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ORIGINAL RESEARCH

Men's experiences of recontact about a potential increased risk of prostate cancer due to Lynch Syndrome: "Just another straw on the stack"

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Abstract

The practice of recontacting patients to provide new health information is becoming increasingly common in clinical genetics, despite the limited research to evidence the patient experience. We explored how men with Lynch Syndrome (LS) understand and experience being recontacted about a potential increased risk of prostate cancer. Sixteen men with LS (*Mean* age 51 years) were recruited from an Australian screening study to undergo a semi-structured interview. A modified grounded theory approach was used to guide data collection and thematic analysis. Qualitative coding was shared by the research team to triangulate analysis. The practice of recontact was viewed by participants as acceptable and was associated with minimal emotional distress. The majority of men understood that they may be above population risk of prostate cancer, although evidence was still emerging. Men reported high engagement with personal and familial health, including regular screening practices and familial risk communication. Findings suggest that men's carrier status and beliefs about the actionability of the new cancer risk information influence their response to recontact. Recontact practices that include the offer of risk management strategies may lead to improved patient outcomes (e.g., reduced cancer worry and increased health engagement), if perceived as valuable by recipients.

KEYWORDS
clinical genetics, Lynch Syndrome, male, prostate cancer, recontact

1 | INTRODUCTION

As genomic technologies are continually introduced into clinical genetics, there is an increasing need to recontact individuals with new health information. In cancer genetics, recontact has implications for individuals diagnosed with a familial cancer syndrome (e.g., Lynch Syndrome [LS]). Recontacting patients with new cancer risk information can have personal implications for the individuals' treatment and surveillance, as well as implications for other family members.

There has been much debate on the ethics and feasibility of recontact, and whose role it is to deliver additional information based on genomic data in clinical practice (Carrieri et al., 2017b, 2016; Johns et al., 2017; Letendre & Godard, 2004; Otten et al., 2015; Sirchia et al., 2018). Despite the lack of professional consensus, health care professionals continue to recontact patients in various settings (Dheensa et al., 2017; Forrest & Young, 2016; Johns et al., 2017; Otten et al., 2015; Sirchia et al., 2018). Recontacting patients

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with new genetic information will become more common. As genetic counselors will be central to this process, it is important to understand patient responses to inform genetic counseling practice and professional guidelines. Patients' experiences of this process remain understudied and there is little research available to inform clinical practice guidelines (Carrieri et al., 2017a, 2016; Otten et al., 2015; Sirchia et al., 2018). In cancer genetics, the need for patient-focused research is rapidly growing in tandem with advances in genomic technology and the increased occurrence of recontact in clinical practice.

Lynch Syndrome is an inherited cancer predisposition syndrome caused by pathogenic variants in DNA mismatch repair genes (e.g., *MLH1*, *MSH2*, *MSH6*, or *PMS2*) (Stoffel & Boland, 2015). LS is characterized by early onset colorectal cancer and other extra-colonic tumors: for example, endometrial, stomach, and ovarian (Stoffel et al., 2009). Risks differ depending on the mismatch repair gene involved, although the main cancer risk for individuals with LS is colorectal cancer, with an estimated lifetime risk of 70% for men and 40% for women, both peaking in the 50s (Jenkins et al., 2015; Stoffel et al., 2009). An increased incidence of prostate cancer in LS has implications for risk management guidelines, including additional screening for men as prostate-specific antigen (PSA) screening is available for high-risk individuals (Catalona, Antenor, Roehl, & Moul, 2002; Schröder et al., 2012).

The international IMPACT clinical study (Identification of Men with a genetic predisposition to ProstAte Cancer: Targeted screening in men at a higher genetic risk and controls; 05/MRE07/25) is currently investigating PSA screening in men aged 40–69 years with pathogenic variants in *BRCA1/2* or selected LS-associated genes (*MLH1*, *MSH2*, *MSH6*). The LS-specific arm aims to identify if men with LS have an increased risk of prostate cancer, whether PSA screening is effective, and to identify novel biomarkers for the early detection of prostate cancer. Recruitment to the IMPACT clinical study involved clinical genetic services recontacting men with LS by letter to notify them of a potential increased risk of prostate cancer and offering research-based PSA screening. As part of the Australian arm of this multicenter study (HREC 06/31) at the Peter MacCallum Cancer Centre (PMCC) in Melbourne, we conducted a psychosocial study exploring the impact of being recontacted about a potential increased prostate cancer risk for men enrolled in the IMPACT clinical study with LS.

To date, very few studies have focused on patients' views of recontact, especially in cancer genetics. Using a hypothetical methodology, Griffin et al., (2007) found that the majority of 354 individuals who attended colon cancer risk assessment clinics had positive attitudes toward recontact by genetics service providers (Griffin et al., 2007). Participants' preferences for recontact were highest for the delivery of new information with implications for individual health (84%); familial cancer risk (78%); individual cancer risk (77%); and clinical screening options (64%). Approximately 42% of participants did not identify a situation in which they would not like to be recontacted (Griffin et al., 2007).

A recent interview study on general views of recontact showed that patients and parents felt that recontact was desirable when the information is clinically actionable or could provide a diagnosis (Carrieri et al., 2017a; Dheensa et al., 2017). Only four participants, of the 41 who took part in the study, had been recontacted by a genetics service (Carrieri et al., 2017a; Dheensa et al., 2017), highlighting the limited empirical evidence base for this practice. Recently, a small number of studies suggested that the majority of individuals recontacted for a variety of conditions have positive attitudes to recontact, although some individuals may be wary of the additional information because of the potential for negative emotions (Bernard, McGillivray, Van Allen, Friedman, & Langlois, 1999; Beunders, Dekker, Haver, Meijers-Heijboer, & Henneman, 2018; Romero Arenas et al., 2018; Sexton, Sahhar, Thorburn, & Metcalfe, 2008). These studies highlight that the experience of recontact is psychologically complex and can lead to ambivalent responses (Carrieri et al., 2017a).

There is a lack of research addressing how patients experience being recontacted, especially in cancer genetics. Consequently, the psychosocial impact of recontact, including respect for the right not to know, and its influence on health behaviors, including screening practices and familial interactions, remain understudied (Letendre & Godard, 2004). In order to inform clinical practice and assist in developing clinical guidelines for recontact, this study aimed to explore how men with LS understand and experience recontact about a potential increased prostate cancer risk.

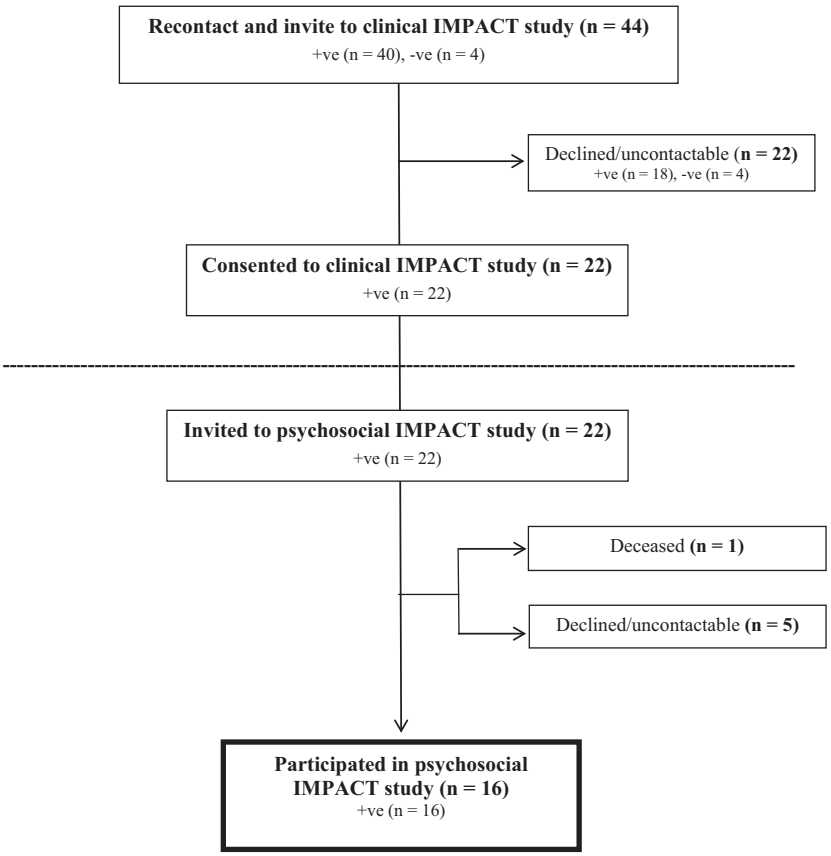
2 | MATERIALS AND METHODS

2.1 | Participants

Participants were recruited from the Familial Cancer Centre (FCC) at the PMCC, Melbourne, Australia. Between 2014 and 2016, all men aged 40–69 years known to the FCC who tested positive or negative for a pathogenic variant in *MLH1*, *MSH2*, or *MSH6*, or those not tested and at 50% risk were recontacted by letter and invited to participate in the clinical IMPACT study (n = 44; Figure 1).

The recontact letter explained a potential increased risk of prostate cancer for men from families with LS and the PSA screening study. Recruitment was also carried out in person by FCC clinical staff if potential participants were attending an appointment; follow-up phone contact was made to all non-responders. Twenty-two men subsequently consented to participate in the clinical IMPACT study. There was no statistical difference between IMPACT clinical study participants and decliners based on age (p = 0.136) or for relative socioeconomic advantage and disadvantage (p = 0.407) (Australian Bureau of Statistics, 2016b; see Table 1).

We purposively sampled all men who: (a) tested positive for an LS-associated pathogenic variant; and (b) had consented to the clinical IMPACT study (n = 22), and invited them to the IMPACT psychosocial study (Figure 1). Ethics approval for this study was obtained from the PMCC Human Research Ethics Committee (LNR/15/PMCC/114).



Abbreviations: +ve = *MLH1, MSH2 or MSH6* pathogenic variant detected; -ve = No variant detected.

FIGURE 1 Flowchart of men participating in the IMPACT psychosocial study

2.2 | Data analysis

Data were collected via in-depth semi-structured interviews conducted between April and August, 2016. Interviews lasted 25–90 min and were conducted by R.F.S either in person or by telephone. An interview guide based on the literature and the research aims was used and explored previous experiences of LS, experiences of being recontacted about a potential prostate cancer risk, and engagement with individual and familial health. Interviews were audio recorded and transcribed verbatim; all identifying information was removed and pseudonyms are used throughout. Further recruitment was available as the IMPACT clinical study continued, but thematic saturation was reached for key research questions following 16 interviews and further recruitment ceased.

A modified grounded theory approach was used to guide concurrent data collection and analysis (Grbich, 2007; Strauss & Corbin, 1990). The data were analyzed using the method of constant comparison, which allows for systematic identification, comparison, and coding of themes within and across interviews (Strauss & Corbin, 1990). Members of the research team (V.R, R.F.S, and M.A.Y) coded the transcripts to triangulate data analysis and achieve greater analytical rigor (MacQueen, McLellan, Kay, & Milstein, 1998). Coding inconsistencies were settled by group discussion and analytical justification. NVivo v11.2.2 (QSR International Pty Ltd) was used for data management.

3 | RESULTS

Twenty-two men with LS who enrolled in the IMPACT clinical study were invited to participate in the IMPACT psychosocial study. Five men declined to participate and one was deceased. Overall, 16 men consented to participate (Figure 1). Participant demographic characteristics are summarized in Table 2. Mean age was 51 years (range 43–68 years); mean time since recontact and the interview was 1.5 years (range 0.9–1.7 years); mean time since genetic testing for LS was 8.7 years (range 2–15 years). A quarter had previously been diagnosed with cancer at least once at a mean age of 43 years. The majority were partnered (69%) and had children (81%). Six (38%) had graduated from university.

Thematic analysis revealed three major themes related to: (a) response and integration of information provided as a result of recontact; (b) prostate cancer screening as a mechanism to moderate psychological distress; and (c) appraising and understanding a potential increased risk of prostate cancer. Importantly, all three themes are significantly contextualized by participants' previous lived experiences

of LS. The majority of men had undergone genetic testing for LS on average 9 years prior to the research interview and were well adapted to their high-risk status, a quarter also having developed cancer at least once. Overall, participants perceived genetic testing for LS as useful and a catalyst for positive lifestyle changes (e.g., quitting smoking, reducing alcohol intake, and increasing exercise) and engagement in regular colorectal screening. Each theme is reported below with supporting quotes in text and supplementary quotes in Tables 3 and 4.

3.1 | Theme 1: Response and integration of recontact information

Regardless of mode of recontact, participants reported low levels of worry in response to being recontacted and learning about a potential increased prostate cancer risk. Many men described themselves as "relaxed," "comfortable," and "not fazed" following the notification (Table 3, quote 1).

> The prostate one didn't have much of an impact on me at all to be honest.
>
> (Ben, 43, unaffected, recontacted by letter)

The low level of worry expressed by participants and acceptance of information of a potential increased risk of prostate cancer was intertwined with a number of factors. Men who had a previous cancer diagnosis expressed acceptance toward learning of their potential prostate cancer risk. One man described prostate cancer risk as a minor addition to his strong cancer narrative.

> Too used to it [cancer]. I've been dealing with it [cancer] for a long time, so as I said I wasn't surprised, and you know, [it] couldn't be any worse than what I've been through already... So we've been through a fair bit in the last 15 years, so it [prostate cancer risk] was just another straw on the stack.
>
> (Dylan, 46, previous cancer diagnosis, recontacted by letter)

Peter, who had previously developed multiple cancers, coped with his potentially increased cancer risk by including prostate cancer as one of a number he is at risk of.

TABLE 1 Participant and decliner information for the IMPACT clinical study

	Participants	Decliners	*p*-value
n (%)	22 (50)	22 (50)	
Gene status, *n* (%)			
+ve	22 (100)	18 (82)	
−ve	0 (0)	4 (18)	
Mean age (*SD*)	49.59 (7.7)	53.59 (9.62)	0.136[a]
SEIFA quintile[b]	Observed (%)		
1	2 (9)	6 (27)	
2	1 (5)	2 (9)	
3	3 (14)	2 (9)	
4	7 (32)	3 (14)	
5	9 (41)	9 (41)	
Total	22	22	0.407[c]

Note. +ve: *MLH1*, *MSH2* or *MSH6* pathogenic variant detected; −ve: no variant detected; IMPACT: Identification of Men with a genetic predisposition to ProstAte Cancer; SEIFA: Socio-Economic Indexes for Australia, index of relative socioeconomic advantage and disadvantage.
[a]*t* test (unpaired). [b]Pooled as quintiles from deciles: lower quintile values indicate areas with lower relative socioeconomic advantage/disadvantage (i.e., people's access to material and social resources, and their ability to participate in society; Australian Bureau of Statistics, 2016b). [c]Fisher's exact test (two-tail).

TABLE 2 Participant characteristics for the IMPACT psychosocial study (*n* = 16)

Characteristic	Mean (range)
Age (years)	51.2 (43–68)
Age at first cancer diagnosis (years)	42.75 (33–54)
Time since genetic testing for LS and interview (years)	8.7 (2–15)
Time since recontact and interview (years)	1.5 (0.9–1.7)
	n (%)
Mode of notification	
Letter	15 (94)
Follow-up phone call	8 (50)
Clinic appointment	1 (6)
Previous cancer diagnoses	
Yes (at least 1)	4 (25)
No	12 (75)
Remoteness of residence[a]	
Major city	11 (69)
Inner regional	5 (31)
Relationship status	
Partnered	11 (69)
Un-partnered	5 (31)
Children	
Yes	13 (81)
No	3 (19)
Highest level of education[b]	
Secondary school[c]	7 (44)
Trade qualification	2 (12.5)
Undergraduate qualification	4 (25)
Postgraduate qualification	2 (12.5)

IMPACT: Identification of Men with a genetic predisposition to ProstAte Cancer; LS: Lynch Syndrome.
[a]Remoteness classification as per the Australian Statistical Geography Standard (Australian Bureau of Statistics, 2016a). [b]One participant did not provide education information. [c]Secondary school collectively refers to completing up to year 10/12.

> I've got Lynch Syndrome, I'm going to get cancer… it's easier in my head if I say that, I'm going to get it, I'm going to get it back. Where? Prostate is one of the areas. That's it for me.
> (Peter, 54, previous cancer diagnosis, recontacted by letter)

Other men expressed low levels of prostate cancer worry because they had anticipated their prostate cancer risk in the context of getting older or having LS. Men's age had an impact on the expected nature of their prostate cancer risk: "I'm getting to that age bracket where I should be aware of prostate risks anyway" (Ben, 43, unaffected, recontacted by letter). Some felt being at risk of prostate cancer was a logical step in the context of having LS: "It just felt scientifically logical, and that if there were half a dozen others [cancers] already identified, that prostate would also be part of [LS]" (Tom, 60, unaffected, recontacted by letter). Others reported a limited emotional response because the information did not impact their "day-to-day" and there was "no point going on about it" because ultimately the information was probabilistic not absolute.

> Well I suppose medically speaking I haven't got it [prostate cancer] … so why stress about something that you don't have?
> (Oscar, 48, unaffected, recontacted by letter)

One participant described his low levels of prostate cancer worry by comparing the differences between colorectal and prostate cancer risk management. LS for him was a "critical condition," whereas prostate cancer was managed by a simple annual blood test (Table 3, quote 2). Similarly, another participant with a strong family history of colon cancer described colon cancer risk as being "more real" than his risk of prostate cancer.

3.2 | Theme 2: Utility of prostate cancer screening in containing psychological distress

Some men described elevated prostate cancer-specific distress after being recontacted. However, the participants described how this distress was moderated through their enrollment in regular screening:

> It [prostate cancer risk] stays with me all the time. Probably a week doesn't go by when I don't think about it. I think that I'm looking forward to the next part of the testing; to make sure I'm still covered.
> (Steve, 62, previous cancer diagnosis, recontacted by letter)

Being enrolled in prostate-specific screening was a critical mediator of men's cancer worry, coping, and acceptance. By being offered an immediate option to manage their prostate cancer risk, albeit with an emerging evidence base, many men were at ease with recontact about a potential increased prostate cancer risk (Table 3, quote 3).

TABLE 3 Quotes illustrating Themes 1 and 2

Quote number	Participant	Quote
Theme 1: Response and integration of recontact information		
1	Lachlan, 43, unaffected, recontacted by letter	Oh, look, I think I wasn't particularly fazed by it.
2	Oscar, 48, unaffected, recontacted by letter	One's [LS] a critical condition that I have to deal with each year with the professor, and the other one's [prostate cancer] a test that I just have done each year and I've got no symptoms… When I wait for the results from each one, I suppose I'm a little bit unnerved to be perfectly honest in relation to the Lynch's one: did they take a polyp, is there any issues? Compared to the research one [where] I'm pretty confident there's nothing there because of my age and my previous history.
Theme 2: Utility of prostate cancer screening in containing psychological distress		
3	Graham, 52, unaffected, recontacted by letter	If they weren't monitoring me every year, yeah, I would feel a lot different about it. Because they're doing the monitoring and keeping an eye on me, I know they're going to get something straight away, if anything does come. That's the peace of mind.
4	Tom, 60, unaffected, recontacted by letter	With prostate cancer, as long as you're under regular screening, the likelihood is that it can be stopped. But you know there's always the percentage that can't. So, I was probably comfortable.
Subtheme: Response to mode of recontact		
5	Lachlan, 43, unaffected, recontacted by letter	Oh well, I mean look that's, probably from my point of view, that's the way I'd prefer to receive the information [by letter]. You know, I'm not sure how you could do it any other way really. I mean I certainly wouldn't want someone ringing me up telling me I had an increased risk of prostate cancer [chuckles]. So, no, you know, the information is… you know the information wasn't alarmist, it just sort of said, "Look, there's new information, we're now doing some more studies and research into it," so yeah. No, I was very happy with the way it was presented.

Note. LS: Lynch Syndrome previous dx: previous cancer diagnosis; unaffected: no cancer.

TABLE 4 Quotes illustrating Theme 3

Quote number	Participant	Quote
Theme 3: Appraising and understanding a potential increased risk of prostate cancer		
1	Dylan, 46, previous dx, recontacted by letter	I understand it's [the link between prostate cancer and LS]… still in its formative stages in terms of research. I mean they're still trying to, as I understand it, confirm that there's a higher risk. I mean I understand that they are reasonably certain that there is, but they're obviously trying to confirm that without a doubt.
2	Tom, 60, unaffected, recontacted by letter	There's obviously the primary mutation where it affects you, and then these other ones [cancer risks] are all probabilities… I see now quotes that it's [prostate cancer risk] doubled to five times the probability for someone with Lynch Syndrome… so it looks like it's sort of a race to one or the other at the moment almost, colon or prostate.
3	Peter, 54, previous dx, recontacted by letter	For me it's just where else is it going to come back? …I don't care if it's prostate or my lung again… for me it's I know it's just going to come back… I know it's going to come back; it's just a matter of when… Prostate is just, oh well, what else are you going to lose? You've lost most of it.
4	Oscar, 48, unaffected, recontacted by letter	I guess you've got a chance of getting it [prostate cancer], but you've also got a… you know I do a thousand Ks a week in the car, I've got a good chance of getting hit by a truck, you know what I mean, like sometimes I'm in a very dangerous lane.
5	Finn, 64, previous dx, recontacted by letter	It's not good saying one, and then plus this. It's just easier to put it [cancer risk] all together, and say you're at a higher risk and just be wary of it, that's all.
Subtheme: Understanding research aims		
6	Dylan, 46, previous dx, recontacted by letter	…Well the only way I can be tested [for prostate cancer] … is through a blood test, and I know that that's not always very accurate. So, I haven't had any advice as to how often I should be tested, or how reliable it is… You can get a lot of false negatives and false positives…
7	Phil, 43, unaffected, recontacted by letter	So, I understand there's an increased [prostate cancer] risk, and they're using the [PSA] as a biomarker to detect whether there is an increased risk with people who do have a history of prostate cancer, and have Lynch Syndrome. So that's my understanding of what [IMPACT] was around… For example, [if you have] elevated levels of [PSA] you might go and then have a further physical examination to see if you do actually have an enlarged prostate, and whether or not you need to get a biopsy… So, it's an early stage marker, but it's not considered to be the most reliable one at the moment.

Note. LS: Lynch Syndrome; previous dx: previous cancer diagnosis; unaffected: no cancer.

> I understand there's a risk…I'm at peace with it, because I know that they're monitoring me every year.
> (Graham, 52, unaffected, recontacted by letter)

Woven into this perspective was a realistic awareness that screening could not prevent prostate cancer, although PSA screening nonetheless provided a level of comfort (Table 3, quote 4). Overall, learning of their potentially increased risk of prostate cancer amounted to adding "another straw on the stack," where prostate cancer was just another cancer risk to face as an individual with LS.

3.2.1 | Response to mode of recontact

The majority of participants in the psychosocial IMPACT study were informed about potential increased prostate cancer risk and PSA screening by letter, and felt it was appropriate (Table 3, quote 5). One participant was notified when he attended an appointment for LS risk management with a trusted health professional (Table 2). All participants expressed that these various modes of notification as personally appropriate.

3.3 | Theme 3: Appraising and understanding a potential increased risk of prostate cancer

Participants differed in their understanding of their potential prostate cancer risk. The majority acknowledged they may be at above population risk and recognized there was a link between LS and prostate cancer (Table 4, quote 1).

> Oh, I think it [prostate cancer risk] was heightened. Off the top of my head I believe I've got a heightened risk of it. Somehow, it's [LS and prostate cancer] connected. I don't quite understand the medicine or the science of it, but I understand that I may be in a higher category than somebody else.
> (Oscar, 48, unaffected, recontacted by letter)

Some participants possessed in-depth knowledge, accurately recalling prostate cancer risk estimates in the context of having LS (Table 4, quote 2). In contrast, other participants overestimated their risk to be a "50/50" chance (Graham, 52, unaffected, recontacted by letter) or even

greater, to a near certain future diagnosis. One man expressed a sense of fatalism when learning of his potential prostate cancer risk, describing his understanding in terms of his risk of recurrence: "where else is it going to come back?" (Table 4, quote 3). In general, participants drew on a wide variety of analogies and comparisons when interpreting their risk of developing prostate cancer. For some, prostate cancer was not considered a greater threat than other risks encountered in daily life: for example, driving long distances for work (Table 4, quote 4).

Overall, the men's perceived risk of prostate cancer was inextricably linked to their previous diagnosis of LS. Participants' understanding of LS as a multi-organ cancer predisposition syndrome was a ready means to understand information about a potential prostate cancer risk. Combining all cancer risks associated with LS as one construct was easier to express than stating cancer-specific risk estimates (Table 4, quote 5). In this way, new cancer risk information was integrated into participants' pre-existing understanding of LS-associated cancer risk.

3.3.1 | Understanding research aims

Most participants understood information of a potential increased risk of prostate cancer in the context of the clinical IMPACT study and the limitations of prostate cancer screening (Table 4, quote 6).

> Well the only way I can be tested [for prostate cancer] … is through a blood test, and I know that that's not always very accurate.
> (Dylan, 46, previous cancer diagnosis, recontacted by letter)

Other participants possessed in-depth knowledge about PSA testing and the purpose of IMPACT (Table 4, quote 7). These participants were predominantly university graduates.

Reflecting the range of understanding and misunderstanding among participants, one man confused clinical risk management strategies for LS with the PSA screening being offered as part of the clinical IMPACT study:

> It was explained to me that by giving blood they are able to pick up whether I do have cancer or not. It's apparently… a very thorough process… Sometimes I think if the doctor misses say a polyp, my feeling—you correct me if I'm wrong—but that blood test might show up something, like if I had cancer.
> (James, 52, unaffected, recontacted in clinic)

4 | DISCUSSION

The results of this exploratory study show that recontacting men with LS about a potential increased prostate cancer risk had minimal emotional impact. The use of a letter to both notify participants of their potential risk and invite them to the clinical IMPACT study was considered acceptable and appropriate by the majority of participants. Overall, the findings suggest that participants comprehended the new risk information and integrated it into existing beliefs about their LS carrier status, including their previous personal and familial experiences of LS. The findings contribute to a small body of emerging research assessing the psychosocial impact of undergoing research-based comprehensive screening for multiple malignancies (Barez, Blasco, Fernandez-Castro, & Viladrich, 2009; McBride et al., 2017; Ross et al., 2017).

Most men had adapted well to having LS and their understanding of LS as a multi-organ cancer predisposition syndrome overshadowed the threat of prostate cancer. This was emphasized by the description of the prostate cancer risk as "just another straw on the stack." These results are consistent with previous studies that acknowledge individuals' use of frameworks to understand and cope with risk, rather than treating risk as a stand-alone concept (Butow, Lobb, Meiser, Barratt, & Tucker, 2003; Croyle & Lerman, 1999; Sivell et al., 2008). The findings may be further explained by the study context, which involved recontact from the FCC where participants had previously accessed services including genetic counseling, genetic testing and on-going risk management for LS. Participants' familiarity of interacting with the FCC to manage LS may have mediated the impact of recontact by IMPACT researchers.

Enrollment in a prostate-specific screening program appeared to facilitate men's coping and acceptance of the prostate cancer risk information. The offer of an immediate option to manage their prostate cancer risk, albeit with an emerging evidence base, put most men at ease during recontact and led to low cancer stress several months afterward. Consistent with previous research, these results suggest that recontact is justified and acceptable to patients if the information is actionable or bears personal relevance (Carrieri et al., 2017a; Dheensa et al., 2017; Griffin et al., 2007; Otten et al., 2015). For participants in this study, recontact, including the option to engage in research-based PSA screening, represented a course of action with personal relevance.

For these men, taking action and participating in screening could also provide a sense of safety and perceived control over the threat of prostate cancer. The availability of research-based screening was significant for men in this study who live with multiple cancer risks as a result of their LS status. The perceived benefits may extend beyond reducing morbidity and mortality to include psychological benefits, such as a source of emotional support and containment for patients (Lammens et al., 2010; McBride et al., 2017; Ross et al., 2017).

An important factor influencing men's response to recontact and PSA screening was their experience of long-term engagement with regular colorectal screening resulting in positive outcomes, including early detection of polyps and prolonged periods of living cancer free. Positive prior experiences of colorectal cancer screening may enhance men's sense of control over their prostate cancer risk when adopting a similar targeted approach (i.e., PSA), and explain the apparent low emotional impact associated with recontact. The observed relationship between beliefs about personal control and

cancer worry supports a large body of research demonstrating the role of perceived control in facilitating adjustment to threatening events (Barez et al., 2009; Beckjord, Glinder, Langrock, & Compas, 2009; Benyamini, Nouman, & Alkalay, 2016).

The relationship between research and clinical care is often blurred particularly in cancer genetics where research protocols provide screening that may be also offered in clinical practice (Hallowell, Cooke, Crawford, Lucassen, & Parker, 2009). Recontact with the option of research-based risk management also highlights the possibility of therapeutic misconception: where research participants assume that a research study will provide clinical benefit (Burke, Evans, & Jarvik, 2014). Most participants in this study understood that the research aims built upon emerging evidence linking increased risk of prostate cancer in men with LS, as well as the limitations of PSA screening. This suggests therapeutic misconception was not a major moderator of the men's emotional response to recontact. Rather, the men demonstrated awareness of the research aims and simultaneously hoped to benefit from an incidental aspect of the study: surveillance for prostate cancer risk. The opportunistic approach to enhancing one's clinical care, through research participation, has been labeled "therapeutic appropriation" (McDougall et al., 2016).

In the present study, the perceived benefits associated with therapeutic appropriation were found to extend beyond the individual. Similar to previous research on motivations to participate in genetics research, men in this study were motivated to participate in IMPACT for a combination of interdependent personal and altruistic motives (Hallowell et al., 2009). Participating in IMPACT offered them prostate cancer screening that simultaneously benefited themselves, their family and science in general. Therapeutic appropriation is important to consider when recontacting patients as it may be an important source of motivation for research participation.

4.1 | Study limitations

The sample characteristics and study context places limitations on the generalizability of results for recontact in other familial cancer populations. In particular, the men in this study had been diagnosed with LS several years prior to being recontacted. This meant that they all had a history of interactions with familial cancer services, including clinical research participation. The men's clinical trajectories suggest a consistent pattern of engaging in health services and seeking information and support.

4.2 | Research recommendations

The findings from this study would be strengthened by including the perspectives of LS carriers who chose not to participate in IMPACT and may therefore, have different responses to cancer threats and risk management. Furthermore, the study context involved recontact by researchers located within an FCC where participants received clinical care. This aspect may have increased participants' sense of trust in the information and beliefs about the efficacy of the research-based screening. While this scenario is not uncommon in familial cancer contexts, it may not be replicable to recontact procedures in other disease settings.

4.3 | Practice implications

This study demonstrates that recontact about new cancer risk information is an acceptable practice that may be delivered by an FCC with minimal emotional distress to patients. Low levels of cancer worry and high engagement with screening were positive outcomes reported by participants and were associated with beliefs about the actionability of the information. The men felt that they had both personal and clinical actions available to them to lower prostate cancer risk thereby, minimizing the threat. Engaging in screening in particular, although research-based, was a source of emotional support and containment for participants. Findings suggest that recontact to provide new cancer risk information should therefore be offered together with risk management strategies that are perceived as valuable by patients. This strategy may be sufficient for overcoming previous cautions in the literature regarding the psychological threat posed by recontact (Letendre & Godard, 2004). Recontact is likely to become more prevalent as genomic technology continues to be translated into clinical genetics practice. The results are important for genetic counselors who will increasingly be involved in recontacting patients with new, clinically significant information regarding their diagnosis or previous genetic testing. The low emotional impact experienced by men in this study provides further evidence for genetic counselors that for many patients recontact is not emotionally distressing and may be beneficial. The study findings may be used to aid in the development of clinical guidelines that are informed by the patient experience and facilitate positive patient outcomes.

AUTHOR CONTRIBUTIONS

Victoria Rasmussen made substantial contribution to the acquisition (e.g., recruitment) and analysis of qualitative data and interpretation of results; drafting of the manuscript and subsequent revisions based on co-author feedback. Rowan Forbes Shepherd made substantial contribution to the acquisition (e.g., recruitment and interviews) and analysis of qualitative data and interpretation of results; drafting of the manuscript and subsequent revisions based on co-author feedback. Laura Forrest collaborated on the design of the study; made substantial contribution to the interpretation of the data for the manuscript; and drafting of the work, including providing thoughtful feedback to inform revisions. Paul James made substantial contribution to the drafting of the work, including providing thoughtful feedback to inform revisions. Mary-Anne Young designed the study; made substantial contribution to the analysis of qualitative data and interpretation of results; and drafting of the work, including providing thoughtful feedback to inform revisions, granting final approval of the version to be published; and giving her agreement to be accountable for all aspects

of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interest

Victoria Rasmussen, Rowan Forbes Shepherd, Laura Forrest, Paul James, and Mary-Anne Young declare that they have no conflict of interest.

Human studies and informed consent

All procedures followed were in accordance with the ethical standards of responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from all individuals for all being included in the study.

Animal studies

This article does not contain any studies with animals performed by any of the authors.

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