Author's response to reviews

Title: Health behaviour modelling for prenatal diagnosis in Australia: A geodemographic framework for health service utilisation and policy development

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Health behaviour modelling for prenatal diagnosis in Australia:
A geodemographic framework for health service utilisation and policy development

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Authors’ response to reviews

Dear Editors,

We thank the reviewers for their considered review of the above manuscript.

We addressed the comments of Babak Khoshnood first as our response resulted in a number of improvements made to the textual content of the manuscript. Following the suggestions of Joan K Morris we have modified Figures 1-4 and we believe that this has enhanced the presentation of the data greatly. We addressed the comments of Jane McElroy last as a number of points raised indicated some misunderstanding of elements of the paper. We hope that the revised manuscript together with our response have clarified the concerns of this reviewer.

Please find our detailed response to each reviewer below, and the revised manuscript with highlighted changes attached for further consideration. We look forward to hearing from you soon.

Sincerely,

Evelyne Muggli
Public Health Genetics
We thank the reviewer for their salient comments and helpful suggestions on the above manuscript. Please find our response below.

General

With respect to the comment made regarding the analysis, which did not take into account individual socioeconomic and segment geodemographic characteristics, we have included this point more explicitly in the limitations and conclusions sections of the manuscript as suggested. The relevant sections now read as follows (with changes highlighted):

Limitations section:

However, even if street addresses had been available, the geodemographic software system only allowed for the study of groups of people. Effects of individual level socioeconomic factors cannot be disentangled from those factors related to place of residence. Our findings need to be interpreted within these limitations and inference about the behaviour and characteristics of individuals cannot be drawn.

Conclusions (abstract):

Classifying data to lifestyle segments allowed for practical comparisons of the geodemographic characteristics of women having prenatal diagnosis in Australia at a population level.

Specific comments:

1. We have incorporated the reviewer’s suggestions to expand on the introduction as follows:

Despite the wide availability of prenatal diagnosis and recent advances in antenatal screening to identify pregnancies at high risk for Down syndrome (DS), the extent to which prenatal screening and diagnostic techniques have resulted in changes in live birth prevalence of DS has been variable. A number studies have reported an unchanged live birth prevalence of DS or suggest that a substantial proportion of cases with DS continues to result in a birth, [1-5] while others have found a decrease in the live birth prevalence of DS [6] or that there is variation across countries. [7] Maternal socioeconomic and demographic factors will be contributing to these findings through their role in influencing the utilisation of prenatal diagnosis. [8-12]
2. Again in the introduction, we have expanded on Victoria’s prenatal screening and diagnosis policy as suggested:

**Second trimester maternal serum screening and routine ultrasound are available to all pregnant women.** Pregnant women aged 37 years or over, and younger women with an increased risk screening test for chromosomal abnormalities are eligible for prenatal diagnosis free of charge. First trimester combined maternal serum and nuchal translucency screening was introduced in Victoria in 2001 and is currently available to women through private providers with minimal government subsidy. Pregnant women under the age of 37 years who wish to have prenatal diagnosis without a recognised indication can do so through a private provider at their own expense.

3. To answer the reviewer’s question on the availability of prenatal screening in rural areas we have extended the relevant sentence as follows:

**While second trimester maternal serum screening is available throughout Victoria, all pregnant women residing in rural regions have few options but to travel to a metropolitan centre to have a prenatal diagnostic test.**

4. With respect to the reviewer’s suggestion to clarify “a range of sociodemographic characteristics”, we did not include specific characteristics as variables as is done in a traditional multivariate analysis. The aim of this study was to identify specific lifestyle segments that may be associated with uptake of testing and to describe these using the underlying demographic data.

In a second point the reviewer rightly pointed out that we did not formally test the applicability of consumer behaviour modelling and have amended the relevant text in the abstract and introduction:

> The objective of this study was to investigate the geodemographic characteristics of women who have prenatal diagnosis in Victoria, Australia, and to examine a potential relationship between uptake of diagnostic testing and Down syndrome birth prevalence by applying a novel consumer behaviour modelling technique in the analysis of health data.

5. We have changed the wording in the relevant paragraphs to clarify the system of notification to the registers and if terminations post 20 weeks are included:

*Births/confinements:* The Perinatal Data Collection Unit, Public Health, Victorian Government Department of Human Services (PDCU) has mandatory reporting of every birth (including pregnancy terminations) at or after 20 weeks gestation and data are collected from all maternity hospitals (and homebirths) in Victoria on the Perinatal Statistics Form.

*Birth Defects Register:* The Victorian Birth Defects Register has multiple sources of notification. It collects information on all births from the PDCU where there has been a
reported birth defect and includes data on pregnancy terminations for birth defects before and after 20 weeks.

6. In response to the reviewer’s comment regarding general principles of the geodemographic segments, we offer to include basic descriptions as online content. We previously made a decision not to include these as they are predominantly used to “showcase” the data for industry clients and do not do justice to the wealth of information available on spreadsheets. However, we had similar concerns about the “black box” and hope that the material offered will be of use:

Basic segment descriptions used to showcase customer behaviour profiles for commercial purposes, are available as online content (see Additional file 1).

7. We hope that the modified sentence answers the reviewer’s query about this point: Expected numbers of live births with DS were calculated by assigning a maternal age-specific risk at term to each birth in 2002, according to the mother’s age. Maternal age-specific risks were taken as published by Reynolds in 1994.

8. It is possible that women giving birth in a private hospital are more likely to have access to nuchal translucency scans and first trimester combined screening through private providers, which may result in higher numbers of women screened/ fetuses diagnosed. (No changes made to the manuscript, a paper on the epidemiology of Down syndrome in Victoria is in progress).

9. The authors agree with the reviewer that it would be valuable to look at the actual maternal age distribution of women having prenatal diagnosis in more detail, as it may explain some of the differences in uptake that we observed. We used the cutoff of 37 years (as per prenatal diagnosis policy in Victoria) to enable modelling of the data to geodemographic segments in two strata and individual level data was not available following the modelling. While age is incorporated in the geodemographic segment characteristics we found this issue to be one of the potential limitations of this method. We also noted this issue on page 12 in the discussion on Down syndrome:

This finding is difficult to explain within the constraints of the data available, but an underlying skewed maternal age distribution and the associated risk for DS may be a contributing factor.

10. We have amended the relevant sentence as follows:

Given that the results were based on modelled data, extrapolated from actual, we discuss only those segments with rates greater or less than 20% variation from State average, (above or below the line in figures) for each region.
11. We agree with the reviewer’s comment that the discussion section includes some straight results. It was difficult however, given the unusual methodology, to present results on the geodemographic segments without elaboration and discussion. The relatively concise results section only refers to data that are visible in the figures and tables (No changes made to the manuscript).

12. As discussed in the relevant paragraph, we believe that this peak is an artefact of the data modelling. High rise rentals in Melbourne, unlike in some other cities in Australia, are interspersed throughout the wealthy suburbs which may have resulted in an artificially high rate of uptake. The characteristics of this segment, which are that of a more disadvantaged community, are given to support the view that the uptake rate may be an artefact. (No changes made in text).

13. We have modified the relevant sentence as below. The term individual opportunity is also referred to in the abstract, where there are examples of its meaning in brackets. Without expanding further in the manuscript, a lack of individual opportunity refers to the person not owning a car or a lack of child minding options or a work situation that does not allow for lengthy doctor’s appointments.

The issue of access to testing for women in this segment more likely relates to a lack of individual opportunity, than their physical location.

14. It has certainly been shown that prenatal screening is a more effective filter for prenatal diagnosis than advanced maternal age alone. [13, 14] Consequently, the relationship between uptake of prenatal diagnosis and live birth rate of Down syndrome may be affected by the extent to which women use prenatal diagnosis with or without consideration of screening results. [15, 16]
Health behaviour modelling for prenatal diagnosis in Australia: A geodemographic framework for health service utilisation and policy development

Reviewer: Joan K Morris

We thank the reviewer for their considered review of, and helpful comments on the above manuscript. Please find our response below.

Major compulsory revision

1. We have modified Table 1 to include additional information on the proportion of births per segment for each region and have provided a SES gradient. This basic gradient is one of average weekly household income, where the lowest income was given as 1, rather than the actual A$ figure. The SES gradient is now comparable between the rural and metropolitan regions. We also modified Figures 1-4 to reflect the gradient accurately along the X-axis.

2. a. Applying a geodemographic framework to our data using mathematical modelling led us to move away from a traditional epidemiological analysis, such as a regression analysis or standardised prevalence. The mathematical modelling, which was done to convert local government data to geodemographic segment data resulted in observations several magnitudes higher than the actual numbers. Consequently, confidence intervals would not be meaningful. Like the reviewer, we initially felt it was necessary to demonstrate a certain validity of our methodology. We have attempted to use a two sample test of proportions to compare the data in a most basic fashion, but realise that this has caused some confusion. After further considerations we have decided not to offer any form of statistical analysis for the comparison of data relating to the geodemographic segments and have presented a purely descriptive analysis.

We have analysed the uptake of prenatal diagnosis using a number of maternal demographic predictors by logistic regression in a manuscript submitted elsewhere. The methodology used in the present manuscript is not of epidemiological rigour, rather it is designed to look at the feasibility of a new framework for policy development and service planning in the context of health services research.

With respect to the reviewer’s comment regarding the state average uptake of prenatal diagnosis, this was 8.4%. This average was across the whole state and did not differ between rural and metropolitan regions. We have now added this figure in the description of the data: “Figures 3 and 4 depict uptake of prenatal diagnosis across all ages compared to State average (8.4%) for metropolitan and rural regions respectively.”
b. We agree with the reviewer that the SES gradient generally does not appear to have a significant influence on the uptake of testing or the live birth ratio of DS and that most of the differences relate to rural vs metropolitan Victoria. However, we believe that visualising the data points along this gradient helps the reader to understand the meaning of the segments and how they compare to each other in their basic socioeconomic position. Following the reviewer’s comments in Point 1, the figures have been greatly improved by making the gradient comparable across rural and metropolitan regions and plotting the segments according to their actual position with regard to their average income.

3. Again, to apply a traditional statistical analysis to our modelled data would not result in valid comparisons and it was not possible to calculate a meaningful standardised live birth prevalence or level of uptake. In this context, we have changed the terminology “live birth prevalence of Down syndrome” to “live birth ratio of Down syndrome” throughout the manuscript and given the following explanation in the text:

A ratio for DS for each segment was created, where the observed rate being equal to the expected rate was defined as 1. A ratio above 1 indicated a higher than expected proportion of DS and a ratio below 1 indicated a lower than expected proportion of DS. This ratio was defined as the live birth ratio of DS.

We are presently working on a manuscript on the epidemiology of Down syndrome in Victoria and hope to examine some of these points of interest in more detail.

4. We agree with the reviewer that some of the differences observed in Table 3 are due to the different maternal age structures in the birth vs live birth of DS groups. However, the findings in the most current time period of the higher proportion of live births in the public sector and in rural regions is not expected to be related to maternal age. In fact, the latter has originally prompted this research. We would like to have Table 3 included in the manuscript as previously.
We thank the reviewer for their review and comments on the above manuscript. Please find our response below.

**General:**

**Separate manuscripts:** In the present manuscript, we examined the geodemographic characteristics of women who have prenatal diagnosis and a potential relationship between uptake of testing and Down syndrome prevalence using a novel methodology. This methodology is of limited epidemiological rigour but may be useful for service planning and policy development. Two other manuscripts are currently underway, one that used a regression analysis to investigate a number of maternal predictors in the uptake of testing and another on the epidemiology of Down syndrome in Victoria.

**Analysis does not match the research questions:** In response to the reviewer’s comment, we hope to have clarified the aims of the study as follows:

The objective of this study was to investigate the geodemographic characteristics of women who have prenatal diagnosis in Victoria, Australia, and to examine a potential relationship between uptake of diagnostic testing and Down syndrome birth prevalence by applying a novel consumer behaviour modelling technique in the analysis of health data.

**Geographic unit:** We believe that a geographic map of Victoria showing postcode and local government boundaries may confuse the reader as the unit of analysis was a combination of geography and demographic factors (geodemographic segments).

**Major Compulsory revision**

**Proportional assignment of postcodes to LGAs based on census data:** For example according to census data postcode 3351 is distributed among LGAs as follows: Ararat 8.3%, Ballarat 14.1%, Golden Plains 61.1% and Pyrenees 16.5%.

**CCDs (census collection districts):** This unit can only be derived if a street address is available.

We believe that we have acknowledged the limitation due to not having actual street addresses for each record in several sections of the manuscript and have cautioned the reader to interpret the findings with this limitation in mind.
Analysis using postcodes or shires: Postcodes cannot be used directly for, or be modelled to, the geodemographic segments used in our study. Our analysis was not a geographic one, but included considerable demographic factors, which are affected by a combination of place of residence and lifestyle.

Effect of non-incorporated areas: French Island is the only non-incorporated area in Victoria and has no recorded births in any given year. It did not affect the analysis.

Geodemographics of the population: We have now made basic geodemographic segment descriptions (as they are used in the commercial sector), available as online content. We have also added the births that occur in each segment as a proportion of all births in each region to Table 1.

Opaque statistics: The mathematical modelling, which was done to convert local government data to geodemographic segment data resulted in observations several magnitudes higher than the actual numbers. Confidence intervals or other traditional statistical comparisons would not be meaningful. We used a 2 sample test of proportions in Tables 2 and 3 as a basic comparison between years. After further considerations we have decided not to offer any form of statistical analysis for the comparison of data relating to the geodemographic segments and have now presented a purely descriptive analysis.

Screening analysis: In the present manuscript we did not examine influences of prenatal screening. With regard to pregnancy terminations following prenatal diagnosis, it is known that most pregnancies are terminated (Mansfield 1999 and Forrester 1999) and unpublished data from the Victorian Birth Defects Register supports this finding. We have acknowledged in the manuscript that the proportion of women who continue their pregnancy is not known for each segment and that choices may contribute to the findings.

Inclusion criteria for women who were expected to deliver in the same year as having prenatal diagnosis: Many women who have prenatal diagnosis (usually done between 10 and 25 weeks gestation) are not expected to deliver until the following year. These women would not be included in the birth cohorts selected for this study (1998 and 2002). In addition, including all women who had prenatal diagnosis in 1998 and 2002 would have considerably complicated the probabilistic record linkage that was undertaken.

Data used in the analysis: Again, we did not investigate prenatal screening in this study. Birth data was used for babies with Down syndrome because in this instance we compared births with births. Data on confinements was used to examine women’s uptake of testing because we looked at women and not births (of which there may be multiple births per woman). In studies such as this, investigators refer to confinements rather than pregnancies because it is not recorded how many pregnancies are miscarried and “lost” to the study data. The years included in the study are described in the study methods and visualised in Tables 2
and 3. The 1997-2002 birth data the reviewer refers to are pooled data on births of babies with Down syndrome.

**Exclusion of segments with less than 1% of births in region:** This is a descriptive analysis of data and we believe that discussing segments that were predominantly non-residential (n=4) or had few births (<500 in metro or <100 in rural regions, n=11)) and even less prenatal tests or births of babies with Down syndrome would not have added value to this manuscript.

**Two sample test of proportions:** We used the two sample test of proportions on data presented in Tables 2 and 3. Total numbers of births and births of babies with Down syndrome were used as denominators, the numerator being the proportions in each category. These statistics can be reproduced from the data provided.

**Expected rates of Down syndrome at term:** We did not reference a model to calculate these rates in the manuscript. The maternal age specific risks of Down syndrome at term were taken as per Reynolds 1994. We calculated the expected rates of Down syndrome at term using a standard technique. We have modified the relevant sentence to clarify this as follows:

*Expected numbers of live births with DS were calculated by assigning a maternal age-specific risk at term to each birth in 2002, according to the mother’s age. Maternal age-specific risks were taken as published by Reynolds in 1994.*

**Modelled observed rates of prenatal diagnosis:** The unit of analysis throughout this manuscript was the geodemographic segment, not geographic areas.

**High risk women (>37) who have opted out of prenatal diagnosis change the screening rate and what does uptake of prenatal diagnosis mean (is it the same as screening?):** We have provided uptake of prenatal diagnosis in women aged 37 years and over in Figure 2. There is no information on the proportion of women who are offered a diagnostic test and opt out. We are unsure about the reviewer’s comment about how this influences the screening rate. With respect to the second comment, prenatal screening is not the same as prenatal diagnosis. Uptake of prenatal diagnosis was defined in the study population section of the manuscript as women who had amniocentesis or chorionic villus sampling. Unlike prenatal screening, which returns only a risk estimate for a potentially affected pregnancy, both these procedures diagnose fetal karyotype abnormalities.

**Table 2 and 3 do not add to the paper:** Tables 2 and 3 show baseline data of the study populations and we believe these should be included in the manuscript.

**Figure 1-4:** We have modified Figures 1-4 to improve their content and labelling. CIs are not presented as this was a descriptive analysis of modelled data. A full list of segment names in the order of appearance in the figures (by SES) is given in Table 1.