1a: A clinical trial of the effectiveness of a dental caries prevention program for Cree mothers and their infants*

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1b: Abstract

Background: The goal of this cluster randomized trial was to test the effectiveness of a counseling approach, Motivational Interviewing, to control dental caries in young Aboriginal children. Motivational Interviewing, a client-centred, directive counseling style, has not yet been evaluated as an approach for promotion of behaviour change in indigenous communities in remote settings.

Methods/design: Aboriginal women from the 9 Cree communities in Quebec recruited expectant and new mothers to the trial, administered questionnaires and delivered the counseling to mothers in the test communities. The goal was for mothers to receive the intervention during pregnancy and at their child's immunization or well-baby visits.

The communities were randomly allocated to test or control group by a random "draw" over community radio. Sample size and power were determined based on an anticipated 20% reduction in caries prevalence. Randomization checks were conducted between groups. Data on children's dental health status and family dental health practices was collected when children were at least 30-months of age.

Results: In the 5 test and 4 control communities, 272 (131 test; 141 control) of the original target sample size of 309 mothers were recruited over a 2½ year period. Demographic and baseline characteristics were similar for the two groups of mothers. At the end of the trial, 241 participating children had follow-up examinations. The actual mean (SD) number of MI interventions per test mother was 2.7 (1.7), range 0-6. Of the 131 test mothers, 93 (71%) had 2 or more MI sessions and 46 (35%) had 4 or more interventions usually of 15-30 minutes duration. When levels of caries of increased severity were examined, an apparent treatment effect emerged. Percentages of children with caries (untreated or treated) at the level of the dentin or worse were 65% in the control group and only 40% in the test group, which implies an estimated 38% reduction in disease prevalence. However, the assessment of statistical significance of this effect was inconclusive. Results by community suggest that the intervention may have had a caries preventive effect in all but one of the test communities

The implementation of the trial in these remote communities, the problems of enlisting dedicated project staff and the unexpected changes in philosophies about research by the territory's governing body heightened the project's challenges.

Conclusions: Overall, the results do not provide compelling evidence for a caries preventive effect of the MI intervention in these Cree communities. Certainly, the overall prevalence of caries in both treatment groups remained alarmingly high. However, there are suggestions that the intervention may have an impact on the extent or severity of disease. These results are not conclusive due to effects of clustering of children within communities and the small number of communities.
Undoubtedly our results will be useful for generating specific hypotheses that can be tested in future trials and for informing protocols, approaches and methods in future randomized controlled trials in high-risk and difficult to access communities.

Trial registration: This trial is registered as ISRCTN41467632.

Introduction

2a: Scientific background and explanation of rationale

The disease: dental caries in young children

The poor dental health of Aboriginal children in Canada is a major public health issue. Early childhood caries or ECC is the term used for dental caries in the young child. This condition can affect children before their first birthday [1]. ECC can be a painful condition, influencing a child's ability to eat properly, sleep through the night [2], grow and develop normally [3] and thus achieve full potential. Furthermore, caries in the primary (baby) teeth has a significant and positive association with caries and malalignment of the permanent teeth [4]. In some Canadian Aboriginal communities, the prevalence of ECC exceeds 90% [5]. Furthermore, the financial burden of treating ECC is enormous, and even more so for young Aboriginal children from remote communities who often must travel vast distances for comprehensive treatment [6].

Dental caries (tooth decay) occurs when "cavity-causing" bacteria, foods usable to the bacteria and susceptible teeth are in contact with each other long enough to allow bacterial by-products to demineralize the enamel of the teeth [4]. A universally effective, caries-prevention program with predictable long-term results for young Aboriginal children has yet to be found. Because ECC is a disease that is multi-factorial in origin, any preventive program must include a variety of strategies [7]. The microorganisms implicated in the initiation of dental caries are transmitted from mother to child [8] [9] [10]. The inoculation of bacteria received by the child appears to be related to maternal oral health status, diet and oral hygiene practices [10]. Once the baby's primary teeth begin to erupt, brushing the teeth regularly with toothpaste containing fluoride is essential. Another means of introducing fluoride is regular application of fluoride varnish.[11]. Given that poor dietary behaviours also contribute to ECC, dietary modification is an important component of an infant oral health promotion strategy. A sleep-time bottle, constant daytime sipping from a bottle or sippy cup containing anything other than water and frequent snacking are practices linked to the development of extensive caries [12].

Most caries-prevention strategies require that a parent change an existing behaviour, for example bottle-feeding, or adopt a new behaviour, for example regular toothbrushing. However, providing knowledge alone to parents rarely leads to long-term changes in preventive behaviours [13]. Conversely, behavioural techniques that structure change and assist parents in the process of change may lead to long-term change in behaviours.

The population of interest

The Cree, an Aboriginal nation of North America, are the largest First Nations group in Canada with over 200,000 members. The Quebec Cree nation is called Eeyou Istchee - Cree for *Land of the People*, and lies to the east and southeast of James Bay. The Quebec Cree number some 14,000 people and live in nine distinct settlements.

In 1985, a survey of children in the eastern James Bay community of Chisasibi revealed that the mean number of decayed, extracted and filled primary tooth surfaces or the "defs" for 4-6 year old girls was 19.3 and for boys was 24.2 [14]. A more recent community-wide survey of 1079 Cree children between the ages of 12 months and 12 years [15] found that 30.4% of 12-24 month olds had dental caries. By comparison, only 4% of similarly aged non-Aboriginal children in

Quebec had caries [15]. The amount of dental disease in these young Cree children is disturbing particularly given the fact that each Eeyou Istchee community has a clinic for provision of dental services and treatment is free-of-charge. However, it is unlikely that there will ever be enough dental clinics and dental practitioners in Eeyou Istchee to manage the amount of disease. Clearly, existing health services need to be supplemented with a population-based approach to promote improved child oral health.

Community consultation:
Oral health intervention strategies must be sensitive to the intended target population because of the important role that culture plays in shaping health-related attitudes and behaviours in Aboriginal communities [16]. Extensive community consultation was a prerequisite before the trial was even considered. Thus, extensive discussions throughout the Cree territory were undertaken. While disturbances in the balance between bacteria, substrate and host are the local factors associated with ECC, health determinants such as economics, social norms and housing conditions also have a substantial impact on the development of the disease [17]. Therefore, any intervention had to be planned with early and continuous community collaboration and consultation and needed to be sensitive to existing beliefs and traditions about parenting practices, child comforting and infant health. The project investigators, one of whom was the public health dentist for the territory, undertook two years of consultation with community leaders, health care workers and families from Eeyou Istchee to discuss appropriate strategies prior to beginning the trial. In addition, the Research Committee of the CBHSSJB collaborated on this project from its conception.

When the project began, relatively few community-based initiatives to improve the oral health of young children had been undertaken [18] [19] and scant few had been conducted as randomized controlled trials [20] [21]. The concept, advantages and disadvantages of a randomized controlled trial were discussed at length to ensure that communities understood that, during the trial, "test" communities were to have a more intensive program than "control" communities. The consensus from the consultation was that, since no infant oral health promotion program was currently in place, it was acceptable to participate in a trial to definitively determine the most effective approach.

2b: Study-research questions
Primary question: Is there any difference in the dental health status of young Cree children whose mothers have participated in a client-centred, one-on-one, preventive counseling intervention, Motivational Interviewing (MI), compared with children whose mothers received oral health information in the form of an educational pamphlet?

This question will be answered by testing the hypothesis that the prevalence of caries in 30 month old children will be lower in the experimental communities than in the control communities.

Secondary questions: Are Cree mother's knowledge and beliefs about child dental health issues, their dental health practices and child feeding and comforting practices altered by participation in an intervention based on principles of Motivational Interviewing (MI)?

Additional questions: Will children whose mothers participate in the MI interventions have fewer negative health outcomes related to poor dental health, for example pain or problems eating, than control children? Does mothers' participation in these interventions decrease the probability of their children requiring dental treatment under general anesthesia or with sedation?

These questions will be answered by testing the null hypothesis that there will be no difference between the two groups.

The goal of the trial was to test the effectiveness of an intensive one-on-one preventive counseling intervention, Motivational Interviewing (MI). Issues related to design of the trial; training and recruitment of staff; articulation with the existing organizational structure of health care delivery; implementation and evaluation of outcomes of the trial will be reported.

Methods

3a: Study-design

The research was approved by the Behavioural Ethics Review Board of the University of British Columbia and by the Cree Board of Health and Social Services of James Bay (CBHSSJB).

The trial was designed as a single-blind study with cluster randomization by community and two treatment groups. Randomization of individual mothers within each community was inappropriate for this study because of the close-knit nature of the communities and the risk of contamination. Therefore, participants were allocated to treatment conditions by community using cluster randomization. A cluster randomization design reduced the risk of cross-contamination but did not eliminate it completely. A characteristic of this project that reduced contamination was the fact that the nine "clusters" (communities) were distinct and well separated by considerable distance in many cases. Furthermore, each community had its own health clinic. In addition, when the trial began, mothers of young children seldom traveled much between communities because of the prohibitive cost and the difficulty of traveling with young children.

3b: Changes to methods after trial commencement

1. Project staff

Cree dental assistants who worked part-time in community dental clinics were ideal personnel to work on this project. The study protocol indicated that these dental assistants be recruited and employed in a new capacity as Dental Health Representatives, or DHRs, to work on the project in test communities and also to recruit mothers from neighboring control communities. Training and calibration in recruitment procedures and in the MI technique were to be provided to the DHRs in workshops facilitated by an expert in MI. DHRs would learn how to frame questions, help structure change, apply fluoride varnish and demonstrate toothbrushing. Instruction and practice in administering the survey instruments would also take place.

Surveillance, promotion, prevention, protection, regulation, research and training relating to the health of the Cree population in Eeyou Istchee are managed by the Public Health Department of the CBHSSJB. Given the support for this project provided by the CBHSSJB, it was essential that the project articulate with the structure and organization of health care

services within Eeyou Istchee. The Department's long-term goal related to this specific project was that, if successful, the intervention would become a component of the Department's health promotion activities. To that end, their inclination was for the day-to-day work of the project to be undertaken by existing Health Department staff rather than new staff, the so-called "Dental Health Representatives" (DHRs), who were to be specific to the research project. After considerable deliberation, it was decided that existing department staff, women currently employed as Community Health Representatives (CHRs) would do the recruiting of mothers and also be the project interveners.

The Project Manager who was hired was a dental hygienist from the Cree nation who was born, raised and had worked for several years in Eeyou Istchee.

2. Staff engagement

For a variety of reasons, CHRs in many of the communities were challenged by the process of subject recruitment and delivering the intervention. The negative side of engaging these existing personnel in a research project was their sense of being overworked and not sufficiently rewarded for this addition to their daily workload. This project was yet another burden for the already busy and in-demand CHRs. Furthermore, the CHRs may have found that expectant women and new mothers were too busy to spend an additional amount of time completing the project's many documents or were simply not interested in participating in a research project. Even though the project investigators were diligent in following recommended principles of involving Aboriginal communities in research, recruitment of participants was a struggle. Thus, in 2 of the 5 test communities and in 2 of the 4 control communities, recruitment and, for the test communities, delivery of the intervention was eventually completed by the Project Manager. One of the project-specific DHRs achieved success in one test-community and eventually took over the work of a retiring CHR in another test-community. A CHR, aided by the Project Manager, completed and continued the project's work in the 5th test community. Local women who were not CHRs completed recruitment in the remaining 2 control communities. Recruitment was closed after 2½ years when the number of mothers recruited, though short of the original goal, was sufficient to maintain the power of the trial.

Needless to say, the inconsistency of project staff's enthusiasm for this research project affected the implementation of the intervention in some of the test communities. On the positive side, in 3 of the test communities, almost all of the mothers had at least one MI intervention and, in many cases, had up to 5 MI interventions. However, in a 4th test community where CHRs were quite indifferent to the project, 8/18 or 40% of participating women never received an MI intervention at all. Expectant women were recruited to the project but assistance from the Project Manager was rebuffed and thus only 60% of subjects ever had an MI intervention. In another test community only 11/15 (73%) mothers had an MI intervention. This community had several unsettling events over the period of the project that consumed the CHR's time and energy and the dental health prevention project became a secondary concern. The Project Manager was by this time working only half-time on the research project because of family issues. She attempted to help these struggling communities but had difficulties managing staff's ability and desire to work on the project amidst their communities' other numerous challenges

3. Participant recruitment

Recruitment of pregnant women to the study began in January 2005. As the Project Manager worked with CHRs to support them in recruiting, the question arose as to whether mothers who had newly delivered could also be included in the trial (see 4a). No scientific or logistical reasons were apparent for excluding newly-delivered mothers so inclusion criteria at all sites were extended to include mothers of "predentate" infants.

4a: Eligibility criteria

All women in the 9 Cree communities who were pregnant were to be asked to participate in the study. Any woman knowing of an impending, permanent move out of her community was excluded. Even children born with a medical concern or a congenital anomaly, for example cleft palate were included, if that was the family's wish. Information about the health of the child will be gathered at the outcomes assessment and will be managed in the statistical analysis.

4b: Settings where data were collected

Women were recruited for the study when they presented to their community's health clinic for prenatal or ongoing well-child care. Given that the project took place in 9 different communities, awareness of who was pregnant or who had just delivered varied amongst clinics. In some clinics, the CHR with the help of the clinic receptionist had access to a current list of eligible women. In other communities, especially the smaller communities, recruitment was more by "word-of-mouth" as it was usually common knowledge who was pregnant or who had just delivered.

The MI interventions were also delivered in a variety of settings; usually in whatever space was available in the health clinic or the adjacent trailers that were often present to provide extra clinic space. It is important that note is made of the challenges of "space availability" for research projects in remote communities. Often the project staff would arrive but would have to take whatever space or room was available depending on what other programs were scheduled during the day. The project did not have an assigned space, but just had to "make the best of it." This problem of battling for space with other programs is a chronic problem for research projects in remote communities. A private space for the counseling intervention was the ideal but simply was not always possible. Furthermore, often when the project staff arrived, no suitable overnight lodging was even available.

The same challenge existed when the outcomes assessments were scheduled. These assessments were done in the health clinic or the adjacent trailer. Home visits, though infrequent, were sometimes done. Many of the children had their dental exam when they were at the Day Care Centre and their mothers were followed up to answer the questionnaires either in person or on the telephone at a later date.

5. Study intervention: Motivational Interviewing (MI)

Patient-centred, personalized approaches that avoid direct persuasion have been shown to produce good results in promotion of behaviour change [22] [23] The intervention in this trial followed the principles of Motivational Interviewing or MI [24], a client-centred but directive counseling style. With this approach, the motivation for change comes from the client, but the

counselor helps create, by questioning and reflection, the expectation of change. Feedback and advice are offered within the context of acknowledgement of the client's right to choose. Many possible paths to a solution are provided. Client and counselor agree upon a menu of effective behaviours. This strategy fits well with the philosophy of the Cree who are more comfortable if someone suggests ways to think of taking a different approach rather than tells them directly how to act. It also fits well with a recommendation from a gestational diabetes program in Eeyou Istchee that, for an intervention to be effective, women should be given the opportunity "to reflect on their roles as women and mothers, caregivers and providers" [25].

MI has been successful in the management of addictive behaviours such as smoking and non-addictive behaviours associated with conditions like diabetes [22, 23]. Furthermore, brief MI interventions have produced good results [26]. MI has been previously successfully applied in a trial to reduce ECC in 6-18 month old IndoCanadian children in western Canada [27]. At the end of the 2-year trial period, the MI children had a 46% lower rate of tooth surfaces affected by caries than did control children.

In this project called "I wish my child would have beautiful teeth" or, in the Cree language, *Kimaa Miywaapitet Nitawaashiim* experimental group mothers were scheduled to have an MI session during pregnancy and several more MI sessions until their child was around two years of age. Mothers received appropriate resources at each MI visit to enable them to implement selected behaviours (infant toothbrushes, toothpaste, sippy cups.) Fluoride varnish was offered after the age of one-year.

The control group mothers received a culturally-appropriate educational pamphlet describing healthy dental care practices for young children. Pamphlets were mailed to mothers when their child was 6 months of age and again at 18 months of age. The pamphlet titled "Protect Baby Teeth: Circle of Smiles" had been previously produced in 2000 by the Nursing Caries Committee of the St. Theresa Point First Nation of Manitoba, Canada and is available from them on request [28]. Fluoride varnish was available to control children at local dental clinics.

A detailed two-day training workshop for CHRs in the 5 intervention or "test" communities was held within the first year of the project. The project's MI consultant provided a template for the MI script and menu that would be used in the counseling sessions with the mothers. By engaging the CHRs in discussion and critique, both the script and the menus were extensively modified to suit the language and style of the Cree. Following the workshop, the menus were finalized, printed on flipcharts and sent with explanatory notes to each CHR in each of the test communities. The MI consultant followed up some months later with an "MI-coaching conference call" to problem-solve MI with the CHRs. A second follow-up workshop was held the next year. Following this workshop, the PM visited each of the communities individually to problem-solve recruiting and MI challenges.

The MI scripts were based on the work of Weinstein [29] and on scripts developed for a previous trial [27, 30, 31]. One script was created for pregnant and new mothers (Appendix 1) and another slightly modified script was developed for mothers after their infant's first tooth had erupted until their child was about 2-years of age. An important aspect of improving infant oral health is enhancing the oral health of the mother to prevent transmission of cavity-causing bacteria from

mother to infant. With this in mind, pregnant and new mothers were given "Privilege Cards" that allowed them expedited dental services at their community's dental clinic.

6a: Outcomes

Questionnaires on oral health practices

Women enrolled in the study completed instruments that included items on demographics, their personal oral hygiene practices and dental knowledge. An instrument called the Readiness Assessment of Future Parents concerning Infant Dental Decay or RAFPIDD was also completed. This 47-item instrument was a modification of a validated instrument developed by Weinstein and Riedy to assess a mother's stage of change with regard to her child's dental health [32]. The instrument was enhanced with items specific to Cree mothers and pre-tested for internal consistency with pregnant women in Eeyou Istchee.

For women in the test communities, information about their MI interventions was recorded at each visit and what transpired during the MI visit was noted, for example, length of time for the MI, specific resources given, application of fluoride varnish.

Child dental health and dental health behaviours at end-point

Clinical data was collected by a dental examination of each participating child when he/she was at least 30 months of age. Because women were recruited for the project over a 2½ year period as they became pregnant or had recently delivered, the assessments of the children have had to be done over a similarly extended period. The assessments began in April 2008 and were finally completed in November 2010.

Three calibrated dental hygienist examiners who had no association to the trial did the dental assessments. The hygienist examiners were masked to the community's treatment assignment. The examiners were calibrated in the use of the indices at Eeyou Istchee health clinics by a gold standard dentist examiner. Radiographs were not used. The clinical detection of caries involved visual/tactile examinations with explorers (to remove plaque), front surface mirrors, cotton rolls and a dental flashlight. Criteria for caries detection were those described by Pitts [33]. Initial caries (color change but no substance loss), enamel caries (substance loss), dental caries, pulpal caries, restorations and absence of a tooth due to caries or for other reasons were recorded. Data on types of dental treatment (for example, restorations, extractions) that a child had received in the dental clinic was also recorded. The examinations were carried out with the child in a supine position or, for very young children, on the lap of the caregiver in the knee-to-knee position with the dental examiner.

Information about dental health knowledge, home-care behaviours and caries-related child quality of life was also collected from the parent by questionnaires administered at the assessment visit. These instruments were administered by the recorder who was assisting the examiner; the recorder was usually either the Project Manager or one of the DHRs hired for the project. For children who were examined in a day care setting where the parent may not have been present, the questionnaires were completed by the parent at a later visit, during a home visit or by telephone. The survey questions had been validated in a previous survey of 301 children undertaken in community health clinics in Quebec [15].

Economic evaluation
The objectives of the economic evaluation (see Appendix 2 for report) will be to estimate the net incremental cost of the intervention strategy and to estimate an incremental cost-effectiveness ratio. Estimation of the net incremental cost of the program will involve a comparison of costs of dental resources utilized by each arm of the study. The cost analysis will involve a study of the time and materials required to carry out each of the intervention visits. Salary costs for the project staff will be assessed based on their compensation rate and disposable materials will be valued at acquisition costs. Value of the mother's time to take her child to the clinic for well-baby care and for MI will be determined by age and gender-matched wages and subjected to sensitivity analyses. If the intervention is effective, children in the intervention arm should have lower rates of use of dental-related health services during the period of the trial. Information concerning types, amount and cost of care received as well as the time lost by caregivers in pursuing this care will be collected. The net cost of the program, then, will include the intervention costs minus any savings in dental services. The incremental cost-effectiveness ratio at follow-up will be based on these net incremental costs and the difference in the primary outcome measure, number of decayed, extracted or filled primary tooth surfaces.

6b: Changes to trial outcomes
The original study protocol aimed to do follow-up clinical examinations and assessments of oral health behaviours when each child was around 30 months of age. Delays in reaching parents to attend for an outcomes assessment, missed appointments, challenges that occurred in the "life" of the community and the imposed hiatus [as described in 7b] in completion of assessments resulted in children being older than 30 months of age at outcomes assessments. However, mean child age at outcomes, though greater than the original protocol, was no different between test and control children.

7a: Sample size
The target sample size of 309 mothers was determined based on the need to have high power to detect a 20% reduction in caries prevalence. This magnitude of reduction was similar to that observed in a community-based oral health promotion program implemented in Native American villages that demonstrated a 25% decrease in "baby bottle tooth decay" in participating communities after 3 years [18].

The power was calculated using the method of adjustment for intra-class correlation by the variance inflation factor [34] applied to the standard formula for calculating power for comparison of proportions from independent samples. The intra-class correlation coefficient was estimated to be 0.0090 by applying the analysis of variance method to the preliminary data on caries prevalence by village, which produced a variance inflation factor of 1.35. The control-group caries prevalence was estimated to be 0.86 using a weighted average of these individual prevalences weighted by sample size per village. Based on the above projections, a total sample size of 265 mother and child pairs would yield power of 82% to detect a 20% reduction in caries from 0.86 to 0.69.

A reported infant mortality rate of 15 deaths per 1000 births was considered [35]. Overall loss to follow-up, including mortality, was estimated to be 5% per year for the 3 years that mother and baby were in the study. Thus, the sample size was determined to be 309 mother and child pairs.

Based on a 75% participation rate, 412 births were required to achieve a sample size of 309. Other researchers working in Eeyou Istchee with new mothers have reported an 80% participation rate which was similar to our expectations. The anticipated number of children enrolled in each community was estimated using the birth data from 2001 and a 75% participation rate in each village, for a total sample size of 258 accrued per year. Thus, it was estimated that 14 months would be required to enroll 309 mothers.

7b: Interim analyses

No interim analyses were performed because it is considered extremely unlikely for there to be evidence of a treatment effect prior to collection of the final outcome data on the entire sample. It is worthy of note that due to changes in territorial governance and the new administration's desire to review all research that was underway in Eeyou Istchee, the outcomes assessments were unexpectedly curtailed for an 8-month period from October 2008 to July 2009. Because of this hiatus, the original calibrated dental hygienist examiners took on other employment. A new hygienist was recruited and calibrated by Dr. Veronneau to the examination protocol in February 2010.

8a, b and 9: Randomization

The advantages and disadvantages of testing the intervention as a randomized controlled trial were discussed and debated at length during a 2-year community consultation process. Those who participated in the discussions during the consultation phase were reassured that the randomization process would be open, impartial and unbiased. Therefore, the randomization was done publicly during a daytime broadcast over community radio. The territory's community radio station with its extensive listening audience was an accepted and trusted way of conveying local news and information in Eeyou Istchee. A community radio broadcast of the randomization process was well-suited to the remoteness of Eeyou Istchee and the distances between communities.

There were two "rounds" of a constrained randomization process: one round for the 2 larger communities and a second round for the 7 smaller communities. Two baskets had been prepared for the "on-air" randomization: one basket for the large communities and another basket for the smaller communities. Each basket contained envelopes marked "test" or control"; the larger community's basket contained 2 envelopes (1 test; 1 control) and the smaller community's basket contained 7 envelopes (4 test, 3 control). Communities were randomized in each round by alphabetically ordering the communities' names. For example, for each round the first name on the alphabetical list of communities was announced followed by the drawing of an envelope from the basket; the next name was announced followed by another draw until all envelopes were allocated. The draw was done "live" on afternoon radio by a radio station employee who was not associated with the research. Of the 9 communities, 5 were allocated to the test condition and 4 to the control condition. The decision was made to allocate one more test than control community to allow a more robust exploration of intervention effects e.g. analyses according to number of MI sessions attended. Communities were not aware of their allocation until their name was drawn. No concerns about the randomization process have been expressed by any community. Randomization was done prior to recruitment of subjects.

10: Participant enrolment

For a variety of reasons, CHRs in many of the communities were challenged by the process of subject recruitment and delivering the intervention. The negative side of engaging these existing personnel in a research project was their sense of being overworked and not sufficiently rewarded for this addition to their daily workload. This project was yet another burden for the already busy and in-demand CHRs. Furthermore, the CHRs may have found that expectant women and new mothers were too busy to spend an additional amount of time completing the project's many documents or were simply not interested in participating in a research project. Even though the project investigators were diligent in following recommended principles of involving Aboriginal communities in research, recruitment of participants was a struggle. Thus, in 2 of the 5 test communities and in 2 of the 4 control communities, recruitment and, for the test communities, delivery of the intervention was eventually completed by the Project Manager. One of the project-specific DHRs achieved success in one test-community and eventually took over the work of a retiring CHR in another test-community. A CHR, aided by the Project Manager, completed and continued the project's work in the 5th test community. Local women who were not CHRs completed recruitment in the remaining 2 control-communities. Recruitment was closed after 2½ years when the number of mothers recruited, though short of the original goal, was sufficient to maintain the power of the trial.

11: Blinding to interventions

Given that the intervention was a type of behavioural counseling, the mothers and the interveners (the CHR and the Project Manager) were aware of their community's allocation. The dental hygienists who did the clinical examinations at follow-up were kept blinded to the allocation and were completely unfamiliar with the details of the intervention. In addition, the biostatistician and the health economists were blinded to allocations.

12a, b: Statistical analysis

Primary statistical analysis

The primary statistical analysis was a comparison of caries prevalence in intervention and control groups, using a permutation test [36] with test statistic equal to the difference between caries prevalences in the two groups. A significance level was determined using the exact permutation distribution of the test statistic, which was computed by enumerating all possible random assignments of communities to intervention or control conditions according to the randomization scheme. The use of the permutation test accounts for intra-class correlation between outcomes on children in the same community and also addresses concerns over the small-sample performance of statistical methods such as Generalized Estimating Equations [37], [38, 39]which rely on asymptotic theory. An exact confidence interval for the treatment effect (difference in caries prevalences) was computed using the usual procedure for inverting the permutation test. Peterson et al provide an illustration of the application of this procedure to a group-randomized trial [40].

The main analysis was not able to be initiated until the end of the study (December 2010) when all outcomes data was collected and entered. However, a preliminary frequency analysis of the data helped the investigators to develop an understanding of the characteristics of the sample.

Subgroup analyses
Within the experimental communities, outcomes were compared looking at factors such as total number of MI sessions attended and individual community.

Results
13: Participant flow (see Flow Chart, Figure 1)
For test and control groups, the random allocation of communities, the number of mothers who received an MI intervention and the number of mother-child pairs who were assessed at outcomes are shown in the flow chart. Subjects who moved away, withdrew or were lost to follow-up are also presented.

The average annual birth rate in the Cree territories is about 335 live births per year. Based on this birthrate, about 840 infants were born during the time of the trial. The mothers recruited represent about one-third of the potential subjects.

14a: Periods of recruitment and follow-up
Two hundred and seventy-two mothers (131 test, 141 control) were recruited over a 2½ year period from January 2005 to October 2007. Because of the cluster-randomized design of the trial, the power of the trial was determined not only by the total sample size but also by the number of communities and by having sufficient numbers of subjects recruited in each of the communities. Whereas the original projected sample size of 309 women would have yielded 82% power to detect a 20% reduction in caries prevalence from 0.86 to 0.69, power calculations using the actual sample sizes show that the resulting power was 79% to detect this treatment effect. These calculations assumed a loss to follow-up of 5% per year. By maintaining an attrition fraction of 4% per year, the power will be 80%. Power will be very high (>90%) to detect a 25% reduction in caries prevalence.

Six months after the follow-up assessments began in April 2008, an 8-month interruption in data gathering ensued (previously described in 7b). As a result of intensive lobbying and strategizing by the project's Principal Investigators and the Research Director of the CBHSSJB, the assessments resumed in July 2009 and were completed in November 2010.

15: Comparison of treatment groups on baseline characteristics (Table 1)
Demographic and behavioral characteristics at baseline were compared between groups in order to assess the success of randomization in producing comparable treatment groups (Table 1). The distributions of most variables was very similar for the two randomized groups of mothers, specifically for mother's mean age (25.5 for Test vs. 25.7 for Control), stage of pregnancy for those who had not yet delivered (22.9 weeks vs. 24.0 weeks), age of child for those who had delivered (6.8 vs. 6.8 weeks), mean knowledge score (3.1 vs. 2.9), percentage with no other children (35.7% vs. 34.3%), toothbrushing (92.3% vs. 92.2%), and seeing the dentist in the prior two years (71.5% vs. 73.8%). However, despite the random assignment of communities to treatment conditions, group differences existed for percentage of mothers who had already delivered at the time of enrollment (19.2% vs. 40.0%), prior tooth extractions for other children in the family among those women with other children (34.1 vs. 48.9%), and the reason for prior visit to the dentist being for toothache among mothers who had a dental visit (35.5 vs. 50.0%).

Testing of differences between randomized groups on baseline variables is not particularly informative, because any differences found must by definition be type I errors. In fact, a difference that could be deemed clinically important may result within the range of normal sampling variability in the cluster-randomized setting. Furthermore, such a difference might be less likely to occur in an individual-randomized design with the same number of participants. Statistical significance aside, what matters is that the group differences (particularly for tooth extraction) may be large enough to be associated with clinically meaningful differences in health outcomes at follow-up in the enrolled children. Therefore, secondary analyses of outcomes will be done with regression adjustment to control differences in whether or not the mother had delivered already, prior tooth extraction for other children, and the reason for prior dental visit being toothache. An unadjusted analysis will be performed as the primary analysis; the results of the adjusted and unadjusted analyses will be compared to determine the impact of the baseline group differences on the results.

16, 17: Results: primary and secondary outcomes and effect size

Test group mothers were to have a total of 6 MI sessions, beginning in pregnancy and regularly thereafter at immunization or well-child clinic visits until the child's 2nd birthday. The actual mean (SD) number of MI interventions per test mother was 2.7 (1.7), range 0-6. Of the 131 test mothers, 93 (71%) had 2 or more MI sessions and 46 (35%) had 4 or more interventions usually of 15-30 minutes duration. Home visits were offered, but overall fewer than 10% of the interventions were delivered at home. During pregnancy 76% (81/106) of mothers had an MI intervention, but by the child's second birthday only (33%) 43/131 of mothers attended for an intervention. All mothers received appropriate resources at each visit to enable implementation of selected behaviours (infant toothbrushes, fluoride toothpaste, sippy cups). As per the trial protocol, control mothers were mailed the child dental health pamphlets when their infant was 6 and 18 months

Behavioral outcomes were similar between the two treatment groups (Table 2). A good proportion of mothers in both groups completed the behavioural outcomes questionnaire with the proportion being slightly higher in the control group than the test group (94 vs. 87%). The average age of the children at assessment was similar. Children in the test group were more likely to be assessed in the health clinic than children in the control group (44.1 vs. 26.9%). A large proportion of parents in both test (95.5%) and control (93.8%) communities reported cleaning their child's teeth at least once a day with fluoride toothpaste. Overall, most behavioral outcomes were similar for the two groups with no overt indication that the intervention had a large effect on dental health-related behaviors. Given the obvious similarities for the behavioural outcomes, p-values were not generated.

Given that attendance at a dental office is an important component of maintaining good dental health, parents were asked about their child's visits to the local dental clinic (Table 3). More control community parents (51.9%) than test parents (38.9%) reported dental clinic visit for their child. Treatment at the clinic, as reported by parents, did not differ between test and control children. Parents were queried about how toothache or tooth pain may have affected their child's quality of life. More control parents than test parents replied that dental pain had affected their child adversely and more control parents reported taking their child to the dentist for tooth pain.

The average number of pain-related dental visits per control child was 32 visits/131 children or 0.24 visits per child compared to 17/110 or 0.15 visits per test child.

The primary outcome was the presence of untreated and treated caries at follow-up (Table 4). Caries was recorded for each surface of each tooth present if it was in enamel (non-cavitated or cavitated), in dentin, into the pulp or had been treated with a filling or by extraction. Similar to the behavioral outcomes assessment, children in the test group were more likely to have the dental exam in a health clinic than children in the control group (42.9 vs. 26.0%). The average age of the dental exam was similar for the two groups (35.0 vs. 35.4 months).

Caries prevalence was extremely high; almost all children had evidence of caries (Table 4). The prevalence of caries at the initial enamel level or greater was similar in the two treatment groups (96 vs. 98%). When levels of caries of greater severity were examined, an apparent treatment effect emerged. Percentages of children with caries (untreated or treated) at the level of the dentin or worse were 65% in the control group and only 40% in the test group, which implies an estimated 38% reduction in disease prevalence. However, the assessment of statistical significance of this effect was inconclusive. Results were compared for Generalized Estimating Equations (GEE) analysis as well as using a bias-corrected version of the GEE that makes adjustments to account for small sample sizes, as well as the permutation test as originally proposed. These results suggest that the evidence for a treatment effect on caries into the dentin or worse is weak. Similar large reductions in disease prevalence were observed when disease was measured as occurrence of a filling or extraction (19% vs. 9%, 52% reduction in risk). As before, the statistical significance of this treatment effect is marginal.

Additional analyses were performed using surface-level and tooth-level outcomes. These results suggest that the intervention may reduce the extent of disease. For example, although almost all children in both groups had disease, there was observed a 30% reduction in the average number of diseased surfaces per child (21.5 vs. 14.6, ratio 0.7). Even larger reductions were observed for more severe levels of caries. As for the previously stated outcomes, there is no strong evidence for the existence of a treatment effect. Results for tooth level outcomes gave similar results although the percentage reductions in the test group were slightly smaller than for surface-level outcomes.

It is our intent to do further analysis focused on the extent of *untreated* tooth decay in young Cree children; as yet only a cursory analysis of this variable has been done. Initial caries into the enamel which is defined as “color change but no loss of (enamel) substance” usually is not restored although it can either progress or “reverse”. Thus, those children with either only initial enamel caries or teeth treated with fillings and extractions were not counted so that the proportion of children in test and control communities with either cavitated enamel, dentinal or pulpal caries could be determined. The proportion of children with untreated decay (at the level of enamel cavitation or worse) in both groups was similar: 57.3% and 56.3%. However, for untreated, irreversible caries that had extended into the dentin, the proportion for control communities was (53/131) 40.4% compared to a proportion in the test communities of (34/110) 30.9%. These results will be further tested and analyzed for surface and tooth level outcomes. However, what is apparent is the unfortunate numbers of young Cree children are living with untreated, extensive decay. Given the wait lists at the dental clinics and the challenges of

managing the behaviour of young children during dental treatment, this observation is not surprising.

18: Results: Subgroup and adjusted analyses

Results by community (Table 5) suggest that the intervention may have had a caries preventive effect in all but one of the test communities (Test #5). This community had disease rates that were higher than those of the other test communities and similar to those of the control communities. Furthermore, test community #5 did not administer the 2nd and 3rd interventions to any of the participants. These results provide additional evidence suggestive of an intervention effect. However, these findings could not be demonstrated definitively in this study due to the large community-to-community variation in the administration of the intervention and in the outcomes.

Additional analyses that examine treatment effects within subgroups based on demographic or other characteristics as well as analyses that provide adjustment for baseline covariates and account for intervention delivery remain to be done prior to submission of an article for publication. These results will have to be interpreted cautiously due to the chance for false positive findings when performing multiple hypothesis tests.

19: Unintended effects:

No adverse events or side effects of any aspect of the intervention, including the fluoride varnish applications, were reported by caregivers.

20: Discussion

This project with Cree mothers in northern Quebec aimed to evaluate the effectiveness of a specific behavioural intervention, Motivational Interviewing, to decrease the prevalence of early childhood caries in their children. The implementation of the trial in these remote communities, the problems of enlisting dedicated project staff and the unexpected changes in philosophies about research by the territory's governing body heightened the project's challenges. Overall, the results do not provide compelling evidence for a caries preventive effect of the MI intervention in these Cree communities. Certainly, the overall prevalence of caries in both treatment groups remained alarmingly high. However, there are suggestions that the intervention may have an impact on the extent or severity of disease. These results are not conclusive due to effects of clustering of children within communities and the small number of communities. Undoubtedly our results will be useful for generating specific hypotheses that can be tested in future trials and for informing protocols, approaches and methods in future randomized controlled trials in high-risk and difficult to access communities.

One of the first steps in launching the project was staff engagement ("buy-in"). Because of the decision to enlist existing staff (CHRs) as recruiters and interveners in the project, job-postings and hiring did not slow down the start of the project. However, 3 of the 9 communities did not have a CHR on staff at the project's start; therefore, local women (as per the original proposal) were hired to begin recruitment.

In order for research trials to be implemented within an organization's existing structure and framework, administrators may request that existing personnel work on the project. However,

these individuals may simply not have the time, the personal resources or the interest to participate, despite the provision of extensive in-service training and ongoing support. Therefore, any clinical trial that is implemented within an existing organizational framework must have the appropriate human and financial resources that can be easily redistributed to ensure continuation of the project. From the beginning of the project and throughout, the Project Manager visited the communities and met with the project's staff to problem-solve challenges and review procedures. She also maintained regular telephone contact with the staff in each community who were working on the project. Despite her ongoing contact and other support from the investigators, there was large community-to-community variation in the administration of the intervention which had an effect on the outcomes.

The observed differences between test and control communities in the locations where the behavioural and clinical outcomes assessments were done underscore the logistical challenges of undertaking controlled trials in remote communities. This issue was also a challenge in the test communities for project staff trying to administer the counseling intervention. Office and clinic space is limited and constantly overbooked in remote First Nations communities. Simply finding a private space for doing the MI interventions, the follow-up interviews and the dental examinations on rambunctious toddlers was an ongoing challenge. Follow-up was often in a different venue every time the project staff came to do assessments. Because of the changes in venues from visit to visit, sometime families attempted to come for a follow-up appointment, but mistakenly went to the wrong location.

While the intervention did not appear to have a striking effect on oral health behaviours, as reported by parents, some findings warrant further discussion. Knowledge about dental health matters was similar across communities as were reports by parents of regularly brushing their child's teeth. Providing snacks during the day for young children was common practice but test moms "repertoire" of snacks appeared to be the more "dentally-healthy": milk, cheese and crackers. The Northern stores in all of the 9 communities stocked similar grocery items, so awareness of healthy snacks rather than availability likely explains the differences. The proportion of children who went to sleep with a bottle was also less in the test communities.

Given the fact of the vertical transmission of cavity-causing bacteria from mother to child, we gave pregnant and new mothers in test communities "Privilege Cards" that would allow them to go to the "top of the wait list" at the local dental clinic. Indeed, 33/103 or 32% of test mothers remembered using the Privilege Cards. Unfortunately because of the turnover of dentists and dental reception staff in the communities not all dental clinics honoured the cards. Regular, ongoing "in-person" visits to providers by research project staff are essential so that all health care professionals, especially those newly employed, know about an ongoing research project and understand their contribution to its success. It should be noted that a series of newsletters were circulated by the Project Manager to keep communities abreast of progress.

The reports from parents of taking their child for a dental visit and the treatment received (Table 3) were corroborated by the restorations ("fillings") and extractions that were noted at the outcomes dental examinations. Control children had evidence of having had more dental treatment than test children. An increased prevalence of extractions in control children at outcomes was also what mothers reported at baseline, i.e. more control mothers had taken their

other children to a dentist for an extraction. All communities have a dental clinic in their community; whether access for treatment for young children differs amongst communities is not known but will be explored further. This increased amount of treatment likely parallels a greater need for treatment by control children.

When caries extends beyond the dentin and into the pulp of the tooth, the resulting discomfort and difficulty eating may affect a child's quality of life. Overall, fewer test than control children were reported by their parents to have lost sleep, cried, missed preschool or seen a dentist because of pain. These differences were not significant. Thus, the effect of the intervention on quality of life was in a positive direction, but thus far inconclusive. Similarly inconclusive, but displaying a positive trend, was the effect of the intervention on severity of caries (Table 4). Further evidence of the intervention's contribution to the "control" of caries in young childre is evident in the results by communities. In the challenging test community #5, the staff only managed to deliver an intervention to 8/18 or 40% of mothers and failed to deliver the intervention at the 2-4 month or 6-9 month visits (about the time of eruption of the first primary tooth). Not surprisingly, this community's disease rates were similar to those of the control communities.

Given the small number of communities and the variation amongst communities in delivering the interventions, it is not surprising that individual components of the results are not overwhelmingly supportive of a positive effect of MI intervention in controlling caries. However, the positive nature of the trends observed in the test communities, e.g. fewer child quality of life disruptions and less invasive caries suggests that MI has a place in the planning of interventions to control caries in First Nations children. Slowing down the process of caries is indeed a positive outcome as the possibility of delivering appropriate and needed dental treatment combined with a positive dental experience increases as a child matures.

We are doing further analyses that examine treatment effects within subgroups based on demographics or other characteristics as well as analyses that provide adjustment for baseline covariates and account for the differences in intervention delivery. These further analyses will provide helpful information regarding the generalizability of our results.

23: Trial registration

This trial is registered as ISRCTN41467632.

24: Funding

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List of tables, figures and appendices

Figure 1 – Flow diagram of the progress of the communities and of the participants from Eeyou Istchee through the phases of the randomized controlled trial.

Table 1 – Pre-randomization characteristic of study sample mothers by treatment group

Table 2 – Behavioural outcome assessments by treatment group

Table 3 - Dental treatment and caries-related child quality of life (QofL) by treatment group

Table 4 – Caries incidence by treatment group

Table 5 – Intervention delivery and outcomes by community.

Appendix 1 - Motivational Interviewing Script

Appendix 2 – Cost-effective analysis of a perinatal Motivational interviewing program for the prevention of early childhood caries in Cree communities Quebec

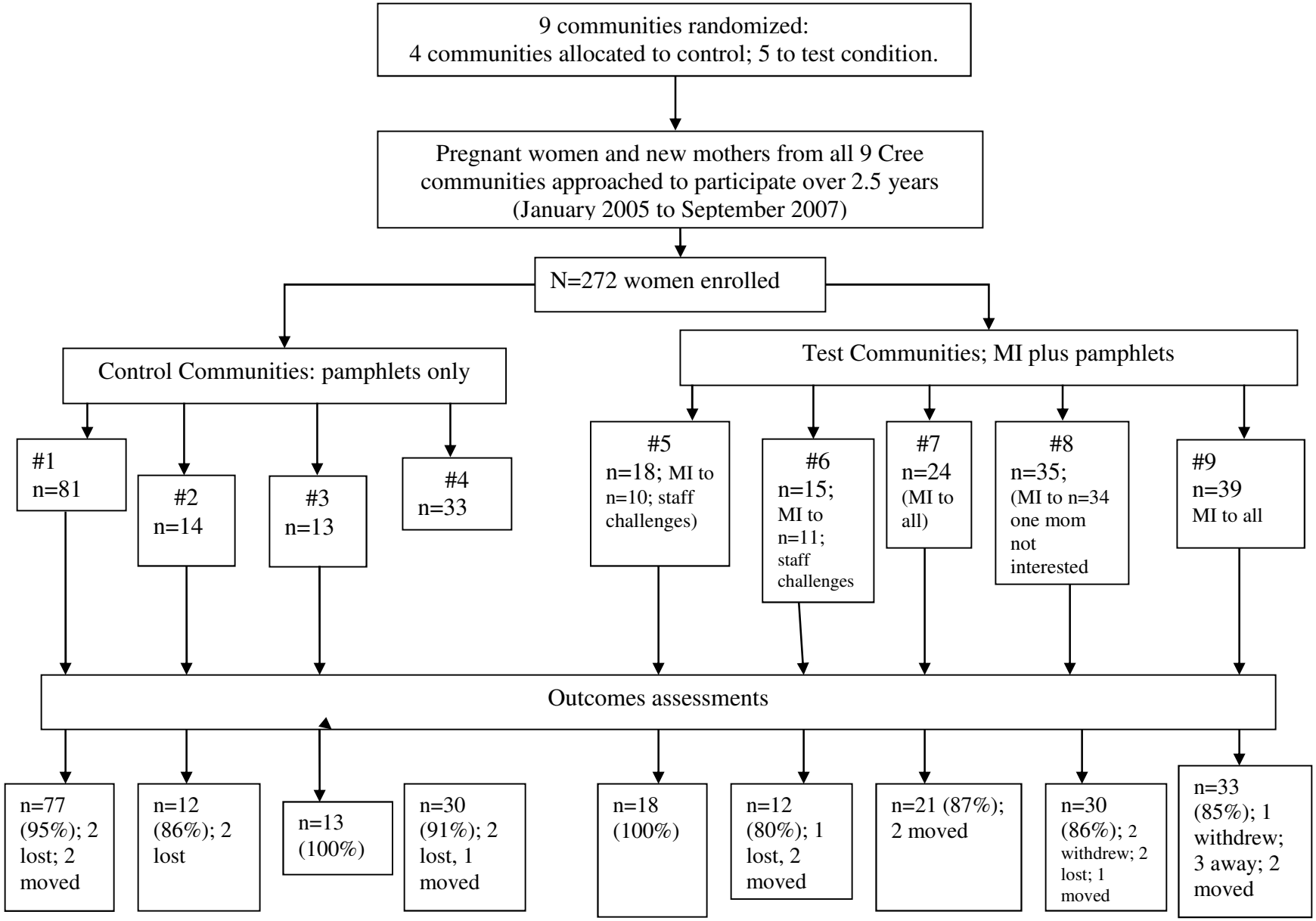

Figure 1. Participant flow-chart

Table 1. Pre-randomization characteristics of study sample mothers by treatment group. Results shown are mean (SD), minimum – maximum for quantitative variables, and number (%) positive for binary variables.

	Test		Control	
Variable	**N**		**N**	
Age (years)	130	25.5 (6.4), 15 – 44	140	25.7 (5.8), 15 – 39
Delivered already	25	(19.2%)	56	(40%)
Stage of pregnancy for those who had not delivered (weeks)	105	22.9 (9.0), 6 – 40	84	24.0 (10.0), 6 – 40
Age of child for those who had delivered (weeks)	18	6.8(7.1),0.4-29.7	51	6.8(6.4),0.0-26.0
Knowledge score	129	3.1 (1.2), 0 – 5	141	2.9 (1.4), 0 – 5
Other children's dental health				
No other children	129	46 (35.7%)	140	48 (34.3%)
Other child had prior tooth extraction*	82	28 (34.1%)	92	45 (48.9%)
Other child had prior fillings or caps*	82	47(57.3%)	92	57(62.0%)
Other child stayed in the hospital*	83	15(18.1%)	92	18(19.6%)
Brushes with fluoride toothpaste	130	120 (92.3%)	141	130 (92.2%)
Saw dentist < 2 years ago	130	93 (71.5%)	141	104 (73.8%)
Reason for visit: toothache, problems	93	33(35.5%)	104	52(50.0%)

* Only responses of mothers with other children are included

Table 2. Behavioral outcome assessments by treatment group

Variable	Test N (%)	Control N (%)
Child age in years: mean(SD)	3.1(0.9),1.2-5.5	3.0(0.7),1.8-4.8
Parent's assessment of child's dental health		
Excellent to very good	29(26.4%)	31(24.4%)
Good or fair	63(57.3%)	77(60.6%)
Poor	18(16.4%)	19(15.0%)
Dental knowledge assessment (correct response)		
What causes tooth decay	66(59.5%)	76(59.4%)
Cavity-causing bacteria transfer	42(37.8%)	36(27.9%)
Tooth decay prevention	80(72.1%)	89(70.1%)
When to start cleaning child's teeth	102(91.9%)	115(89.1%)
Do cavities hurt	81(73.0%)	98(76.0%)
Baby teeth contribute to adult teeth	72(65.5%)	75(58.1%)
Mother chews gum		
At least once a day	47(43.9%)	58(45.4%)
Rarely	60(56.1%)	70(54.7%)
Type of child's regular snack (multiple responses allowed)		
Milk	71(65.7%)	67(51.1%)
Fruit drink	54(50.0%)	55(42.0%)
Cheezies or chips	20(18.5%)	7(5.3%)
Cheese	61(56.5%)	58(44.3%)
Crackers	49(45.4%)	46(35.1%)
Child sleeps with bottle (among those using bottle)	14/21(67%)	9/10(90%)
Age bottle stopped		
Never had a bottle	7(8.1%)	14(12.1%)
1 year or less	41(47.6%)	61(52.6%)
1-2 years	35(40.7%)	38((32.3%)
Older than 2 years	4(4.7%)	3(2.6%)
Back to sleep		
Cuddle and talk	30(27.8%)	38(29.0%)
Juice	6(5.6%)	11(8.4%)
Toy	3(2.8%)	2(1.5%)
Water	11(10.2%)	8(6.1%)
Bring into bed	15(13.9%)	13(9.9%)
Milk	29(26.9%)	24(18.3%)
Other	12(11.1%)	15(11.5%)

Table 3. Dental treatment and caries-related child quality-of-life (QoL) by treatment group

Variable	Test (N=110)	Control (N=131)
Dental treatment reported by parent		
Clinic visit	42(38.9%)	68(51.9%)
Treatment received at clinic:		
"Yellow" fluoride varnish	44*(40.1%)	20(29.4%)
Check-up for cavities	35(31.8%)	52(40.0%)
Teeth cleaning	24(21.8%)	28(21.4%)
Filling or cap	7(6.4%)	12(9.2%)
Tooth pulled	6(5.5%)	10(7.6%)
Other	11(10.0%)	5(3.8%)
"Quality of life (QoL)" questions		
Answered "yes" to ≥1 QoL question below	21(19.4%)	37(28.2%)
1. Had tooth pain/toothache	21(18.9%)	26(20.2%)
2. Lost sleep due to tooth pain	17(15.3%)	18(14.1%)
3. Cried due to tooth pain	10(9.0%)	7(5.4%)
4. Missed preschool due to tooth pain	5(5%)	3(2%)
5. Seen a dentist due to tooth pain	13(11.7%)	22(17.5%)
Number of visits to dentist for tooth pain**	17	32
Had fillings or caps	7(6%)	19(15%)
Had a tooth pulled	7(6%)	13(10.1%)
Given medicine while teeth being fixed	5(5%)	10(8%)
Gone "south" for a general anesthetic	3(3%)	3(2%)

* Note: 2 parents said their child received varnish but child not recorded as having a clinic visit. Varnish actually had been administered as part of MI intervention

** Number of times a parent said their child went to the dentist because of tooth pain. Some children had more than one visit.

Table 4. Caries incidence by treatment group

Variable	Test (N=110)	Control (N=131)	RR (95% CI)***	GEE P-values (two-sided)	Bias-corrected GEE P-values (two-sided)	Permutation P-values (one-sided)
Age in years, mean(SD) 0.7 - 5.8 1.8 - 4.7	3.0(0.9)	2.9(0.7)				
Binary Outcomes ((N (%))						
Any caries: enamel (initial or cavitated) dentinal, pulpal; filling, or extraction	100(98%)	119 (96%)	1.03 (0.99, 1.06)	0.067	0.436	0.957
Enamel cavitated, dentinal, pulpal caries; filling or extraction	72 (84%)	100 (86%)	0.95 (0.87, 1.08)	0.253	0.427	0.285
Dentinal, pulpal caries; filling or extraction	44 (40%)	85 (65%)	0.62 (0.47, 0.81)	0.022	0.087	0.071
Pulpal caries; filling or extraction	20 (18%)	32 (24%)	0.83 (0.43, 1.39)	0.487	0.600	0.286
Filling or extraction	10 (9%)	25 (19%)	0.48 (0.27, 0.84)	0.003	0.052	0.057
Extraction	5 (5%)	14 (11%)	0.35 (0.19, 0.76)	0.008	0.100	0.157
Surface Outcomes (mean (SD))						
Surfaces with any caries	14.6(15.7)	21.5(20.6)	0.70(0.52,0.95)	0.022	0.090	0.114
Surfaces with enamel cavitated, dentinal, pulpal caries; filling, or extraction	7.7(11.8)	13.3(16.2)	0.65(0.38,1.10)	0.105	0.212	0.156
Surfaces with dentinal, pulpal caries; filling, or extraction	4.9(9.4)	9.5 (14.5)	0.58(0.32,1.08)	0.087	0.193	0.157
Surfaces with pulpal caries; filling, or extraction	1.5(5.6)	3.5(10.4)	0.48(0.17,1.33)	0.156	0.300	0.200
Surfaces with filling or extraction	1.0(5.0)	3.0(10.0)	0.27(0.10,0.73)	0.010	0.075	0.100
Surfaces extracted	0.5(2.8)	1.5(5.5)	0.27(0.11,0.66)	0.004	0.115	0.228
Tooth Outcomes (mean (SD))						
Teeth with any caries	8.0(5.3)	9.4(5.8)	0.84(0.79,0.90)	0.000	0.040	0.100
Teeth with enamel cavitated, dentinal or pulpal caries; filling, or extraction	4.1(4.4)	6.3(5.4)	0.65(0.47,0.89)	0.009	0.042	0.071
Teeth with dentinal or pulpal caries; filling or extraction	2.3(3.4)	4.0(4.7)	0.56(0.38,0.84)	0.004	0.031	0.043
Teeth with pulpal caries; filling, or extraction	0.5(1.4)	1.1(2.5)	0.47(0.22,1.01)	0.052	0.154	0.143
Teeth with filling or extraction	0.3(1.1)	0.8(2.3)	0.31(0.15,0.65)	0.002	0.025	0.071
Teeth extracted	0.1(0.6)	0.3(1.1)	0.26(0.11,0.62)	0.002	0.104	0.214

*** Risk ratio (RR) is preferable to odds ratio (OR) because it gives an interpretation in terms of percentage reduction in risk, eg, a RR of 0.85 means that the intervention is associated with a 15% reduction in risk.

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Table 5. Intervention delivery and outcomes by community.

Variable	Control#1 (N=81)	Control#2 (N=14)	Control#3 (N=13)	Control#4 (N=33)	Test#5 (N=18)	Test#6 (N=15)	Test#7 (N=24)	Test#8 (N=35)	Test#9 (N=39)
Age at dental exam years; mean(SD)	2.8 (0.6)	3.2 (1.0)	2.7 (0.2)	3.2 (0.8)	3.3 (0.9)	2.6 (0.7)	2.9 (1.3)	2.7 (0.7)	3.2 (0.9)
Intervention Delivery (N (%))									
Intervention 1	NA	NA	NA	NA	11(61%)	8(53%)	14(58%)	17(49%)	31(79%)
Intervention 2	NA	NA	NA	NA	0(0%)	5(33%)	13(54%)	27(77%)	18(46%)
Intervention 3	NA	NA	NA	NA	0(0%)	6(40%)	9(38%)	29(83%)	16(41%)
Intervention 4	NA	NA	NA	NA	5(28%)	8(53%)	4(17%)	31(89%)	9(23%)
Intervention 5	NA	NA	NA	NA	3(17%)	5(33%)	6(25%)	29(83%)	9(23%)
Intervention 6	NA	NA	NA	NA	5(28%)	3(20%)	3(13%)	22(63%)	10(26%)
Patient-Level Caries Outcomes (N(%))									
Any caries[1]	69(95%)	12(100%)	13(100%)	25(96%)	18(100%)	9 (100%)	17 (100%)	28(100%)	28(93%)
Enamel caries[2]	58(85%)	9(75%)	12(92%)	21(84%)	15 (88%)	6(75%)	13 (93%)	19(83%)	19(70%)
Dentinal caries[3]	51(66%)	5(42%)	10(77%)	19(66%)	11(61%)	4(36%)	9(43%)	12(41%)	8(26%)
Pulpal caries[4]	20(26%)	3(25%)	4(31%)	5(17%)	7(39%)	2(18%)	5(24%)	2(7%)	4(13%)
Fillings[5]	16(21%)	3(25%)	2(15%)	4(14%)	4(22%)	1(9%)	1(5%)	2(7%)	2(6%)
Extractions[6]	10(13%)	0(0%)	2(15%)	2(7%)	2(11%)	1(9%)	1(5%)	0(0%)	1(3%)
Surface Count Caries Outcomes (mean(SD))									
Any caries[1]	23.6(21.7)	14.8(13.4)	27.1(22.1)	16.9(18.9)	21.9(15.0)	9.3(12.6)	19.8(21.6)	12.2(12.7)	12.3(13.9)
Enamel caries[2]	15.1(18.2)	7.8(7.2)	16.6(15.2)	9.8(13.3)	15.1(13.5)	6.0(12.6)	12.7(14.9)	5.2(7.9)	4.2(7.7)
Dentinal caries[3]	11.2(16.3)	2.5(4.0)	12.9(14.3)	6.8(11.5)	9.8(13.1)	3.1(6.0)	8.5(12.9)	3.4(6.7)	2.5(6.3)
Pulpal caries[4]	4.6(12.4)	1.4(3.4)	1.9(3.7)	2.6(8.5)	5.4(11.9)	1.7(5.2)	2.3(5.8)	0.1(0.7)	0.4(1.9)
Fillings[5]	3.7(11.9)	1.4(3.4)	1.5(3.8)	2.4(8.4)	3.7(11.1)	1.3(5.2)	0.8(4.1)	0.1(0.7)	0.4(1.9)
Extractions[6]	2.1(6.9)	0.0(0.0)	1.5(3.8)	0.6(2.6)	1.0(3.1)	1.3(5.2)	0.8(4.1)	0.0(0.0)	0.3(1.6)
Tooth Count Caries Outcomes (mean(SD))									
Any caries[1]	9.7(6.0)	8.3(5.1)	11.7(5.2)	8.1(5.7)	9.0(4.6)	5.8(4.2)	8.3(6.8)	7.9(5.0)	8.0(5.3)
Enamel caries[2]	6.4(5.5)	5.6(4.4)	8.2(5.3)	5.5(5.4)	5.9(4.3)	3.2(4.2)	5.5(5.9)	3.5(3.4)	2.7(3.5)
Dentinal caries[3]	4.4(4.8)	2.3(2.7)	5.4(5.4)	3.7(4.6)	3.9(3.9)	1.5(2.1)	3.1(4.7)	2.0(3.0)	1.3(2.6)
Pulpal caries[4]	1.2(2.8)	1.0(2.1)	0.6(1.0)	0.9(2.5)	1.3(2.3)	0.5(1.2)	1.0(1.9)		
Fillings[5]	0.9(2.5)	1.0(2.1)	0.4(0.8)	0.7(2.3)	1.0(2.1)	0.4(1.2)	0.4(1.0)	0.1(0.3)	0.2(0.9)
Extractions[6]	0.4(1.4)	0.0(0.0)	0.3(0.8)	0.1(0.5)	0.2(0.5)	0.3(1.0)	0.2(0.8)	0.0(0.0)	0.1(0.3)

Legend (Table 5)

1. Enamel (not cavitated or cavitated), dentinal or pulpal caries; filling or extraction
2. Enamel cavitated, dentinal or pulpal caries; filling or extraction
3. Dentinal or pulpal caries; filling or extraction
4. Pulpal caries; filling or extraction
5. Filling or extraction
6. Extraction

Appendix 1

"I wish my child would have beautiful teeth"
Kimaa Miywaapitet Nitawaashiim
Motivational Interviewing Script
(For pregnant woman or new moms before first baby tooth comes in)

You might need to say to a mom-to-be or new mom at the first session:

- ❑ *"It may seem early, but I would like to talk about your new baby's teeth"*
- ❑ *"What we say to each other will be confidential"*
- ❑ *"I will be looking at my guidebook and taking notes as we talk" or, if mom will be more comfortable, make notes after she leaves*

- ❑ **Visit #1: During pregnancy**
- ❑ **Visit #2: At 2- or 4-months immunization appointment**

Background for pregnancy and new moms before first baby tooth comes in

Why is it so important for mom-to-be or new mom to have a healthy mouth?

For a cavity to start, at least 3 factors are required

1. Sugary foods and/or drinks
2. Cavity-causing bacteria (germs)
3. Susceptible teeth

Babies are not born with cavity-causing bacteria (germs*) in their mouth

**These germs are usually passed from a mom to her baby*

How does a mom spread these germs to her baby?

If mom has cavities that have not been fixed, or does not keep her teeth as clean as she should, she will have cavity-causing germs on her teeth and in her saliva (spit)

Therefore, whenever a mom
-kisses her baby
-licks a soother to make it clean
-blows on her baby's food
-or, any of those good things that we expect moms to do, she is at risk of passing on these germs to her baby.

How does a mom stop spreading these germs to her baby?

A mom should have all her cavities fixed and brush her teeth regularly. Cutting down on sugary foods and drinks and chewing xylitol-containing gum will help stop new cavities from forming in her mouth.

That is why the themes of the counseling that you will do ***before*** a baby gets any teeth are:

1. Improving Mom's oral health

2. Stopping the spread of cavity-causing bacteria (germs) from mom to child

1. ASK QUESTIONS: **WHY ASK QUESTIONS?**
    1. To show your concern or empathy for mom and her new baby
        - Counseling, like motivational interviewing, works best when you have developed a relationship with a mom
            - when you know her, even a little bit
    2. To get mom to talk, so you are not doing all of the talking
        - You should do more listening than talking

❑ Only ask the type of questions that you are comfortable with
An indirect question might be best! For example
- *"Some parents have had problems with their own teeth, so they worry about their children's teeth…what about you?*

❑ Or you could ask other questions, just to get mom talking
- "*Do you have other children*?" (write down children's names and ages)
- Comment about other children, for example, *"It must be hard for you to have time for yourself as well as look after……"*
- Encourage the mother to tell you about the pregnancy. For example,
    - *"Are you sleeping and eating okay?"*
    - *"Do you work outside of the home?"*
- Respond to the mother by nodding and paraphrasing. Encourage her *"Tell me more."* Write down important points in her folder.

➢ At our workshop, we decided not to dwell on the past, but look to a positive future.

➢ You also felt that moms may not want to talk (with you) about their own, or their other children's teeth especially if there have been "problems"

➢ So, we will move on to asking what a mom wants for her new baby's teeth

❑ Ask mom what she wants for her child's teeth
- *"If we could change the future, leaving the past behind, tell me what you would want for the dental health/teeth of your child?"*
- *"Tell me more."* or *"Anything else?"*_____.
- Repeat back to mom what she has just told you. This lets her know that you have been listening!
    - *"Let me be sure that I understand. You would like your child to……"*
- Write down what she wants:

_____

2. BE POSITIVE ABOUT HOW GOOD A PARENT SHE IS TO WANT THESE THINGS FOR HER NEW BABY.
   - *"You are/will be a really good mom"*
   - *"You really love that baby."*
   - *"You are really good at…."*

3. SAYING IT AGAIN MAKES IT SO!
   - that is, say things to motivate mom
   - "*I think I heard you say you want your baby to have great teeth. Did I get it right?"*
   - *"Your child will thank you (for doing this)."*

4. REFLECT, LISTEN AND SUMMARIZE!
   - summarize dental health wish here
   - *"Please tell me again what you want for your child's dental health, so I can be sure I get it right"*

5. TIME TO MOVE ON TO THE MENU

Providing information to a parent who is ready to do something:

- *"In order to (repeat again mom's dental wish), I want to share with you some things we have recently learned."*
- *"Would you like to hear about them?"*

Ask her permission!

- *"We have spoken to many moms about what we can do to help our children have healthy teeth. They suggested that we talk to other moms during pregnancy or before the baby gets her/his first tooth. That is why we are with you now."*
- *"Some of the steps that were suggested for moms are on a list (menu) that I would like to show you."*
  - Explain to mom that if her teeth are healthy, her baby will have a better chance of healthy teeth**. (Background in box on page 1.)**
  - Emphasize choice: no need to choose everything*!*

- Show mom the menu.

Menu for pregnant woman or new mom before first baby tooth comes in

1. Brush my own teeth with toothpaste at least once a day.
2. If it has been more than 6 months since I went to the dental clinic, I will make an appointment for a check-up.
3. I will have healthy meals and snacks in my diet
4. I will drink less pop or drink pop only with meals.
5. I will only chew sugar-free gum.
6. I will always hold my baby during feeding, then lay him/her down to sleep and, if he/she wakens, I will give water, not formula or juice
7. I will sometimes give my new baby a soother if he/she is fussy, not a bottle.
8. Other_____

If time permits, do the following or go on to #6 and do this next visit:

a. Review the menu items

"Let's look at the items on the menu, talk about each of them briefly, and you decide which ones are for you."

The following is an example:
"Is it possible for you to brush your teeth with toothpaste at least once a day?
"What problems would you face if you tried to do this?" E.g. lack of time

b. Review the rest of the list:
   - *"What problems would you face if you tried to do any of the others that you have chosen?"* Examples of problems below
     - fear of the dentist,
     - lack of time
     - hard to get an appointment at the clinic
     - good food is too expensive*!*

c. Say, "*Maybe you have some ideas of your own on what to do?*
Be positive about any idea. "*That is a great idea!*"

d. Identify additional benefits
   - *"Let's talk about the additional benefits of each item you have chosen."*
   - *"Any other good things that would happen when you for example,*
     - Change snacking habits— I will be healthier (less weight problems, diabetes, heart problems);
     - Cut down on pop (I will save money)
   - "*Is this what YOU want to do?"* _____

6. INCREASE MOM'S BELIEF IN HERSELF THAT SHE CAN DO THIS
   - ❑ Say supportive words: ***"I can tell that you are/will be a great parent/mom…"***
   - ❑ Highlight her competence and abilities: ***"I can tell that you are/will be a great parent/mom…"***
   - ❑ Encourage her to tell you again what she wants for her child: ***"Tell me again what you want for your child's teeth/smile?***
   - ❑ Remind her of outcome if she did nothing: ***"If someone chose not to do anything to help a child have healthy teeth, what would happen?"***

7. IF MOM IS "RESISTANT": TIME TO CHANGE YOUR APPROACH!
   - ❑ If a mom argues, interrupts, or shows reluctance. Do not argue with her
     - Try something different, for example,
       - Emphasize choice.
       - Agree with mother: *"It's your choice. I'm not here to make your decision"*
     - Shift into reverse—that is, go back to the beginning
       - Work on rapport and trust: *"I really believe in you, your child will have great teeth because of you"*
       - Ask questions, listen
     - If mother is not ready to make commitment do not press her; say,
       - "*If you are not quite ready yet to take the steps we have gone over, I do not want you to go ahead and make a commitment. This is too important to decide now.*
       - *"Go home and think it over."*
       - *"I will see you when you bring in your new baby for his/her 2-month immunization, and we can talk about it further."*
       - *"I hope everything goes well for you."*

8. WHAT TO SAY WHEN YOU HEAR:

"Baby teeth are not important."
"All kids get cavities; it is normal…."
"My family doesn't have strong teeth; our teeth decay/go bad easily….."

- Listen to what mom is saying.
  "I understand what you are saying; we used to believe that too…"
- Say, as we discussed "new knowledge exists":
  "Recently research has given us new information/approaches…"
- Remind mom about what you said about mom spreading germs to baby, etc.

9. ENDGAME

❑ Give a copy of the menu to mom
"I am going to give you a copy of your menu"
"The items that you chose are checked off"
"You know where to contact me if you have more questions"

❑ Anticipate problems
"Not everything goes the way we plan.
"There are always problems."

❑ Encourage contact with CHR
"Feel free to call me if you have any problems with your men"
"I appreciate your situation and your willingness to try."

Appendix 2

Cost effectiveness analysis of a perinatal Motivational Interviewing program for the prevention of early childhood caries in Cree communities in Quebec

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Abstract

Background:

Early childhood caries are prevalent in remote Canadian First Nations communities. A community randomized 'Motivational Interviewing (MI)' program with expectant mothers was hypothesized to reduce burden of the disease in a group of remote Cree communities in Quebec, Canada. Our objective was to evaluate the cost-effectiveness of this program.

Methods:

We performed a cost-effectiveness analysis with a health system perspective, relative to standard care in the area. Analysis of health effects is limited to the trial follow up period (2-5 years), while consideration of future health care costs arising from program effects is performed in a sensitivity analysis. Outcome measures include averted DMFTS, averted ECC cases, dental treatment costs, and incremental cost-effectiveness ratios ($2006 CDN).

Results:

The MI program was associated with improved dental health and decreased dental treatment costs, but an increase in overall dental expenditures during the trial period. Cost-effectiveness ratios are estimated at $81/averted DMFTS and $3900/averted ECC case by net benefit regression using the in-trial data, or $48 and $2250 with assumed future dental treatment expenditures and a pragmatic estimate of program costs upon implementation.

Conclusion:

Given the severity of ECC in the trial communities, the MI program appears to be a cost-effective way to improve health relative to standard practice.

Introduction

The aim of health economic evaluation is to maximize the benefits produced by scarce healthcare resources by identifying the costs, benefits, and opportunity cost of interventions intended to improve health, and promote treatments that offer the greatest value for money. Ideally an evaluation will include all of the relevant costs and benefits arising from an intervention, subject to the perspective taken for the analysis, and without temporal or measurement limitations. A second major focus is on uncertainty, whether the result of variation in parameter estimates or the result of structural assumptions made by an analyst that may impact whether an intervention is considered to be cost-effective (Drummond, 2005).

Trial based economic models are notable for having a number of common theoretical limitations (Sculpher et al., 2006). Economic evaluations ideally consider all relevant (non-dominated) treatment comparators and assess cost effectiveness incrementally against the best comparator, while trials engineered to demonstrate treatment effectiveness often take a placebo, standard treatment, or no treatment as their reference point. Trials commonly have limited follow up periods, and may collect intermediate outcomes rather than the terminal outcomes best suited to generalizable economic evaluations. Finally, trials may not collect all the parameters of relevance to a health economic evaluation, or may be powered to demonstrate effectiveness but not cost-effectiveness, which generally requires larger sample sizes due to correlation between morbidity and healthcare expenditure (Glick et al,. 2007).

Decision analytic modeling stands as an alternative and complement to the economic analysis of clinical trials, but ultimately depends on trial data for parameter estimates. The current recommendation for best practice in economic evaluation alongside clinical trials is for an incremental, iterative process to be taken, with a pre-trial model created on the basis of a meta-analysis of existing evidence, with the results of that model informing experimental design as to maximize the return on investment of research spending on decision uncertainty as assessed via value of information and value of sample information analysis (Sculpher et al., 1997). The pre-trial model is then to be updated with trial results, and if economically warranted, reevaluated periodically to update parameter estimates with new information from a growing literature.

This approach to economic evaluation presupposes an evidence base, and is of questionable relevance to novel treatments, or applications of treatments to clinically unique patient groups. Treatment of early childhood caries (ECC) represents one such situation. The condition has a number of documented (though not quantified or parameterized) effects and comorbidities, including stunted growth, pain, child abuse, and long term effects on dental health, which speak to the seriousness of the condition, but are not particularly amenable to current methods of economic analysis (Li & Wang, 2002; Casamassimo et al., 2009). No estimates of QALY decrement have been found for ECC, while significant methodological challenges may inhibit the valid collection of estimate due to the young age of affected children (Griebsch et al., 2005).

Modeled economic evaluations of caries prevention have been undertaken (Ramos Gomez & Shepard, 1999; Quinonez et al., 2006; Warren et al., 2010), but are based almost entirely on assumed effectiveness and treatment utilization rates, and do not provide convincing evidence of the cost-effectiveness of any particular intervention. Likewise, Canadian evaluations of ECC prevention programs have not been localized to the target population in Northern Quebec, and have not collected economic data (Lawrence et al., 2008; Peressini et al., 2004; Harrison et al., 2007).

This trial provides valuable evidence of MI program effects, ECC incidence in the trial communities, and the cost associated with ECC treatment, but suffers from common limitations in trial-based data collection. Some of these limitations may be mitigated, while others cannot and must be noted as caveats for the analysis. The comparator in the trial is current standard practice in the area,

consisting of standard dental treatment plus the provision of an ECC prevention pamphlet to expectant mothers after program enrollment. Using an incremental approach, a variety of ECC prevention programs could be devised, including the current option (MI + Fluoride application), only MI, only fluoride application, a community information night, provision of health information videos, development of a website, etc. These alternatives may or may not be effective or cost-effective, but they stand as potential comparators to the evaluated MI program. If standard practice is not the most cost effective treatment amongst these comparators, then evaluation against it biases the evaluation in favor of the experimental treatment.

The trial is also characterized by a limited set of outcome measures. The limited follow up period (2-5 years, depending on the child) may not be sufficient to capture important program outcomes, especially if improved dental health is measured as a result of improved dental health habits which have an ongoing effect, or if childhood dental health has an impact on adult dental health (as in: Li & Wang, 2002). Other potential program effects are largely immeasurable, including the value of potential future averted days of school truancy due to improved dental health, and (hypothetical) family and community 'good dental health practice' externalities. Limitations in the current analysis, and efforts to control for them, will be discussed to follow.

Methods

Three related economic models have been adopted to evaluate the trial data, with consideration of the potential factors underlying determination of a relevant cost-effectiveness threshold and sensitivity analysis concluding this report.

As a starting point we adopt a deterministic cost effectiveness model, which assumes effective randomization on the part of the trial, and is presented here with selected scenario based sensitivity analyses. Attention is paid to both cost-effectiveness of the MI program within the trial, and to cost-effectiveness upon implementation in a non-trial setting. Cluster robust tests of significance (clttest in STATA 11: Donner & Klar, 2000) are calculated for trial outputs and model covariates, but not for cost effectiveness, which is calculated on the basis of an incremental cost effectiveness ratio (ICER: Equation 1). The assumption of normality that t-tests require is obviously violated in case of many of the outcome variables, which have skewed and bounded distributions, but common non-parametric tests of significance fail to provide inference on the distribution of arithmetic mean values, which is the measure of central tendency of interest in economic evaluation (Glick et al., 2007), and cluster bootstrap methods have been criticized when the number of experimental clusters is small (Donner & Klar, 2000).

$$ICER = \frac{Costs_{Experiment} - Costs_{Control}}{Health\ Benefits_{Experiment} - Health\ Benefits_{Control}}$$

Equation 1

The assumption of effective randomization and the attribution of the full observed treatment effect to the intervention is relaxed in the second model, in which regression analysis is performed to assess the impact of covariates on measured values of dental health and treatment utilization. Separate regressions are performed for DMFTS, cases of early childhood caries (defined as DMFTS≥4), local treatment costs, and total dental care costs, which includes estimated dental treatment expenditures within and external to the local communities, and per capita counseling costs for the experimental groups. All regressions are run with 'general linear models' (glm) in STATA 11, with clustered standard errors on the basis of community ID using the Huber-White sandwich estimator. GLM family and link parameters are optimized using the modified Park's test (Manning & Mullahy, 2001), and link diagnostics (Pregibon, 1980) contained in the 'glmdiag' program published for STATA by Glick et al. (2007). The 'recycled predictions' method is used to estimate the effect of the intervention on the outcome variable: predicted values of the outcome variable are estimated with each observation in the sample coded as being in the control group, and then in the intervention group. The mean difference in predicted values provides the regression estimate of the outcome variable independent of covariate differences in the control sample and intervention sample. Recycled prediction estimates of incremental cost and effect replace unadjusted point estimates of cost and effect in the ICER calculation for a covariate adjusted cost-effectiveness estimate.

The third method involves individual level net benefit regressions. Stinnett and Mullahy (1998) proposed the net benefit framework as an alternative to ICERs, which have a number of inconvenient statistical properties, including asymptotic tendencies as measured benefits approach 0, and ambiguous meaning for the negative values ascribed to dominated treatments. Net Monetary Benefit (NMB) is measured by a simple rearrangement of the ICER formula (Equation 2) :

$$NMB = \left(Health\ Benefits_{Exp.} - Health\ Benefits_{Control}\right)(\lambda) - \left(Costs_{Exp.} - Costs_{Control}\right)$$

Equation 2

Under this rearrangement, an intervention is considered to be cost effective when NMB>0, which occurs when the assumed monetary value of incremental gains exceeds incremental costs. Hoch et al. (2002) proposed net benefit regression, in which net monetary benefit is generated as an outcome variable on the basis of observed costs, effects, and an assumed cost effectiveness threshold (λ). If NMB is used as a regressand, the standard errors calculated by a well specified model can be used to calculate the confidence interval of the cost-effectiveness estimate, and implied probability that the intervention will be cost effective for a given level of λ. This is not true of split regression models for cost and effect, for which bootstrap evaluation of cost-effectiveness is necessary. Flynn and Peters (2005) and Bachmann et al. (2007) argue that bootstrap estimates of cost effectiveness confidence intervals are less reliable than NMB regressions with cluster robust standard errors when the number of clusters is small, as is the case for this trial (9 communities).

We use OLS regression for this report to estimate net monetary benefit and to inform cost-effectiveness acceptability curves (Fenwick et al., 2001) using the method in Hoch et al. (2006). When

finalized cost estimates are available, NMB regressions will be optimized with GLM models, as for the cost and benefit regressions.

The three model approaches can thus be seen to have complementary functions. The deterministic model (Model1) is meant to be illustrative, but does not adequately express uncertainty in the cost effectiveness estimate. Cost and effect regressions (Model 2) serve to refine treatment estimates in Model 1 on the basis of imperfect randomization and the effect of covariates. Net benefit regression serves as the primary model and best estimate of the uncertainty arising around the cost effectiveness estimate. A health system perspective has been adopted for the base case analysis, with a 3% discount rate on costs and effects. All monetary estimates are expressed in $ CDN (2006).

Model Structure

A simple decision analytic model has been adopted on the basis of recorded intervention costs, outcome dental health exams, local dental treatment cost records, and parental reports of dental care utilization (Table 1).

	Control Group	Experimental Group
Costs	- Within community dental treatment expenditures - Parental reports of caries related physician access, anesthetic use, and dental treatment in major centers	- Control group expenses **and** - Training program for local MI staff - MI staff time - Costs of fluoride application in MI Sessions
Benefits		Inc. Healthy Tooth Surfaces (HTS) = Inc. averted DMFTS -Incremental averted ECC Cases

Table 1: Model Parameters

The incremental cost effectiveness ratio (Equation 3) is calculated on a per-capita basis to account for the disparate size of the treatment and control groups, and is calculated as the net present value (2006) of:

$$ICER = \frac{(MI\ Training\ Costs + MI\ Running\ Costs + Treatment\ Costs_{Exp.} - TC_{Cont.})}{Healthy\ Tooth\ Surfaces_{Exp} - Healthy\ Tooth\ Surfaces_{Cont.}}$$

Equation 3

Program Benefits

The Cree ECC trial will be assessed using a cost-effectiveness analysis, with Cost per Averted Decayed Missing or Filled Tooth Surface (DMFTS) as the primary outcome measure. Cost utility analysis is generally a preferred method in health economic evaluation, as it facilitates greater comparability of healthcare interventions (Drummond et al., 2005), however, a number of factors make utility based measures such as the QALY unsuitable for analysis of this intervention. First, utility weights for early childhood caries have not been surveyed; estimation of those values would be hindered due to both existing methodological difficulties in assessing QALYs for children (Greibsch et al., 2005).

Additionally neither QALYs nor Quality Adjusted Tooth Years (QATYs) (Birch, 1986) are used widely within the dental economic evaluation literature and it is likely the case that the cost-effectiveness measure $/DMFTS will provide greater comparability within the field. Lastly, the value of a generalizable health measure exists only amongst interventions between which institutional priority setting decisions may be made. It is not clear that funds may be transferred from pediatric dentistry in northern Cree communities to other authorities within the healthcare system. If budget setting is limited to the dental care system a field-specific measure may be more sensitive to clinical effects and thus preferred. DMFTS can be assessed given the data collected within the trial, while calculation of QATYs requires time-specific data that has not been collected, while neither measure has an explicit or consensus cost-effectiveness threshold in Canada or internationally. For these reasons, a cost-effectiveness model based on cost/averted DMFTS has been chosen.

For the purposes of this economic analysis, DMFTS have been defined as a tooth surface score of 2-5 on the dental health outcome examination, or a tooth score of 6. Surface score '1', corresponding to color change on the tooth but no substance loss, has not been included in the DMFTS estimates.

Cost/averted case of ECC has been adopted as a secondary outcome measure, where a case of ECC is defined by a DMFTS score of 4 or greater (as in Perressini et al., 2004). DMFTS appears to be the most prominent (and thus comparable) measure in the ECC literature, however, Casamassimo et al (2009) argue that is an insufficient outcome measure for ECC, as it provides no indication of the serious side effects that may accompany ECC. Casamassimo et al do not explicitly address the appropriateness of using DMFTS for the economic evaluation of programs that impact ECC, with uncertainty; we translate their argument into 'health economist' as one of the following:

1) The DMFTS is an incomplete measure of the burden of ECC, however as long as the measure is used solely to compare ECC interventions it is valid as it does not bias analysis of similar interventions. If DMFTS from ECC are equated to adult DMFTS, this is invalid, due to the extra burden of ECC.
2) The DMFTS is an incomplete measure of the burden of ECC, and the burdens of ECC are not linearly related to DMFTS (there is a threshold effect or a multiplicative/exponential relationship). At some level of X, X individuals with 1 DMFTS bear a smaller burden than 1 individual with X DMFTS. DMFTS are thus not appropriate as an outcome measure without

severity weighting (if effect is multiplicative), or consideration of the population above the threshold value (if a threshold effect is observed).

No DMFTS weighting algorithms have been found, however a number of ECC classification systems exist. For the purposes of this analysis we adopt the DMFTS ≥4 = ECC definition of Perressini et al. (2004), which was used for similar purposes in the estimation of ECC incidence in a First Nations population in Northern Ontario.

Program Costs

The economic model includes fixed training costs for MI staff, salary costs for program staff delivering the MI interviews, resource costs for fluoride application and dental gifts delivered as a part of the MI program (fluoride>1 year after birth), and public expenditures on dentistry for trial participants. Costs relating to the MI counseling program are relatively certain as estimates of the total cost of the training program for local MI staff and local staff wage rates have been provided by senior members of the research staff. Local dental treatment costs have been estimated on the basis of the 2006 Quebec dental fee schedule used by program staff (Régie de l'Assurance-Maladie du Québec, 2008). Assumed model inputs are presented in Table 2.

Parameter	Value	Source	Sensitivity Min.	Sensitivity Max.
CHR (Counselor) Wage	$25/Hour	Program Staff	20	30
Fluoride Application Cost	$5.00	Local Dentistry Cost Charts		
Discount Rate	3.00%	Author	0	5.00%
Health System Cost for a Physician Visit	$100.00	Author		
Guardian Reported Anesthesia Use	$60.00	Local Dentistry Cost Charts		
Guardian Reported Multi-Day Travel Event for Pediatric Dental Treatment	$4,000.00	Author	$2650 (Milnes et al., 1993): 1988	$5,000

			Manitoba estimate in $2007 (CPI Inflation)	
Staff Training Costs/Child		See Appendix 1	See Appendix 1	

Table 2: Cost-Effectiveness Input Values and Sources

Treatment Costs

Overall follow-up in the trial was very good, with dental outcome exams on 241 of 272 patients. However, dental treatment costs on enrolled children have not been as thoroughly documented. Local patient cost records were transcribed for 224 patients, though these records are believed to be deficient in a couple respects. It is known that out of community treatment is not recorded on local records, while it is expected (but still rare) that severe ECC cases be treated in major medical centers under general anesthetic. This medical procedure caries non-negligible risk, and has been identified as a major cost in treating ECC in remote Canadian First Nations communities (Milnes et al., (1993). Out of community treatment costs relating to severe caries cases (as expressed by guardians in the outcome survey as non-accident related trips to a doctor, use of dental anesthetic, or major travel event for non-accident related dental health problems) have been inferred, on the basis of local costing charts (Régie de l'Assurance-Maladie du Québec, 2008) and expert judgment.

Also of concern is discordance between local cost records, parental reports, and the dental health exam. In 173 of the 224 dental health records there is a general agreement between the dental exam and cost records, however in 51 cases the cost records indicate no treatment ($0 overall cost), in opposition to parental report or the dental record, which may include a code 5 for a filled tooth surface, or 6 for a caries related extracted tooth. Underreporting may exist throughout the data set but not be as obvious. Potential 'false 0 values' are clustered in community 1, a control community, and communities 8 and 9 in the intervention group. It is generally unclear whether low reported costs correspond to good dental health, low utilization, or missing data.

Estimation of treatment costs outside the trial follow up period which are attributable to dental health deficiencies incurred in the trial period is also necessary. Approximately 80% of DMFTS in the trial population (2200 of 2750) were untreated at the time of the dental exam. It is unclear whether low utilization represents a temporal lag in care seeking behavior (caries are filled or teeth are extracted at a scheduled check-up, or when they become problematic) or permanently low utilization. It is also unclear whether or not this distinction matters to decision makers.

If a decision maker is cost-minimizing, he may consider low utilization advantageous, and thus have a weaker incentive to prevent the development of disease in low utilization populations. However,

this line of thinking has obvious deficiencies, and is open to criticism both on equity and health maximization grounds. If dental treatment is cost-effective a health maximizing decision maker should aspire to full utilization, as by definition the costs of treatment are proportionately less than the health gain. Public funding for children's dental health is common internationally (Birch & Anderson, 2005), but has not been subject to widespread economic evaluation. Common practice could represent flawed conventional wisdom (as was the case for common medical procedures exposed by the advent of the evidence based medicine movement), or treatment with such clear positive outcomes and reasonable cost that economic analysis is redundant. For the purposes of this report we assume the latter.

When future cost data is compiled, it is expected that 'full treatment' will be the primary result, with 'local expected utilization' as a potential sensitivity analysis. In the interim, a 'full treatment scenario' has been prepared as a sensitivity analysis below. We divide the total dental treatment expenditure by the number of treated DMFTS in our analysis sample to find the average cost per treated DMFTS ($30), which is used to estimate the future costs of treatment for existing untreated DMFTS.

Training Costs

Given current estimated treatment costs the largest expense associated with the MI program was training for program staff. A 3 day workshop was held prior to implementation of the program with an outside consultant and research staff in order to train local staff on the MI program, adapt it to match local norms, and prepare local staff for research requirements. A 2 day follow-up workshop was held prior to the second year of the program.

The measured effects of medical treatments are commonly greater in controlled experiments than in common use (Godwin et al., 2003), an effect often attributed to increased training of staff, greater attention paid to paid to individual patients, or higher rates of patient compliance due to increased respect for the treatment program. Likewise, intervention costs may be higher within an experimental setting, both due to increased requirements for data collection for research, and due to an intensified focus on treatment quality. Where possible, costs relating solely to research requirements should not be included in an economic analysis. In this case, staff costs relating to data collection and the baseline subject interview have been excluded from the analysis. However costs related to atypical investments in the training of program staff must be included in the analysis, as their exclusion could impart a strong bias in favor of the intervention, which would be evaluated on the basis of full experimental program effect, and a pragmatic estimate of cost.

It is recognized that the per capita costs associated with the MI training program may be artificially high so a series of scenarios have been drafted to correspond to alternate staff training methods. Analysis surrounding each scenario concerns both the cost difference associated with a less intense training program, and the proportion of the measured treatment effect (in reduced dental treatment costs and dental health gains) that must be preserved for the strategy to match the cost-effectiveness estimate under trial conditions. These training scenarios are:

1. The training program utilized in the experiment, with staff from each intervention community traveling to a central location for intensive training annually with an outside consultant.

2. Training program 1), but assessed for all 9 experimental communities, so that fixed training costs are amortized over a larger population, and with an annual 2 day workshop. This may be considered the 'best estimate' of implementation cost.

3. A senior staff member (costs based on the research coordinator) travels to each community annually to train staff members for 1 day. No outside consultant.

4. An annual teleconference with the senior staff member and local staff to provide training, assuming resource costs of $1000/year.

Sample Size

The cost and cost effectiveness analyses presented in this report are based on the subgroup of children (171) for which cost estimates are without obvious deficiency (excluding universal issues regarding expenses on parental reports of anesthetic use, physician access and travel for treatment). Following clinical estimates of treatment expenses, or regression based cost imputation (STATA's multiple imputation process), final analysis is expected to be based on the sample of 241 with dental exam data.

Results

OUTCOMES	Control Mean	Intervention Mean
DMFTS	15.23	11
Prob. Of ECC CASE	0.65	0.56
Prob. Of DMFTS≥10	.5	.41
Caries Related Lost Teeth/Child	0.35	0.06
Local Treatment Expenditure	$73	$32
Total Treatment Expenditure	$188	$101
MI Counseling & Fluoride Costs	0	$32
MI Training	0	$318
Total Dental Expenditure	$189	$451

COVARIATES	Control Mean	Intervention Mean	SE of Difference	2 sided P=	ICC
Mother's Age at Birth	25.6	25.4	1.34	0.86	0.04
Child's Age at Dental Exam	2.88	2.89	0.17	0.94	0.05
Maternal Education: In High School	0.11	0.07	0.06	0.6	0.02
Did not complete High School	0.48	0.38	0.19	0.61	0.19
Finished High School	0.27	0.33	0.14	0.67	0.11
Some College	0.1	0.14	0.11	0.71	0.15
University	0.01	0	-	-	-
Education Unknown	0.03	0.03	0.04	0.42	0.03
Maternal Behavior:					
Brushed Teeth in Last Day	0.92	0.94	0.02	0.24	-0.04
Visited Dentist in the past 2 years?	0.71	0.67	0.09	0.66	0.27
Was the last dental visit a routine checkup?	0.4	0.52	0.13	0.4	0.07
Initial Dental Knowledge Score (of 5)	2.93	3.08	0.44	0.74	0.14
Mean RAFPIDD (of 5, 1= fewest challenges to pediatric dental health)	2.29	2.29	.06	.52	.02

Table 3: Sample Statistics (n=173)

Summaries of sample statistics in the experimental population are presented in Table 3. Lower mean DMFTS scores and local dental treatment costs are measured in the intervention group, though total expenditures on dentistry are higher in the owing to MI costs. A 9% reduction is observed in the proportion of the intervention group with DMFTS≥4 and DMFTS≥10 relative to the control group.

No significant differences (p=.05) are observed in the measured covariates, though differences in mean scores with potential relevance to the economic evaluation are observed in maternal education, the proportion of the sample whose last visit to the dentist was for a routine checkup, and in the initial dental knowledge score. The initial ICER point estimates are $60.25/ averted DMFTS and $2800/averted ECC case. Figure 1 illustrates the DMFTS distribution by intervention group. Both DMFTS distributions are right skewed, the distribution of the intervention group is characterized by a greater proportion of recorded zeros, and fewer children with a very large number of DMFTS (>40).

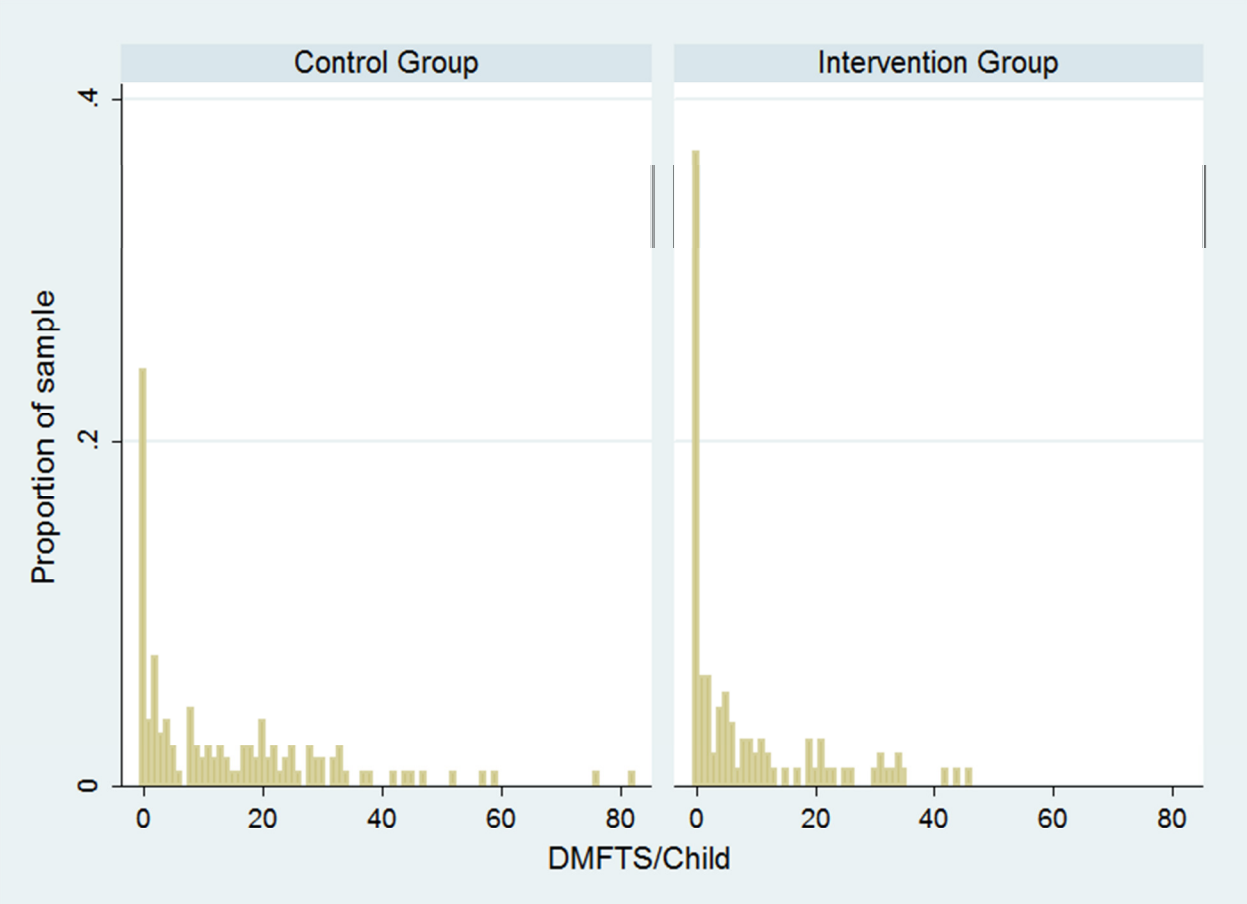

Figure 1: DMFTS distributions by Intervention Group

Incremental Point estimates of Cost, Effect, and Cost Effectiveness (X_{EXP}-$X_{CONTROL}$): per capita		Incremental Regression estimates of Cost and Effect, and implied Cost Effectiveness (X_{EXP}-$X_{CONTROL}$): per capita	
Healthy Tooth Surfaces	3.9	Healthy Tooth Surfaces	3.2
Averted ECC Cases	0.09	Averted ECC Cases	0.076
Incremental Expenditure	$253.05	Incremental Expenditure	$326
$/HTS	$60.25	$/HTS	$102
$/Averted ECC Case	$2,812	$/Averted ECC Case	$4071

Table 4: Regression Estimates of Intervention Attributed Costs and Effects

Changes in mean outcome estimates attributed to the intervention and corresponding effects on mean cost effectiveness ratios are presented in Table 4 on the basis of cost and effect regressions. Estimated mean incremental cost, effectiveness, and cost-effectiveness are less favorable for the intervention when controlling for influential covariates which are not evenly distributed between the intervention and control groups in the sample. Approximately ¼ of the observed incremental effect on DMFTS and 1/5 of the observed reduction in ECC risk are associated with covariate imbalances. Use of adjusted point estimates of incremental cost and effects increases ICER estimates to $102/ Averted DMFTS and $4071/averted ECC case.

Averted DMFTS		
Cost Effectiveness Threshold	**Net Benefit / Child**	**Probability MI is Cost Effective**
0	-255	0.006
30	-161	0.163
50	-98	0.327
81	-1	0.499
100	59	0.563
200	373	0.7065
300	688	0.7525
400	1002	0.7745
500	1316	0.7875
10000	31188	.833
Averted Case of ECC		
Cost Effectiveness Threshold	**Net Benefit / Child**	**Probability MI is Cost Effective**
0	-251	0.006
100	-244	0.0085
500	-219	0.0335
1000	-185	0.105
3000	-53	0.429
3900	6	0.5065
5000	78	0.5675
10000	409	0.6785

Table 5: Net Benefit Estimates

Current net benefit regression estimates were computed with cluster robust GLM (Gaussian family, identity link) and are reported in Table 5. Net monetary benefit can be interpreted as the per capita value of health gains attributed to the intervention at the specified cost effectiveness threshold, subject to the incremental costs associated with the intervention. The intervention is cost effective when net benefit ≥0.

Similar effectiveness is implied for risk reductions in ECC risk when diagnosed by a DMFTS score≥4 or ≥10, though the appropriate cost effectiveness threshold may differ for the two outcomes. Figures 2 and 3 are cost-effectiveness acceptability curves: they show the probability (given the NB regression results for the specified model) that the MI intervention would be cost effective relative to standard treatments, given varying estimates of the cost-effectiveness threshold. Subject to the constraints of this analysis, the MI program is not expected to be cost-saving. Critical cost effectiveness thresholds are observed at $81/averted DMFTS and $3900/ averted case of ECC. If the cost effectiveness threshold is above these critical values, then expected net benefit will be positive, and the program may be considered cost effective.

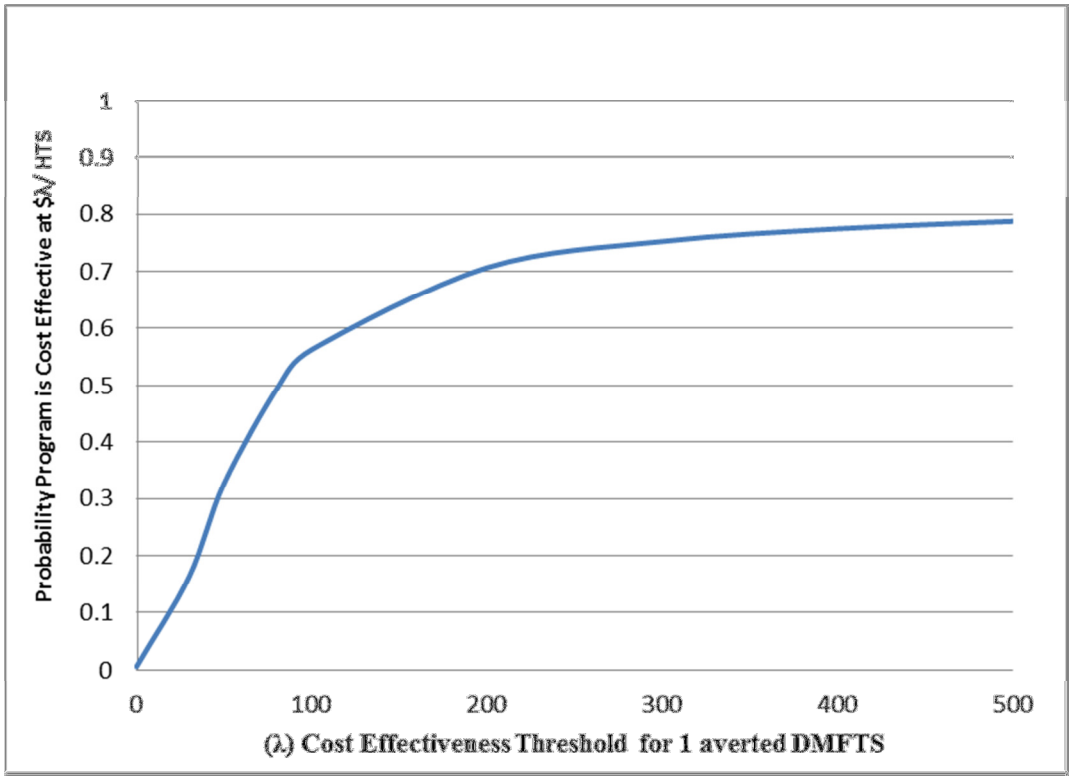

Figure 2: Cost Effectiveness Acceptability Curve: $/DMFTS

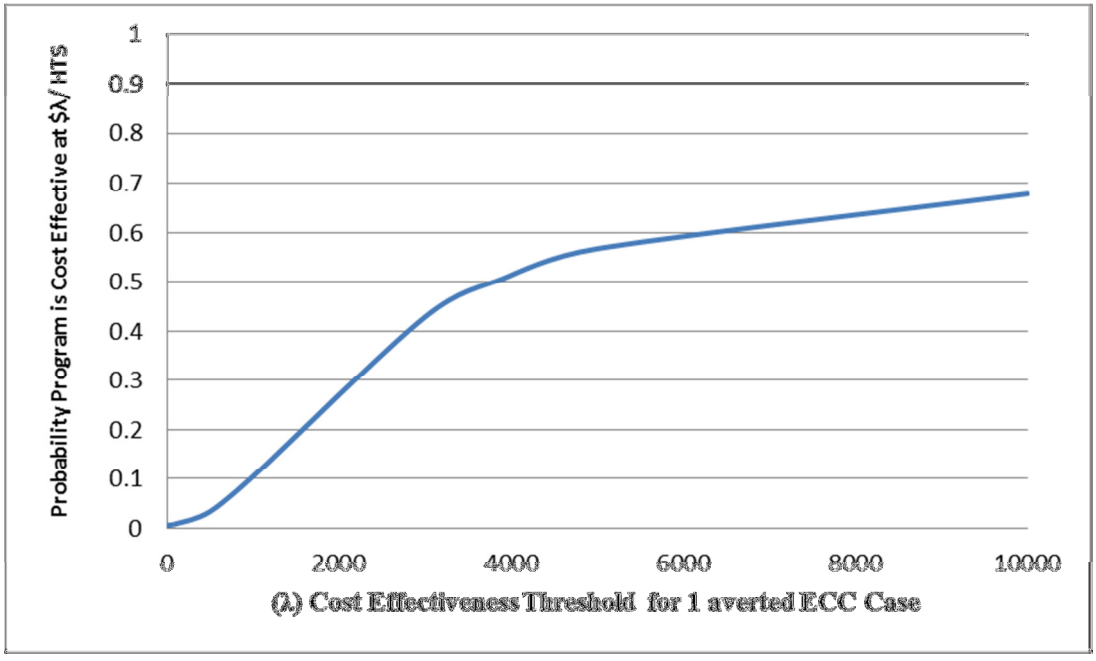

Figure 3: Cost Effectiveness Acceptability Curve: $/ Averted ECC Case

No meaningful 95% ICER (λ) confidence interval can be computed, as program effect is not significant at the p=.05 level within the regression framework (Glick et al., 2007). Even if decision makers are willing to pay $10,000/ averted DMFTS there is still only an 83% that the intervention will be cost-effective, not due to high incremental costs, but due to uncertainty in effect outcomes (a one sided p value of 0.17 is implied).

		$/Averted DMFTS	$/Averted ECC
Baseline CE Estimate		102	4070
Discount Rate	5%	104	4112
(base 3%)	0%	100	4016
	50	110	4610
Staff Wage Rate	30	103	4345
(base $25)	20	100	4213
	0	94	3948
Travel Treatment Cost	$5000	98	4128
(Base $4000)	$2650	107	4489
'Full Treatment' of in-trial caries	$30/DMFTS	84	3552

Table 6: Sensitivity Analysis for deterministic model, with Model 2 estimates of incremental cost and effect

Table 6 presents one-way sensitivity analyses for selected model inputs, using regression estimates of cost and effect. Assumptions regarding the discount rate used in the analysis and the public cost of trips to larger cities for hospital based dental treatment both, and the staff wage rate all appear to be insignificant.

Expenditures on dental treatment and on the training program for MI staff were the largest expenses associated with the trial. If all untreated DMFTS measured at the outcome exam (coded 2,3 or 4) were treated with the same average cost/DMFTS ($30) as those teeth coded as treated (DMFTS=5, or Tooth=6), the $/DMFTS ICER estimate falls from $102 to $84. If this treatment occurs in the future, discounting to the present day would raise the ICER estimate, while inclusion of assumed estimates of future reductions in dental decay or unmeasured major program effects (reductions in orthodontic expenditures) would improve the cost-effectiveness estimate. Of particular note is that even at full estimated dental utilization the MI program is not expected to be cost saving.

		$/Averted DMFTS	% of observed DMFTS effectiveness necessary to maintain CE estimate with less intense training program
Training Costs	Scenario 2	81	80%
	Scenario 3	48	47%
	Scenario 4	19	21%

Table 7: Alternate MI Staff Training Programs

Table 7 outlines ICER sensitivity to alternate staff training scenarios (page 9 of this report), including an estimate of the proportion of the observed treatment effect (on DMFTS/child) that would need to be observed under less intensive training programs for the resulting cost effectiveness estimate to match that of the trial. The baseline results describes the training program for the experiment, while scenarios 2-4 describe less expensive training programs that may be implemented. Under a low-cost teleconference training method (Scenario 4), only 21% of the observed experimental effect (a reduction of 0.6 caries/child) would need to be observed to match experimental cost effectiveness, though it very well could be the case that less-intensive training would result in an intervention that is not effective at all. Scenario 1 (experimental costs) is taken as the baseline for this analysis, though there is good reason to consider Scenario 2, which describes the same training program implemented for all 9 trial communities.

	Crit. CE Threshold $/ Averted DMFTS	Probability Scenario is Cost Saving	Crit. CE Threshold $/ Averted ECC
Baseline	81	.006	3900
'Full Treatment'	68	.065	3300
'Training Scenario 2: Program Rollout to all Cree Communities'	58	.03	3000
Both FT + S2	48	.14	2250

Table 8: Sensitivity analysis for Net Benefit Model

Sensitivity analysis on the net benefit model is presented in Table 8. Under the first alternate scenario, the 'full treatment' assumption is made: critical cost effectiveness thresholds fall to $68/ averted DMFTS and $3300 averted ECC case. If baseline treatment costs are maintained, and 'Training Scenario 2' is implemented, the critical cost effectiveness thresholds are at $58/averted DMFTS and $3000/averted ECC case. If both pro-trial assumptions are adopted: $48/ averted DMFTS and $2250/averted ECC case, with a 14% estimated probability on the basis of sampling and regression

uncertainty that the program would be cost saving. The associated cost effectiveness acceptability curve for this 'pro-MI' scenario is presented in Figure 4.

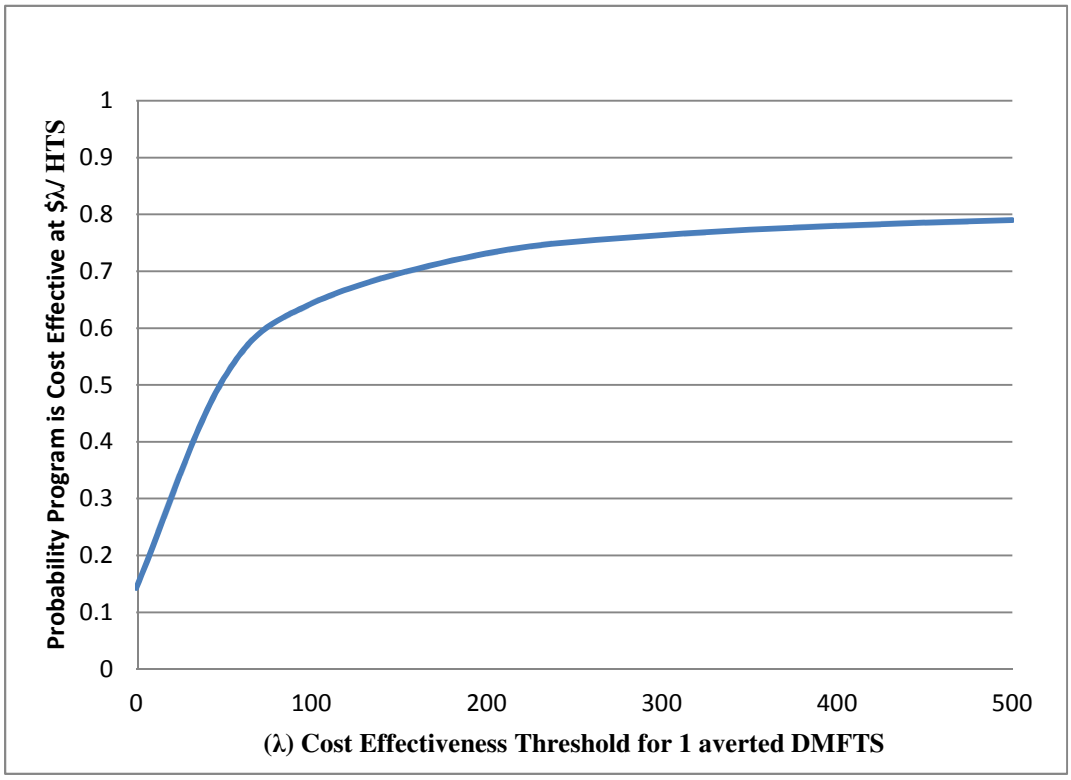

Figure 4: CEAC for Pragmatic Training Costs and Full treatment utilization

Discussion

Health economic evaluation of competing interventions becomes a relatively simple exercise when one comparator in an evaluation 'dominates' the others: it produces more health and entails lower overall costs. In this event, determination of the cost effective comparator is clear, the relevant question is whether the evidence base is strong enough to adopt the intervention, or further research should be required (Griffin et al., 2010).

This is not the case for the MI counseling program, which given the constraints of this analysis can be expected to improve the dental health of children and to increase the total costs of dental care provision over the first 3 years of children's lives relative to the comparator intervention. Without reference to some external standard it is not possible to say whether the program is cost effective or not. Under the decision making framework (Claxton, 1999) which is currently predominant in the UK (McCabe et al., 2008; NICE, 2009) calculation of critical cost effectiveness thresholds and ICERs is the role of an analyst, while judgment of program cost-effectiveness and appropriate willingness to pay levels for program health benefits is left to the relevant decision maker.

The chief advantage to performing a cost-utility analysis using the Quality Adjusted Life Year (QALY) as an outcome measure is comparability with other healthcare programs assessed using the same metric, and established (if informal) estimates of the cost effectiveness threshold for QALYs. Laupacis et al. (1992) introduced a cost effectiveness range of $20,000 - $80,000/QALY into the Canadian literature, while the $50,000/QALY estimate of the cost effectiveness of renal dialysis by Klarman et al. (1968) still hold disproportionate influence as a rule of thumb in Canada and the United States. Welfarist health economists , exemplified by Shiroiwa et al. (2009,) argue that the the cost effectiveness threshold to be used by decision makers ought to match public willingness to pay for health and health services. Alternatively it is argued that health maximizing decision makers should set the cost effectiveness threshold to match the marginal productivity of the healthcare system, such that adoption of cost effective interventions displaces less efficient health producing activities (McCabe et al., 2008).

Public willingness to pay for ECC prevention is not known, nor is the marginal productivity of the healthcare system in James Bay communities, Quebec, or Canada. Appeal to established $/QALY estimates may provide the best reference point against which to judge the cost effectiveness of the MI program despite the fact that no estimates of ECC QALY decrement exist. ECC caries the potential for severe acute and chronic effects, including pain, anxiety, poor sleep and developmental disruption. If $50,000/QALY is taken as a representative estimate of cost-effectiveness thresholds in Canada, the mean QALY decrement for an ECC case need only be 0.05-0.01 time discounted QALYs for the MI program to be considered cost effective relative to standard treatment. Given the severity of the disease, and the potential for systemic chronic health effects, this seems very plausible.

Additionally, common practice in cost effectiveness analyses to date has been to not explicitly consider public equity preferences with regards to health outcomes. To fill this void, it is common for decision making bodies (see: NICE, 2009 for an example) to allow informal adjustment of cost effectiveness rules to account for disparate preferences for equity or innovation not addressed by QALY maximization.

Relative to the median Canadian myriad equity claims may be made on behalf of children suffering from ECC in Cree communities in Northern Quebec. These claims are all value based, but depending on the preferences of decision makers and the public, claims could be plausibly justified on the basis of: the young age of affected children, their socioeconomic disadvantage and expected future health outcomes, the fact that ECC is a condition that almost certainly has poverty and socioeconomic disadvantage as a root cause, challenges relating to access to effective treatment for dental health decrements in remote communities, the special legal status of First Nations health in Canada, and the fact that health gains arise from information provision, which may be considered to be a prerequisite for rational decision making on the part of parents.

Birch and Gafni (1992; Gafni & Birch, 2006) argue that health funding is not often fungible, and urge explicit consideration of the local opportunity cost of program implementation in order to determine whether adoption of a intervention will increase health given existing budgets, and can thus

be considered to be cost effective in practice. If funding for the MI program is the product of a reallocation of resources within local health authorities, such that adoption of the program 'crowds out' other local activity, then the aforementioned equity claims may no longer apply, as equally disadvantaged groups may be affected by the removal of affected services. Likewise, if the MI program displaces time spend on effective prevention programs by community CHRs, the opportunity cost may be greater than the health gain provided by the program.

In practice the health gain attributable to the program and appropriate cost effectiveness threshold may depend on the source of program funding. It should also be noted that standard treatment may not represent the most cost-effective comparator for the MI program. Application of fluoride treatment to infant children (Lawrence et al., 2008) has recently been tested as a preventive measure in Canadian First Nations communities, while incremental consideration of the experimental MI program may identify a more cost effective implementation strategy.

Further Analysis

Further work is required before this analysis can be considered complete, including estimation of future dental treatment costs attributable to program effects (ie. Orthodontics), while the current estimate of a 'full dental treatment' scenario does not have clinical verification, and does not allow cost variability for extreme cases. Restriction of this analysis to a subsample (173) of the study population increases the potential for follow-up bias to impact the results, a risk that is increased due to the higher proportion of intervention cases excluded. Future work will use cost estimates from clinical staff and imputation strategies to perform analysis on the sample with dental exams (n~240), if not the entirety of the sample. Even under this scenario, not all potential program effects will be analyzed, including potential gains in dental health and resulting reductions in dental treatment costs past the follow-up period, adoption of better dental health habits within families, and program effects on non-health related public authorities.

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