Whole-body MRI surveillance in TP53 carriers is perceived as beneficial with no increase in cancer worry regardless of previous cancer: Data from the Swedish TP53 Study

Meis Omran MD1,2 | Hemming Johansson BA1 | Claudia Lundgren MD3 | Gustav Silander MD4 | Marie Stenmark-Askmalm MD, PhD5 | Niklas Loman MD, PhD6,7 | Annika Baan MSc, RN8 | Jamila Adra MD8 | Ekaterina Kuchinskaya MD, PhD9 | Lennart Blomqvist MD, PhD10,11 | Emma Tham MD, PhD10,12 | Svetlana Bajalica-Lagercrantz MD, PhD1,2,12 | Yvonne Brandberg PhD1 | on behalf of the Swedish Clinical TP53 Study Group (SweClinTP53)

1Department of Oncology-Pathology, Karolinska Institutet, Stockholm, Sweden
2Cancer Theme, Karolinska University Hospital Solna, Stockholm, Sweden
3Department of Immunology, Genetics and Pathology, Uppsala University Hospital, Uppsala, Sweden
4Department of Radiation Sciences, Umeå University, Umeå, Sweden
5Division of Clinical Genetics, Department of Laboratory Medicine, Office for Medical Services, Skåne University Hospital, Lund, Sweden
6Division of Oncology and Pathology, Department of Clinical Sciences, Lund University, Lund, Sweden
7Department of Hematology, Oncology and Radiation Physics, Skåne University Hospital, Malmö, Sweden
8Department of Oncology, Sahlgrenska University Hospital, Gothenburg, Sweden
9Department of Clinical Genetics, Linköping University Hospital, Linköping, Sweden
10Department of Molecular Medicine and Surgery, Karolinska Institutet, Stockholm, Sweden
11Department of Imaging and Physiology, Karolinska Institutet, Stockholm, Sweden
12Department of Oncology-Pathology, Karolinska Institutet, Stockholm, Sweden

Abstract

Background: To evaluate the psychosocial consequences of surveillance with whole-body MRI (WB-MRI) in individuals with the heritable TP53-related cancer (hTP53rc) syndrome, also known as the Li-Fraumeni syndrome, with regard to cancer worry, perceived benefits and risks to surveillance and overall health.

Patients and methods: Since 2016, the national Swedish TP53 Study (SWEP53) has offered surveillance with WB-MRI to all individuals with hTP53rc syndrome. Seventy-five individuals have been included in the study. Sixty consecutive participants fulfilled a base-line evaluation as well as an evaluation after 1 year with structured questionnaires including the Cancer Worry Scale (CWS), perceived benefits and risks of surveillance, and the 36-item Short Form Survey (SF-36). Individuals with or without previous personal cancer diagnosis were enrolled and results at baseline and after 1 year of surveillance were compared. For SF-36, a comparison with the normal population was also made.

Results: Participants with previous cancer tend to worry more about cancer, but both individuals with and without cancer had a positive attitude toward surveillance with no differences regarding perceived benefits and barriers to surveillance. Participants with a previous cancer scored significantly lower on some of the SF-36 subscales, but between-group differences were found only for social functioning after 1 year.
INTRODUCTION

Heritable TP53-related cancer (hTP53rc) syndromes include the Li-Fraumeni syndrome (LFS) and are characterized by an increased risk of early onset female breast cancer, sarcomas, brain tumors, and adrenocortical carcinomas as well as various other tumors at different body sites. The life time risk of cancer development is estimated to be 70%–100%. Estimates of population prevalence vary, but one of the most recent publications indicates it to be 1:4500. The cause behind LFS is a germline disease causing variant, and individuals are henceforth referred to as TP53 carriers. Carriers are at risk of developing cancers at substantially younger ages than noncarriers with the same cancer types. A recent article estimated that TP53 carriers have a nearly 24 times higher incidence of any cancer compared with the general population, especially from childhood to 30 years of age. In addition, individuals with hTP53rc also have a high risk of multiple primary tumors as well as developing secondary tumors after irradiation. Thus, it is not unusual that these families have multiple affected family members simultaneously. A recently published review on several psychosocial aspects of being a TP53 carrier in surveillance identified need for psychosocial support for these families. Because of the high cancer risks, the European guidelines from 2020 recommend surveillance with whole-body MRI (WB-MRI) to carriers of all ages, although in many countries, including Sweden, this has not yet been implemented in routine care. In the Swedish TP53 Study (SWEP53), adult carriers are offered routine surveillance with WB-MRI including MRI of the breasts and brain. Our first publication indicated 30 imaging findings in 19 of 61 participants (31%) of which 21 lesions in 16 individuals (84%) were incidental and nonmalignant. Until now, studies evaluating the psychosocial impact of surveillance with WB-MRI have been reported from Australia, Great Britain, Germany, and the United States, including 17–49 study participants. In general, more studies evaluating the psychosocial aspects are needed.

The aim of the present study is to describe cancer worry, perceived benefits, and barriers of participation in the WB-MRI surveillance within SWEP53 at baseline and after 1 year. In addition, we explore health-related quality-of-life (HRQOL) at both time points, and compare these variables between participants with and without a previous cancer diagnosis. In this article, results from the first 60 eligible individuals are reported.

MATERIALS AND METHODS

Sample and procedures

The national SWEP53 Study started inclusion in April 2016. Individuals with a verified likely pathogenic (class 4) or pathogenic (class 5) TP53 variant according to the American College of Medical Genetics and Genomics classification are invited to participate regardless of previous cancer history. In this study, that is currently recruiting, participating adults 18 years of age and older undergo yearly WB-MRI screening and an annual clinical check-up that includes a physical examination according to a specific protocol (Supporting Information S1).

Participants are recruited through the hereditary cancer units at the six cancer genetics clinics in Sweden (i.e., Umeå University Hospital, Linköping University Hospital, Sahlgrenska University Hospital [Gothenburg], and Skåne University Hospital [Lund]). All known eligible TP53 carriers in Sweden are invited to participate in the study. By consenting to the SWEP53 Study one may participate in (1) registration in the Swedish registry, (2) participation in...
surveillance with WB-MRI, (3) participation in the psychosocial evaluation of surveillance, and (4) participation in collection of cell-free DNA. Because the psychosocial evaluation is connected to the WB-MRI surveillance, both parts 2 and 3 must be consented to for the study participant to be included in this article.

Participants were handed questionnaires by their local caregiver at the baseline visit and at the 1-year follow-up visit. Completed questionnaires were collected by local coordinators and sent to the national study coordinator (Meis Omran). The questionnaires were completed before information of the results from the WB-MRI. All questionnaires for baseline and the 1-year visits received from the different study sites from April 2016 to October 2021 were included for analysis in this article.

Psychosocial measures

Cancer Worry Scale

To measure cancer-specific distress, the Cancer Worry Scale (CWS) was used. CWS is an eight-item questionnaire with a response format ranging from 1 to 4 or 5 ("Not at all" to "A lot" or to "Almost always"). The original six items assess own cancer worry, with the later addition of two items concerning worries about family members and worries regarding future cancer operations. A higher score indicates more cancer worry. A cut-off score of ≥14 has been reported to correlate with clinical significant cancer worry.

Perceived benefits and barriers

The questionnaire measuring perceived benefits and barriers of participating in the surveillance program consists of 11 items based on previously performed work by other researchers. Both these questionnaires and the CWS were developed in English and translated to Swedish by a forward and backward translation process, although none of them have been formally validated in Swedish.

The 36-Item Short Form Survey

Health-related quality of life (HRQOL) was assessed with the 36-Item Short Form Survey (SF-36) version 1.0. SF-36 consists of eight subscales, and higher levels on each scale indicate higher HRQOL. The Swedish version of SF-36 has undergone formal validation.

Sociographic and clinical data

The participants’ sociodemographic details and personal history of cancer diagnoses were obtained from the study specific questionnaire. Family history of cancer diagnoses and death due to cancer among family members were acquired from pedigrees. Personal history of cancer diagnoses was also confirmed in medical records. There were no restrictions regarding recency of previous cancer diagnoses.

Statistical analysis

Descriptive statistics such as mean, range, counts, and percentages were used to describe sociodemographic and clinical data as well as psychosocial measures. Differences in ordinal items were tested using the Mann-Whitney test. For the eight subscales included in SF-36, linear regression was used to estimate and test group differences. Results from the regression models are presented as mean differences together with 95% confidence intervals and Wald p values. Indirect standardization using the age and sex distribution in the cohort together with normative data was used to calculate expected (population) mean scores for the SF-36 subscales. Cronbach’s coefficient α was calculated to determine internal consistency reliability for the different subscales in SF-36. The other questionnaires are presented as items and not aggregated as scales. Stata Version 17 was used for all statistical analysis. All statistical analysis were performed by Hemming Johansson, statistician.

The study was conducted according to the guidelines of the 1964 Declaration of Helsinki and approved by the regional ethical review board in Stockholm (approval number 2015/1600-3 with the amendments 2017/1527-32 and 2018/1690-32). Written informed consent was obtained from all participants. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee. Written informed consent has been obtained from the participants to publish this article.

RESULTS

Out of 75 included participants in SWEP53, 60 (39 women and 21 men) were eligible for the study (Figure 1). Of these, 56 (93%) underwent WB-MRI scans both at baseline and 1 year after. Ten participants (of whom three had a personal history of cancer) were already performing clinical WB-MRI before study inclusion. Regarding previous cancer, 32 participants had a history of cancer before inclusion. The median time from the latest cancer diagnosis was 5 years, ranging from 3 months to 23 years before inclusion. Baseline demographic characteristics are shown in Table 1, with further details in Table S1. After undergoing a baseline WB-MRI, three asymptomatic participants were diagnosed with cancer.

CWS

The response rate for CWS was 97% (58 of 60) at baseline and 67% (40 of 60) at year one. There was no systematic difference (regarding
sex, age, and previous cancer) between those who responded and nonresponders at the two time points. At baseline, there were statistically significant differences among individuals with previous cancer history in comparison with those with no personal cancer history for all items, except for the item on cancer worry concerning a family member (Table 2). Higher proportions of individuals with previous cancer reported that their mood was affected by thoughts about cancer. Worry about getting cancer, or getting cancer again, and for future operations were reported more frequently among participants with previous cancer and these thoughts were also reported to be more of a problem in this group compared to participants without previous cancer.

Changes in CWS items from baseline to 1-year follow-up according to “No previous cancer” and “Previous cancer” at baseline are presented in Table 3. No statistically significant changes were found with one exception regarding whether the thoughts about the risk of getting cancer has affected the ability to perform daily tasks: 95% (18 of 19) of individuals with “No previous cancer” reported no changes at all, whereas 38% (8 of 21) of those with “Previous cancer” reported lower levels of worry. Responses at the 1-year follow-up are presented in Table S3.

### Benefits and barriers to participation in surveillance

Regarding the benefits and barriers to participation in surveillance, the baseline results reflect the responders’ expectations of participation, whereas the results at 1 year are an evaluation of the perceived benefits and barriers after the first year of surveillance.

#### Benefits

The response rate for “benefits of participation” was 95% (57 of 60) at baseline and 83% (50 of 60) at year one. No differences were seen in the perception of benefits among participants when compared to...
### TABLE 2  
Cancer Worry Scale: baseline results comparing the groups “No previous cancer” with “Previous cancer”

<table>
<thead>
<tr>
<th>Question</th>
<th>Previous cancer</th>
<th>n</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>p*</th>
</tr>
</thead>
<tbody>
<tr>
<td>How often have you thought about your chances of developing cancer (again)?</td>
<td>No</td>
<td>28</td>
<td>1 (4)</td>
<td>4 (14)</td>
<td>16 (57)</td>
<td>7 (25)</td>
<td>0 (0)</td>
<td>.009</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>1 (3)</td>
<td>1 (3)</td>
<td>11 (37)</td>
<td>14 (47)</td>
<td>3 (10)</td>
<td></td>
</tr>
<tr>
<td>Have these thoughts affected your mood?</td>
<td>No</td>
<td>28</td>
<td>7 (25)</td>
<td>18 (64)</td>
<td>3 (11)</td>
<td>0 (0)</td>
<td>x</td>
<td>.002</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>3 (10)</td>
<td>13 (43)</td>
<td>9 (30)</td>
<td>5 (17)</td>
<td>1 (3)</td>
<td></td>
</tr>
<tr>
<td>Have these thoughts interfered with your ability to do daily activities?</td>
<td>No</td>
<td>28</td>
<td>24 (86)</td>
<td>3 (11)</td>
<td>1 (4)</td>
<td>0 (0)</td>
<td>x</td>
<td>.004</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>15 (50)</td>
<td>9 (30)</td>
<td>5 (17)</td>
<td>1 (3)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>How concerned are you about the possibility of developing cancer (again) one day?</td>
<td>No</td>
<td>28</td>
<td>2 (7)</td>
<td>11 (39)</td>
<td>11 (39)</td>
<td>4 (14)</td>
<td>x</td>
<td>.028</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>0 (0)</td>
<td>8 (27)</td>
<td>10 (33)</td>
<td>12 (40)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>How often do you worry about developing cancer (again)?</td>
<td>No</td>
<td>28</td>
<td>3 (11)</td>
<td>8 (29)</td>
<td>15 (54)</td>
<td>2 (7)</td>
<td>0 (0)</td>
<td>.046</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>1 (3)</td>
<td>7 (23)</td>
<td>12 (40)</td>
<td>10 (33)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>How much of a problem is this worry?</td>
<td>No</td>
<td>28</td>
<td>15 (54)</td>
<td>12 (43)</td>
<td>1 (4)</td>
<td>0 (0)</td>
<td>x</td>
<td>&lt;.001</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>4 (13)</td>
<td>16 (53)</td>
<td>8 (27)</td>
<td>2 (7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>How often do you worry about the chance of family members developing cancer?</td>
<td>No</td>
<td>28</td>
<td>4 (14)</td>
<td>7 (25)</td>
<td>8 (29)</td>
<td>8 (29)</td>
<td>1 (4)</td>
<td>.226</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>1 (3)</td>
<td>4 (13)</td>
<td>14 (47)</td>
<td>11 (37)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>How concerned are you about the possibility that you will ever need surgery (again)?</td>
<td>No</td>
<td>28</td>
<td>9 (32)</td>
<td>13 (46)</td>
<td>6 (21)</td>
<td>0 (0)</td>
<td>x</td>
<td>.030</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>30</td>
<td>3 (10)</td>
<td>15 (50)</td>
<td>10 (33)</td>
<td>2 (7)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

x = the response alternative is not included in the question.

The Mann-Whitney U test.

### TABLE 3  
Cancer Worry Scale: change in response category at 1 year compared with baseline according to “No previous cancer” and “Previous cancer” at baseline

<table>
<thead>
<tr>
<th>Question</th>
<th>Previous cancer</th>
<th>n</th>
<th>Lower at 1 year, No. (%)</th>
<th>No change at 1 year, No. (%)</th>
<th>Higher at 1 year, No. (%)</th>
<th>p*</th>
</tr>
</thead>
<tbody>
<tr>
<td>How often have you thought about your chances of developing cancer (again)?</td>
<td>No</td>
<td>19</td>
<td>4 (21)</td>
<td>9 (47)</td>
<td>6 (32)</td>
<td>.179</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>8 (38)</td>
<td>10 (48)</td>
<td>3 (14)</td>
<td></td>
</tr>
<tr>
<td>Have these thoughts affected your mood?</td>
<td>No</td>
<td>19</td>
<td>4 (21)</td>
<td>11 (58)</td>
<td>4 (21)</td>
<td>.219</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>7 (33)</td>
<td>13 (62)</td>
<td>1 (5)</td>
<td></td>
</tr>
<tr>
<td>Have these thoughts interfered with your ability to do daily activities?</td>
<td>No</td>
<td>19</td>
<td>0 (0)</td>
<td>18 (95)</td>
<td>1 (5)</td>
<td>.014</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>8 (38)</td>
<td>11 (52)</td>
<td>2 (10)</td>
<td></td>
</tr>
<tr>
<td>How concerned are you about the possibility of developing cancer (again) one day?</td>
<td>No</td>
<td>19</td>
<td>5 (26)</td>
<td>9 (47)</td>
<td>5 (26)</td>
<td>.075</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>12 (57)</td>
<td>7 (33)</td>
<td>2 (10)</td>
<td></td>
</tr>
<tr>
<td>How often do you worry about developing cancer (again)?</td>
<td>No</td>
<td>19</td>
<td>7 (37)</td>
<td>9 (47)</td>
<td>3 (16)</td>
<td>.356</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>4 (19)</td>
<td>14 (67)</td>
<td>3 (14)</td>
<td></td>
</tr>
<tr>
<td>How much of a problem is this worry?</td>
<td>No</td>
<td>19</td>
<td>4 (21)</td>
<td>12 (63)</td>
<td>3 (16)</td>
<td>.974</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>6 (29)</td>
<td>10 (48)</td>
<td>5 (24)</td>
<td></td>
</tr>
<tr>
<td>How often do you worry about the chance of family members developing cancer?</td>
<td>No</td>
<td>19</td>
<td>6 (32)</td>
<td>9 (47)</td>
<td>4 (21)</td>
<td>.271</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>9 (43)</td>
<td>11 (52)</td>
<td>1 (5)</td>
<td></td>
</tr>
<tr>
<td>How concerned are you about the possibility that you will ever need surgery (again)?</td>
<td>No</td>
<td>19</td>
<td>8 (42)</td>
<td>7 (37)</td>
<td>4 (21)</td>
<td>.557</td>
</tr>
<tr>
<td></td>
<td>Yes</td>
<td>21</td>
<td>5 (24)</td>
<td>13 (62)</td>
<td>3 (14)</td>
<td></td>
</tr>
</tbody>
</table>

Using the individual changes, the Mann-Whitney U test.
those without previous cancer at baseline (Table S4), and there were no significant differences in changes of response category after 1 year. In general, both groups scored high on these items, indicating a positive view of the benefits with surveillance (Table S5).

Barriers

The response rate for “barriers to participation” was 93% (56 of 60) at baseline and 80% (48 of 60) at year one. There were no differences at baseline (Table S6) or after 1 year in perceived barriers to surveillance between participants with or without cancer. Overall, few participants reported perceived barriers to participation (response category ≥3). Most individuals did not score differently after 1 year of surveillance (Table S7).

SF-36

The response rate for SF-36 was 93% (56 of 60) at baseline and 83% (50 of 60) at year one. Comparisons of outcomes in SF-36 at baseline and at 1 year (Figure 2) were compared with normative age- and gender-adjusted population data (Figure 3).29 Generally, Cronbach’s α indicated a good internal consistency for the different subscales; physical functioning (10 items, α = 0.89), role limitations due to physical functioning (four items, α = 0.9), general health (five items, α = 0.8), role emotional (three items, α = 0.86), social functioning (two items, α = 0.74), bodily pain (two items, α = 0.93), vitality (four items, α = 0.87), and mental health (five items, α = 0.86). When the study population as a whole was compared with the normative age-and gender-adjusted population data for SF-36, the study participants scored clinically significantly lower on the items regarding vitality and emotional role functioning (role limitations due to mental health difficulties). Participants with a previous personal cancer scored significantly lower on some of the SF-36 subscales, such as “physical functioning,” “role limitations due to physical functioning,” and “general health” at inclusion in the SWEP53 study. At the 1-year follow-up, between-group differences were found for social functioning and for physical function.

DISCUSSION

This study presents the views of participants in surveillance with WB-MRI focusing on two groups of TP53 carriers – individuals with and without a previous personal cancer diagnosis.

In terms of sociodemographic characteristics, our cohort reflects the previously reported studies of surveillance in TP53 carriers with regard to median age (40 years old in our study and 38–43 years old in other studies13–17) and the female dominance of 65% in SWEP53, compared with 61%–94% in the other studies, except one reporting 47%.15 In SWEP53, 53% reported a previous personal cancer history, slightly less than most of the abovementioned studies that report 65%–88%, with the exception of Bancroft et al.15 reporting 34%. We also included families that were identified through mainstream genetic testing due to an index with breast cancer, which probably explains this difference. In summary, our cohort consists of more participants unaffected by cancer in comparison with previous reports, but is otherwise comparable. The cohort size of 60 participants is quite small, but in the light of the rareness of hTP53rc, it constitutes, to our knowledge, the largest published psychosocial evaluation of surveillance to date.

![Figure 2](https://acsjournals.onlinelibrary.wiley.com/doi/10.1002/cncr.34631)

**Figure 2.** SF-36 at baseline and at year 1: comparison between the general population and the study cohort. *Indicates clinically significant differences (≥10 points difference in mean value34) between TP53 carriers compared to the population (matched for age and gender). Numbers show mean values at baseline.
Participants with previous cancer reported in general more cancer worry than carriers without previous cancer according to the responses at both assessment points. Notably, there were no between-group differences regarding worry about a family member being affected. This might be a consequence of the fact that the majority of the participants (85%) had a first-degree relative with cancer and 60% had experienced cancer-related death in a first-degree relative. At the 1-year follow-up, the only remaining difference between the two groups was for the item “Have these thoughts interfered with your ability to do daily activities?” where 45% (18 of 40) without previous cancer scored the same as at baseline in contrast to the ones with previous cancer, with 20% (8 of 40) scoring lower than at baseline. It is reasonable that cancer worry increases with time and this is also supported by other articles regarding patients with other cancer predisposing syndromes. 

Bancroft et al. showed no differences in cancer worry between persons with a personal cancer history compared with those without, but they only included patients with previous cancer that had occurred more than 5 years ago. Rippinger et al. reports no significant difference in attitudes toward surveillance or not with regard to previous cancer diagnosis or affected family members. They also reported that 61% (30 of 49) of their participants had an adequate risk perception of getting cancer from “quite high” to “high” regardless of familial cancer history or own cancer history. The other studies have not performed comparable quantitative analyses. Cancer worry might also be affected by the need of further workup after the WB-MRI. Our cohort is too small to allow for a comprehensive statistical analysis in this first presentation. Other researchers present somewhat conflicting data. Bancroft et al. reports no significant differences between TP53 carriers who had to undergo further investigations due to radiological findings at baseline in comparison with carriers who had a normal scan. Another study by McBride et al. only two of 17 participants reported that the burden of screening was increased due to further examinations. Clearly, this aspect needs to be further investigated with larger cohort sizes.

Professional teams around TP53 carriers should be aware and prepared for cancer worry regardless of previous cancer history. It could be valuable to evaluate the use of repeated cancer worry scales as clinical intervention to monitor the need for psychosocial support during surveillance.

The majority of the participants in our study expressed positive attitudes toward the surveillance program and reported few barriers to participation. Except for having to take time off from work and possibly some travel expenses, the imaging itself was performed within the study and did not come at an extra cost for the participants. With regard to benefits and barriers to surveillance participation, the results seem to be in line with previous work reporting mainly a benefit of taking part in surveillance programs with WB-MRIs despite more worry and anxiety connected to the hereditary risk of cancer and anxiety related to the scans and awaiting the results. The fact that most participants in our study reported no difference in perceived benefits at baseline compared with the year-one assessment could be a ceiling effect, where persons had a high score already in the first evaluation. After 1 year, most participants expecting the barriers to be few at baseline had not changed their mind and continued to score as they did at baseline. In addition to the ceiling effect, 10 of the participants were already performing WB-MRIs before the study inclusion and therefore would reasonably already have a positive attitude toward surveillance. Another possible reason for the positive attitudes expressed in our study may be that only those who participated in the surveillance program...
responded to the questionnaires. People tend to defend their actions and engage in behaviors that they find valuable.\textsuperscript{34}

Results from the SF-36 indicated that participants with a previous cancer diagnosis scored lower on some subscales. These results are expected and in line with a previous report,\textsuperscript{14} indicating that being a TP53 carrier negatively affects one's view of vitality and emotional role functioning in comparison with noncarriers.

A standardized surveillance protocol, as suggested by Rippinger et al.,\textsuperscript{15} is now offered within the SWEP53 study to all individuals with hTP53rc in all regions in Sweden, indicating a strength of this study as other countries might be limited in offering study participation secondary to insurance status or where the person resides geographically.\textsuperscript{15,16} In our study, we did not come on any significant findings indicating major inconvenience or insurance problems. The latter could be a reflection of the publicly available Swedish health care system. Despite being informed of the risks of further workup after WB-MRI as well as uncertainty of survival benefits, the participants held a beneficial attitude toward surveillance regardless of having a previous cancer or not.

Considering that this cohort is focusing on a rare cancer predisposition syndrome, distinctive strengths of this national multicenter study were the relatively large cohort size (60 participants), a high compliance of questionnaire completion (95% response rate on average for all questionnaires at baseline, and 79% at year one), the use of standardized questionnaires and the longitudinal study design (1 year), with comparisons of the results over time. Limitations include a lower response rate at year 1 for the CWS. This might be due to administrative failure regarding the collection of the CWS as there were no differences between participants with/without cancer regarding nonresponses. Another possible limitation to our study may be a selection bias toward participating TP53 carriers that may have an overall more positive view of surveillance because they accepted participation in the surveillance protocol. This limitation is shared with previous publications and the aforementioned studies. Although eligible carriers who do not consent to the study are not registered, our experience is that the vast majority are included. From the first set of TP53 carriers who were offered inclusion, 15 of 16 consented to participation. Randomization between surveillance and no surveillance is considered unethical in individuals with a known high life time risk of developing cancer and is especially difficult because of its rarity. This would differentiate the follow-up among members of the same family, especially when there are reports of early cancer detection\textsuperscript{35} and a survival benefit.\textsuperscript{36} Therefore, it is reasonable to continue to offer surveillance programs to all TP53 carriers within a study format to ensure further evaluation of clinical and patient-reported outcomes.

Cancer predisposition syndromes are rare, requiring multi-professional teams with high syndrome-specific knowledge. By taking part in nationally coordinated studies like SWEP53, the participants are ensured easier access to the health care system, including psychosocial support. This might also affect the individuals’ view of study participation, as has been discussed by other researchers.\textsuperscript{13,14,17}

In conclusion, WB-MRI appears beneficial for all carriers and does not appear to increase cancer worry. Surveillance with WB-MRI is feasible from a psychosocial point of view, both among TP53 carriers with as well as those without a previous history of cancer.

**AUTHOR CONTRIBUTIONS**

Emma Tham: Conceptualization, formal analysis, funding acquisition, project administration, resources, supervision, and writing–review and editing. Lennart Blomqvist: Conceptualization, formal analysis, supervision, and writing–review and editing. Meis Omran: Data curation, formal analysis, project administration, writing–original draft, and writing–review and editing. Svetlana Bajalica-Lagercrantz: Conceptualization, formal analysis, funding acquisition, project administration, supervision, and writing–review and editing. Yvonne Brandberg: Conceptualization, formal analysis, supervision, and writing–review and editing. Annika Baan: Resources and writing–review and editing. Claudia Lundgren: Resources and writing–review and editing. Ekaterina Kuchinskaya: Resources and writing–review and editing. Gustav Silander: Resources and writing–review and editing. Jamila Adra: Resources and writing–review and editing. Marie Stenmark-Aksman: Resources and writing–review and editing.

**ACKNOWLEDGMENTS**

We thank all study participants and the Swedish Clinical TP53 Study Group for collaborations within the SWEP53 Study, Helene Carson for translating the questionnaires from Swedish back to English, and all the coordinators for collecting the questionnaires (Annika Baan, Madeleine Dewerand, Monica Emmertz, Margareta Karlsson, Anne Kinhult Ståhlbom, Susanna Lindström, Carin Nylander, Barbro Silfverberg, and Sofia Åslund). Funding has been received from the Cancer Research KI at Karolinska Institutet, the Cancer Research Funds of Radiumhemmet (201052), the Rare Disease Research Foundation, Stockholm County Council (SLL20180046 and SLL500306), the Swedish Cancer Society (CAN 2016/775), and the Swedish Childhood Cancer Fund (TJ2018-0054 and TJ2021-0125). The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript; or in the decision to publish the results.

**CONFLICTS OF INTEREST**

Lennart Blomqvist is cofounder of Collective Minds Radiology. Ekaterina Kuchinskaya reports consulting fees from Karolinska University Hospital. Niklas Loman reports consulting fees from Lund University Medical Faculty Foundation. Meis Omran reports grant/contract funding from Radiumhemmets Forskningsfonder and Rare Disease Foundation. Emma Tham reports grant/contract funding from Barncancerfonden and Stockholms Läns Landsting. The other authors made no disclosures.
REFERENCES


33. Arver B, Haegermark A, Platten U, Lindblom A, Brandberg Y. Evaluation of psychosocial effects of pre-symptomatic testing for breast/...


SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

How to cite this article: Omran M, Johansson H, Lundgren C, et al. Whole-body MRI surveillance in TP53 carriers is perceived as beneficial with no increase in cancer worry regardless of previous cancer: data from the Swedish TP53 Study. Cancer. 2023;129(6):946-955. doi:10.1002/cncr.34631