
Family and living expenses, n (%)	
No	27 (21.1)
Yes	101 (78.9)
Caregiving expenses, n (%)	
No	56 (43.8)
Yes	72 (56.3)
Personal loss of wages, n (%)	
Not applicable	40 (31.0)
No	40 (31.0)
Yes	49 (38.0)
Caregiver loss of wages, n (%)	
Not applicable	38 (29.9)
No	62 (48.8)
Yes	27 (21.3)
Logistical burdens	
Coordination of frequent medical appointments, n (%)	
No	69 (53.5)
Yes	60 (46.5)
Completion and submission of paperwork, n (%)	
Not applicable	20 (15.4)
No	76 (58.5)
Yes	34 (26.2)
Submission of medical bills, n (%)	
Not applicable	28 (21.5)
No	61 (46.9)
Yes	41 (31.5)
Arrangement of time off work, n (%)	
Not applicable	53 (40.8)
No	36 (27.7)
Yes	41 (31.5)
Arrangement of childcare, n (%)	
Not applicable	88 (67.7)
No	27 (20.8)
Yes	15 (11.5)

and travel expenses (87.7%), accommodation and meal expenses (76.6%), family and living expenses (78.9%), caregiving expenses (56.3%) and personal loss of wages (38%). Logistical burdens are also very significant for some participants. These included coordination of medical visits (46.5%), arrangement of time off work (31.5%) and arrangement of childcare when applicable.

Table 3 Reasons for trial participation

Reason	N=130 N (%)
I believe that the study offers the best treatment available.	65 (61.9)
I want to contribute to scientific research.	83 (79.0)
I believe that the quality of care I receive would be better as part of this study.	42 (40.0)
I trust the doctor treating me.	79 (75.2)
I believe the benefits of participating would outweigh any negative side effects.	53 (50.5)
I believe the results from the study could benefit other patients in the future.	82 (78.1)
I believe that I would be monitored more closely as part of this study.	42 (40.0)
I think my cancer will get worse unless I participate in this study.	1 (1.0)
I had a positive experience in a previous research study.	6 (5.7)
Other (specify).	0 (0)

The SAFETY trial: reasons to participate and views on cancer research

A summary of patients' perceptions on cancer research and the SAFETY trial specifically are outlined in tables 3 and 4. The most common reasons for agreeing to participate in cancer research represented trust in the healthcare team and altruism: 'I want to contribute to scientific research' (79%), 'I trust the doctor treating me' (75%), "I believe the results from the study could benefit other patients in the future" (78.1%) and 'I believe that the study offers the best treatment available' (61.9%). With respect to overall views and perceptions of cancer research, approximately two-thirds of participants (68.7%) feel that they have a good understanding of clinical research. Notably, only about half (53.5%) are generally comfortable with the process of randomisation, in which their treatment or surveillance arm could be determined by chance. However, an overwhelming majority of participants (128/130, 98.5%) strongly agree or agree that cancer research will help doctors better understand and treat cancer. In addition, 93.9% of respondents strongly agree or agree that the primary reason cancer research is done is to improve the treatment of future patients with cancer. Interestingly, over half of respondents (68/130, 52.3%) strongly agree or agree that they would not benefit directly from participating from cancer research.

A total of 106 of 124 respondents that answered the question 'Would you participate in the SAFETY trial if eligible?' reported that they would agree to participate (85.5%). Those that believed they would not agree to participate reported that they would decline for the following reasons: (1) 'I do not believe that I can currently cope with the additional requirements of a research study' (eight, respondents, 44.4%), (2) "I have concerns about possibly being followed

Table 4 Views on cancer research

View	N=130 N (%)
I am interested in participating in clinical research related to my cancer.	
Strongly agree	63 (49.2)
Agree	51 (39.8)
Neither agree nor disagree	11 (8.6)
Disagree	2 (1.6)
Strongly disagree	1 (0.8)
I have a good understanding of clinical research.	
Strongly agree	31 (24.2)
Agree	57 (44.5)
Neither agree nor disagree	31 (24.2)
Disagree	3 (2.3)
Strongly disagree	6 (4.7)
Some clinical research determines by chance what treatment a patient receives (randomisation). I am comfortable with being randomly assigned (randomised) to receive a treatment.	
Strongly agree	24 (18.6)
Agree	45 (34.9)
Neither agree nor disagree	35 (27.1)
Disagree	15 (11.6)
Strongly disagree	10 (7.8)
Cancer research will help doctors better understand and treat cancer.	
Strongly agree	102 (78.5)
Agree	26 (20.0)
Neither agree nor disagree	2 (1.5)
Disagree	0 (0)
Strongly disagree	0 (0)
The primary reason cancer research is done is to improve the treatment of future patients with cancer.	
Strongly agree	86 (66.2)
Agree	36 (27.7)
Neither agree nor disagree	3 (2.3)
Disagree	3 (2.3)
Strongly disagree	2 (1.5)
I will not directly benefit from participating in cancer research.	
Strongly agree	26 (20.0)
Agree	42 (32.3)
Neither agree nor disagree	31 (23.8)
Disagree	28 (21.5)
Strongly disagree	3 (2.3)
Patients who participate in research studies should be told the results when the study is compete.	
Strongly agree	46 (35.4)
Agree	62 (47.7)
Neither agree nor disagree	20 (15.4)
Disagree	1 (0.8)
Strongly disagree	1 (0.8)
I would agree to participate in the SAFETY trial if eligible (n=124).	
Yes	106 (85.5)
No	18 (14.5)

SAFETY, Surveillance AFter Extremity Tumor SurgerY.

less intensively in this study" (four respondents, 22.2%), (3) "I have concerns about additional radiation exposures from CT scans" (four respondents, 22.2%) and (4) "I believe that the quality of care I receive would be inferior to what I would receive if I did not participate" (three respondents, 16.7%). Other less common reasons to decline the study included "I do not believe that the study offers the best treatment available", "My family is not keen for me to participate" and travel and religious reasons. One respondent reported a negative experience with a previous trial.

DISCUSSION

Summary of findings

This study explored the perceptions of international patients with extremity sarcoma on cancer surveillance. We found that patients endure significant financial and logistical burdens associated with sarcoma care and follow-up. In general, patients are very interested in participating in clinical research, and specifically in cancer surveillance research. The reasons for participating in research include the desire to help future patients and the perception that their care would be improved in the context of a clinical trial. However, some participants expressed a lingering concern with leaving their care and/or surveillance to chance (randomisation) and several indicated that they believe that they would not participate in research due to feeling overwhelmed with their cancer diagnosis and treatment. Overall, the results of this study will help inform the SAFETY trial and guide approaches to eligible patients when obtaining consent.

Strengths and limitations

This study has several strengths. First, we used a rigorous process for the development of the patient questionnaire and extensive piloting of the survey. This stepwise process created a questionnaire that was acceptable for patients and sufficiently clear and comprehensive to provide a robust dataset. Second, we surveyed patients across Canada, the USA and Spain. Although this required translation of English documents into French and Spanish, it provided a more global picture of patients' perceptions. The SAFETY trial is an international endeavour, and therefore international

participation in the background survey was critical. Finally, this survey study represents an important step in engaging patients in randomised controlled trial development and inception, thus improving the patient-centred nature of cancer research.

Our study also had some limitations to consider. First, there may have been selection bias in that those who agreed to participate in the survey study are also more likely to participate in research in general. This would overestimate the acceptance rate of the SAFETY study and interest in clinical research. However, our response rate was 92%, somewhat mitigating these concerns. Second, the survey was not a validated survey; however, it allowed us to determine the proportion of participants who would theoretically consent to participating specifically in the SAFETY trial, as well as investigate patients' views on the burden of cancer care and on cancer research in greater detail than would have been possible with standardised questionnaires. Third, the demographics of the respondents were not diverse with respect to race (82.3% white) and continent of residence (93.1% from North America). The incidence data collected in the Surveillance, Epidemiology and End Results database of the National Cancer Institute demonstrate similar rates of sarcomas between white and black populations.[22–25] This is also inconsistent with the overall North American demographic data, as black individuals comprise approximately 13% of the North American population.[26 27] These demographic discrepancies somewhat limit the external validity of the findings with respect to Europe and other international sites. And while it is not uncommon for non-white racial/ethnic groups to be under-represented in cancer clinical trials, the race demographics of this survey have highlighted an important gap to address in our recruitment strategy for the SAFETY trial.[28–30] Fourth, while the survey addressed indirect costs of sarcoma surveillance (such as the cost of travel or missed work to attend a clinic visit), it did not address the direct costs of surveillance (such as the cost to patients of different thoracic imaging techniques or additional imaging and clinic visits). However, postoperative sarcoma surveillance is considered standard of care despite being highly varied among orthopaedic oncologists with respect to thoracic imaging and frequency.[31–33] Therefore, direct costs should not apply to most patients as a wide spectrum of surveillance care regimens are within the range of standard practice and should be covered by the patients' federal, provincial/state or private health insurance.[34] Nevertheless, these cost data would likely prove valuable when considering trial participation of patients without private health insurance in countries without socialised healthcare such as the USA. Finally, the survey did not evaluate the optimal timing and method to approach patients to participate in the SAFETY trial.

Relevance to previous research

The exploration of patients' perceptions of sarcoma surveillance in the context of a randomised surveillance trial has not, to our knowledge, previously been reported. However, as far back as 1979, researchers interviewed patients with sarcoma to determine reasons for acceptance of randomisation in treatment-related clinical trials.[35] The authors of this study concluded that patient acceptance of participation in treatment-related clinical trials was associated with treatment factors such as burden of care and drug toxicities. Within the field of orthopaedic surgery, Creel *et al*[36] surveyed patients with meniscal tears and determined willingness to participate in a trial in which they would be randomised to operative versus non-operative treatment. The authors found that lack of strong treatment preferences and male gender were significantly associated with willingness to participate in such a trial. Only 46% of patients reported that they would be definitely willing or probably willing to participate.

A large survey study of 1227 Swiss patients in which four different clinical trial vignettes were described found that all studies were not equally acceptable to patients. A higher willingness to participate was found when a new drug was considered safe, no extra logistical burden of care was required, results were openly available to the public and the project was approved by a research ethics committee. In contrast, use of placebo controls, and random allocation to study arms were associated with a lower likelihood of participation.[37] Similarly, Halpern *et al* found that in patients with hypertension, inconvenience, fear of known side effects and the possibility of receiving placebo were the most common concerns for patients in clinical trials.[38] Similar to the orthopaedic trial outlined above, only 47% of patients would be willing to participate in a placebo-controlled trial.

Implications

In this study, we found that a high percentage of patients with sarcoma would be willing to participate in surveillance research. In comparison to other published patient survey studies of treatment-related RCTs, the willingness to participate identified in this study is significantly greater. This has positive implications for sarcoma surveillance research in general, and specifically for the SAFETY trial. However, survey responses do not necessarily align with actual participation. Moreover, the sense of being overwhelmed with the diagnosis of sarcoma and the need for intensive treatment can deter patients from accepting an additional dimension to their care in the form of a trial. Nevertheless, the patient engagement strategy used in this study is likely to increase enrolment in the SAFETY trial and help guide study implementation.[39]

CONCLUSIONS

The results of this patient survey demonstrate that the majority of participants would be willing to participate in an RCT that evaluates different postoperative sarcoma surveillance regimens. Participants' motivations for trial participation included trust in the healthcare system and altruistic reasons to help future patients. Those that would decline the study for the most part would do so because of the overwhelming burden of a cancer diagnosis. These results will help inform the development of patient-centred clinical trial protocols in cancer surveillance research and specifically the implementation of the SAFETY trial.

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Ethics approval Each author certifies that his or her institution approved or waived approval (McGill University Health Centre Research Ethics Board) for the human protocol for this investigation and that all investigations were conducted according to the ethical principles of research. The Methods Centre received approval from the Hamilton Integrated Research Ethics Board (HiREB) (Protocol No. 2954). Approval from each of the local ethics committees was obtained in writing prior to the local commencement of the study.

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Data availability statement The datasets generated during this study are not publicly available, but are available from the corresponding author on reasonable request.

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Participant ID ☐☐ – ☐☐☐☐

Completion Date ☐☐ (DD) ☐☐ (MM) 2 0 ☐☐ (YYYY)

Surveillance AFter Extremity Tumor SurgerY (SAFETY) Protocol Study

PATIENT QUESTIONNAIRE

Thank you for agreeing to complete this questionnaire. Your responses will help orthopaedic oncology researchers better understand whether sarcoma patients are willing to participate in research evaluating different post-operative follow-up schedules. This questionnaire should take you approximately 15 minutes to complete. A participant ID number will be assigned to track completion of the questionnaires. A master list linking the ID number will be maintained during the data collection phase. Once all questionnaires from each round have been received, the list will be destroyed and your responses will be anonymized.

Some of the questions may be uncomfortable for you to answer. However, we ask that you try your best in answering all of the questions. Your participation is important to us and those whom may benefit from this research.

Part A: DEMOGRAPHICS

This section asks a few basic questions to let us know a little bit more about you.

1. What is your age?

   _____ **years**

2. What is your gender?

   ☐ **Male** ☐ **Female**

   ☐ **Other (specify):** _____

3. What is your race/ethnicity?

   ☐ **Caucasian** ☐ **Native/Aboriginal**

   ☐ **African/Caribbean** ☐ **East Asian**

   ☐ **Hispanic/Latino** ☐ **South Asian**

   ☐ **Middle Eastern** ☐ **Other (specify):** _____

   ☐ **Mixed (specify):** _____

4. Where do you live?

   ☐ **Canada** ☐ **Spain**

   ☐ **Netherlands** ☐ **USA**

   ☐ **Other (specify):** _____

5. What is your first language?

   ☐ **Arabic** ☐ **French** ☐ **Korean** ☐ **Spanish**

   ☐ **Cantonese** ☐ **German** ☐ **Mandarin** ☐ **Urdu**

   ☐ **Dutch** ☐ **Hindi** ☐ **Portuguese** ☐ **Vietnamese**

   ☐ **English** ☐ **Italian** ☐ **Russian** ☐ **Other (specify):** _____

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6. What is your marital status?

☐ **Single** ☐ **Separated** ☐ **Divorced** ☐ **Common Law** ☐ **Married** ☐ **Widowed**

7. What is your highest level of education?

☐ **Did Not Complete High School**
☐ **High School Diploma**
☐ **College/Trade Diploma**
☐ **Undergraduate Degree**
☐ **Masters Degree**
☐ **Doctorate Degree**
☐ **Professional Degree**
☐ **Other (specify):** _____

8. Are you currently employed?

☐ **Yes** → If *yes*, what is your current occupation? _____
☐ **No** → If *no*, please specify why:

☐ **Retired**
☐ **Homemaker**
☐ **Student**
☐ **Unemployed**
☐ **Doctor's Advice/Disability**
☐ **Other (specify):** _____

9. Do you have a medical history of any of the following diseases?
Please select ALL that apply.

☐ **None**	☐ **Diabetes (Type I)**	☐ **Inflammatory Bowel Disease**	☐ **Peripheral Vascular Disease**
☐ **Addiction**	☐ **Diabetes (Type II)**	☐ **Kidney Transplant**	☐ **Psychoses**
☐ **AIDS/HIV**	☐ **Heart Disease**	☐ **Liver Failure**	☐ **Pulmonary Circulation Disorder**
☐ **Anemia**	☐ **Hepatitis**	☐ **Neurological Disorders**	☐ **Renal Failure**
☐ **Cardiac Arrhythmia**	☐ **Hypertension**	☐ **Obesity**	☐ **Rheumatoid Arthritis**
☐ **Chronic Pulmonary Disease**	☐ **Hyperthyroidism**	☐ **Osteoarthritis**	☐ **Systemic Lupus Erythematosus**
☐ **Depression**	☐ **Hypothyroidism**	☐ **Osteoporosis**	☐ **Other (specify):** _____

10. Do you smoke?

☐ **Never** ☐ **Former Smoker** ☐ **Current Smoker**

11. Do you routinely use recreational drugs?

☐ **Never** ☐ **Former User** ☐ **Current User**

12. How much alcohol do you drink on a weekly basis?

_____._____ **Drinks/Week**

*If you live in **Canada** or the **USA**, please proceed to **Page 3**.*
*If you live in the **Netherlands** or **Spain**, please proceed to **Page 4**.*

Participant Initials ☐☐ Participant ID ☐☐ – ☐☐☐☐

PLEASE COMPLETE THIS PAGE IF YOU LIVE IN **CANADA** OR THE **USA.**

13. What is your yearly household income before taxes?

- ☐ **Less than $20,000**
- ☐ **$20,000 to $39,999**
- ☐ **$40,000 to $59,999**
- ☐ **$60,000 to $79,999**
- ☐ **$80,000 to $99,999**
- ☐ **$100,000+**

14. Please answer 14A if you live in **Canada**. Please answer 14B if you live in the **USA**.

(A) For **Canadian** patients, do you have any *additional* medical insurance coverage outside of your provincial health insurance plan?

- ☐ **No**
- ☐ **Yes** → If *yes*, please indicate what type of additional medical insurance coverage:
  - ☐ **Employer-Provided Insurance**
  - ☐ **Personally-Purchased Insurance**
  - ☐ **Military/Veteran**
  - ☐ **Other (specify):** _____

(B) For **American** patients, do you have medical insurance coverage?

- ☐ **No**
- ☐ **Yes** → If *yes*, please indicate what type of additional medical insurance coverage:
  - ☐ **Employer-Provided Insurance**
  - ☐ **Personally-Purchased Insurance**
  - ☐ **Medicare**
  - ☐ **Medicaid**
  - ☐ **Military/Veteran**
  - ☐ **Other (specify):** _____

*Please proceed to **Part B** on **Page 5**.*

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PLEASE COMPLETE THIS PAGE IF YOU LIVE IN THE **NETHERLANDS** OR **SPAIN.**

13. What is your yearly household income before taxes?

- ☐ **Less than €14,500**
- ☐ **€14,500 to €28,999**
- ☐ **€29,000 to €43,499**
- ☐ **€43,500 to €57,999**
- ☐ **€58,000 to €71,999**
- ☐ **€72,000+**

14. Do you have any *additional* medical insurance coverage outside of your state health insurance plan?

- ☐ **No**
- ☐ **Yes** → If *yes*, please indicate what type of additional medical insurance coverage:
  - ☐ **Employer-Provided Insurance**
  - ☐ **Personally-Purchased Insurance**
  - ☐ **Military/Veteran**
  - ☐ **Other (specify):** _____

*Please proceed to **Part B** on **Page 5**.*

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Part B: CANCER HISTORY

This section asks questions about your cancer and cancer treatment. If you have been diagnosed with more than one cancer, please answer the following questions considering only the cancer you are in clinic for today.

15. What type of cancer do you have?

- ☐ **Chondrosarcoma**
- ☐ **Ewing's sarcoma**
- ☐ **Fibrosarcoma**
- ☐ **Fibrous histiocytoma**
- ☐ **Giant cell tumor of bone**
- ☐ **Leiomyosarcoma**
- ☐ **Liposarcoma**
- ☐ **Non-osteogenic sarcoma of bone**
- ☐ **Osteosarcoma**
- ☐ **Rhabdomyosarcoma**
- ☐ **Synovial sarcoma**
- ☐ **Other (specify):** _____
- ☐ **Not Sure**

16. Where is your cancer located?

- ☐ **Arm**
- ☐ **Leg**
- ☐ **Not Sure**
- ☐ **Other (specify):** _____

17. When were you diagnosed with cancer?

☐☐ **DD** ☐☐ **MM** ☐☐☐☐ **YYYY**

18. How long have you been a cancer patient at the center where you are for your current treatment?

☐ **Less Than 2 Weeks** ☐ **2 - 4 Weeks** ☐ **1 - 6 Months** ☐ **Over 6 Months**

19. How has your cancer been treated so far?
Please select ALL that apply.

- ☐ **Chemotherapy**
- ☐ **Radiation therapy**
- ☐ **Physiotherapy**
- ☐ **Other (specify):** _____

20. How many times have you seen your orthopaedic oncologist (cancer surgeon)?

☐ **First Visit** ☐ **Once Before** ☐ **2 - 3 Times** ☐ **Over 3 Times**

21. How long does it *typically* take you get from home to the hospital for a cancer appointment?

☐ **Less Than 30 Minutes** ☐ **30 - 59 Minutes** ☐ **1 - 1.5 Hours** ☐ **1.5 - 2 Hours** ☐ **Over 2 Hours**

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22. How do you *typically* travel to the hospital for a cancer appointment?

- ☐ **Public Transit**
- ☐ **Taxi**
- ☐ **Foot**
- ☐ **Relative/Friend's Vehicle**
- ☐ **Personal Vehicle**
- ☐ **Bicycle**
- ☐ **Hospital Transportation**
- ☐ **Other (specify):** _____

23. Who is your primary caregiver?

A primary caregiver is the person who assumes the most responsibility in caring for your health and wellbeing.

- ☐ **Myself**
- ☐ **Parent**
- ☐ **Child**
- ☐ **Friend**
- ☐ **Spouse/Partner**
- ☐ **Sibling**
- ☐ **Grandchild**
- ☐ **Other (specify):** _____

Part C: IMPORTANCE OF CANCER RESEARCH

This section asks questions about your previous participation in research and your opinion on cancer research. For each opinion question, please rate your level agreement with each statement.

24. I am interested in participating in clinical research related to my cancer.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

25. Have you previously participated in any other research studies?

- ☐ **No**
- ☐ **Yes** → If *yes*, how many other research studies have you previously participated in?

☐	☐	☐	☐
1	**2**	**3**	**Over 3**

26. How many different research studies have been discussed with you over the course of your cancer treatment?

☐	☐	☐	☐	☐
0	**1**	**2**	**3**	**Over 3**

27. I have a good understanding of clinical research.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

28. Some clinical research determines by chance what treatment a patient receives (randomization). I am comfortable with being randomly assigned (randomized) to receive a treatment.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

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29. Cancer research will help doctors better understand and treat cancer.

☐ Strongly Agree	☐ Agree	☐ Neither Agree Nor Disagree	☐ Disagree	☐ Strongly Disagree

30. The primary reason cancer research is done is to improve the treatment of *future* cancer patients.

☐ Strongly Agree	☐ Agree	☐ Neither Agree Nor Disagree	☐ Disagree	☐ Strongly Disagree

31. I will not directly benefit from participating in cancer research.

☐ Strongly Agree	☐ Agree	☐ Neither Agree Nor Disagree	☐ Disagree	☐ Strongly Disagree

32. Patients who participate in research studies should be told the results when the study is complete.

☐ Strongly Agree	☐ Agree	☐ Neither Agree Nor Disagree	☐ Disagree	☐ Strongly Disagree

Part D: FINANCIAL BURDEN OF CANCER CARE

This section asks questions about some of the costs you may have incurred as a result of your cancer treatment and whether they are a financial burden to you. A financial burden is any cost or fee that is difficult to pay.

33. Are transportation and travel expenses incurred due to your cancer care paid by you/your family?
Some examples of transportation and travel expenses include costs from gas, tolls, parking, taxis, and public transportation fares.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden these costs are to you:

☐ Unmanageable Burden	☐ Significant Burden	☐ Somewhat of a Burden	☐ Slight Burden	☐ No Burden

34. Are accommodation and meal expenses incurred due to your cancer care paid by you/your family?
Some examples of accommodation and meal expenses include costs from hotel stays and meals at restaurants.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden these costs are to you:

☐ Unmanageable Burden	☐ Significant Burden	☐ Somewhat of a Burden	☐ Slight Burden	☐ No Burden

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35. Are family and living expenses incurred due to your cancer paid by you/your family?
Some examples of family and living expenses include costs related to running your household, childcare, and housekeeping.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden these costs are to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

36. Are caregiving expenses incurred due to your cancer care paid by you/your family?
Some examples of caregiving expenses include costs from hiring a person to prepare meals or drive you to appointments, extended nursing care, homecare, and personal support workers.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden these costs are to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

37. Have you experienced a loss of your own wages due to your cancer care?

☐ **Not Applicable** → I was not employed prior to my cancer diagnosis.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden this loss of income is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

38. Has your primary caregiver experienced a loss of wages due to your cancer care?

☐ **Not Applicable** → My primary caregiver was not employed prior to my cancer diagnosis.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a financial burden this loss of income is to your primary caregiver:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

Part E: LOGISTICAL BURDEN OF CANCER CARE

This section asks questions about some of the tasks you may have to manage as a result of your cancer treatment and whether they are a logistical burden to you. A logistical burden is any task that involves the coordination of many details or people that is difficult to manage.

39. I find that coordinating frequent medical appointments for my cancer care is a logistical burden.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a logistical burden coordinating medical appointments is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

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40. I find that completing and submitting paperwork related to my cancer care is a logistical burden.

☐ **Not Applicable** → I do not have any additional paperwork to complete related to my cancer care.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a logistical burden completing additional paperwork is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

41. I find that processing medical bills related to my cancer care is a logistical burden.

☐ **Not Applicable** → I do not have any additional medical bills related to my cancer care.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a logistical burden processing additional medical bills is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

42. I find that arranging for time off work to attend medical appointments for my cancer care is a logistical burden.

☐ **Not Applicable** → I am not currently employed.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a logistical burden arranging for time off work is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

43. I find that arranging childcare to attend medical appointments for my cancer care is a logistical burden.

☐ **Not Applicable** → I do not have children OR I do not have children that currently require childcare.

☐ **No**

☐ **Yes** → If *yes*, please indicate how much of a logistical burden arranging childcare is to you:

☐	☐	☐	☐	☐
Unmanageable Burden	**Significant Burden**	**Somewhat of a Burden**	**Slight Burden**	**No Burden**

Part F: THE SAFETY TRIAL

Please review the Patient Information Sheet for the SAFETY Trial before answering the following questions. For questions asking your opinion, please rate your level of agreement with each statement.

44. The post-operative follow-up schedule described below is standard care for my type of cancer.

For the first two years after your surgery, your doctor will see you every three months to see if the tumor will grow back where you had your surgery or in your lungs. After that, your doctor will see you for the same reasons every six months for three years. At five years after surgery, your doctor will see you once a year. You will have a CT scan of your lungs for the first two years. Otherwise, you will only have a chest x-ray at each visit.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

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45. The post-operative follow-up schedule described above has been scientifically proven to be the best for my type of cancer.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

46. Compared with the standard follow-up schedule, none of the other study follow-up schedules carry any additional risks or discomforts.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

47. I have concerns about being followed by my orthopaedic oncologist less frequently.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

48. I have concerns about my exposure to radiation from additional CT scans or x-rays.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

49. I have concerns that CT scans will miss any cancer nodules that weren't detected on a chest x-ray.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

50. Compared with the standard follow-up schedule, fewer follow-up appointments would ease the financial burden of my cancer care.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

51. Compared with the standard follow-up schedule, fewer follow-up appointments would ease the logistical burden of my cancer care.

☐	☐	☐	☐	☐
Strongly Agree	**Agree**	**Neither Agree Nor Disagree**	**Disagree**	**Strongly Disagree**

52. Would you discuss this research study with anyone before deciding to / not to participate in this study?

☐ **No**

☐ **Yes** → If *yes*, please specify who:

☐ **Spouse/Partner** ☐ **Parent**

☐ **Sibling** ☐ **Child**

☐ **Friend** ☐ **Grandchild**

☐ **Family Physician** ☐ **Other (specify):** _____

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53. Would you search for any additional information before deciding to / not to participate in this study?

☐ **No**

☐ **Yes** → If *yes*, please specify where:

- ☐ **Internet**
- ☐ **Hospital Resources**
- ☐ **Other Organization (specify):** _____
- ☐ **Literature (books/journals)**
- ☐ **Patient Support Group(s)**
- ☐ **Other (specify):** _____

54. Would you participate in the SAFETY trial?

☐ **Yes** ☐ **No**

55. My decision to / not to participate in this research study was easy.

☐ **Strongly Agree** ☐ **Agree** ☐ **Neither Agree Nor Disagree** ☐ **Disagree** ☐ **Strongly Disagree**

56. Please answer 56A if you *would* participate in the SAFETY trial. Please answer 56B if you *would* ***not*** participate in the SAFETY Trial.

(A) Why would you agree to participate in this research study?
Please select ALL that apply.

- ☐ **A. I believe that the study offers the best treatment available.**
- ☐ **B. I want to contribute to scientific research.**
- ☐ **C. I believe that the quality of care I receive would be better as part of this study.**
- ☐ **D. I trust the doctor treating me.**
- ☐ **E. I believe that the benefits of participating would outweigh any negative side-effects.**
- ☐ **F. I believe the results from the study could benefit other patients in the future.**
- ☐ **G. I believe that I would be monitored more closely as part of this study.**
- ☐ **H. My family is keen for me to participate.**
- ☐ **I. I think my cancer will get worse unless I participate in this study.**
- ☐ **J. I had a positive experience in a previous research study.**
- ☐ **K. Other (specify):** _____

(B) Why would you choose not to participate in this research study?
Please select ALL that apply.

- ☐ **A. I do not believe that the study offers the best treatment available.**
- ☐ **B. I do not want to contribute to scientific research.**
- ☐ **C. I believe that the quality of care I receive would be inferior to what I would receive if I did not participate.**
- ☐ **D. I do not trust the doctor treating me.**
- ☐ **E. I have concerns about possibly being followed less intensively in this study.**
- ☐ **F. I have concerns about the additional radiation exposure from CT scans.**
- ☐ **G. My family is not keen for me to participate.**
- ☐ **H. I believe that this study would cause issues with my insurance coverage.**
- ☐ **I. I do not believe that I can currently cope with the additional requirements of a research study.**
- ☐ **J. I had a negative experience in a previous research study.**
- ☐ **K. Other (specify):** _____

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Participant Initials ☐☐ Participant ID ☐☐ – ☐☐☐☐

57. Which of the reasons above was the most important reason for you deciding to / not to participate in the SAFETY trial?

_____

58. Additional Comments: _____

_____

_____

_____

_____

Thank you for completing this questionnaire!

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