From normal response to clinical problem: Definition and clinical features of fear of cancer recurrence

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Abstract

PURPOSE: Research to date on fear of cancer recurrence (FCR) shows that moderate to high FCR affects 22-87% of cancer survivors and is associated with higher psychological morbidity [1]. Despite growing research interest in FCR, lack of consensus on its definition and characteristics when it reaches a clinical level have impeded knowledge transfer into patient services.

METHODS: In order to address these gaps, expert researchers, policy makers, trainees, and patient advocates attended a two-day colloquium at the University of Ottawa in August 2015. A Delphi method was used to identify the most relevant definition of FCR and attendees generated possible diagnostic characteristics of clinical FCR.

RESULTS: After three rounds of discussion and voting, attendees reached consensus on a new definition of FCR: "Fear, worry, or concern relating to the possibility that cancer will come back or progress". Regarding clinical FCR, five possible characteristics were proposed: 1) high levels of preoccupation, worry, rumination or intrusive thoughts, 2) maladaptive coping, 3) functional impairments, 4) excessive distress, and 5) difficulties making plans for the future.

CONCLUSIONS: The new proposed definition of FCR reflects the broad spectrum in which patients experience FCR. A consensual definition of FCR and the identification of the essential characteristics of clinical FCR are necessary to accurately and consistently measure FCR severity and to develop effective interventions to treat FCR. We hope this broad definition can encourage further research and the development of inclusive policies for all cancer patients and survivors who are struggling with this issue.

Keywords: Fear of Cancer Recurrence, clinical Fear of Cancer Recurrence, Delphi study, expert opinion, definition, psychosocial oncology

Introduction

Fear of cancer recurrence (FCR) has become a popular topic of research in recent years, as evidenced by the recent publication of four reviews on this topic [1-4] and practice guidelines [5]. FCR is often defined as "the fear that cancer could return or progress in the same place or another part of the body" [6]. However, there is no consensus among researchers that this definition adequately describes the phenomenon of FCR. For example, it does not refer to the multidimensional nature of this fear, which may encompass triggers, emotions, thoughts, physiological reactions, and coping strategies [7]. Also, while there is consensus that FCR ranges from "normal" to "clinical", there is no agreement about what constitutes a clinical level of FCR. Discussion between researchers and clinicians is required to agree on the diagnostic characteristics of clinical FCR to enable accurate and consistent measurement [4]. Lack of agreement on a definition of FCR and clinical FCR was identified as the most urgent research need by a panel of FCR experts, policymakers and consumer advocates [8].

Methods

Procedure

Experts ~~with known FCR research programs~~ in the field of FCR were invited to attend a two-day colloquium on FCR in Ottawa, Canada, on August 3-4, 2015. Experts were selected by having published on the topic of FCR in the past 5 years and/or being involved in on-going trials of FCR interventions. These experts were asked to nominate colleagues who met these criteria. Nineteen researchers were approached, 15 accepted and 12 attended. Attendees had: 5-35 years of experience in FCR research, conducted between 2-11 studies on this topic across several disease sites, and were predominantly psychologists who focused on FCR rather than fear of disease progression (FoP). Researchers who could not attend were asked for their input about the content and methodology of the colloquium. In addition, 10 trainees with research experience in FCR (nominated by the experts), a representative of the Canadian Partnership Against Cancer (CPAC; an independent government-funded organization to accelerate action on cancer control), and two patient advocates (who were nominated by CPAC and purposefully selected based on their interest, gender, and experience with chronic and acute diagnoses) attended and contributed their perspectives on research directions and practice priorities (see [8] for a complete list of recommendations).

The Delphi method was used to reach consensus on a FCR definition. This method elicits consensus opinions from experts using an iterative process known as rounds [9]. The number of rounds used in the Delphi process varies but is usually 2-3 rounds. The Delphi is complete when a point of diminishing returns is reached [9]. We started the first round using the four most common FCR definitions cited in the literature [6, 10-12], and a new definition generated by attendees. Each attendee received a copy of the possible definitions and was asked to rate, anonymously, each definition on a scale ranging from 1 (not relevant) to 5 (extremely relevant), using a live voting website (Mentimeter.com). The three most valued definitions were retained and discussed followed by a second round, again consisting of anonymous, consensus live voting. The definition with the most votes was

retained and further discussed within the group to assess for further modifications deemed necessary and was submitted to a third round of live voting to obtain a final rating.

For the diagnostic characteristics of clinical FCR, participants' views on core characteristics were elicited on the live voting website. Because several other research priorities were addressed during the colloquium [8] and more time than allowed by the two days was needed to properly analyze the data we collected, we limited ourselves to a preliminary list of statements that will constitute the first step of a future formal Delphi study. These preliminary statements were sorted and responses with similar content were grouped together ~~classified~~ into categories using content analysis with NVivo 10. The classification of statements into categories was performed by B.M. and moderated by S.L. Disagreements were noted and resolved to achieve consensus by discussion among the two researchers.

Results

In round 1 of the Delphi, attendees were asked to formulate a new definition of FCR in addition to considering four commonly cited FCR definitions [6, 10-12]. They suggested "Fear, worry, or concern relating to the possibility that cancer will come back or progress". This new definition was proposed to reflect the broad spectrum in which patients experience FCR and to be inclusive of new primaries, chronic forms of cancer with on-going active treatment, recurrences of the same cancer, metastases, and progression of incurable disease. This definition reflects the fact that FCR is a normal reaction to the cancer experience which at a certain point becomes problematic or pathological. The four commonly cited definitions plus the new one were then rated by all participants (see Table 1).

In round 2, the three most highly rated definitions were retained, discussed and then rated. The new definition obtained the highest rating and was slightly reworded, followed by a third and final round of voting. The final proposed definition was "Fear, worry, or concern about cancer returning or progressing" (see Table 1).

In the second part of the study, attendees were asked to submit elements of clinical FCR based on their experience (personal, clinical and/or research) via live voting (completion of text fields entered into via the web-based system). This resulted in 112 elements that were categorised into five possible characteristics of clinical FCR: high levels of preoccupation, worry, rumination or intrusive thoughts (e.g. "excessive rumination that is not easily dismissible," "persistent and or frequent anxiety or worry," "perpetual preoccupation/rumination surrounding cancer returning"; 37 entries), maladaptive coping (e.g., as "avoidance", "reassurance seeking", "body checking"; 36 entries), functional impairments (e.g. "fears are interfering with daily living such as work"; "fears are impacting their relationships with others"; 22 entries), excessive distress (e.g. "a high level of distress and disruption associated with the FCR," "significant emotional distress"; 13 entries), and difficulties making plans for the future ("feeling paralyzed about the future," "patient not planning for the future"; 4 entries).

Discussion

To our knowledge, the proposed definition "Fear, worry, or concern about cancer returning or progressing" is the first expert-based definition of FCR including details of the generation process. It

focuses on the cognitive and emotional aspects of FCR. The exact wording was carefully debated and chosen to encompass what is traditionally referred to in the literature as FCR as well as FoP [12]. This is a deliberate consensus effort to propose an inclusive definition and reflect the fact that the concepts of FCR and FoP are often viewed as identical constructs [1-4, 12]. Nonetheless, we recognize that additional research is needed to examine if and how the phenomenon of FCR differs between those who have curable disease and fear a recurrence versus those who have incurable disease and fear the progression of their illness.

The five categories of possible characteristics of clinical FCR have been previously proposed in the literature [7,11,12] but seldom been empirically validated [13,14]. The current study gathered international expert consumers, clinicians and researchers, but still represents a fairly small group, which may not be fully representative of the wider expert community. Consensus processes do not necessarily represent truth and the results need to be tested empirically. Nevertheless, this represents an important initial milestone to further clarify this area of research and practice. The statements that were identified in the present study will be refined and submitted in the future to a larger group of researchers and clinicians with experience with FCR, using an extended Delphi survey. Specifically, we will select this group as follows. First, we will invite known research experts who will be defined as having published in the past 5 years in the field, or be involved in an ongoing FCR intervention trial. Second, clinicians who self-identify as having 5 years of experience working with patients with FCR will also be solicited. A snowballing technique will be used to ask the identified participants to invite colleagues who they know would meet the criteria to join the survey. All participants will be asked to confirm they meet eligibility criteria prior to participation. These experts will rate the five categories of possible characteristics, suggest modifications and provide new features that will be submitted to further rounds. Some of the coping strategies identified by experts in the present study may be adaptive to a point (e.g. body checking and doing monthly breast self-exam) and need to be refined in the extended Delphi survey. Time frame as a potential indicator of clinical FCR as being in a clinical FCR was not raised by the experts but should be considered in future studies of clinical features of FCR.

Table 1 Results of the Delphi study on FCR definitions

Definitions	Rating of definition relevance (1 = not relevant, and 5 = extremely relevant)		
	Round 1	Round 2	Round 3
Degree of concern reported by subjects about the chances of cancer returning at a future time [10]	1.83		
The fear that cancer could return or progress in the same place or another part of the body [6]	2.88	2.25	
The fear or worry that cancer could return or progress in the same place or another part of the body [11]	3.67	2.92	
We define fear of disease progression (FoP) as patients' fear that the illness will progress with all its biopsychosocial consequences, or that it will recur. This is a reactive, non-neurotic fear response patients are fully aware of. The fear is based on the personal experience of a life-threatening or incapacitating illness. Like other anxieties, FoP is experienced in emotional, behavioral, and physiological qualities. Basically, FoP is an appropriate response to the real threats of diagnosis, treatment, and illness course. In our view, the level of FoP can range between functional and dysfunctional ends [12]	2.54		
Fear, worry, or concern relating to the possibility that cancer will come back or progress[a]	4.54	4.29	4.50[b]

Note [a] Definition the colloquium attendees proposed; [b] after the second round, the definition was modified to "Fear, worry, or concern about cancer returning or progressing" and was submitted to a third round for a final rating

References

1. Simard S, Thewes B, Humphris G, Dixon M, Hayden C, Mireskandari S, et al (2013) Fear of cancer recurrence in adult cancer survivors: a systematic review of quantitative studies. J Cancer Surviv 7:300-322. doi: 10.1007/s11764-013-0272-z
2. Crist JV, Grunfeld EA (2013) Factors reported to influence fear of recurrence in cancer patients: a systematic review. Psychooncology 22:978-986. doi: 10.1002/pon.3114
3. Koch L, Jansen L, Brenner H, Arndt V (2013) Fear of recurrence and disease progression in long term (≥5 years) cancer survivors – a systematic review of quantitative studies. Psychooncology 22:1-11. doi: 10.1002/pon.3022
4. Thewes B, Butow P, Zachariae R, Christensen S, Simard S, Gotay C (2012) Fear of cancer recurrence: a systematic literature review of self-report measures. Psychooncology 21:571-587. doi: 10.1002/pon.2070
5. Cancer Australia (2014) Recommendations for the identification and management of fear of cancer recurrence in adult cancer survivors. Retrieved from: http://guidelines.canceraustralia.gov.au/guidelines/fear_of_recurrence/ch01.php. Accessed 1 December 2015
6. Vickberg SM (2003) The Concerns About Recurrence Scale (CARS): a systematic measure of women's fears about the possibility of breast cancer recurrence. Ann Behav Med 25:16-24. doi: 10.1207/S15324796ABM2501_03
7. Lee-Jones C, Humphris G, Dixon R, Hatcher MB (1997) Fear of cancer recurrence – a literature review and proposed cognitive formulation to explain exacerbation of recurrence fears. Psychooncology 6:95-105
8. Lebel S, Ozakinci G, Humphris G, Thewes B, Prins J, Dinkel A, Butow P (in press) Current state and future prospects of research on fear of cancer recurrence. Psychooncology.
9. Iqbal S, Pipon-Young L (2009) The delphi method. Psychologist 22:598-601
10. Northouse LL (1981) Mastectomy patients and the fear of cancer recurrence. Cancer Nursing 4: 213-220
11. Simard S, Savard J (2009) Fear of cancer recurrence inventory: development and initial validation of a multidimensional measure of fear of recurrence. Support Care Cancer 17:241-251. doi: 10.1007/s00520-008-0444-y
12. Herschbach P, Dinkel A (2014) Fear of progression. In: Goerling U (ed) Psycho-Oncology. Recent results in cancer research, Vol. 197. Springer-Verlag , Berlin, pp 11-29
13. Custers JAE, Gielissen MFM, Janssen SHV, de Wilt JHW, Prins J B (2015) Fear of cancer recurrence in colorectal cancer survivors. Support Care Cancer. doi: 10.1007/s00520-015-2808-4
14. Simard S, Savard J, Ivers H (2010) Fear of cancer recurrence: specific profiles and nature of intrusive thoughts. J Cancer Surviv 4:361-371. doi: 10.1007/s11764-010-0136-8