**CORRESPONDENCE**

**Perceptions of future health and cancer risk in adult survivors of childhood cancer: implications for engagement in follow-up care**

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on behalf of the ANZCHOG Survivorship Study group\*

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Gibson and colleagues recently evaluated the risk perceptions of adult survivors of childhood cancer.(1) The authors concluded that a subset of survivors at risk of treatment-related late effects lacked concern about developing subsequent neoplasms (35%) and about their future health (24%), potentially reducing their likelihood of engaging in follow-up care.

Data from our ANZCHOG Survivorship Study, involving childhood cancer survivors from Australian and New Zealand primary treating centres, support these findings and provide further insight into how survivors’ health concerns may impact follow-up engagement.(2) We collected data on survivors’ i) perceived risk, and ii) amount of worry (“No/low” vs “Some/high”) about subsequent neoplasms (i.e. relapse or new cancers) and late effects, including time they spent thinking about them (“few seconds” to “many hours”) and frequency of the thoughts (“a few times a year” to “every day”). Survivors reported their current follow-up care engagement and levels of general anxiety/depression. We also summed the number of late effects survivors reported ever experiencing as well as those which they believed posed a future health risk.

In our study, 402 long-term survivors completed surveys, (43.3% male; median age 25.0 years [range=16-61]; median 18.0 years since diagnosis [range=5-59]). Forty-two percent were currently engaged in follow-up care. Sixty-two (15.9%) were treated for relapse.

Many survivors in our study (61.3%) believed that they were not at risk of developing subsequent neoplasms. Survivors reported an average of 3.8 treatment-related late effects (SD=3.6, range 0-19), most commonly fatigue (41.9%), memory/learning difficulties (33.7%), and emotional issues (33.4%). We found 60.3% of all survivors reported not being worried about subsequent neoplasms, somewhat more than the 40% who lacked concern in Gibson’s cohort. Many survivors in our cohort (48.6%) did not believe they were at risk of developing late effects. Survivors perceived themselves at risk of developing an average of 4.2 future late effects (SD=3.7, range=0-16). Of all survivors in our study, 56.9% reported not being worried about developing future late effects, compared with 30.7% in Gibson’s study.

Relevant to Gibson’s hypothesis regarding the potential link between future health concerns and engagement in follow-up, in multiple regression, our survivors were more likely to be engaged in follow-up if they had higher perceived risk of future late effects (OR=1.797, 95%CI=1.104-2.925, *p=0.018*), seldom worried about subsequent neoplasms (OR=0.532, 95%CI=0.295-0.962, *p=0.037*), reported lower education levels (OR=0.412, 95%CI=0.181-0.940, *p=0.035*), received a bone marrow transplant (OR=2.876, 95%CI=1.114-7.426, *p=0.029*). No other demographic, clinical, or psychological factors (including perceived risk of subsequent neoplasms or worry about late effects) were significantly associated with follow-up engagement (all p>0.05).

To further explore Gibson’s hypothesis that levels of concern may influence follow-up engagement, we examined the interaction between survivors’ risk perceptions and level of worry about future health problems, on engagement. We found no strong evidence of an interaction effect between risk perceptions and worry about subsequent neoplasms (p=0.120) or about late effects (p=0.633), on follow-up engagement.

Our study included survivors up to 59 years after their primary diagnosis, compared with 31 years post-diagnosis in Gibson’s cohort. A higher proportion of survivors in our study lacked concern for future health problems than in Gibson’s, suggesting concern may decrease with time since diagnosis. Decreasing risk perceptions with increasing age is worrying as late effects risk actually increases as they age,(3) and only 23% in our population sought cancer-related care from their general practitioner.(4) More innovative interventions are required to re-engage survivors who are lost to follow-up, to promote ongoing surveillance and the early detection of treatment-related sequelae.(5)

Our data provides some understanding of the factors which may influence survivors’ engagement, including survivors’ future health concerns. Our analyses highlight the influence of future health concerns on follow-up engagement, particularly survivors’ levels of perceived risk and frequency of worrying about their future health. Future efforts should focus on establishing optimal ways of communicating survivors’ personal risks to promote risk-appropriate engagement, without creating unnecessary anxiety.

Adding to Gibson’s findings, our data show that a significant proportion of survivors in our cohort still worried about the risk of developing health problems, despite reporting low perceived risk of subsequent cancers and late effects. The relationship between worry and follow-up engagement appears complex. Echoing Gibson’s suggestion regarding the importance of cancer-related health worries, general anxiety/depression did not appear to influence survivors’ follow-up engagement. Rather than worry about health concerns, survivors’ beliefs about their risk of future late effects more consistently predicted their follow-up engagement. Together with Gibson’s study, our data highlight the need to better educate survivors about their risk of future health problems and the importance of follow-up engagement.

**References**

1. Gibson TM, Li C, Armstrong GT, Srivastava DK, Leisenring WM, Mertens A, et al. Perceptions of future health and cancer risk in adult survivors of childhood cancer: A report from the Childhood Cancer Survivor Study. *Cancer*. 2018.

2. Vetsch J, Fardell JE, Wakefield CE, Signorelli C, Michel G, McLoone JK, et al. “Forewarned and forearmed”: Long-term childhood cancer survivors’ and parents’ information needs and implications for survivorship models of care. *Patient Education And Counseling*. 2017;100(2):355-63.

3. Diller L, Chow EJ, Gurney JG, Hudson MM, Kadin-Lottick NS, Kawashima TI, et al. Chronic disease in the Childhood Cancer Survivor Study cohort: a review of published findings. *Journal of Clinical Oncology.* 2009;27(14):2339.

4. Signorelli C, Wakefield CE, Fardell JE, Foreman T, Johnston KA, Emery J, et al. the role of primary care physicians in childhood cancer survivorship care: multiperspective interviews. *The Oncologist.* 2018(23):1-10.

5. Signorelli C, Wakefield CE, Johnston KA, Fardell JE, Brierley M-EE, Thornton-Benko E, et al. ‘Re-engage’pilot study protocol: a nurse-led eHealth intervention to re-engage, educate and empower childhood cancer survivors. *BMJ Open.* 2018;8(4):e022269.