



CHÉOS

Centre for Health Evaluation
& Outcome Sciences

STAKEHOLDERS' PREFERENCES & COST-EFFECTIVENESS OF NON- INVASIVE PRENATAL TESTING IN BRITISH COLUMBIA, CANADA

ABOUT CHÉOS

The Centre for Health Evaluation and Outcome Sciences (CHÉOS) produces high-quality evidence to make informed changes to the health care system. Bridging the gap between data, research, and care, CHÉOS is a collaboration between cross-disciplinary scientists and expert research staff evaluating the effectiveness of health interventions at the population level. From assessing the cost-effectiveness of a new drug or treatment option, to informing policy decisions that change how care is delivered, CHÉOS seeks to improve health outcomes for all.

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Background

Prenatal screening programs are routinely employed in primary maternity care worldwide. Different methods that assess the risk of fetal abnormalities, including Down syndrome (DS), and trisomy 13 and 18, provide many options for prospective parents^(1,2). In recent years, non-invasive prenatal testing (NIPT), a blood test that measures cell-free fetal DNA circulating in maternal serum, has shown greater sensitivity (>99%) and specificity (≥99%) for DS as compared to conventional screening tests⁽³⁾. When health care policymakers develop prenatal screening

programs, they must determine which tests to implement, in what sequence, and for whom.

In Canada, a publicly funded health care system is in place and screening guidelines vary by province⁽⁴⁾. In the case of British Columbia (BC), universal coverage is provided for conventional prenatal screening that includes prenatal serum screening. Women may also choose to pay out-of-pocket for options that are not publicly funded, including NIPT for those who are at low risk of chromosomal abnormalities—not eligible for public coverage⁽⁵⁾.

Why this research matters



THE FUTURE OF SCREENING

New and expanded prenatal screening options are giving rise to considerations that need to be investigated in jurisdictions with varying prenatal screening protocol and health care coverage.

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THE CHOICE INVOLVED

Since prospective parents may be faced with difficult choices surrounding prenatal screening and diagnostic results, individual preferences will influence test uptake and must be investigated.

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THE COST OF CARE

The cost-effectiveness of new screening methods must be assessed to provide decision-makers with adequate information to prioritize and allocate funding for optimal screening strategies.

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In collaboration with BGI, scientists from CHÉOS proposed a series of health economic analyses to investigate patient preferences and cost-effectiveness of NIPT in the BC setting. Findings from our studies have important

implications for guiding prenatal screening program design and implementation in BC and other publicly-funded, high-income jurisdictions. Our findings can also be used to assess future technology that results in improved attributes.



Attitudes Toward Prenatal Screening for Chromosomal Abnormalities: a Focus Group Study

BACKGROUND

While discrete choice experiments (DCEs) are well established methods to ascertain patient preferences, there is limited literature describing use of qualitative methods in DCE design.

AIM

This article provides a case study of the qualitative research process for developing the conceptual attributes for a DCE for prenatal screening and diagnosis.

METHODS

Participants were recruited through posters and social media. Four in-depth, semi-structured focus groups with pregnant women and their partners/support people were conducted in Metro Vancouver.

FINDINGS

Our analysis indicates that choosing prenatal screening and diagnosis involves four intertwined decisions: whether to undergo screening and testing, which screening test to take, which diagnostic test to take, and what to do with a positive diagnosis. The factors that are important to women and their partners vary depending on the decision and include: time of diagnosis, information on conditions tested, false positives, cost, the invasiveness of the test, and potential harm to woman and baby.

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This analysis addresses the gap in reporting of qualitative methods in DCE survey development. With the emergence of new prenatal testing options, it is essential to understand women's preferences for screening to inform guidelines and resource allocation.

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DISCUSSION

Findings suggest that certain attributes were more salient for screening versus diagnostic tests. Preferences were often shaped by a woman's perceived ability to care for a child with a genetic anomaly, personal risk factors, parity, views on termination, and perceptions on public or private coverage. Participants valued mental well-being and demonstrated a willingness to trade-off on certain attributes in order to minimize stress or anxiety during pregnancy.

CONCLUSION

Study findings will be used to inform DCE attributes, levels, and choice questions. Findings will be important for policy decisions surrounding prenatal testing.

Full article: Munro S, Sou J, Zhang W, Mohammadi T, Trenaman L, Langlois S, Anis AH. Attitudes toward prenatal screening for chromosomal abnormalities: A focus group study. *Women Birth*. 2019 Aug;32(4):364-371. doi: 10.1016/j.wombi.2018.09.006. Epub 2018 Sep 27.



A Hierarchical Bayes Approach to Modeling Heterogeneity in Discrete Choice Experiments: an Application to Public Preferences for Prenatal Screening

BACKGROUND

Previous studies assessing preferences for prenatal screening have focused on preferences of the affected population and have largely assumed homogeneous preferences. We aimed to estimate public preferences and willingness to pay for prenatal screening and diagnosis from a Canadian general population sample and model preferences at the individual level.

METHODS

A discrete choice experiment was used to elicit preferences for different aspects of prenatal screening and diagnostic strategies. Strategies differed in five attributes: timing of the results, false negative rate, false positive rate, risk of miscarriage, and out-of-pocket cost. Respondents made forced and unforced choices using a dual response approach. Hierarchical Bayes analysis was applied to estimate individual-level part-worth utilities. Individual probability and expected uptake of prenatal screening under different scenarios were also assessed. Subgroup analyses were conducted using individual-level preferences.

RESULTS

The final analyses were based on a sample of 4,601 respondents. Results showed that the two most important attributes were false negative rate and miscarriage risk. There was significant heterogeneity in preferences among respondents. Individuals' perception of the risk of pregnancy with chromosomal abnormalities affected their preferences for screening. The relatively high

uptake of safe and accurate screening among all groups of respondents indicated people's desire for information about the health of their unborn baby regardless of their decision to continue the pregnancy.

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Since false negative rates and risk of miscarriage are shown to be more important to the general public, improvement in the overall detection rate and safety can increase public acceptability of these programs.
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CONCLUSION

Our findings are consistent with previous studies based on affected-population-preferences. This concordance should be reassuring from a policy perspective and can inform the design of publicly-funded prenatal screening programs.

Full article: Mohammadi T, Zhang W, Sou J, Langlois S, Munro S, Anis AH. A Hierarchical Bayes Approach to Modeling Heterogeneity in Discrete Choice Experiments: An Application to Public Preferences for Prenatal Screening. *The Patient: Patient-Centered Outcomes Research*. Accepted on Nov 25, 2019.



Cost-Effectiveness of Prenatal Screening and Diagnostic Strategies for Down Syndrome: a Microsimulation Modeling Analysis

OBJECTIVES

Down syndrome (DS) is the most frequently occurring fetal chromosomal abnormality and different prenatal screening strategies are used for determining risk of DS worldwide. New non-invasive prenatal testing (NIPT), which uses cell-free fetal DNA in maternal blood can provide benefits due to its higher sensitivity and specificity in comparison to conventional screening tests. This study aimed to assess the cost-effectiveness of using population-level NIPT in fetal aneuploidy screening for DS.

METHODS

We developed a microsimulation decision-analytic model to perform a probabilistic cost-effectiveness analysis (CEA) of prenatal screening and diagnostic strategies for DS. The model followed individual simulated pregnant women through the pregnancy pathway. The comparators were serum-only screening, contingent NIPT (i.e., NIPT as a second-tier screening test) and universal NIPT (i.e., NIPT as a first-tier screening test). To address uncertainty around the model parameters, the expected values of costs and quality-adjusted life-years (QALYs) in the base case and all scenario analyses were obtained through probabilistic analysis from a Monte Carlo simulation.

RESULTS

Base case and scenario analyses were conducted by repeating the micro-simulation 1,000 times for a sample of 45,605 pregnant women per the population of British Columbia, Canada (N=4.8 million). Preliminary results of the sequential CEAs showed that contingent NIPT

was a dominant strategy compared to serum-only screening. Compared with contingent NIPT, universal NIPT at the current test price was not cost-effective with an incremental cost-effectiveness ratio over \$100,000/QALY. Contingent NIPT also had the lowest cost per DS case detected among these three strategies.

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These results may aid policymakers in prioritizing and allocating funds toward a cost-effective prenatal screening strategy.
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CONCLUSION

Including NIPT in existing prenatal screening for DS is shown to be beneficial over conventional testing. However, at current prices, implementation of NIPT as a second-tier screening test is more cost-effective than deploying it as a universal test.

Full article: Zhang W, Mohammadi T, Sou J, Anis AH. Cost-effectiveness of prenatal screening and diagnostic strategies for Down syndrome: a microsimulation modeling analysis. PLOS ONE. 14(12): e0225281. <https://doi.org/10.1371/journal.pone.0225281>

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