



OBSERVATION ARTICLE

REVISED Respondent driven sampling of wheelchair users: A lack of traction? [version 2; referees: 3 approved]

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Abstract

Background: Internationally, wheelchair users are an emerging demographic phenomenon, due to their increased prevalence and rapidly increasing life-span. While having significant healthcare implications, basic robust epidemiological information about wheelchair users is often lacking due, in part, to this population's 'hidden' nature. Increasingly popular in epidemiological research, Respondent Driven Sampling (RDS) provides a mechanism for generating unbiased population-based estimates for hard-to-reach populations, overcoming biases inherent within other sampling methods. This paper reports the first published study to employ RDS amongst wheelchair users.

Methods: Between October 2015 and January 2016, a short, successfully piloted, internet-based national survey was initiated. Twenty seeds from diverse organisations were invited to complete the survey then circulate it to peers within their networks following a well-defined protocol. A predetermined reminder protocol was triggered when seeds or their peers failed to respond. All participants were entered into a draw for an iPad.

Results: Overall, 19 people participated (nine women); 12 initial seeds, followed by seven second-wave participants arising from four seeds. Completion time for the survey ranged between 7 and 36 minutes. Despite repeated reminders, no further people were recruited.

Discussion: While New Zealand wheelchair user numbers are unknown, an estimated 14% of people have physical impairments that limited mobility. The 19 respondents generated from adopting the RDS methodology here thus represents a negligible fraction of wheelchair users in New Zealand, and an insufficient number to ensure equilibrium required for unbiased analyses. While successful in other hard-to-reach populations, applying RDS methodology to wheelchair users requires further consideration. Formative research exploring areas of network characteristics, acceptability of RDS, appropriate incentive options, and seed selection amongst wheelchair users is needed.

Open Peer Review

Referee Status:

	Invited Referees		
	1	2	3
version 2 published 26 Aug 2016	 report	 report	 report
	↑	↑	↑
version 1 published 26 Apr 2016	 report	 report	 report

- 1 **Jesse Kokaua**, University of Otago New Zealand
- 2 **John F Smith**, Kohn Kaen University Thailand
- 3 **A James O'Malley**, Geisel School of Medicine at Dartmouth USA

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Grant information: This paper reports on a section of JAB's doctoral research, which was supported by a University of Canterbury Doctoral Scholarship and a research scholarship from the Burwood Academy of Independent Living.

The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Competing interests: No competing interests were disclosed.

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REVISED Amendments from Version 1

In response to reviewers' suggestions, new information has been included briefly describing the fundamental assumptions of respondent-driven sampling, and the concept of sampling equilibrium: what it is, and why it was not reached in our study. New information has also been included clarifying recruitment and response rates. Finally, the Discussion has been expanded to consider how the strength of wheelchair users' social networks might have impacted our study, and more detailed recommendations have been included regarding the selection, and education, of seed participants.

See referee reports

Introduction

Robust epidemiological research generally requires data collection from representative samples of the population of interest, and effective modes of sampling contact are essential¹. Such effective modes can be difficult in hard-to-reach populations where no (or inadequate) sampling frames exist. Traditional chain-referral sampling approaches are inherently biased in their participant selection methods; a bias that is compounded as recruitment waves continue. An appealing alternative, Respondent Driven Sampling (RDS), was developed to counter these biases by employing specific data collection and statistical analysis methods which enable the derivation of valid population-based estimates²⁻⁴. RDS has traditionally been used to sample from 'hidden' populations with inadequate sampling frames, such as those with greater risk of HIV, including injecting drug users^{5,6}.

In brief, RDS is initiated by recruiting a handful of individuals who serve as 'seeds'. After completing the survey, seeds are then invited to recruit their peers to complete the same survey. To enhance recruitment, RDS employs 'dual incentives' whereby individuals are rewarded (usually monetary⁷) for both their participation, and the participation they can elicit from the person(s) they recruit². For RDS to provide unbiased population estimates, several assumptions need to be satisfied. Unlike convenience sampling, RDS requires that recruitment chains are traceable (via recruitment codes), that participants can provide an estimate of their network size (the number of people a person knows in the target population), and that participant's recruit randomly from their networks⁴. If these RDS assumptions are met, at some point sampling will reach a state of equilibrium, and useable unbiased data – independent from the initial seeds – can be gleaned.

Equilibrium is deemed to have been reached when there is relatively little variation in the sample proportions of key participant characteristics (such as age or sex) between successive measurement waves. The threshold for variation tolerance is determined prior to RDS implementation and a value of 2% is commonly employed^{8,9}. Equations for calculating equilibrium are described by Heckathorn (2002)¹⁰. Data produced before sampling equilibrium is reached are termed 'out-of-equilibrium' and are normally discarded due to their inherent biases. Unbiased population-based estimates use 'in-equilibrium data', data generated after equilibrium is reached so that recruitment theoretically represents a probabilistically determinable sample of network members.

Due to its appeal, the use of RDS has rapidly increased in two decades, with over 120 RDS studies reported in more than 20 countries with over 30,000 participants⁷. Despite wider adoption of RDS, and its successful application in many topic areas, concerns have been raised regarding whether RDS estimates hold in practice. For instance, some RDS estimates are more variable than expected¹¹, and some sampling patterns appear to violate core RDS assumptions^{5,12}.

The prevalence of wheelchair users has rapidly increased over the last half century due, in part, to advancing medical care, ageing populations, increasing community supports, increased prescription of wheelchairs, and changes in attitudes to disablement such that people may feel less stigmatised about using a wheelchair^{13,14}. Despite this, robust epidemiological research with this group in New Zealand and Australia is scant^{15,16}. Contacting wheelchair users in the community is challenging. Recruitment approaches are often limited to using disability organisations and personal contacts, which can differentially exclude many wheelchair users¹⁷. Consequently, wheelchair users may constitute a 'hidden population', under-researched and excluded from population estimates¹⁸. Furthermore, many countries, including New Zealand, have yet to establish registries of wheelchair users which could provide a reliable sampling frame^{16,18}.

Here we report our experience of applying a RDS methodology to a survey of wheelchair users in New Zealand. To our knowledge this is the first time RDS has been applied to people who use wheelchairs, and could potentially offer a significant new sampling approach in epidemiology and disability fields.

Methods

After a successful pilot with wheelchair users, this study employed a short internet-based national survey which was open from October 2015 until January 2016. Administered through the SurveyMonkey™ website, an information sheet and video were embedded within the survey preamble (see [Supplementary material](#)). The information sheet stated that informed consent was implied through the voluntary participation in the survey. Ethics approval was obtained from the University of Canterbury Human Ethics Committee (reference HEC 2015/117). Eligibility criteria included: wheelchair use as the primary form of mobility; being a New Zealand resident; aged 16 years or more; being able to read English; having internet access; and, having an operational email account.

Invitations seeking 'seed' participants were circulated to various national disability organisations serving members with a range of impairments that lead to wheelchair use. People expressing interest in being seeds contacted the researcher, who confirmed eligibility and then sent a recruitment code and a link to the survey website. Once a participant completed the survey, they were thanked and emailed three unique recruitment codes. Participants were asked to recruit a maximum of three other wheelchair users, following Heckathorn's (1997) recommendations². This limit of three was premised on two primary reasons: to ensure that a broad array of participants are recruited; and, to prevent the emergence of semi-professional recruiters. Participants were asked to email one code

and the survey link to three other persons they knew who were likely to satisfy the eligibility criteria. This process was envisaged to continue for multiple recruitment waves. Participation was incentivised (an entry into a draw to win an iPad); one entry for completing the survey, and another when each person they recruited completed the survey. Recruitment chains were tracked through tracing the recruitment codes. A predetermined reminder protocol was triggered when seeds or their peers failed to respond.

Results

Twenty wheelchair users expressed interest in participating as seeds, of whom 12 completed the survey (60% response rate). All 12 seeds were asked to recruit a maximum of three wheelchair users. Only four seeds were successful in recruiting further participants (three seeds each recruited two participants, and one seed recruited one participant), accumulating in a total of seven first wave participants. Despite all seven first wave participants being asked to recruit a maximum of three wheelchair users, using a clearly stated invitation and reminder protocol, no second wave participants completed the survey. Thus, the final sample was composed of 19 wheelchair users. Mean age of participants was 55.6 years (range: 28–73 years), and nine were women. Survey completion time ranged between 7 and 36 minutes.

Our recruited sample of 19 wheelchair users, however, failed to satisfy the requirements needed to reach equilibrium; the point at which the sample composition becomes independent of the initial seeds, thereby enabling the calculation of unbiased population estimates⁴. This failure stems from the study's premature termination, where only one measurement wave was completed. Even in the best-case scenario where equilibrium is reached in the smallest possible number of waves, namely one, no useable data could be produced after equilibrium. Furthermore, when only a single wave is conducted, all participants are within a single degree of separation from the seed participants accessed by the researcher. Such a sample lacks what Heckathorn terms 'sociometric depth' and it would, in all likelihood, fail to be representative of the entire hidden population¹⁰. For these reasons, an empirical assessment of equilibrium was not formally undertaken here, as it was both conceptually and statistically impossible for the data captured within this study to be in-equilibrium.

Discussion

Despite a rigorous recruitment process and offering incentivising participation, our use of RDS failed as an effective sampling approach amongst wheelchair users in New Zealand. There are a number of possible explanations as to why this occurred. The target population of the study was novel compared with hidden populations generally targeted by RDS studies. Research using RDS typically samples stigmatised populations, such as those with greater risk of HIV, including injecting drug users⁵, men who have sex with men^{12,19}, and sex workers²⁰. Wheelchair users have experienced increased integration into many societies in recent years and are arguably less stigmatised when compared to populations traditionally sampled using RDS. Populations experiencing greater

stigma may have a tendency to establish stronger social and internal networks, helping to facilitate the RDS requirement that the population being sampled has sufficiently strong internal networks which enable the random recruitment of other members of the population. With no literature to our knowledge regarding the internal networks of wheelchair users, it is unknown whether wheelchair users would satisfy the random recruitment criteria of RDS. Although the precise mechanism by which perceived stigma might affect RDS participation is unknown it, nonetheless, remains noteworthy. Second, the use of an unguaranteed reward (entry into a draw for an iPad) for survey completion has not been previously reported in RDS studies. This lack of guaranteed reward may have influenced participation. In addition, RDS studies often offer participants additional non-monetary free services related to the mitigation of HIV risk through counselling and educational material⁷.

Exploring the areas of network characteristics, acceptability of RDS, appropriate incentive options, and seed selection have all been suggested as important for assessing the feasibility and appropriateness of RDS in certain populations²⁰. Here, critical feedback on the appropriateness of the incentives, RDS methodology, elicitation mechanism and platform, and the survey itself was obtained from the pilot group – but not from the seeds. Formative research regarding specific seed selection is warranted. First, we recommend judiciously selecting diverse seeds who have large social networks, which should facilitate positive growth of recruitment chains. This increases the chances of participants with diverse characteristics being recruited and helping to avoid the exclusion of isolated subpopulations and individuals. It also helps to increase the speed at which sampling equilibrium can be reached⁹. Indeed, one RDS study exploring people who inject drugs in Sydney Australia reported that 80% of their participants resulted from one seed²¹. Second, meeting with seeds to provide greater education regarding goals and protocol of the survey might have improved the recruitment rates of our survey. Providing greater education to seeds might have increased their commitment to the goals of the survey, increasing the chances that seeds will report favourably about the survey, accurately explain the survey goals, and be motivated to pass on all three recruitment codes⁹. Despite the traditionally low response rates and impersonal nature of electronic surveys¹, administering surveys electronically is becoming more feasible and successful with populations who use wheelchairs²², and having informed and enthusiastic seeds might have encouraged greater response rates. Until such time as these and other factors, and their implications for recruitment, are better understood we feel that using RDS for recruiting wheelchairs users may have limited merit, and recommend formative research to optimise success.

Conclusions

Wheelchair users are an increasingly prevalent population in society who often lack an adequate sampling frame, and sampling approaches enabling valid population based estimates are becoming increasingly necessary. This paper reported the failure

of RDS to survey wheelchair users. Despite the unsuccessful recruitment in this study, further research exploring the application of RDS with wheelchair users is recommended before discounting this sampling approach in this population.

Data availability

Data are available upon request from the corresponding author to protect participant identity. Demographic data will be pooled to protect participant identity, as individual-level demographic data could be theoretically traceable due to the small sample size, and suspected small national population of wheelchair users.

Consent

All participants were informed that the voluntary completion of the survey implied informed consent, including for the publication of survey data.

Author contributions

All authors contributed to the design of the study. JAB conducted the data collection and prepared the first draft of the manuscript. All authors contributed to subsequent drafts and the final manuscript.

Competing interests

No competing interests were disclosed.

Grant information

This paper reports on a section of JAB's doctoral research, which was supported by a University of Canterbury Doctoral Scholarship and a research scholarship from the Burwood Academy of Independent Living.

The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Supplementary material

Internet-based national survey.

[Click here to access the data.](#)

Information sheet.

[Click here to access the data.](#)

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[Reference Source](#)

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Open Peer Review

Current Referee Status:



Version 2

Referee Report 22 September 2016

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Jesse Kokaua

Pacific Islands Research & Student Support Unit, University of Otago, Dunedin, New Zealand

The changes the authors have made have addressed the recommendations I had made for the first version.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Competing Interests: No competing interests were disclosed.

Referee Report 12 September 2016

doi:[10.5256/f1000research.10247.r16242](https://doi.org/10.5256/f1000research.10247.r16242)



A James O'Malley

Department of Biomedical Data Science and The Dartmouth Institute of Health Policy and Clinical Practice, Geisel School of Medicine at Dartmouth, Lebanon, NH, USA

I have read the revised article and feel that the authors have responded to my concerns as well as possible given the space limitations. As such, I am willing to change my decision to Approved.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Competing Interests: No competing interests were disclosed.

Referee Report 05 September 2016

doi:[10.5256/f1000research.10247.r16076](https://doi.org/10.5256/f1000research.10247.r16076)



John F Smith

Postgraduate Tropical Medicine Program, Faculty of Medicine, Kohn Kaen University, Khon Kaen, Thailand

I agree with the revisions, and therefore change my recommendation to Approved.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Competing Interests: No competing interests were disclosed.

Version 1

Referee Report 11 July 2016

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A James O'Malley

Department of Biomedical Data Science and The Dartmouth Institute of Health Policy and Clinical Practice, Geisel School of Medicine at Dartmouth, Lebanon, NH, USA

Title and Abstract: The title is catchy and appropriate given the content of the article.

Article content: The article is very well written and is easy to read and follow. The article would be much improved if it included an introduction to respondent drive sampling (RDS) with particular emphasis on the assumptions required for RDS to work (reaching equilibria and otherwise) written in clear terms to a lay audience. Describe what it means for the sampling process to be in equilibrium. Currently, readers do not have the necessary information to reach an informed conclusion as to why RDS did not work well in this instance or to assess whether it is applicable to their own work.

The rationale for RDS assumes the existence of a social network in which the probability of a tie between a wheelchair user and a randomly selected wheelchair user exceeds the probability of a tie between a wheelchair user and a nonuser wheelchair user. What evidence that this is true? Do wheelchair users have sufficient social and other relationships importantly enough for them to be able to name sufficient other wheelchair users for the next round of sampling. Is it possible that many wheelchair users are isolates (in the network sense) because they are highly functional in everyday life and thus have reduced need for a community of wheelchair users? I don't know the answers to these questions and still don't as the needed background information is not provided in the article.

It would have been helpful to have been told the current state of knowledge about social networks among wheelchair users along with a connection of such knowledge to the assumptions under which RDS can yield unbiased population-level inferences. I would like to see some discussion of how RDS handles scenarios such as isolates in the wheelchair user network. Presumably, the only way such individuals make it into the sample is if they were randomly selected in the first (i.e., seed) wave.

Please add some general intuition for how RDS yields unbiased population-level estimates. Discuss the role of and derivation of individuals' sampling probabilities and thus sampling weights for computing population estimates. For example, the work of Thompson (2006a, 2006b, 2003, 2000 – see below – the 2012 text on Sampling and recent arXiv contributions) should be reviewed and used to inform the article.

References:

1. Thompson, Steven K. Targeted random walk designs. *Survey Methodology* 2006. 32:11-24

2. Thompson, Steven K. Adaptive web sampling. *Biometrics* 2006. 62:1224-1234
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Data (if applicable): Details about the design/data for the study are lacking. It is stated that 20 wheelchair users were the initial seeds of which 12 agreed to participate (60% response rate). It then appears as though 4 of these 12 participants culled 7 further participants in the second stage of sampling. Question: Did the remaining 8 seeds not generate any wheelchair users (i.e., does the enrolment rate for the second stage equal 7/12) or was the plan to only ask 33% of the initial seeds for links to trace (in which case the enrolment rate is 7/4)? In the third stage, there was no addition to the sample-size. Were the 7 participants who entered at the second stage each asked to name other wheelchair users; is the enrolment rate at the third stage 0/7 or 0/x, where $x < 7$? Was there any limit as to how many wheelchair users a seed to subsequently enrolled person could name? A lot of these questions would be answerable if the design of the RDS was described in the article!

Conclusions: The discussion makes a number of good points that have appear sound and logical. The primary point made is that the current study may have failed because the wheelchair user population has different characteristics from populations where RDS has been used successfully. This is a valid point. But shouldn't this point have been considered when the study was design? Was there any prior information to inform the parameters of the RDS design? Maybe RDS would work quite well for the wheelchair user population as long as the number of initial seeds is much greater than 20. A more positively-framed Discussion might leave readers with a more balanced appreciation for using RDS in their own studies of wheelchair users or other new populations.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.

Author Response 23 Aug 2016

Philip Schluter, University of Canterbury

Thank you greatly for your prompt, encouraging, and constructive feedback on our paper. Together with the comments made by the other two reviewers, your feedback has resulted in what we feel is a much improved paper.

We would like to take this opportunity to make two notes. [1] The original submission was restricted to 1000 words in total – but, with permission from the journal, this constraint has been relaxed and we are able to include more material. This means we are able to broaden discussion of relevant issues and thus strengthen the manuscript. [2] This paper was submitted to the journal as an 'observation article', defined as: *Observation Articles allow the description of a novel observation that may be unexpected, and possibly currently without explanation. An observation can be a phenomenon that has been identified in field work, in the laboratory or through experimental analysis* (see: <http://f1000research.com/for-authors/article-guidelines/observation-articles>).

Title and Abstract: *The title is catchy and appropriate given the content of the article.*

Thank you for the positive comment.

Article content: *The article is very well written and is easy to read and follow. The article would be much improved if it included an introduction to respondent drive sampling (RDS) with particular emphasis on the assumptions required for RDS to work (reaching equilibria and otherwise) written in clear terms to a lay audience. Describe what it means for the sampling process to be in equilibrium. Currently, readers do not have the necessary information to reach an informed conclusion as to why RDS did not work well in this instance or to assess whether it is applicable to their own work.*

We thank the reviewer for describing our article as well written and easy to read and follow. We agree that an introduction to RDS would improve the article. Our original submission was limited by word restrictions and article scope, as outlined above. However, in this revision, we have included more details. Specifically, we have included text in the Introduction section (see second paragraph) providing a brief introduction to RDS, and describing equilibrium (see third paragraph). We have expanded the Results section (second paragraph) to describe why equilibrium was not reached in our study.

The rationale for RDS assumes the existence of a social network in which the probability of a tie between a wheelchair user and a randomly selected wheelchair user exceeds the probability of a tie between a wheelchair user and a nonuser wheelchair user. What evidence that this is true?

We had no evidence to suggest this was true or false and felt on balance that the use of RDS in our study was warranted as there was a reasonable enough chance that it could be true without evidence to the contrary. Conducting a revised search on PubMed, Google scholar, and Science Direct, we could find no literature explicitly addressing this issue (we have included a sentence in the first paragraph of the Discussion section describing this).

Do wheelchair users have sufficient social and other relationships importantly enough for them to be able to name sufficient other wheelchair users for the next round of sampling. Is it possible that many wheelchair users are isolates (in the network sense) because they are highly functional in everyday life and thus have reduced need for a community of wheelchair users? I don't know the answers to these questions and still don't as the needed background information is not provided in the article.

This is indeed possible. The fact that a person uses a wheelchair does by no means infer that they know other people who use wheelchairs, let alone have strong internal networks with other wheelchair users. With respect to these questions we have included new text in the Discussion section (see first paragraph) regarding stigma and whether or not wheelchair users have sufficiently strong internal networks that enable random recruitment of other wheelchair users.

It would have been helpful to have been told the current state of knowledge about social networks among wheelchair users along with a connection of such knowledge to the assumptions under which RDS can yield unbiased population-level inferences. I would like to see some discussion of how RDS handles scenarios such as isolates in the wheelchair user network. Presumably, the only way such individuals make it into the sample is if they were randomly selected in the first (i.e., seed) wave.

As noted above, the current state of knowledge regarding the social networks among wheelchair users is unknown. As a result we cannot know if some wheelchair users are isolates. RDS theory assumes that the population being sampled comprises of a complete social network component, so in theory every person within their population has a probability of being sampled. In practice, to overcome isolated subpopulations, it is recommended that seeds from diverse subpopulations be selected. We have included a sentence to this effect in the second paragraph of the Discussion section.

Please add some general intuition for how RDS yields unbiased population-level estimates. Discuss the role of and derivation of individuals' sampling probabilities and thus sampling weights for computing population estimates.

Due to original word count, we had not included information clarifying that question two of our survey asked participants to provide an estimate of their network size (a requirement of RDS). Had our study produced in-equilibrium data (see new text in second paragraph in Introduction section, and new text in second paragraph of the Results section), sampling weights would have been allocated accordingly with those with smaller social network sizes being given a higher weight.

For example, the work of Thompson (2006a, 2006b, 2003, 2000 – see below – the 2012 text on Sampling and recent arXiv contributions) should be reviewed and used to inform the article.

References:

1. Thompson, Steven K. Targeted random walk designs. *Survey Methodology* 2006. 32:11-24.
2. Thompson, Steven K. Adaptive web sampling. *Biometrics* 2006. 62:1224-1234.
3. Chow, Mosuk, Thompson, Steven K. Estimation with link-tracing sampling designs - A Bayesian approach. *Survey Methodology* 2003. 29:197-205.
4. Thompson, Steven K., Frank, Ove. Model-based estimation with link-tracing sampling designs. *Survey Methodology* 2000. 26:87-98.

Thank you greatly for suggesting these references, which were reviewed and indeed helped with our thinking. Because our study failed to produce in-equilibrium data that warranted further analysis and consideration of sampling weights and sampling probabilities, substantial engagement with the topics in these papers was felt to be beyond the scope of our paper.

Data (if applicable): *Details about the design/data for the study are lacking. It is stated that 20 wheelchair users were the initial seeds of which 12 agreed to participate (60% response rate). It then appears as though 4 of these 12 participants culled 7 further participants in the second stage of sampling. Question: Did the remaining 8 seeds not generate any wheelchair users (i.e., does the enrolment rate for the second stage equal 7/12) or was the plan to only ask 33% of the initial seeds for links to trace (in which case the enrolment rate is 7/4)?*

All 12 seeds were asked to recruit 3 peers and for links to trace. Despite this, 8 seeds did not generate any more participants. The enrolment rate was 7/12. To clarify recruitment/response rates, text has been included in the first paragraph of the Results section.

In the third stage, there was no addition to the sample-size. Were the 7 participants who entered at the second stage each asked to name other wheelchair users; is the enrolment rate at the third stage 0/7 or 0/x, where $x < 7$?

The enrolment rate was 0/7. All 7 participants were asked to recruit 3 peers and for links to trace, but no participants were recruited. To clarify recruitment/response rates, text has been included in the first paragraph of the Results section.

Was there any limit as to how many wheelchair users a seed to subsequently enrolled person could name? A lot of these questions would be answerable if the design of the RDS was described in the article!

Thank you for raising this issue. All seeds and subsequent participants were asked to recruit a maximum of 3 peers and for links to trace. Heckathorn (1997) recommends that each respondent is limited to 3 ID codes to pass on to others, to ensure that a broad array of participants is recruited, and to prevent the emergence of semi-professional recruiters. We have now included two sentences describing this in the second paragraph in the Methods section.

Conclusions: *The discussion makes a number of good points that have appear sound and logical. The primary point made is that the current study may have failed because the wheelchair user population has different characteristics from populations where RDS has been used successfully. This is a valid point. But shouldn't this point have been considered when the study was designed?*

Yes, a valid point. In retrospect the potential impact of this point could have attracted greater consideration. However, we had no evidence to suggest it would be detrimental to the success of our employment of RDS. We also acknowledged that our application of RDS with wheelchair users was unprecedented and novel, and many outcomes were simply unknown. In the absence of any reliable sampling frame or registry, we believed the potential success amongst this population (on balance) was greater than the risks.

Was there any prior information to inform the parameters of the RDS design? Maybe RDS would work quite well for the wheelchair user population as long as the number of initial seeds is much greater than 20. A more positively-framed Discussion might leave readers with a more balanced appreciation for using RDS in their own studies of wheelchair users or other new populations.

Since our employment of RDS, a number of points, that may have improved our experience, have been considered. These include selecting more diverse seeds, and meeting with seeds to provide more information to them. To summarise these points, new text has been included our revised manuscript (see second paragraph in Discussion section).

Competing Interests: No competing interests to disclose.

Referee Report 13 June 2016

doi:10.5256/f1000research.9259.r14016



John F Smith

Postgraduate Tropical Medicine Program, Faculty of Medicine, Kohn Kaen University, Khon Kaen, Thailand

I support indexing this article as it appears to be a first-time test of the utility of Respondent Driven Sampling (RDS), a relatively novel sampling procedure, on a new "hidden" (hard to easily access) population--wheelchair riders. In this case it was not successful in recruiting sufficient respondents, but reporting that, and possible reasons for that outcome, could be useful for refining usage of the procedure.

I think the title and abstract are clear and provide good insight into the major points of the paper.

In addition to the possible reasons for low returns provided by the author I think it would be useful to include consideration of;

1. The typically low returns from internet surveys compared to face to face and pencil and paper surveys (see 1,2,3). It is possible that promoting an internet survey was just insufficient to motivate the "seeds", or the second wave recruits, to recruit more members into the network, even with an iPad reward incentive.
2. Elaborating on the "stigmatization" issue raised in the discussion. The "mechanism by which perceived stigma may effect RDS sampling" rather than being unknown, may well be that other groups e.g., drug users, "men who have sex with men"(MSM), sex-workers etc as a result of stigma, may have stronger social networks and obvious gathering spaces and be easier to access via seeds. Seeds with these groups are often "leaders", advocates who are "out", support group leaders, health service liaison/volunteer workers who move easily within those communities, social gatherings, entertainment or work spaces. There are not the same social presses for wheelchair riders to have these group social connections or common activities.
3. Methodological point. The article focuses on RDS's value for accessing representative population samples for research seeking normative and epidemiological data and concomitant importance of avoiding/controlling sample selection bias. However, RDS could also be valuable for accessing qualitative data e.g., wheelchair riders' views/experiences with public policies/services etc where as wide a range of responses (variability) should be expected, indeed welcomed, rather than seen as requiring some statistical weighting or control procedure. Other forms of criterion for data "completeness" e.g., saturation could be used here. Another use for RDS is just to collect as large a sample as possible in hidden groups regardless of their representativeness. For example, RDS "seeds" have been used to recruit as many MSM as possible in "test and treat" outreach campaigns for blood testing for HIV and ante-retroviral treatment programs.

Overall a potentially useful addition to methodological literature on sampling procedures.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.

Author Response 23 Aug 2016

Philip Schluter, University of Canterbury

Thank you greatly for your prompt, encouraging, and constructive feedback on our paper. Together with the comments made by the other two reviewers, your feedback has resulted in what we feel is a much improved paper.

We would like to take this opportunity to make two notes. [1] The original submission was restricted to 1000 words in total – but, with permission from the journal, this constraint has been relaxed and we are able to include more material. This means we are able to broaden discussion of relevant issues and thus strengthen the manuscript. [2] This paper was submitted to the journal as an ‘observation article’, defined as: *Observation Articles allow the description of a novel observation that may be unexpected, and possibly currently without explanation. An observation can be a phenomenon that has been identified in field work, in the laboratory or through experimental analysis* (see: <http://f1000research.com/for-authors/article-guidelines/observation-articles>).

I support indexing this article as it as it appears to be a first-time test of the utility of Respondent Driven Sampling (RDS), a relatively novel sampling procedure, on a new "hidden" (hard to easily access) population - wheelchair riders. In this case it was not successful in recruiting sufficient respondents, but reporting that, and possible reasons for that outcome, could be useful for refining usage of the procedure.

Thank you for recognising the novelty of this approach, together with your encouraging and supportive comments regarding our paper.

I think the title and abstract are clear and provide good insight into the major points of the paper.

Thank you.

In addition to the possible reasons for low returns provided by the author I think it would be useful to include consideration of;

- 1. The typically low returns from internet surveys compared to face to face and pencil and paper surveys (see 1,2,3). It is possible that promoting an internet survey was just insufficient to motivate the "seeds", or the second wave recruits, to recruit more members into the network, even with an iPad reward incentive.*

Yes, this is indeed a valid consideration. In response, we have included a sentence the second paragraph in our revised discussion specifically referring to how more informed and enthusiastic seeds could have encouraged greater response rates, acting as a potential mitigation strategy against the impersonal nature of electronic surveys.

- 1. Elaborating on the "stigmatization" issue raised in the discussion. The "mechanism by which perceived stigma may effect RDS sampling" rather than being unknown, may well be that other groups e.g., drug users, "men who have sex with men"(MSM), sex-workers etc as a result of stigma, may have stronger social networks and obvious gathering spaces and be easier to access via seeds. Seeds with these groups are often "leaders", advocates who are "out", support group leaders, health service liaison/volunteer workers who move easily within those communities, social gatherings, entertainment or work spaces. There are not the same social presses for wheelchair riders to have these group social connections or common activities.*

Yes, this is valid and interesting point, and was also raised by another reviewer. In response, we have elaborated on the stigma issue by including new text in the first paragraph in the Discussion

section addressing the area of perceived stigma, and whether or not wheelchair users have a sufficiently strong internal network that enable the random recruitment of other wheelchair users.

1. *Methodological point. The article focuses on RDS's value for accessing representative population samples for research seeking normative and epidemiological data and concomitant importance of avoiding/controlling sample selection bias. However, RDS could also be valuable for accessing qualitative data e.g., wheelchair riders' views/experiences with public policies/services etc where as wide a range of responses (variability) should be expected, indeed welcomed, rather than seen as requiring some statistical weighting or control procedure. Other forms of criterion for data "completeness" e.g., saturation could be used here. Another use for RDS is just to collect as large a sample as possible in hidden groups regardless of their representativeness. For example, RDS "seeds" have been used to recruit as many MSM as possible in "test and treat" outreach campaigns for blood testing for HIV and ante-retroviral treatment programs.*

The value of using the RDS in qualitative research is a very interesting concept, and could be raised in the Discussion. However, on balance we feel it may be beyond the scope of our paper here, which was to discuss our experience of unsuccessfully satisfying the requirements needed to produce a theoretically representative sample of the population with no sampling frame.

Overall a potentially useful addition to methodological literature on sampling procedures.

We appreciate these words and the additional references offered.

Competing Interests: No competing interests to disclose

Referee Report 09 May 2016

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Jesse Kokaua

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I think this paper has merit as a scientific publication, in that it adds to the body of research about RDS by its application to a "hidden" population, that by all accounts should be reasonable, but turned out to be not effective.

The abstract begins with a sentence that ends with a repeated point "rapid increasing life-span" and "accelerated general population ageing". One of those statements is redundant. Paragraph 2 of the introduction starts with international findings but does the second sentence beginning with "Despite this . . ." refer to the NZ experience, or outside of the US and UK?

I feel that some points could be expanded upon:

The authors could expand, in the introduction, how the method improves the validity of, and reduces biases in, population-based estimates over other methods.

Examples of typical target populations for this method that are introduced in the discussion should move

to the introduction, as it supports the novelty of this application.

I feel the results are two-fold. Firstly, the sample as described. I know its a moot point but I would be interested to know how big a sample is required to reach equilibrium.

Secondly, since this is a paper on the RDS, I would have liked to see a description of the total numbers of people approached in each wave, with eligible and ineligible participants. Thus reporting if the failure was a result of response, eligibility, or simply a lack of invitation to participate in subsequent waves.

This is an interesting application of a novel method of sampling from a group of people made hard-to-reach, it seems, as a result of their successful integration into contemporary New Zealand society. One characteristic of strongly stigmatised groups in New Zealand, such as those identified in the paper, are their tendency to establish strong internal networks, both informal and formal, that I feel would be useful to this type of methodology. It would be difficult for individuals with small networks to fulfill the random selection criteria for RDS. I take it that this may have influenced the less stigmatised wheelchair user population but it wasn't clearly stated if this was the case.

The paper reports a good example of where a useful method may not apply, in its initial format, to a particular interest group. However, it wasn't totally discounted as a potential approach but the reader is left without any suggestion as to how it might be modified to improve its take up, except for the suggestion of formative research, while fulfilling the assumptions required for RDS to provide unbiased population-wide estimates.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.

Author Response 23 Aug 2016

Philip Schluter, University of Canterbury

Thank you greatly for your prompt, encouraging, and constructive feedback on our paper. Together with the comments made by the other two reviewers, your feedback has resulted in what we feel is a much improved paper.

We would like to take this opportunity to make two notes. [1] The original submission was restricted to 1000 words in total – but, with permission from the journal, this constraint has been relaxed and we are able to include more material. This means we are able to broaden discussion of relevant issues and thus strengthen the manuscript. [2] This paper was submitted to the journal as an 'observation article', defined as: *Observation Articles allow the description of a novel observation that may be unexpected, and possibly currently without explanation. An observation can be a phenomenon that has been identified in field work, in the laboratory or through experimental analysis* (see: <http://f1000research.com/for-authors/article-guidelines/observation-articles>).

I think this paper has merit as a scientific publication, in that it adds to the body of research about RDS by its application to a "hidden" population that by all accounts should be reasonable, but turned out to be not effective.

Thank you for your positive comments and summation. This, for us, was the basis for opting for the “novel observation” article type.

The abstract begins with a sentence that ends with a repeated point "rapid increasing life-span" and "accelerated general population ageing". One of those statements is redundant.

Although the intention was to make two important points, the first referring to wheelchair users and the second referring to the general population, we agree that this is unnecessarily confusing. We have revised this sentence to: “Internationally, wheelchair users are an emerging demographic phenomenon, due to their rapidly increasing life-span.”

Paragraph 2 of the introduction starts with international findings but does the second sentence beginning with "Despite this . . ." refer to the NZ experience, or outside of the US and UK?

This appears to be an international trend. However, to be clearer we have included “in New Zealand and Australia” and inserted an apposite reference.

I feel that some points could be expanded upon: The authors could expand, in the introduction, how the method improves the validity of, and reduces biases in, population-based estimates over other methods.

Agreed – indeed, a similar point was raised by another reviewer. Our original submission was limited in scope due to the word restrictions, as outlined above. However, in this revision, we have included more information and discussion. Specifically, we have included text providing a brief introduction to RDS (See second paragraph in Introduction section).

Examples of typical target populations for this method that are introduced in the discussion should move to the introduction, as it supports the novelty of this application.

Thank you for this comment; it indeed strengthens our introduction. In our revised manuscript we have included the following sentence with paragraph one of the Introduction: “RDS has traditionally been used to sample ‘hidden’ populations with inadequate sampling frames, such as those with greater risk of HIV, including injecting drug users.”

I feel the results are two-fold. Firstly, the sample as described. I know it's a moot point but I would be interested to know how big a sample is required to reach equilibrium.

We also felt that this information would strengthen our paper, but omitted it initially – due to the word count restrictions. In this version, within the Introduction, we now explicitly add an explanation about the concept of equilibrium and how it is reached (see third paragraph), while the second paragraph in the Results section explains specifically why our sample failed to satisfy the threshold for in-equilibrium data.

Secondly, since this is a paper on the RDS, I would have liked to see a description of the total numbers of people approached in each wave, with eligible and ineligible participants. Thus reporting if the failure was a result of response, eligibility, or simply a lack of invitation to participate in subsequent waves.

These details would be hard, if not ethically impossible, to determine. Consistent with RDS

protocol, an email was sent to people asking them to send on the invitation to three people, but there was no obligation for us (the researchers) to receive confirmation about whether all three recruitment codes were successfully allocated and distributed. Nevertheless, more information has been included in the Methods section (second paragraph) regarding how participants were asked to recruit three others, and the first paragraph in the Results section, which clarifies recruitment and response rates.

This is an interesting application of a novel method of sampling from a group of people made hard-to-reach, it seems, as a result of their successful integration into contemporary New Zealand society. One characteristic of strongly stigmatised groups in New Zealand, such as those identified in the paper, are their tendency to establish strong internal networks, both informal and formal, that I feel would be useful to this type of methodology. It would be difficult for individuals with small networks to fulfil the random selection criteria for RDS. I take it that this may have influenced the less stigmatised wheelchair user population but it wasn't clearly stated if this was the case.

Yes, this is valid and interesting point and was also raised by another reviewer. In response, we have included two sentences in the first paragraph of the Discussion section addressing the area of perceived stigma and whether or not wheelchair users have sufficiently strong internal networks that enable the random recruitment of other wheelchair users.

The paper reports a good example of where a useful method may not apply, in its initial format, to a particular interest group. However, it wasn't totally discounted as a potential approach but the reader is left without any suggestion as to how it might be modified to improve its take up, except for the suggestion of formative research, while fulfilling the assumptions required for RDS to provide unbiased population-wide estimates.

Thank you for this comment that will strengthen our paper. In response we have included more information in the second paragraph of the Discussion section about potential ways to improve similar studies, namely, selecting diverse seeds with larger networks and providing greater information and education to those seeds.

Competing Interests: No competing interests to disclose