

BMJ Open Online survey to assess parents' experience and preferences for follow-up visits for children living with type 1 diabetes in Quebec, Canada: a study protocol

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ABSTRACT

Introduction It is accepted that although patients may initiate a visit to a healthcare provider, follow-up visits are often based on recommendations from providers. This suggests that follow-up care, since not initiated by patients, may not reflect patients' perception of a need for care. However, few studies have examined the burden of regular follow-up care and patients' perceived value of such care. For parents of children with type 1 diabetes (T1D), follow-up visits are scheduled regardless of how well controlled the diabetes is. Our study examines how benefits and burden from the parents' perspective could affect their preferences in regard to the frequency of regular follow-up care.

Methods We aim to develop an online patient survey to be distributed to parents of children living with T1D in the province of Quebec, Canada. The survey will be available in French and English, and distributed through diabetes clinics, on social media groups and forums for parents of children with T1D. The survey will be developed in collaboration with parents of children with T1D to ensure that it appropriately reflects the services in regular follow-up care and that the language is understandable and clear.

Ethics and dissemination All participants will be informed of the requirements and objectives of the survey at the beginning of the questionnaire and that the data collected will remain anonymous and confidential. Ethics approval for the study was obtained from the research ethics committee of the CHU de Québec-Université Laval. Results of the study will be shared with relevant stakeholders with the aim of improving practices and better meeting patients' and families' needs.

INTRODUCTION

Although patients may initiate a visit to a healthcare provider, follow-up visits are often based on recommendations from providers which may be determined by clinical guidelines, their clinical judgement and incentives.¹⁻³ Generally, the most recommended approach is to schedule a clinical visit with the endocrinologist every 3 months.¹⁻³ Since follow-up care is not initiated by patients,

Strengths and limitations of this study

- This is the first study to examine parents' experience and preferences for follow-up care for children living with type 1 diabetes.
- A patient was engaged in the study design and in the questionnaire development to increase relevance and ease of completion.
- Online bilingual survey design enables diversity in the participation on a geographically spread territory.
- Ensuring participation is representative of actual population may represent a challenge.

service utilisation may not reflect patients' perception of a need for care or their preferences,⁴ but instead what physicians believe to be best.⁵ However, the best care may require adapting guidelines to patients' preferences in the clinical setting.^{6,7} There is indeed a growing evidence of the value of involving patients in chronic disease management and in treatment decisions, which is associated with better adherence to treatment, better health outcomes and a more appropriate utilisation of health services.⁸⁻¹¹ Although shared decision-making is becoming increasingly common in adults, it is less so in paediatric populations. In children, the effects of a chronic condition are on the children affected and on the parents, who play an important role in the disease management as informal caregivers and bear a large part of the burden. Many studies have associated caregiver burden with poor health outcomes for both the recipient and the caregiver.¹²⁻¹⁴ As such, recent studies have examined parents' preference in long-term follow-up. In cancer, preferences were found to vary for the type of provider (paediatric oncologist vs family doctor) that should be providing the follow-up.¹⁵ In a study on type 1 diabetes



(T1D), parents expressed their desire for more tailored care.¹⁶ Based on their findings, authors of a recent study about parents' preferences regarding education for long-term complications of T1D recommended that parents' role be acknowledged and that they be provided with the level of information that meets their needs, which includes an appropriate timing of information provision and credible resources.¹⁷ These recommendations highlight the need to individualise follow-up care rather than taking a 'one size fit all' approach. Such recommendations are aligned with findings from a systematic review on parents' learning needs for management of their children's chronic disease: information needs differ from one to the other in terms of amount, content, communication modality, source of information and parents' motivation for wanting information.¹⁸ Hence, it is important to consider how chronic disease management could be adapted to preferences of parents of children with chronic diseases.

After initial phases of diagnosis, education on diabetes management and adaptation of a family's routine comes the routine follow-up care. Children have scheduled visits every 3 months, typically with an endocrinologist, but sometimes also with other health professionals, to support appropriate management and control of the diabetes. The frequency of these visits is generally higher than with adults, which could be due to lower diabetes control in children. However, regular follow-up care is also a disruptor for both parents and children, for instance, in terms of stress¹⁹ and of absenteeism from work for parents²⁰ and from school for children.²¹ These visits are scheduled regardless of their value, which may differ depending on the child's condition and the usefulness of information received, but also on the burden that these visits impose on parents. Some parents continue to feel stressed years after their child's diagnosis²² and may need more frequent visits to be reassured. Our study examines the benefits and the burden from the parents' perspective and how these could affect their preferences in regard to the frequency of follow-up visits with an endocrinologist. The goal of this study is to understand the potential heterogeneity of families' preferences and eventually translate these findings into practice to better meet patients' needs.

Conceptual framework

The study is based on Grossman's theoretical framework of demand for healthcare.²³ Grossman²³ defines the demand for healthcare as derived from the demand for health. As such, consumption of healthcare services would reflect a perception by the consumers that these services are beneficial to maintain or improve their health. The benefits that people estimate receiving from consuming healthcare services may vary depending on their demographic characteristics (age, sex), their socioeconomic status (education, income) and their health status. The decision to consume services also depends on the costs associated with such consumption. Such costs include the

direct medical (such as copays) and non-medical (such as transportation to the appointments) costs, and the indirect costs (absenteeism from work and loss of revenue). Hence, consumption of services would result from an estimation of the perceived costs and benefits. Such estimates may be subject to a high degree of uncertainty in many healthcare situations. However, as individuals gain experience with a chronic condition, they may acquire a better ability to self-manage. Their capacity to appropriately estimate the benefits from using services and the associated costs may also improve.

According to Grossman's model, even individuals with the same characteristics could make different choices about their health. Here, the knowledge of a person is an important factor when making a decision about health.

In the context of diabetes management, results from empirical studies support Grossman's theoretical model in regard to the effect of health literacy,^{24 25} and that people who have a better capacity to self-manage will spend less time on activities related to the disease yet with a better health outcome than a person with less knowledge about the disease.²³

In the context of our study, a parent's knowledge and capacity to self-interpret results from regular diabetes tests could also affect their perception of the benefit of having a visit with an endocrinologist, which generally includes discussion of test results. Based on Grossman's model, we hypothesise that parents would want to lengthen the time between visits in the following situations: (1) their child's condition is under control and (2) they have the ability to adjust insulin doses independently. Costs associated with visits and household income could also affect parents' preferences. Household income may however be correlated with higher health literacy and ability to manage the child's T1D, which may translate in a lower value for the visit.

METHODS AND ANALYSIS

Study context

The study examines the experience of parents in the regular follow-up care of their child with T1D in Quebec. With over eight million people, Quebec is the second most populous province in Canada. Its population is predominantly French-speaking, with French being the only official language. Most residents live in urban areas between Quebec City and Montreal, with about half of the province's population living in the Greater Montreal Area.

Quebec has a universal public health insurance (called Régie d'assurance maladie du Québec (RAMQ)) that covers hospital and medical services for all citizens and permanent residents, as required by the Canada Health Act.²⁶ Covered services are free at the point of service. Public coverage for non-medically necessary services and for prescription drugs varies across provinces. The RAMQ administers over 40 programmes for specific needs, and

there is an insulin pump reimbursement programme for children living with diabetes.

Sample and setting

The study will consist of a cross-sectional survey. The survey will be available to potential participants via a link to the online page where the survey will be hosted. This distribution method and the online mode of data collection will make participation simple and will avoid downfalls associated with a paper version of the survey, such as data entry errors. There is currently no registry of children diagnosed with T1D, rendering contacting eligible participants directly impossible.

The study population will consist of parents of a child living with T1D residing in Quebec and receiving long-term follow-up from any healthcare organisation in Quebec. Participants must be able to read and understand either written English or French, comprehend the information provided and have access to the internet (the survey will only be available online). In Canada, about 1 in 300 children live with diabetes (type 1 and type 2).²⁷ A national estimate of T1D is not available, but it is assumed that approximately 90% of children living with diabetes have type 1. In the absence of data on the exact number of children with diabetes in Quebec, we will assume that the prevalence is similar across provinces. According to Statistics Canada, there are 1 725 842 children in Quebec.²⁸ Based on the prevalence of diabetes in children in Canada of 0.3%,²⁸ we estimate the number of children with diabetes (all types of diabetes included) at 5177. Assuming that only one parent would answer the survey for each child living with T1D and that there is no more than one child living with T1D per family, the study will require a sample of 358 participants, based on a $\pm 5\%$ margin of error, at the 95% confidence level.

Survey development and validation

Questionnaire development will be referring to the principles of the Consumer Assessment of Healthcare Providers and Systems (CAHPS)²⁹ to certify the quality, comparability and consistency of questions. The CAHPS is considered a reference for patient experience surveys. First, questions must be consistent with the care path being studied. Thus, the questions will be developed with reference to the current clinical guidelines. Also, several measures will be taken to ensure that the respondents to the questionnaires have experienced the care path studied. Therefore, recruitment will target patients of organisations that are related to the study population (details on the modalities of recruitment can be found in the Recruitment section). In addition, the survey will include questions about the location of follow-up (to eliminate potential respondents receiving care outside of Quebec) and about the type of diabetes of the child (parents whose children have type 2 diabetes will be excluded from the study). Type 1 and type 2 are clinically different and the clinical guidelines for follow-up differ. Moreover, the aspects that will be evaluated are meant

to be aligned with those that would be important for the respondents, such as the impact on daily activities (work, school) and on the well-being of the child.

The questionnaire will be developed by a research team composed of a health services researcher, a patient-partner who received specific training and who is a parent of a child living with T1D, and a physician. The survey development will be an iterative process, combining information on care processes and on disease burden from the literature (peer-reviewed publications, grey literature and clinical guidelines), with the experience of the patient-partner. Health workers and other professionals working in a field relevant to the study (camp, school, associations and so on) will also be consulted informally. The structure of the questionnaire will be aligned with the elements from the conceptual framework.

The validation of the items will be an iterative process. First, the research team will review each question to ensure the coverage of all aspects to be investigated and to exclude irrelevant content. The process should ensure that no important questions are omitted (confirm the validity of the content) and that the questionnaire follows the principles of the CAHPS for the development of patient survey questionnaires.

Then, the questionnaire will be tested with parents who have a child living with T1D who meet the inclusion criteria of the study. The purpose is to ensure that the meaning of each question is clear, the response items are comprehensive and there are no irrelevant items (intelligibility).

The survey will be developed in French and then translated into English. The list of items will be composed mostly of closed questions (yes/no items, Likert scales and nominal items) and a few open questions (free text box) to simplify data analysis. Revisions will be made to ensure that the final version is easy to understand for respondents.

There is no similar instrument for which the patient experience in the long-term follow-up of T1D in children survey could be compared against; therefore, it will not be possible to measure convergent validity. An online version of the survey will be generated for this study and we will use SurveyMonkey to distribute the survey online.

Recruitment

To avoid selection bias, multimodal recruitment strategies will be used to inform potential participants of the study.

The first strategy is to recruit participants by directly communicating with parents of children living with T1D through a patient who is an integral member of the research team. The family involved in this study has a child living with T1D and a network of parents who have a child with a T1D diagnosis in Quebec. These parents have informal meetings, as well as other means of communication where the patient can promote the study. She also attends events organised for people concerned with T1D.

The second strategy is indirect recruitment. To do this, organisations with a patient contact list who meet the

inclusion criteria of the study will be contacted to participate in the distribution of the survey. In Quebec, mainly three university hospital centres (CHU) provide regular follow-up care of children with T1D: the CHU Sainte-Justine and Montreal Children's Hospital, both located in Montreal, and the CHU de Québec-Université Laval, located in Québec City. For these organisations, meetings will be scheduled with different clinicians (endocrinologists and paediatricians) to obtain permission to hang posters in the waiting rooms of diabetes clinics. We will ask diabetes clinics for their support in disseminating the information about the survey. Other organisations that will be contacted include associations such as Diabetes Quebec, the Diabetes Summer Camps (the Diabetic Children's Foundation) and the Juvenile Diabetes Research Foundation. Organisations will be invited to share a link to the survey on their websites and to share an email about the study to their contact list.

The third strategy is to promote the study on websites and forums for patients and parents that are concerned with T1D. We will search for groups on Facebook with keywords such as 'diabetes type 1' and 'juvenile diabetes', in French and in English, with no geographical limit. A few questions at the beginning of the survey will be used to ensure that respondents meet all the inclusion criteria.

The survey time frame

The study was planned to begin in the spring of 2018 and completed in the fall of 2019. Questionnaire development is planned over a 3-month period. An initial review of the literature and clinical guidelines will be conducted to identify (1) current practices for T1D management, (2) identify clinical outcomes relevant to T1D management, (3) individual characteristics that could affect T1D management, and (4) measures and costs and benefits for patients and families. A first version of the questionnaire will be developed and shared with the patient-partner and will be followed with a think aloud meeting. Additional rounds of revisions with think aloud meetings will be conducted until a consensus is reached. Testing with parents of children with T1D to ensure clarity of the wording and understandability of the questions is planned to occur in the month following the development phase. Once the questionnaire is finalised, it will be translated and transferred on an online platform. Online data collection will be continued for a 6-month period or until a satisfactory number of respondents is reached. A data cleaning and analysis phase will follow.

Planned analysis

Once the survey is closed, survey data will be exported from the SurveyMonkey platform into a statistical analysis software such as Stata for data cleaning and statistical analysis. Respondents who do not meet the inclusion criteria will be excluded.

The study will include a descriptive component and an analytical component. Our main outcome of interest is the preferred time (in months) that separates two visits with

the endocrinologist. Other outcomes of interest are the burden and the benefits of follow-up visits, as perceived by parents. Benefits refer to the value of the information provided by the endocrinologist to manage the disease. The value may be indirectly measured through the number of years with the condition, where a family with more experience is expected to perceive less benefit and hence prefer spacing out visits. The burden will be estimated from direct cost data (eg, travel to appointments) and from indirect costs (eg, opportunity cost, stress).

First, we will provide descriptive information on the study sample. By comparing the descriptive information from the study participants with descriptive data on the target population, we will know how representative our sample is. Data on the target population will be derived from the population 2016 census data for the province of Quebec.

The descriptive data will include average and standard deviations (SD) on various elements collected through the survey in terms of the burden of follow-up care (direct and indirect costs) and in terms of benefits (usefulness of information).

Second, we will analyse how perceived benefits and burden could affect parents' preferences on the time laps between follow-up appointments. The nature of the outcome suggests that we may be using an ordered Probit regression. However, careful examination of the characteristics of the data will be needed to ensure that appropriate methods are used.

Patient and public involvement

The idea for this study originated from comments of parents of children with T1D. The comments were made at a symposium on T1D in Québec City in Spring 2018, in which authors of the present study participated. A parent of a child with T1D, hereafter referred to as a patient-partner, was involved from the initial steps of the study. The research team, including the patient, was having a preliminary meeting to discuss future research, learning from the experience of the symposium. The theme of the follow-up care and specifically the regular appointments with the endocrinologist came up. From the patient-partner's point of view, it appeared that the way in which this follow-up care was structured and in place was not responsive to patients' needs. In a back-and-forth of discussions, the research team identified questions of interest and a study design. The patient-partner was involved throughout the development of the study design and the questionnaire, as well as in the recruitment strategy for participation.

The patient-partner will have a crucial role in the recruitment and in the dissemination of the results, thanks to a strong network in the T1D community, including with local clinicians and with local groups of parents of children living with T1D. Once the results are available, the research team will work on developing a product, which could take the form of a Prezi presentation or a video, in lay language that the patient-partner will be able to share

on social media groups, with diabetes associations and with clinicians.

ETHICS AND DISSEMINATION

All participants will be informed of the requirements and objectives of the survey at the beginning of the questionnaire and that the data collected will remain anonymous and confidential. No personally identifiable information will be collected. Completion of the survey will be considered as provision of voluntary and informed consent. A revision of the study protocol with the final version of the questionnaire will be submitted for approval. Data collection will begin when the amendment to the study protocol with the questionnaire is approved by the research ethics committee. Results will be disseminated through scientific and non-scientific media. First, the research team will present results in relevant scientific congresses and will submit a manuscript to a peer-reviewed journal. Second, the team will develop a product that is specifically designed to key stakeholders in paediatric diabetes, including patient associations.

Contributors ML and MR codesigned the study, including the research question and the methodology. MR has a strong network in the paediatric diabetes community in Quebec. MB, as a trained MD, has medical knowledge of diabetes. MB wrote the initial version of the protocol's methods and analysis. ML wrote the introduction, the patient and public involvement, and the ethics and dissemination sections. MR provided revisions until the final version, which was reviewed and approved by all authors.

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Competing interests None declared.

Patient consent for publication Not required.

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Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement There are no data in this work.

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