Reviewer's report

**Title:** Consent for use of personal information for health research: Do people with potentially stigmatizing health conditions and the general public differ in their opinions?

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**Reviewer:** Damien Jolley

**Reviewer's report:**

Short Statistical Review

Willison and colleagues address the ethical question of access to personal information and they seek to measure the extent to which the views of people with potentially stigmatizing health conditions vary from those of “the general public”.

They define “stigma”, and nominate four health conditions for which they have evidence of level of stigmatization, and three others with likely stigmatization. Their reference group was “people recruited through Harris Interactive who had none of the target health conditions”. The predictor variable is thus an ordered categorical collection of eight levels, ordered by increasing stigmatization from zero (“general public with no target health condition”) through low (hypertension, diabetes), high (alcoholism, lung cancer) to extreme (AIDS).

Participants were recruited through a variety of sources, some from lists maintained by Harris Interactive (HI), a commercial market research company active in US and Canada, and some through health care clinics, both primary and specialist, local to the authors’ neighbourhood. There is a strong likelihood that the subjects recruited through HI will be biased in their views, relative to truly randomly sampled Canadians, since the lists maintained by HI are self-selected. There is insufficient information to infer the population coverage of the sample recruited through Hamilton clinics. However, since the main objective is to estimate an association, rather than a prevalence or incidence measure, these departures from population representativeness are not major drawbacks to the design.

The outcome measure was collected by a cross-sectional survey administered either by internet (for HI recruits) or by the participants’ choice of internet or telephone (for clinic-based recruits). The authors not only checked possible differences in administration modality using a HI omnibus phone survey, but also incorporated a modality indicator in their eventual regression analysis (which did indeed reveal a consistent bias between the internet and phone collections).

The principal outcome variable collected by the survey was a repeated-measure “consent choice”, which was an ordered categorical response to one of a set of five unordered hypothetical scenarios related to personal information disclosure.
The ordinal response to each scenario ranged from 1 (no concern) to 5 (complete refusal).

Measured potential confounding variables included demographics, self-reported health and “attitudinal variables” – a general disclosure concern measure, and a composite measure of the general benefits of medical research & treatment.

The statistical methods used were appropriate for these data. The authors used a GEE method in SAS, based on multiple linear regression, with an exchangeable correlation structure. The GEE method is valid for repeated measures from the same subjects, and the exchangeable correlation is valid for the unordered set of scenarios applied to each respondent. Use of linear regression to model the ordered categorical consent choice outcome variable might be questioned, since the measure is not inherently intervally-scaled, however, the authors address this satisfactorily by reference to earlier work which showed equivalence.

However, the decision to use linear regression rather than multinomial logistic or ordinal regression has consequences for the interpretation of results. As a linear score, the magnitude of the reported effect sizes (Table 2) must be interpreted as fractions of a “step” on the ordinal scale from “no concern” to “complete refusal”. The authors fail to make this connection anywhere in this manuscript.

The principal statistical result was the apparently small influence of underlying health condition on the outcome of consent choice. This null result is supported by a series of large P-values (for each of the seven health conditions, relative to the HI “general public” level). When this was modelled separately and together with the confounders of attitudinal variables, the results (Table 2) show that the latter variables are the dominant predictors of consent choice. This is a clever and informative modelling strategy, and clearly supports the authors’ conclusions.

There are two refinements and improvements to the reported modelling which I recommend the authors consider:

1) It may be worth commenting on the non-linear pattern of coefficients within Health Condition in both the full model and reduced model 1 in Table 2. The pattern appears, without having sketched the graph, to be almost J-shaped: low stigma conditions are LOWER than referent, then climbing to high stigma which are ABOVE referent. Lung cancer seems to be out of place in the stigma scale.

2) Some interpretation of the CI widths from Table 2 is necessary, particularly since the sample size calculations avoid any mention of effect size (just what is “stability of the model”, anyway?). For example, the CI for AIDS is -0.2 to +0.4; hence, patients with AIDS may be up to +0.4 of a “step” on the consent choice scale above the general public. Is this clinically important?

To summarise, I commend the authors for a thorough and robust statistical approach to their estimation of the association between stigmatization and consent for disclosure of personal information. Mostly, their methods are valid.
and appropriate. I remain unconvinced by their justification of sample size, and, in the light of the “surprising” null result, I urge them to interpret the confidence intervals from Table 2, since these hold the key to the power of the study.

**Level of interest:** An article of importance in its field

**Quality of written English:** Acceptable

**Statistical review:** Yes, and I have assessed the statistics in my report.

**Declaration of competing interests:**

I declare that I have no competing interests