Format and readability of an enhanced invitation letter did not affect participation rates in a cancer registry based study: A randomised controlled trial

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ABSTRACT

Objective: Assess effectiveness of an "enhanced" invitation letter in increasing participation in an Australian cancer registry-based study; and assess representativeness of the study sample.

Study design and setting: 800 haematological cancer survivors, diagnosed within the last 3 years and aged 18 to 80 years at recruitment, were selected from one Australian state-based cancer registry. Half were randomly allocated to receive the standard invitation letter (control group). The remaining half received a modified invitation letter, incorporating content and design characteristics recommended to improve written communication (intervention group).

Results: Of 732 eligible survivors 268 (37%) returned a completed survey. There was no difference in participation between the intervention (n=131, 36%) and control groups (n=137, 38%; p=0.53). Participants were representative of the population for characteristics assessed, except for age-group at diagnosis. Survivors 50 years or older at diagnosis had higher odds of returning a completed survey, 50 to 59 (OR 2.53; 95% CI 1.47, 4.35), 60 to 69 (OR 2.69; 95% CI 1.58, 4.58) and 70 to 80 (OR 1.90; 95% CI 1.07, 3.35), than survivors aged 15-39 years at diagnosis.

Conclusions: An enhanced invitation letter was not effective in increasing participation of haematological cancer survivors in an Australian cancer registry study. The study sample was moderately representative on variables assessed, with age-group at diagnosis the only variable associated with participation. Research should evaluate strategies to increase participation in registry studies, and focus on tailoring techniques to patient's age.

KEYWORDS Patient information, Patient recruitment, Cancer registry, Patient letter

Trial registration Australian New Zealand Clinical Trial Registry; ACTRN12611000892910

Running title: Format and readability of an enhanced invitation letter did not affect participation rates

What is new?

Key Findings:

- An enhanced invitation letter did not affect participation rates, with a similar percentage of survivors who received the standard invitation letter (38%) returning a completed survey as those who received the enhanced letter (36%).
- However, low response rates may not have substantially affected study representativeness, with age at diagnosis the only variable assessed, that differed between participants and non-participants.

What this adds to what was known?

- This study emphasises the difficulties in recruiting patients from cancer registries.

What is the implication, what should change now?

- Strategies that effectively increase study participation, which can easily be adopted into standard registry recruitment methods should be identified.
- The representativeness of a study sample should be assessed on as many variables as possible to allow for identification of potential bias, particularly when faced with a low response rate.

INTRODUCTION

In Australia and many other countries, it is a legal requirement that all cancer diagnoses are notified to the relevant cancer registry [1-3]. Population-based cancer registries collect demographic and disease information relating to all cancers diagnosed in a defined location [2, 4]. Cancer registries thus provide an opportunity to recruit large, representative and unbiased samples of cancer patients for empirical research [2, 5]. However, studies have reported low response and participation rates when utilising cancer registries for recruitment [6-8].

Why use written communication to increase participation rates?

Written communication (i.e. invitation letters, information sheets) is utilised in most research studies to inform and invite potential participants. Despite a number of guidelines and recommendations on how to improve written communication, health related information is often written above an eighth grade reading level (approximately 13 years[9]) [10-13], which has been suggested as an appropriate reading grade level for written health communication [14]. Study materials, such as consent forms, used for health research have been shown to be complex and difficult for patients to understand [15]. Their length and complexity has been suggested to be, in part, influenced by the regulations and requirements set out by institutions relating to the level and type of detail that must be included in these documents [10, 16]. This may also be true for standard invitation letters that are sent from cancer registries to patients. In an unpublished analysis performed by the authors, it was found that patient invitation letters designed for a larger study using the standard template of several Australian state-based cancer registries had an average reading grade level of 12.8 (over 17 years[9]), included long sentences with a mean of 22.1 words and did not contain headings. Patient

communication needs to be coherent and comprehensive to ensure that it is easily understood by the intended population. If communication is not understood by the target audience they may be less likely to pay attention to the material [17], to understand key points of the research and therefore may be less likely to agree to participate. Consequently, altering the content and/or presentation of written communication sent to potential participants may help to increase participation rates.

Does "enhanced" written communication influence behaviour?

Certain design and content characteristics of written information have been suggested to increase readability and comprehension [17-23]. Design characteristics are those that relate to document design such as layout, font and use of visual material [23, 24]. Content characteristics include the use of active voice, short words and sentences, and are argued to reduce the complexity of written materials [18, 23].

Questions still remain as to whether the design and content characteristics of written communication influence people's behaviour [17]. Studies investigating the influence of written communication on behaviour in real world settings have produced mixed and often unfavourable results [10, 25, 26]. For example, several studies attempting to increase cancer screening behaviour by providing participants with enhanced or simplified brochures have been unsuccessful [10, 26]. However, we are aware of only a few published studies that have examined the effect of incorporating such design and content characteristics to improve the readability and comprehension of study invitation letters on study participation rates [27-31]. None of these identified studies have been conducted in the area of health.

Are high participation rates the only thing to consider when recruiting from population-based cancer registries? In theory population-based cancer registries should offer access to an entire population of cancer survivors, however certain sub-groups of cancer patients, including younger patients, older patients, men and racial and ethnic minorities, have been under-represented in previous studies [7, 8, 32]. Regardless of the response rate if a study sample is not representative of the population being researched the validity and generalizability of the study results to the wider population are reduced. While a high response rate increases the chance of obtaining a representative sample it is not a guarantee. For example, several cancer registry-based studies that have recorded modest response rates above 60% have evidence of potential response bias, with differences found between some responder and non-responder characteristics [32-34]. Therefore, in addition to trying to increase response rates to cancer registry based studies it is important that researchers strive to obtain a representative sample. While this is not always possible studies should attempt to assess the representativeness of their sample on as many characteristics as possible. Doing so will provide an understanding of the limitations of the data and allow for appropriate conclusions to be drawn.

There is a need for research to identify strategies that are effective in increasing participation in registry studies which can easily be adopted into standard recruitment procedures. Enhanced written communication may be an appropriate avenue for investigation, particularly as written invitation letters are a required component of the recruitment process for most Australian registry-based studies. It is also important that a study sample is representative of the population being investigated. In a bid to address these issues, this study aimed to:

1. Evaluate the effectiveness of an enhanced study invitation letter sent from an Australian state cancer registry on participation rates. The enhanced letter incorporated content and design characteristics suggested by the literature to improve readability and comprehension;
2. Assess the representativeness of the study sample by identifying demographic and disease characteristics associated with participation in a cancer registry-based study.

MATERIALS AND METHODS

Subjects

Cancer survivors were identified by the Australian state-based cancer registry and invited to take part in a cross-sectional survey of unmet needs and psychological disturbance of rural and urban haematological cancer survivors. The survey consisted of a number of standardised measures including: the Survivor Unmet Needs Survey (SUNS) [6], the Depression Anxiety and Stress Scale (DASS) [35], the Distress Thermometer [36] and an adapted version of the Control Preferences Scale [37, 38]. Additional author derived questions assessing patient disease, treatment, sociodemographic, service utilisation and internet use were also included. As we aimed to assess the specific outcomes of rural and urban survivors we oversampled cancer survivors from rural locations to ensure we obtained an appropriate sample size of rural participants.

Eligible survivors were diagnosed in the last three years (between 1 July 2007 and 30 June 2010) with a haematological cancer (including: leukaemias, lymphomas and myelomas) and aged between 18 and 80 years at the time of recruitment.

Development of the intervention

A checklist based on content and design characteristics commonly identified from the literature as improving written communication was developed. The checklist contained 12 features. Seven content characteristics included: short sentences [14, 19, 20, 22, 23] (15 words or less) [17], active voice [14, 17, 18, 20-23], written in the second and/or first person [17-19], conversational style [14, 17], objective clearly stated at the beginning [17], information presented in a question answer format [17, 19, 22, 23] and a lower reading age (eighth grade (13 years [9]) or lower) [14]. Five design characteristics entailed the use of: headings [14, 17, 20, 21, 23], headings in bold [17, 18, 22, 23], simple typeface [17, 20, 23], size 12 font [14, 18, 21-23], one idea per sentence [18] and one idea per paragraph [20, 23]. Based on the checklist we modified the standard invitation and reminder letters used by the cancer registry to create an enhanced version.

To ensure the enhanced letter was written at a lower reading age than the standard invitation letter the Flesch-Kincaid readability formula [39] was used to assess the reading age of the main content of both letters. This formula evaluates the reading difficulty of a piece of text based on the average number of syllables per word and average number of words per sentence [40]. A number of computer software programs, including Microsoft Office can automatically calculate this formula, returning a readability score based on United States school grade level [40, 41].

Six health behaviour experts were invited to rate the degree to which the enhanced invitation letter complied with each item on the checklist on a three-point scale (‘not at all’, ‘somewhat but needs improvement’ and ‘yes’). The letters were then further refined based on the

experts' feedback to try and incorporate the content and design characteristics to a greater extent than the standard invitation letter.

Eight health behaviour researchers then rated the final enhanced invitation letter and seven rated the standard invitation letter, using the same checklist and three-point scale described above. Six of these researchers rated both the enhanced and the standard letters. From this comparison four of the 12 characteristics were rated by a high percentage of the researchers as being incorporated into the standard invitation letter, suggesting that the standard letter template used by the cancer registry was already perceived as incorporating these features. Consequently, these four items, 'written in the second and/or first person,' 'written in conversational style,' 'objective clearly stated at the beginning' and 'one idea presented per sentence' were removed from the list. In comparison to the standard letter, the enhanced invitation letter was rated by a higher percentage of researchers as incorporating the final 8 content and design characteristics (as shown in Table 1). Both the enhanced and standard invitation letters were printed on the cancer registry letterhead and contained the signature of the Manager of the Cancer Registry. Survivor's physicians were not involved in patient contact or recruitment for this study.

INSERT TABLE 1 HERE

Procedure

On behalf of the researchers, registry staff used random number allocation to randomize survivors into one of two groups:

1. *Intervention group:* prospective subjects were mailed an enhanced invitation letter; or

2. *Control group:* prospective subjects were mailed the standard registry invitation letter.

Patients were contacted directly by the cancer registry without consent, as permitted by legislation and Human Research Ethics Committee approval. Initial contact involved the mailed invitation letter along with a study package that contained: an information statement, survivor questionnaire, non-participation form, a brochure explaining the cancer registry, reply-paid envelope and a questionnaire package for their principal support person. The patient's physician was not involved in patient contact or recruitment. Survivors were assured that their decision to take part in this study was entirely their choice and their decision would not affect their access to care. Non-responders were mailed a reminder letter and an additional study package approximately 4 weeks later. Return of the survey was taken as voluntary consent to participate.

This project received ethics approval from the University of Newcastle Human Research Ethics Committee and the Human Research Ethics Committee responsible for the cancer registry. The randomised control trial has been registered with the Australian New Zealand Clinical Trial Registry; registration number ACTRN12611000892910.

Measures

Response rates were collected by the cancer registry. Those who returned a survey were recorded as participants. Those who did not agree to participate or did not respond were recorded as non-participants. Participants returned their completed survey to the cancer registry, which was then forwarded to the researchers

For those participants who agreed, details on their age, gender, cancer type, year of diagnosis, postcode and other demographic and disease characteristics were collected from the cancer registry. De-identified data was collected from the registry for non-consenters for age group, gender, cancer type, year of diagnosis and postcode. Postcode was used to categorise survivors into rural or urban location based on the Accessibility and Remoteness Index of Australia (ARIA+) classification. ARIA+ is a continuous variable with values ranging from 0, (representing high accessibility to services) to 15, (representing high remoteness) [42, 43].Using the ARIA+ index the Australian Bureau of Statistics has defined five categories: major cities of Australia; inner regional; outer regional; remote and very remote Australia [42, 43]. For the purpose of this study rural was defined as outer regional, remote and very remote Australia; and urban was defined as major cities of Australia and inner regional Australia [42, 43].

Statistical methods

Baseline sociodemographic and disease characteristics of participants are reported for each of the intervention groups. Chi-squared analysis was used to compare the proportion of survivors who returned a completed survey from the intervention and the control groups. Representativeness of the study sample was assessed using logistic regression analyses to identify survivor disease and demographic variables associated with participation rates. Initially, patient demographic and disease variables, including age group at diagnosis, year of diagnosis, rural/urban location, gender and cancer type were compared between participants and non-participants using Chi-squared analyses. . Variables with a *p*-value of 0.2 or less on Chi-squared analyses were included in the multiple logistic regression analysis [44]. Backwards stepwise method was used to remove variables from the logistic regression model if they had a *p*-value of 0.1 or more on the likelihood ratio test. To control for any differences

between survivors in the intervention and control groups the variable experimental group was included in the final logistic regression model despite having a p-value >0.2 on univariate analysis.

A total of 800 patients were approached to participate in the study, with 400 randomised to receive the enhanced letter and 400 to receive the standard letter. This number would provide at least 80% power, with a 5% significance level, to detect a difference of 10% in participation rates between intervention and control groups and, assuming a consent rate of approximately 30%, would allow detection of differences in characteristics between participants and non-participants of approximately 11%.

RESULTS

Participants

Of the 800 survivors sent a study package, 68 (31 from the enhanced letter group and 37 from the standard letter group) were later deemed ineligible as they were either unable to be contacted (n= 56), had died (n=8) or were misdiagnosed (n=4). Of the 732 eligible survivors, 268 returned a completed survey thus resulting in an overall participation rate of 37%.

Baseline sociodemographic and disease characteristics of the two experimental groups appeared to be reasonably similar, as shown in Table 2.

INSERT TABLE 2 HERE

Effectiveness of the enhanced letter on participation

A total of 131 (36%) participants in the intervention group and 137 (38%) participants in the control group returned a completed survey. The difference in participation rates between

intervention and control groups was not statistically significantly different (χ^2=0.40; df=1; p=0.53).

Factors associated with participation

Univariate analyses (as shown in Table 3) resulted in only 'age group at diagnosis' and 'cancer type' having p-values equal to or less than 0.2, and therefore were included in the logistic regression analysis, along with the variable 'experimental group'. 'Age group at diagnoses' was the only variable statistically significantly associated with participation in the final logistic regression model (Table 3). As shown in Table 3, the odds of participating generally increased with increasing age, but was only statistically significant for the 50 to 59 (OR 2.53; 95% CI 1.47, 4.35), 60 to 69 (OR 2.69; 95% CI 1.58, 4.58) and 70 to 80 (OR 1.90; 95% CI 1.07, 3.35) years age groups, relative to the youngest age group (15-39 year olds).

INSERT TABLE 3 HERE

DISCUSSION

Do enhanced letters improve participation from cancer registries?

The incorporation of commonly endorsed content and design characteristics into a cancer registry invitation letter was not effective in increasing participation by haematological cancer survivors in a self-report pen-and-paper survey. This finding is consistent with several studies which have found that enhanced, easy-to-read written brochures do not affect behaviour [26, 45]. Though improvements to written communication may not be effective in influencing behaviour, patients have previously reported a preference for simpler written materials [10, 46, 47], emphasising the importance of developing study materials that are comprehensive and easy to read.

Are certain patient characteristics associated with participation in cancer registry studies?

Age group at diagnosis was significantly associated with participation in this study, with survivors in the three oldest age groups having higher odds of returning a completed survey compared to survivors in the youngest age group. This finding is consistent with a recent study that reported difficulties in recruiting young adults and adolescent cancer survivors from a population-based cancer registry [48]. In contrast, other cancer registry studies have reported higher participation rates by younger adult cancer survivors compared to older survivors [8, 32]. Several differences between these studies and the current study may account for why younger adults were found to have a higher response than their older counterparts in these previous studies. These factors include recruitment of different cancer types, a definition of 'younger age' that was substantially higher (18 to 54 years and 64 years and younger vs. 18-39 years) and the lack of an upper age limit. Despite these differences age seems to be related to participation in psychosocial cancer research. This is of concern, as differences in some outcomes have been demonstrated between younger and older cancer survivors [49-52]. We suggest that future research investigate ways to tailor recruitment strategies to a patient's age and assess their effect on participation rates.

Was this study sample representative?

Despite a low overall participation rate (37%), the study sample was relatively representative of the target population on variables that were assessed. The only statistically significant difference found between participants and non-participants was age group at diagnosis. This result is consistent with previous research which found that an increased response rate did not affect sample representativeness on most key variables [53]. However, the original response rate of this study was already high at 74% [53]. Our results suggest that a low-response rate

may not impact as strongly on study representativeness as previously thought, at least on the several key demographic and disease characteristics we were able to assess. Instead it may only be certain sub-groups that are affected. However, data on non-responder characteristics in this study were only available on several key variables, including age, sex, postcode at diagnosis, year of diagnosis and cancer type. Consequently we were not able to assess the representativeness of this study sample on other variables such as ethnicity, stage of disease and treatments received, which have been found to be different between responders and non-responders in previous cancer registry studies [32, 33].

As research studies may not be able to assess the full representativeness of their study sample it is still essential that empirical research utilising cancer registries strive to achieve the highest possible participation rate. Doing so will help to ensure that research studies are adequately powered and the likelihood of potential response bias is reduced. We also strongly recommend that studies utilising cancer registries to recruit cancer patients attempt to compare as many demographic and disease characteristics of participants with non-participants as possible. Such analysis is vital to provide an overall indication of the representativeness of a study sample, which is necessary when attempting to generalise study results. With respect to overcoming the misrepresentation of certain sub-groups of a population in cancer research studies, we suggest that future studies consider over sampling participants from specific sub-populations that are known to be misrepresented. Alternatively researchers should evaluate and utilise strategies that specifically aim to increase response rates from under represented sub-populations.

Limitations

Although the enhanced letters used in this study were assessed by several researchers in an attempt to incorporate the chosen design and content characteristics as much as possible, the letters were not assessed by members of the target population i.e. haematological cancer survivors. Consequently the letters may not have been relevant or easy to read from the patient's perspective. To guarantee that study materials incorporate key content and design characteristics from the perspective of the intended audience we recommend that future studies pilot all study materials with the target population [10, 17], including information statements and invitation letters. In addition, four of the original 12 characteristics were rated by a high percentage of the researchers as being incorporated into the standard invitation letter. Consequently, there may not have been enough of a perceived difference between the two letters to have a significant effect. However, the final eight characteristics were rated by a higher percentage of the researchers as being incorporated into the enhanced letter compared to the standard letter. It is also possible that the standard and enhanced letters did not differ on other content and design characteristics that were not assessed in this study. Other features of the invitation letter may be more influential in increasing participation rates, such as personalising the letter and including a hand-written signature [54]. These features did not differ between the enhanced and standard invitation letters in this study.

For this intervention we reduced the average reading age of the main content of the standard registry letter from over an eleventh grade level (16 years [9]) to an eighth grade level (13 years [9]). We chose an eighth grade level as the maximum reading age as it has previously been deemed a satisfactory level [14]. In addition, it also allowed for the use of complex words that were specific to our sample population (i.e. leukaemia, myeloma and lymphoma). A reading age of this level also helped to maintain specific information that complied with institution requirements. Regardless of these efforts, the reading age may still have been too

high for our population to understand. Other recommendations suggest a reading age of no higher than a fifth or sixth grade level (10-11 years [9]) [17, 18]. While we attempted to improve the formatting and reading ease of the main content of the enhanced letter, there were still several characteristics that were difficult to incorporate entirely and therefore could have been improved. For example a number of sentences were longer than 15 words. However, the average sentence length in the enhanced letter was an improvement from the standard letter. In addition the average sentence length of the enhanced letter was below 15 words and the majority of sentences contained less than 20 words, which has been suggested to be an acceptable level by some guidelines [19, 22].

It is conceivable that other factors besides the invitation letter influenced people's decision to take part in this study, including survey length, perceived relevance of the study to participants, the types of questions asked and the number of study documents sent to participants. For example, the length of the survey (28 pages) may have had a stronger impact than the invitation letter, as survey length has previously been found to impact on survey completion rates [54]. Additionally participants may have considered this study irrelevant to their situation, or believed their situation to be of little use to the study. This seems likely as a number of non-consenters indicated that they did not take part as they felt the study was not applicable to their situation. The importance of study relevance and perceived usefulness of a person's situation to the study question has been identified as main reasons for non-participation in previous research studies [55, 56]. We recommend that future research attempts to increase participation rates by emphasising the relevance of research studies to all participants while creating a survey that is as short as possible. Piloting all study materials with the target population may also assist in identifying ways to emphasise the importance/relevance of participation by all sub-groups.

The questions asked in the survey may have also impacted on participation rates; with questions covering sensitive issues having been found to reduce participant response rates [54]. However, it is not believed that the questions were sensitive in nature and previous cancer survivor studies assessing unmet needs and psychological outcomes have reported both higher [57] and similar participation rates [6] to this study. The number of documents sent to survivors (n=5) may have overwhelmed them and resulted in lower participation rates. The inclusion of a survey package for survivor's principle support persons may have reduced participation rates as well, with previous meta-analysis finding that inclusion of a survey for subject's relatives was associated with reduced questionnaire response rates [54]. Finally, it is possible that our sample was not entirely representative of the population on other variables (i.e. stage of the cancer trajectory, marital status) that we were unable to assess as this data was not available for non-participants.

What are the differences between this study and other population-based studies that report a higher participation/response rate?

There are examples of cancer survivor studies recruiting from cancer registries that report higher response rates, of over 50% [33, 34, 57-60]. One of the differences between a number of these previous studies [33, 34, 57, 59, 60] and the current study is the active involvement of the patient's treating clinician in the recruitment procedure of patients. Actively involving a patient's treating clinician, whereby the clinician must consent to the registry contacting their patient or the clinician contacting the patient initially on behalf of the registry, appears to assist in increasing patient response rates. However, actively involving clinicians can often be difficult, timely [48, 61, 62] and may also introduce selection bias [57]. In some studies, clinician non-consent and non-response has resulted in reductions in the number of eligible

patients invited to take part in the research [57, 59, 60]. We believe that identifying effective strategies to increase patient participation rates that are inexpensive, easily implemented into registry recruitment protocols and allow for a random sample of cancer patients from the whole population to be invited to take part in the research, is necessary in reducing both response and selection bias.

Other factors that could account for the lower participation rate of the present study compared to other population-based cancer survivorship studies, include, the number of reminder follow-ups conducted with non-responders and the type of cancer patients investigated. A number of studies have included at least two reminder follow-ups of non-responders [58-60], compared to only one reminder used in this study. The additional contact with non-responders may partly explain the higher participation rates of previous studies. In addition most of these studies did not focus specifically on haematological cancers [57, 59, 60]. Differences in response rates by haematological cancer types compared to other cancer types warrants further investigation. However two studies reported higher response rates by NHL survivors of 82% and 55%, suggesting that a higher participation rate for some haematological cancer survivors may be possible. The first, by Mols et al [34], employed active involvement of the patient's treating clinician in the recruitment of survivors. The second, by Arora et al's [58] included two reminder phone calls to non-responders as well as providing survivors with the option of completing a shortened version of the survey over the phone [58]. Providing the option of completing a shorter survey may be effective in increasing haematological cancer survivor response rates, with almost 22% of survivors from Arora et al's [58] study completing only the abridged version of the survey. Arora and colleagues [58] also included a $20 gift certificate in the questionnaire package sent to all survivors invited to take part in the research. Inclusion of cash incentives has been found to

increase survey response rates [54] and again may account for the higher participation rate in Arora et al's [58] study compared to ours. If possible future research should assess the effectiveness of including cash incentives and the option of survivors completing a shorter questionnaire on the participation rates of haematological cancer survivors recruited from population-based cancer registries.

Conclusion

Population-based cancer registries have the potential to provide large, representative samples of cancer patients for empirical research. However the advantages of cancers registries are often jeopardised by low-participation rates, and underrepresentation of certain sub-groups. Utilising an enhanced study invitation letter appears to have no impact on improving participation rates; however, a low response rate may not necessarily result in a largely unrepresentative sample. If the full potential of cancer registries is to be utilised strategies that are effective in increasing patient participation, particularly those sub-populations who may be misrepresented (i.e. younger survivors), should be identified and incorporated into registry recruitment procedures.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

REFERENCES

1. Australian Institute of Health and Welfare. Australasian Association of Cancer Registries. Canberra2011 [cited 2011 August].

2. Sanson-Fisher R, Carey M, Mackenzie L, Hill D, Campbell S, Turner D. Reducing inequities in cancer care. Cancer 2009; 115(16):3597-605.

3. Coleman MP, Muir CS, Menegoz F. Confidentiality in the Cancer Registry. British Journal of Cancer 1992; 66:1138-49.

4. Parkin DM. The evolution of the population-based cancer registry. Nature Reviews 2006; 6:603-12.

5. Beskow lM, Sandler RS, Weinberger M. Research Recruitment through US Central Cancer Registries; Balancing provacy and scientific issues American Journal of Public Health 2006; 96(11):1920-6.

6. Campbell HS, Sanson-Fisher R, Turner D, Hayward L, Wang XS, Taylor-Brown J. Psychometric properties of cancer survivors' unmet needs survey. Supportive Care in Cancer 2011; 19:221-30.

7. Girgis A, Boyes A, Sanson-Fisher R, Burrows S. Perceived needs of women diagnosed with breast cancer: rural versus urban location. Australia and New Zealand Public Health 2000; 24:166-73.

8. Smith T, Stein KD, Mehta CC, Kaw C, Kepner JL, Buskirk T, et al. The rationale, design, and implementation of the American Cancer Society's studies of cancer survivors. Cancer 2007; 109(1):1-12.

9. U.S. Department of Education, National Center for Education Statistics. Digest of Education Statistics, 2001. Washington, DC2002.

10. Davis TC, Williams MV, Marin E, Parker RM, Glass J. Health literacy and cancer communication. Ca-A Cancer Journal for Clinicians 2002; 52(3):134-49.

11. Shieh C, Hosei B. Printed health information materials: Evaluation of readability and suitability Journal of Community Health Nursing 2008; 25:73-90.

12. Wolf MS, Davis TC, Shrank WH, Neuberger M, Parker RM. A critical review of FDA-approved medication guides Patient Education & Counseling 2006; 62:316-22.

13. Estrada CA, Martin-Hryniewicz M, Higgs V, Collins C, Byrd JC. Anticoagulant Patient Information Material Is Written at High Readability Levels. Stroke 2000; 31:2966-70.

14. Rudd RE, Anderson JE. The health literacy environment of hospitals and health centers: Partners for action Making your healthcare facility literacy-friendly. Boston: Harvard University2006.

15. Rudd RE, Anderson JE, Oppenheimer S, Nath C. Health literacy: an update of medical and public literature In: Comings JP, Garner B, Smith C, editors. Review of adult learning and literacy: connecting research, policy and practice New Jersey: Mahwah; 2007. p. 175-203.

16. Sharp SM. Consent documents for oncology trials: Does anybody read these things? . American Journal of Clinical Oncology 2004; 27(6):570-5.

17. Hoffmann T, Worrall L. Designing effective written health education materials: Considerations for health professionals. Disability and Rehabilitation 2004; 26(19):1166-73.

18. Griffin J, McKenna K, Tooth L. Written health education materials: making them more effective. Australian Occupational Therapy Journal 2003 09; 50(3):170-7.

19. Directorate-General for Translation European Commission. How to write clearly. Brussels & Luxembourg: European Commission Translation and drafting resources; 2010 [cited 2010 June]; Available from: http://ec.europa.eu/translation/index_en.htm.

20. Ministry of Health New Zealand. National guideline for Health Education Resource Development in New Zealand. Wellington: Ministry of Health New Zealand2002.

21. National Cancer Institute. Clear & simple: Developing effective print material for low-literate readers. Bethesda: National Cancer Institute; 2003 [cited 2011 April]; Available from: http://www.cancer.gov/cancertopics/cancerlibrary/clear-and-simple.

22. National Health Service. National Health Service Written information: General guidance. London: National Health Service; 2010 [cited 2011 June]; Available from:

http://www.nhsidentity.nhs.uk/tools-and-resources/patient-information/written-information%3a-general-guidanc.

23. Paul CL, Redman S, Sanson-Fisher R. The development of a checklist of content and design characteristics for printed health materials. Health Promotion Journal of Australia 1997; 7(3):153-9.

24. Clerehan R, Buchbinder R, Moodie J. A linguistic framework for assessing the quality of written patient information: its use in assessing methotrexate information for rheumatoid arthritis. Health Education Research Theory & Practice 2005; 20(3):334-44.

25. Paul CL, Redman S. A review of the effectiveness of print material in changing health-related knowledge, attitudes and behaviour. Health Promotion Journal of Australia 1997; 7(2):91-9.

26. Paul CL, Redman S, Sanson-Fisher RW. Print material content and design: Is it relevant to effectiveness? Health Education Research 2003; 18(2):181-90.

27. Redline C, Oliver J, Fecso R. The effect of cover letter appeals and visual design on response rates in a government mail survey. Annual meeting of the American Association for Public Opinion Research; Pointe Hilton, Tapatio Cliffs, Phoenix, Arizona 2004. p. 4873-80.

28. Gendall PJ. The effect on mail survey response rates of covering letters and questionnaire cover design. Palmerston North: Massey University; 2003.

29. Gendall P. Which letter worked best? Marketing Bulletin 1994; 5:53-6.

30. Mockovak W. The impact of visual design in survey cover letters on response and web take-up rates Washington Bureau of Labor Statistics 2011.

31. Wagner WG, O'Toole WM. The effects of cover letter format on faculty response rate in mail survey research. Educational & Psychological Research 1985; 5(1):29-37.

32. Kelly BJ, Fraze TK, Hornik RC. Response rates to a mailed survey of a representative sample of cancer patients randomly drawn from the Pennsylvania Cancer Registry: a randomized trial of incentive and length effects. BMC Medical Research Methodology 2010; 10(65).

33. Mols F, Oerlemans S, Denollet J, Roukema JA, van de Poll-Franse LV. Type D personality is associated with increased comorbidity burden and health care utilization among 3080 cancer survivors General Hospital Psychiatry 2012.

34. Mols F, Aaronson NK, Vingerhoets AJJM, Coebergh JWW, Vreugdenhil G, Lybeert MLM, et al. Quality of life among long-term non-Hodgkin lymphoma survivors. Cancer 2007; 109:1659-67.

35. Brown TA, Chorpita BF, Korotitisch W, Barlow DH. Psychometric properties of the Depression Anxiety Stress Scale (DASS) in clinical samples. Behaviour Research and Therapy 1997; 35:79-89.

36. National Comprehensive Cancer Network (NCCN). Distress management: Clinical practice guidelines Journal of National Comprehensice Cancer Network 2003; 1:344-74.

37. Degner LF, Sloan JA, Venkatesh P. The control preferences scale. Cancer Journal of Nursing Research 1997; 29:21-43.

38. Carey M, Anderson A, Sanson-Fisher R, Lynagh M, Paul CL, Tzelepis F. How well are we meeting haematological cancer survivors' preferences for involvement in treatment decision making? Patient Education & Counseling 2012; 88(2012):87-92.

39. Kincaid JP, Fishburne RP, Rogers RL, Chissom BS. Derivation of new readability formula for navy enlisted personnel. Branch NR, editor. Millington, TN1975.

40. Friedman DB, Hoffman-Goetz L. A systematic review of readability and comprehension instruments used for print and web-based cancer information. Health Education & Behavior 2006; 33:352-73.

41. Paasche-Orlow MK, Taylor HA, Brancati FL. Readability standards for informed-consent forms as compared with actual readability. New England Journal of Medicine 2003; 348(8):721-6.

42. Australian Bureau of Statistics. Information paper: Outcomes of ABS views on remoteness consultation, Australia. In: ABS, editor. Canberra2001.

43. Australian Institute of Health and Welfare. Rural, regional and remote health. Canberra2004.

44. Hosmer D, Lemeshow S. Applied logistic regression. New York: Wiley; 1989.

45. Davis TC, Berkel HJ, Arnold CL, Nandy I, Jackson RH, Murphy PW. Intervention to increase mammogrphy utilization in a public hospital. Journal of General Internal Medicine 1998; 13:230-3.

46. Davis TC, Fredrickson DD, Aronold C, Murphy PW, Herbst M, Bocchini JA. A polio immunization pamphlet with increased appeal and simplified language does not improve comprehension to an acceptable level. Patient Education & Counseling 1998; 33:25-37.

47. Davis TC, Holcombe RF, Berkel HJ, Pramanik S, Divers SG. Informed consent for clinical trials: a compartative study of standard versus simplified forms. Journal of National Cancer Institute 1998; 90(9):668-74.

48. Clinton-McHarg T, Carey M, Sanson-Fisher R, Tracey E. Recruitment of representative samples for low incidence cancer populations: Do registries deliver? BMC Medical Research Methodology 2011; 11(5).

49. Hall AE, Boyes AW, Bowman J, Walsh RA, James EL, Girgis A. Young adult cancer survivors' psychosocial well-being: a cross-sectional study assessing quality of life, unmet needs, and health behaviors. Supportive Care in Cancer 2011; 20(6):1333-1341.

50. Parker PA, Baile WF, De Moor CC, L. Psychosocial and demographic predictors of quality of life in a large sample of cancer patients. Psycho-Oncology 2003; 12:183-93.

51. Kroenke CH, Rosner B, Chen WY, Kawachi I, Colditz GA, Holmes MD. Functional impact of breast cancer by age at diagnosis Journal of Clinical Oncology 2004; 22(10):1849-56.

52. King MT, Kenny P, Shiell A, Hall J, Boyages J. Quality of life three months and one year after first treatment for early stage breast cancer: influence of treatment and patient characteristics. Quality of Life Research 2000; 9(7):789-800.

53. Bootsma-van der Wiel A, van Exel E, de Craen AJM, Gussekloo J, Lagaay AM, Knook DL, et al. A high response rate is not essential to prevent selection bias: Results from the Leiden 85-plus study. Journal of Clinical Epidemiology 2002; 55:1119-25.

54. Edwards PJ, Roberts I, Clarke MJ, Diguiseppi C, Wentz R, Kwan I, et al. Methods to increase response to postal and electronic questionnaires (review). Cochrane Database of Systematic Reviews 2009(3).

55. Williams B, Irvine L, McGinnis AR, McMurdo ME, Crombie IK. When "no" might not quite mean "no"; the importance of informed and meaningful non-consent: results from a survey of individuals refusing participation in a health-related research project. BMC Health Services Research 2007; 7(59).

56. Boyle T, Landrigan J, Bulsara C, Fritschi L, Heyworth J. Increasing study participation (letter to the editor). Epidemiology 2011; 22(2):279-80.

57. Harrison SE, Watson EK, Ward AM, Khan NF, Turner D, Adams E, et al. Primary health and supportive care needs of long-term cancer survivors: A questionnaire survey. Journal of Clinical Oncology 2011; 29(15):2091-8.

58. Arora NK, Hamilton AS, Potosky AL, Rowland JH, Aziz NM, Bellizzi KM, et al. Population-based survivorship research using cancer registries: a study of non-Hodgkin's lymphoma survivors. Journal of Cancer Survivorship 2007; 1:49-63.

59. Boon H, Stewart M, Kennard MA, Gray R, Sawka C, VBrown JB, et al. Use of complementary/alternative medicine by breast cancer survivors in Ontario: prevalence and perceptions. Journal of Clinical Oncology 2000; 8:2515-21.

60. Courneya KS, Karvinen KH, Campbell KL, Pearcey RG, Dundas G, Capstick V, et al. Associations among exercise, body weight, and quality of life in a population-based sample of endometrial cancer survivors. Gynecologic Oncology 2005; 97:422-30.

61. Affleck P. The challenge of recruitment Nurse Researcher. 2005;13(1):78-84.

62. Dicker BG, Kent DL. Physician consent and researchers' access to patients. Epidemiology 1990; 1(2):160-3.

Table 1: The final 8 content and design characteristics rated by a higher percentage of researchers as being incorporated into the enhanced invitation letter compared to the standard letter

Content Characteristics	Enhanced letter	Standard letter
Short sentences (15 words or less)	✓ (Mean words per sentence = 12.8)	Mean words per sentence = 20.9
Use of active voice	✓	
Information presented in a question answer format	✓	
8th grade or lower reading level	✓ (Flesch-Kincaid Grade level = 8.0)*	(Flesch-Kincaid Grade level = 11.2)*
Design Characteristics	**Enhanced letter**	**Standard letter**
One paragraph per topic	✓	
Headings as questions	✓	
Headings written in bold	✓	
Use of simple typeface with size 12 font	✓ (Garamond font, size 12)	Times New Roman font, size 11

*Readability score determined by the Flesch-Kincaid readability formula [39] which assesses the readability of text based on the average number of syllables per word and average number of words per sentence [40, 41]. A reading age score is produced which is based on the United States school grading levels [40, 41]. A grade level of 8 corresponds to an age of approximately 13 years [9]. A grade level of 11 corresponds to an age of approximately16 years [9].

Table 2: Demographic and disease characteristics of participants from the control and intervention groups

Characteristics	Control (n=137) N (%)	Intervention (n=131) N(%)
Sex		
Female	62 (45)	49 (37)
Male	75 (55)	82 (63)
Location of residency at diagnosis		
Urban	110 (80)	99 (76)
Rural	27 (20)	32 (24)
Year of diagnosis		
2007	16 (12)	21 (16)
2008	45 (33)	42 (32)
2009	53 (39)	46 (35)
2010	23 (17)	22 (17)
Age group at diagnosis		
15-39	14 (10)	10 (8)
40-49	15 (11)	17 (13)
50-59	43 (31)	31 (24)
60-69	43 (31)	45 (34)
70-80	22 (16)	28 (21)
Cancer type		
NHL	63 (46)	71 (54)
Leukaemia	39 (28)	33 (25)
Myeloma	24 (18)	18 (14)
Other lymphoma	11 (8)	9 (7)

Table 3: Results of chi square and multiple logistic regression analyses assessing disease and demographic variables associated with haematological cancer survivors returning a completed survey (participants)

Variables	Chi-squared analysis				Multiple regression analysis	
	Participants n (%) (n=268)	Non-participants n (%) (n=464)	Test statistic (df)	p-value[‡]	Odds Ratio (95% CI)	Likelihood ratio χ^2(df), $p^{\S}$
Experimental group			0.40 (1)	0.529[#]		0.57(1), 0.45[#]
Intervention	131 (49)	238 (51)			1	
Control	137 (51)	226 (49)			1.12 (0.83, 1.52)	
Gender			0.57 (1)	0.449		
Male	157 (59)	285 (61)				
Female	111 (41)	179 (39)				
Location of residency at diagnosis			0.32 (1)	0.573		
Rural	59 (22)	94 (20)				
Urban	209 (78)	370 (80)				
Year of diagnosis			3.18 (3)	0.364		
2007	37 (14)	46 (9.9)				
2008	87 (32)	147 (32)				
2009	99 (37)	180 (39)				
2010	45 (17)	91 (20)				
Age group			16.89 (4)	0.002[*]		17.73 (4), 0.0014
15-39	24 (9.0)	84 (18)			1	
40-49	32 (12)	69 (15)			1.62 (0.87, 3.01)	
50-59	74 (28)	103 (22)			2.53 (1.47, 4.35)	
60-69	88 (33)	115 (25)			2.69 (1.58, 4.58)	
70-80	50 (19)	93 (20)			1.90 (1.07, 3.35)	
Cancer Type			5.01 (3)	0.171[*]		0.45 (3), 0.93[†]
NHL	134 (50)	221 (48)				
Leukaemia	72 (27)	124 (27)				
Myeloma	42 (16)	61 (13)				

Other lymphoma	20 (7.5)	58 (13)

[*] Variables included in the multiple logistic regression analysis

[†] Variable removed during backwards stepwise multiple logistic regression analysis

[‡] Variables with a p-value of ≤ 0.2 were included in the initial multiple logistic regression analysis

[§] Variables with a p-value of ≥ 0.1 on the ratio likelihood test were removed from the logistic regression model

[#] To control for any differences between survivors in the intervention and control groups the variable experimental group was included in the final logistic regression model despite a p-value >0.2 in the univariate analysis and p-value >0.1 in likelihood ratio

What is new?

Key Findings:

- An enhanced invitation letter did not affect participation rates, with a similar percentage of survivors who received the standard invitation letter (38%) returning a completed survey as those who received the enhanced letter (36%).
- However, low response rates may not have substantially affected study representativeness, with age at diagnosis the only variable assessed, that differed between participants and non-participants.

What this adds to what was known?

- This study emphasises the difficulties in recruiting patients from cancer registries.

What is the implication, what should change now?

- Strategies that effectively increase study participation, which can easily be adopted into standard registry recruitment methods should be identified.
- The representativeness of a study sample should be assessed on as many variables as possible to allow for identification of potential bias, particularly when faced with a low response rate.