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CORRESPONDENCE

Correspondence on "Assessing relatives' readiness for hereditary cancer cascade genetic testing" by Bednar et al.

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As Bednar et al. clearly laid out in their study, cascade testing is a valuable component to continuing gains in preventive cancer care, specifically evaluation of genetic cancer predisposition.[1] There are clearly barriers to cascade testing for families that health systems are not addressing. While the authors' motivation for this study is sound, there are some major limitations to the methods and framework for this study, thus the conclusions are hardly actionable.

First, limitations mentioned by the authors were downplayed and could have been addressed with changes in protocol. This study was publicized on social media and advocacy group pages, biasing the study toward participants who are advocating for their health, worried about their predisposition to cancer, and more likely to have told their family. This creates a highly motivated sample, unreflective of families that decline to engage in cascade testing. It is possible that there was not enough time to collect a sample in a clinical setting, but the reason for sampling methods should be addressed. Additionally, the appropriateness of genetic testing is limited for young children or elderly parents over 80 years old. This information should have been collected during the survey for exclusion during analysis and for testing the association between age of the relative and stage of behavior change. Finally, the authors mention that interventions to increase cascade testing differ in effectiveness depending on state privacy laws, but did not evaluate any confounding effect that residence had on their outcome of interest. State of residence, insurance status, and geographic barriers to genetic counseling have been shown to influence cascade testing uptake in a number of studies.[2,3] Considering that family communication is only one factor in deciding to pursue genetic testing, the study here by Bednar et al. is unable to comment on these confounding factors.

A limitation the authors did not thoroughly explore was how the anonymity of participants severely limits conclusions because there is no way to confirm that initiating cascade testing is appropriate. As mentioned, the reporting participant has to assume that their family members know about the genetic diagnosis and remember if each family member has undergone testing. Information about family members' readiness for testing is unreliable for two reasons. First, using second-hand reports of behaviors is subject to misreporting due to memory and possibly due to social desirability bias; this sample is of people who are likely very engaged in their diagnosis and may feel pressure to be an advocate and confirm that they told family members. The outcome of interest is also unreliable because half of the participants underwent genetic testing more than two years ago, and nearly 20% of the participants had testing five or more years ago. This is a significant portion of participants who may have a hard time remembering how discussions with family members transpired, especially in the wake of an emotional genetic test result. Importantly, since genomic status cannot be confirmed there is no way to know how specific test results influence communication to family members. An interesting question to address would have been how communication patterns differ depending on pathogenicity and how communication differs if the participant was concurrently diagnosed with cancer. Left unexamined, characteristics indicative of lower uptake could have helped advocates and physicians create better interventions.

As the authors point out, the transtheoretical model is "a framework for understanding how people change health behaviors over time," but pursuing genetic testing is not necessarily a behavior change, it is a discrete decision.[4] In the introduction the authors state that "[B]ehavioral science theory can aid in understanding the role of *determinants, barriers, and facilitators* of cascade genetic testing within families," but a single question survey cannot uncover any of these factors. In the end, the majority of participants reported that their family members either had already undergone testing (30.5%) or had plans to get tested but not in the next six months (16%). This does not really tell us anything about why they have made that choice and what is stopping family members from participating in cascade testing. Critically, the available answers for "Which of the following describes [the family member's] readiness for genetic testing?" are fairly time sensitive (within six months, within one month, never, maybe in the future). The way this was structured does not actually get to answering the question because almost a quarter of the participants may have told their family members their test result five years ago and there is no way of knowing how that family member feels now unless the participant had recently been discussing their genetic test results. So these participants' answers are suspect within the transtheoretical model stages of change.

It is hard to grasp how the transtheoretical model actually served as a framework for survey methods, analysis, and conclusions. This model is not fit for a cross-sectional study nor for a one-time decision that depends on being informed through familial communication networks. When applied in appropriate health behavior change settings, it has also been recently criticized as having limited utility to create stage-based interventions.[5] It is understandable that the authors were mindful of participant burden but there is not enough information about the participants or their families to draw actionable conclusions or assess behavioral change readiness, even within other frameworks.

Julia Carlson[1] ✉

[1]*Department of Population Health Sciences, University of Wisconsin–Madison, Madison, WI, USA.* ✉*email: jncarlson2@wisc.edu*

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COMPETING INTERESTS

The authors declare no competing interests.

ADDITIONAL INFORMATION

Correspondence and requests for materials should be addressed to J.C.