ECONOMIC CONSIDERATIONS OF PATIENT DECISION AIDS:

A CASE STUDY IN SLEEP APNEA

by

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Abstract

Background: Patient decision aids (PtDAs), tools used to facilitate shared decision-making, help improve patient-physician communication and the quality of healthcare decisions. Over 500 PtDAs are available, yet implementation of these tools has been limited. In order for decision-makers to implement new health care interventions such as PtDAs, they require rigorous economic evidence demonstrating that such interventions provide value for money.

Objectives: To explore the economic consideration of PtDAs by (1) systematically reviewing PtDA trials that have evaluated economic outcomes, (2) exploring the potential cost-effectiveness of a PtDA for individuals with obstructive sleep apnea (OSA), and (3) describing the development of a an OSA PtDA prototype.

Methods: PtDA trials evaluating economic outcomes were systematically reviewed through an electronic search of Medline/PubMed, Embase, CINAHL, and PsycINFO databases. The potential cost-effectiveness of a PtDA for OSA was evaluated through a Markov cohort decision-analytic model, which explored the cost-effectiveness of a PtDA compared to usual care. Finally, an OSA PtDA prototype was developed according to the International Patient Decision Aid Standards (IPDAS) criteria.

Results: Our systematic review found that PtDAs will likely increase upfront administration costs, but may decrease short-term costs by reducing the uptake of invasive treatments. Most studies did not comprehensively capture long-term costs and

health outcomes appropriately. Through our economic modelling of a PtDA for OSA we found it could be a cost-effective use of resources provided it increases adherence to treatment. However there was considerable uncertainty in this estimate, with expected value of information analysis revealing that additional research is warranted. We developed and tested a prototype OSA PtDA, and found no evidence that users became stuck or experienced errors during usability testing. The majority of users found the PtDA easy to use and worthy of recommending to others.

Conclusions: Policy-makers lack sufficient economic evidence to make informed decisions about whether and where to invest in PtDAs. This evidence gap could be a factor contributing to the slow implementation of PtDAs. Using OSA as a case study, this work demonstrates an economic modelling framework that can be used to evaluate the potential cost-effectiveness of PtDAs.

Preface

I was involved in the conceptualization, study design, data collection, analysis, and interpretation of all three studies in this program of research. I performed the systematic review and data synthesis which is part of Chapter 2, with support from Dr. Nick Bansback and Dr. Stirling Bryan. The OSA cost-effectiveness model, described in chapter 3, was originally published by Dr. Mohsen Sadatsafavi to compare treatments for OSA. I updated this model with new evidence with the support of clinicians Dr. Najib Ayas and Dr. Fernanda Almeida, and researchers Dr. Carlo Marra, and Dr. Larry Lynd, and made substantial changes to its structure to include the decision aid in with support from my supervisors, Dr. Bansback, Dr. Bryan and researcher Dr. Dawn Stacey. I then conducted all the analysis. I was also involved in the evidence synthesis and subsequent development of the OSA PtDA and DCIDA software platform upon which it is based. I was supported by Dr. Bansback, Dr. Bryan, Dr. Ayas and Dr. Almeida, software developer James Hicklin, and graduate student Sarah Munro. Usability testing required approval from the UBC Behavioural Ethics Board (H14-01429).

Table of contents

List of tables

List of figures

List of abbreviations

BC: British Columbia
CEAC: cost-effectiveness acceptability curve
CI: confidence interval
CINAHL: cumulative index to nursing and allied health literature
CPAP: continuous positive airway pressure
CV: cardiovascular
CVD: cardiovascular disease
CUA: cost utility analysis
EVPI: expected value of perfect information
EVPPI: expected value of partial perfect information
EQ-5D: euroqol 5-dimensions
GP: general practitioner
HRQoL: health-related quality-of-life
HRT: hormone replacement therapy
ICER: incremental cost-effectiveness ratio
MAIS: maximum abbreviated injury scale
MAS: mandibular advancement splint
MeSH: medical subject headings
MI: myocardial infarction
MVC: motor vehicle crash
OA: osteoarthritis
OSA: obstructive sleep apnea
PtDA: patient decision aid
QALY: quality-adjusted-life-year
QoL: quality-of-life
RAND: research and development
SDM: shared decision making
TJA: total joint arthroplasty
UK: United Kingdom
WTP: willingness-to-pay

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1 Introduction

This thesis considers patient decisions aids (PtDAs) – the key tools for engaging patients to be involved in their own treatment decisions. There are over 500 PtDAs for different clinical decisions, and an increasing interest in their use, as demonstrated by a 611% increase in publications over the past ten-years.(1) Policy-related activities in 13 countries have been designed to implement PtDA across the healthcare continuum,(2) the most notable example being provisions in the Affordable Care Act in the US.(3) However, there is little evidence that these policies have led to an increase in their use. One potential reason for this is the lack of economic evidence to support their use. In this thesis, I explore my interest in both health economics, and patient decision aids. I aim to bring an economic lens to the consideration of PtDA design and implementation. Implementing PtDAs will require upfront costs, and policy makers require evidence that these costs provide value for money. I will begin by exploring the patient-physician relationship and the theoretical rationale for PtDAs, followed by the economic arguments for making resource allocation decisions. This chapter will conclude with an identification of knowledge gaps and a set of research questions.

1.1 The patient-physician relationship

The traditional model of decision-making between the patient and physician is one of paternalism, in which the physician makes treatment decisions based on what he/she feels is in the patient's best interests. Patient involvement is either absent or

"limited to providing consent to the treatment advocated by the physician."(4) Over the past 30 years, there has been shift in the desire for a more patient-centred approach. This has been advocated by patients, physicians, and medical ethicists in recognition of the importance of engaging patients in their own healthcare decisions.(5) The shift away from paternalism is important also given the growing number of 'preference-sensitive' treatment decisions where there is no clear best option, and in the context of emerging evidence indicating that physicians are often poor judges of patient preferences.(6) Examples of preference-sensitive healthcare decisions can include choosing between lumpectomy and mastectomy for treatment of breast cancer, or between heart surgery and angioplasty for chronic angina. Further, modern patients are active and informed 'consumers' of healthcare and are demanding greater patient-centred care.(7)

1.1.1 Information asymmetry and the agency paradigm in healthcare

In healthcare, information asymmetry exists between the patient and physician. Physicians hold knowledge about disease etiology, diagnosis, prognosis, treatment options, and outcome probabilities, while patient have expertise in the experience of their illness, social circumstances, risk attitudes, values, and preferences.(8) Information asymmetry can lead to poor decision-making. For instance, a physician may fail to understand his/her patients' preferences and recommend an inappropriate treatment, a concept known as a 'preference misdiagnosis.'(6) An example from breast cancer found

that while "doctors believe that 71% of patients rate keeping their breast as a top priority, the figure reported by patients is just 7%."(6) Alternatively, a patient may make a poor decision because he or she does not fully understand the potential benefits and harms of the treatment options.

In economic theory, poor decision-making as a result of information asymmetry can lead to a 'market failure,' meaning the market fails to allocate resources efficiently.(9) In healthcare specifically, market failure can result from information asymmetry between provider and patient where the consumer (patient) lacks information about the service (health care) and relies on the supplier (physician) to act in his or her best interest.(10) This imbalance of power, where the physician is in a position to create demand for the service, is a concept known as 'supplier-induced demand.'(11) As stated by Mwachofi and Al-Assaf, "in this case the supply and demand are jointly determined by the same individual at the same time which can result in market failure."(10) One of the most widely cited examples of supplier-induced demand in health care is elective surgery, where rates are known to vary widely and cannot be explained solely by population demographics or differences in informed patient preferences.(12,13) Observers note that these unwarranted variations in care arise because "medical opinion rather than patient preference tends to dominate the treatment choice."(14) Reducing information asymmetry between the patient and physician helps separate the supply and demand sides of the market. It requires that the

physician communicate information about the disease, treatment, and expected outcomes to the patient, and the patient express their preferences to the physician.

'Agency theory' is a paradigm commonly used to explore and explain models of decision-making between patient and physician, and problems associated with these models.(15–18) Agency theory seeks to explain the behaviour of self-interested parties who have conflicting goals under uncertainty and incomplete information.(19,20) Applying the agency paradigm to healthcare is appealing because it is supported by a considerable body of research in the field of economics.(15,18) Three main models have been used to describe the patient-physician relationship, the 'perfect agent,' 'informed decision-making,' and 'shared decision-making' models.(21) These models can be contrasted based on the information exchange between patient and physician, and who makes the final decision (see Figure 1).

Figure 1. Models of patient-physician relationship

Model	Information Exchange	Decision
Perfect Agent	Physician ← Preferences — Patient	Physician alone
Shared Decision Making	Physician ⇄ Medical Information / Preferences — Patient	Physician and patient
Informed Decision Making	Physician → Medical Information → Patient	Patient alone

1.1.2 The physician as 'perfect agent'

The perfect agent model illustrates a paternalistic relationship between physician and patient. In it, the patient delegates decision-making authority to the physician, who elicits the patient's preferences about treatment options and makes a decision on the patient's behalf based on her knowledge and experience.(22) The physician does not make decisions that maximize her individual gains; rather, the physician as a "perfect agent" is committed to fulfilling the patient's goals. As stated by Evans, "if this agency relationship were complete, the professional would take on entirely the patient's point of view and act as if she were the patient."(20)

This model is firmly rooted in theory; however, it is widely accepted that the perfect agency relationship does not exist in health care. In clinical practice, preference elicitation is an imperfect process and often impractical due to time constraints and other factors.(21) Further, even if the physician fully understands his or her patient's preferences, the physician and informed patient may reach different conclusions about which treatment option is best. This divergence may result from the physician and patient's subjective interpretation of clinical risks and benefits. The perfect agency relationship is also influenced by the fact that physicians possess their own self-interests, which may conflict with those of their patients. As stated by Evans, "perfect agency would also require the use of that information solely in the patient's interests, at

complete disregard for her own. In fact, such complete selflessness is rarely found, among professionals or anyone else."(20)

1.1.3 Informed-decision making model

In the informed decision-making model, the patient holds decision-making authority and the physician acts to provide the patient with sufficient information to make an informed choice. While the challenge in the perfect agent model is ensuring the physician elicits and incorporates the patients' preferences into the treatment decision, the challenge in the informed decision-making model is ensuring the physician provides the patient with clear, understandable, and unbiased information.(21) This model has been criticized as it requires the patient to assume decision-making responsibility without guidance from the physician, which may result in increased anxiety for the patient or feelings of abandonment.(23,24)

1.1.4 Shared decision-making

Both the perfect agent and informed decision-making models attempt to engage patients in the decision-making process. In the former, the physician explicitly takes into account patient preferences, and in the latter, the patient retains decision-making authority. However, as noted above, these two models represent theoretical ideals that are limited in their applicability to the 'real-world' clinical encounter. As a result, shared decision-making (SDM) has emerged as the preferred model of care.(3,25,26)

Charles et al. identify four key characteristics of SDM: 1) that at least two participants be involved – the patient and physician, 2) that both parties share information, 3) that both parties take steps to build a consensus about the preferred treatment, and 4) that an agreement is reached on the treatment to implement.(27) As hinted by this definition, shared decision making represents a 'middle-ground' between the perfect agent and informed decision-making model,(28) where the information exchange is bidirectional and the decision is shared between the two parties. It views the exchange as a partnership between two experts, with the physician possessing expertise in the medical condition and treatment options, and the patient in his or her experience of the condition and their preferences.(29) One of the strongest arguments for SDM is based on the ethical imperative that patients should make informed choices free of coercion by deliberating and make trade-offs between the potential benefits, harms, and costs.(30) Beyond the ethical argument, evidence suggests SDM can result in a number of benefits for patients, such as reduced anxiety and decisional conflict, and improved patient-provider communication.(31)

Importantly, studies have demonstrated that a proportion of patients prefer that their physician make the decision for them.(32,33) It is accepted that there is no single model of care that is ideal for all patients and physicians, and "there is nothing wrong with a case where both patient and doctor, after discussing alternative decision-making approaches, agree they prefer a paternalistic treatment decision-making approach."(5) It

is suggested that successful SDM "is part of the ongoing doctor-patient dialogue, one in which doctors introduce patients to greater involvement and respond to patient cues for decisional responsibility."(34) An ongoing dialogue about preferred level of patient involvement can be important as evidence suggests that patient preferences for communication with their physician, and preferences related to the condition and treatment specifically, change over time.(35,36) Evidence suggests that physicians do support the idea of tailoring the consultation to the patient's preferred level of involvement and specific decision context.(34)

1.1.5 Shared decision-making in practice

While SDM may not be relevant for every decision-making context, it is rarely used in practice where it should be relevant.(37,38) SDM is relevant when the scientific evidence on treatment options is limited or conflicting, or if the decision is "preference-sensitive," that is, the benefits and harms of multiple options are fairly balanced and the treatment decision comes down to patient preferences. In the case of end-stage hip and knee osteoarthritis, for instance, patients must choose between conservative management with pain medication and exercise or total joint arthroplasty (TJA). While TJA is more effective at reducing pain and improving mobility, it carries a greater risk of serious surgical complications, such as infection, or even death.(39) As such, the best option for each patient will depend on their preferences.

Despite a supportive policy environment and increasing evidence of health and well-being benefits, is it estimated the SDM only occurs in 10% of consultations.(7) Time is the most significant barrier to SDM, as physicians believe it lengthens the consultation.(40) Medical education is another barrier, as it has failed to adapt to greater patient engagement, with observers noting that there is "no natural place for SDM in their medical interview script."(7) Power dynamics in the physician-patient relationship may also play a role. Patients often feel intimidated, and may be reluctant to engage with their physician for fear of compromising their relationship.(41) Compounding the issue is confusion about what actually constitutes SDM. Physicians often believe they are doing SDM, when in fact they are not.(30)

1.1.6 Patient decision aids (PtDAs)

The most widely used interventions to promote SDM are patient decision aids (PtDAs). They are defined as "tools designed to help people participate in decision making about health care options, with the goal of promoting deliberation between patients, health care providers, and others about those options. They provide information about the options, and help patients construct, clarify, and communicate the personal values they associate with the different features of the options."(42) With respect to information asymmetry, they act both to increase patient knowledge and elicit their preferences. While PtDAs are tools that can be used to support SDM, critics

note that their use alone is insufficient to ensure SDM occurs.(43,44) Importantly, PtDAs may act as barriers to SDM if a patient decides on his/her preferred treatment prior to consulting with his/her physician.

1.1.7 Benefits of PtDAs including the impact on treatment adherence

Despite these criticisms, it is widely accepted that PtDAs improve patient-physician communication. The Cochrane Systematic Review of PtDAs updated in 2014 found that compared to usual care, PtDAs have been shown to increase knowledge, result in more accurate risk perceptions, increase the proportion of people making choices that are congruent with their values, decrease decisional conflict, increase the proportion of people engaged in decision-making, and decrease the proportion of patients who remain undecided.(31) While PtDAs may lessen the time burden of SDM by reducing the need for physicians to provide information to their patients, trials show that compared to usual care, PtDAs increase consultation time by a median of 2.6 minutes, though this ranges from 8 minutes shorter to 23 minutes longer.(31)

It has been proposed that PtDAs have the potential to improve treatment adherence given that they help improve patient knowledge, involvement in decision-making and patient-provider communication.(45) 'Adherence' has replaced the term 'compliance' is recent years, as compliance "betrays a paternalistic attitude towards the patient on the prescriber's part."(46) By comparison, adherence is patient-centred and

views the patient and clinician as collaborators that exchange, discuss and negotiate.(47) It is clear that while compliance is associated with a paternalistic model of care, adherence is aligned with the principles of SDM. Adherence can be used to describe many phenomenon, such as adherence to guidelines, or adherence to a decision. Herein I use adherence to describe adhering to therapy.

It is argued that SDM through the use of PtDAs may increase health system efficiency by reducing unwarranted variations in care.(48) This could involve reducing the use of services that are unwanted or not valued, or increasing the use of services that are. In spite of these and other benefits, resources are required to deliver PtDAs. As such, investments in PtDAs need to be weighed against potential alternatives.

1.2 Resource scarcity

All health care systems operate in an environment of resource scarcity. Demand for health services always outstrips available resources, as even in an environment with infinite financial means, people, time, facilities, and knowledge are scarce.(49) Decision-making in this environment requires consideration of opportunity costs – that is, what "alternative investments could be made with the same health-care resources."(50) In other words, policy-makers are tasked with ensuring that the opportunity costs are lower than the costs and benefits afforded by opportunities that are taken. New

demand for health resources can come by the way of new drugs, technologies, or changes in practise – such as the integration of PtDAs in the context of SDM.

1.2.1 Economic evaluation to inform resource allocation decisions

Resource allocation decisions are challenging, but there are a number of tools available to help. Economic evaluation is one of the most commonly used tools. It is defined as the "comparative analysis of alternative courses of action in terms of both their costs and consequences."(49) Economic evaluations can be performed as part of a trial to provide insight into the potential cost-effectiveness of the program under evaluation. However Sculpher et al. note a number of limitations of trial-based economic evaluations, such as an inability to compare all relevant options, a truncated time horizon, and a failure to incorporate all existing evidence.(51) For this reason, it has been proposed that "the use of decision analytic models, coupled with full evidence synthesis, is the only framework that has the potential to meet all the requirements for economic evaluation for decision making."(51) Decision models enable the combination of different sources of evidence in appropriate time-horizons to examine the impact of hypothetical scenarios on long-term costs and benefits. As such, trial and model based methods should not be thought of as competing alternatives, but rather complementary techniques.

There are a number of different types of economic evaluation, including cost-benefit analysis, which measures costs and benefits in monetary terms, and cost-effectiveness analysis, which measures health benefits in natural units such as life years saved or cancers prevented.(52) However the most widely used method of economic evaluation is cost-utility analysis (CUA) (although it is often referred to as cost-effectiveness analysis).(53) It measures benefits in terms of quality-adjusted-life-years (QALYs), a metric that combines quantity and quality of life.(49)

As stated by Torrance, "the lifetime of an individual can be considered as consisting of two major components: quantity of life and quality of life."(54) While measuring quantity of life is easy, measuring quality of life is challenging. There are a number of tools available to measure health-related quality of life (HRQoL), including general (focused on overall health and well-being) and condition-specific instruments. In order to generate QALYs for economic evaluation, general measures are used because they provide a single value that represents HRQoL, and can be compared across conditions.(55) This value is often elicited through the utility approach to measuring HRQoL, which measures the "desirability or preference that individuals exhibit for the condition."(54) The strength of preference for specific health states is most often measured on a scale between 0 and 1, with 1 representing perfect health and 0, death. This value, multiplied by the quantity of life (amount of time in this health state) produces QALYs. This measure of benefit is incorporated with costs in the

incremental cost-effectiveness ratio (ICER), which is the incremental costs of the alternatives under consideration divided by incremental QALYs.(56)

1.2.2 Decision making using cost-utility analysis

The resulting ICER can be used to inform two types of decisions that policy-makers consider. The first is whether or not the technology or program under evaluation should be adopted. This 'adoption' decision should be based on "the expected (mean) cost-effectiveness of the technology, given the existing information."(57) It considers the incremental cost and QALYs for the program under consideration relative to current practice. Generally speaking, programs providing greater benefit at less cost than the alternative would be adopted, while those providing less benefit at a greater cost would not. The decision is more challenging when a program provides greater benefit at increased cost, or less benefit at less cost. In these cases, policy-makers must consider whether the additional benefit provided is worth the additional cost, or in the latter case, whether the opportunity cost of continuing current practice is greater than potential alternatives.

The adoption decision cannot be made without considering the quality of evidence used in this cost-effectiveness estimate. Despite a program having a favourable cost-effectiveness estimate, there may be considerable uncertainty. The

second decision facing policy-makers is called the 'research' decision, and focuses on whether to "demand further research to support adoption (or rejection)."(57) Often the two decisions are made in parallel. For instance, policy-makers may decide not to adopt on the basis of the CUA results, but suggest that future research is warranted given uncertainty in the estimate.

In an ideal world, all technologies and programs would be evaluated through CUA, and policymakers would simply rank all available options by their respective ICERs from lowest to highest, and fund the programs until their budget runs out. In reality it is unfeasible to evaluate all potential programs and technologies through CUA. As such, decision-making often relies on a cost-effectiveness threshold, with programs falling under the threshold considered cost-effective. The threshold is estimated to be $50,000 per QALY,(58) though in reality no jurisdictions have implemented an explicit threshold(59) as decision-makers make adoption decisions using additional factors such as equity and burden of disease.(60) The threshold approach to determining cost-effectiveness is not without limitations. It has been criticized for assuming perfect divisibility of health care programs, constant returns to scale, and constant marginal opportunity costs.(61) In summary, CUA results are one tool, albeit an important one, that can help decision-makers ensure that resources are deployed in the most efficient manner.

1.3 Overall aim of thesis

Despite the potential benefits offered by PtDAs, implementation of these tools has been sparse.(62) Health service decisions are not made strictly on evidence of effectiveness, but also on whether investments provide value for money. A number of high-profile commentaries in the NEJM and BMJ have suggested that SDM through the use of PtDAs could help improve the quality of health care decision-making while reducing costs.(3,6) In this thesis, I aim to explore the economic evidence of PtDAs. I began by systematically reviewing PtDA trials that have evaluated the influence on economic outcomes of PtDAs, including both costs and benefits measured in QALYs. This identified a number of knowledge gaps, including the fact that the vast majority of economic evidence of PtDAs were trial-based evaluations that are limited by a short time frame, and that contexts where self-management was important had not been widely explored.

To address these gaps, I next explored the potential cost-effectiveness of a PtDA using a model-based economic evaluation. I have chosen to focus on adults with obstructive sleep apnea (OSA) as a case study since it is a condition where physicians often misdiagnose their patients' treatment preferences, and patients' adherence to treatment has profound personal and societal implications. This economic analysis finds a PtDA to be a potentially cost-effective use of health care resources; however there are no PtDAs that have been developed for OSA to date.

Finally, I describe the development of a PtDA prototype for individuals with OSA. PtDA development is a multistage process, and this prototype that will be further developed for evaluation in a clinical study.

1.4 Research questions

1) What is the impact of PtDAs on costs, health outcomes, and adherence, and how does this influence their subsequent cost-effectiveness?
    a. Is there any evidence that PtDAs can improve health outcomes while reducing costs?
2) How can the economic impact of PtDAs best be determined? Using obstructive sleep apnea as a case-study, I describe an economic modelling framework that explores the following question:
    a. Could developing a PtDA for use in adults with obstructive sleep apnea (OSA) be a cost-effective use of resources?
        i. What level of treatment adherence would need to be reached in order for the PtDA to be considered cost-effective?
        ii. Based on expected value of information analysis, is the potential value of performing additional research to reduce the uncertainty in critical parameters justified?

3) How can we improve decision-making for newly diagnosed individuals with obstructive sleep apnea?
    a. In describing the development of an OSA PtDA prototype, I seek to determine: how user-friendly is the prototype?
    b. What are the strengths and limitations of the prototype from the user's perspective?

2 The cost-effectiveness of patient decision aids: A systematic review

2.1 Introduction

There is a clear desire for patient-centred care to be a core feature of all health care systems in the developed world.(25,26) This reflects an understanding that patients' values and preferences are key to making preference-sensitive health care decisions, those where no single option is clearly superior in all respects.(63) The importance of engaging the patient in the decision process is highlighted by evidence indicating that clinicians are often poor judges of what patients value.(6) Failure to incorporate patient preferences in clinical decision making can result in patients receiving inappropriate care, or care that they choose not to use. This misuse of services is wasteful for the healthcare system and may result in poorer health for patients.(63)

Shared decision making (SDM), facilitated through patient decision aids (PtDAs), is one way of incorporating patient preferences into clinical decision making.(48) PtDAs are "…tools designed to help people participate in decision making about health care options, with the goal of promoting deliberation between patients, health care providers, and others about those options. They provide information about the options, and help patients construct, clarify, and communicate the personal values they associate with the different features of the options."(42)

A recent report identified patient centred care as an area where all health care systems needed substantial improvement,(64) however the use of PtDAs has been

low(65) Impetuous for patient centred care and PtDAs has come through prominent commentaries suggesting PtDAs could both improve the quality of medical decisions and reduce costs.(3,6) Given that all health care systems are operating in an environment of resource scarcity, empowering patients while reducing expenditures has great appeal. However, these claims for their potential for savings health care costs are not evidence-based,(66) and it is argued that focussing on costs alone is not the appropriate objective.(67)

The reality is that decision makers consider both costs and benefits to ensure that available resources are deployed appropriately and efficiently to maximize the health and well-being of the population.(49) While helping patients make good healthcare decisions is the primary goal of PtDAs, resources are required to incorporate them into practice. Whether these resources could be allocated better elsewhere is often informed by economic analysis, which examines the opportunity cost of spending by considering costs along-side benefits.

We performed a detailed systematic review of articles that provide evidence on the cost-effectiveness of PtDAs. They findings help identify important gaps in evidence that will be crucial for informing policies to improve the patient-centeredness of care.

2.2 Methods

We searched electronic databases from their inception to January 2013. Databases included Medline, EMBASE, PsycINFO, and the Cumulative Index to Nursing and Allied Health Literature (CINAHL). In addition, the reference lists of included papers were hand-searched to identify additional articles of interest. Search terms used Medical Subject Headings (MeSH) and keywords associated with patient decision aid trials, based predominantly on terms used in a Cochrane review(31) initially combined with terms relating to cost-effectiveness. However, anticipating a paucity of published analyses, we expanded the search to also include terms relating to a) cost, b) adherence/compliance to treatment or c) Quality Adjusted Life Years (QALYs), health utilities or quality of life instruments such as the EQ-5D or SF-36 which can be converted into health utilities. Adherence to treatment was included as an outcome of interest given the potential for PtDAs to improve adherence, and that this has the potential to influence both costs and outcomes. We recognize that adherence to treatment is not a relevant outcome for all decisions, we are solely interested in understanding the impact of PtDAs on adherence in contexts where it is relevant. The search strategy is available on request.

All primary peer reviewed studies, including randomized controlled trial and experimental and quasi-experimental designs utilizing a comparison group. Study selection was undertaken by two reviewers (LT and NB). All studies were reviewed at

the title and abstract level simultaneously, with all selected by at-least one reviewer evaluated at the full-text level. Any discrepancies at the full-text level were resolved through discussion.

2.3 Results

A total of 5,347 records were identified. After duplicate removal, and title and abstract review, 53 papers proceeded to full text review. Hand-searching the reference lists of these articles identified six additional articles. Following full-text review, 29 articles were retained (Figure 2); 9 related to QoL, 16 related to treatment adherence, and 8 related to cost (Table 1).

Only one cost-effectiveness analysis of a PtDA was identified. This three-arm randomized trial was described in two separate articles(68,69). It investigated the use of a PtDA with and without a nurse interview, in women with menorrhagia, compared to usual care. They found that the PtDA plus interview strategy was dominant, by incurring lower costs (£1,030) and higher QALYs (1.582) compared to the PtDA alone (£1,333, 1.567) or usual care (£1,810, 1.574).

To consider the broader economic implications relating to the cost-effectiveness of PtDAs, we conceptualized three cost categories that warrant consideration in PtDA studies (Table 2): those related to administration and delivery of the PtDA; short- term costs (defined as 12-months or less), where competing treatment options can vary

substantially and the potential influence of the PtDA on treatment choice is a central issue; and long-term costs (longer than 12-months) associated with treatment decision. We also considered studies that provide evidence on treatment adherence benefits in terms of QALYs that can be used in economic analyses.(70)

2.3.1 The cost of delivering the patient decision-aid

Five(68,71–74) out of the eight(75–77) studies reporting costs considered the resources required to deliver the PtDA. Costs included the printing of brochures or handouts, training for staff or clinicians, identifying eligible patients and enabling the delivery of the PtDA. If administered in the clinician's office, further costs may be incurred through the additional time spent with a nurse, staff member, or clinician.(78)

Among the studies measuring costs, there was large variation. For example, van Peperstraten et al. found that the cost of printing and mailing the PtDA to individuals considering in vitro fertilization was €9.15, and adding in a telephone counselling session increased costs by €109.50.(74) While in this case the PtDA arm had lower costs overall, two other studies found that the cost of delivering the PtDA (approximately £220-280 per patient) was the driving force behind the intervention arms having significantly higher costs.(72,73)

A proxy measure for increased administration cost is the effect of PtDAs on consultation time with clinicians. The most recent Cochrane review found considerable

variation in the influence of PtDAs on consultation time.(31) It is difficult to generalize on the drivers of this variation, and it is likely a reflection of context specific factors, such as whether the PtDA is delivered at home prior to consultation, or during the clinical encounter. However, it is important to consider how administration of the PtDA will impact costs, and account for these when performing an evaluation.

2.3.2 Short-term costs associated with treatment choice

Of the 7 studies(71–77) that provided only evidence on short-term costs, a majority have focussed on PtDAs for one-off decisions such as mode of delivery for a pregnancy(76), in vitro fertilization(74), or elective surgeries compared to conservative management.(72,75,77) A trend that informed patients typically choose less invasive, and as a result, less costly procedures when fully informed is becoming more apparent.(79) PtDAs have also been shown to decrease healthcare resource use. For example, when PSA screenings detect "low risk" prostate cancers for which there is no clear best treatment option, patients favoured "watchful waiting" when using a PtDA.(80)

The limited evidence, which has so far predominantly focussed on clinical decisions which result in patients choosing a less expensive option has perhaps given a false impression that PtDAs in general will save costs. There are many situations where informed patients will be more likely to choose more expensive options, such as patients

with sleep apnea who choose more expensive dental appliances instead of cheaper airway devices.(81) Increased screening uptake is an example where a PtDA may increase short-term costs.(82,83) While we found no direct evidence of PtDAs in these areas leading to increased costs, the increased resource utilization suggests that short-term costs might be elevated when using a PtDA.

2.3.3 Long-term costs associated with treatment choice

Only one study tracked costs beyond one-year.(68,69) It found that more informed patients were less likely to choose invasive treatment (hysterectomy), which resulted in decreased costs over the following 24-months.

Policy-makers need to consider both the short and long-term implications of using a PtDA. For example, while some patients may choose medication over surgery, this may simply delay surgery until a later point.(75) This delay may lead to short-term cost savings, but it may also possibly lead to a more complicated or expensive surgery at a later date, increasing the total lifetime costs. Choosing the appropriate time frame for analysis is critical. Researchers who found reduced costs over 6-months following the implementation of a PtDA for osteoarthritis noted that "greater longitudinal studies … should be conducted to determine the long-term patient care impact and cost-effectiveness of integrating shared decision making."(79)

2.3.4 Adherence to treatment

Fourteen studies(84–97) evaluated the short-term impact of a PtDA on adherence to treatment. Seven relied exclusively on self-reporting(84–86,89,90,96,97), and only three demonstrated an improvement.(91,96,97)

Two (98,99) PtDA studies have evaluated the long-term impact of a PtDA on adherence to treatment, with neither finding a significant impact. One relied solely on self-reported adherence,(98) while the other used three sources, including plasma measurement.(99)

The lack of evidence supporting improvements in treatment adherence is concerning, but most studies relied on problematic self-reports, and did not link results directly to costs.

2.3.5 Health Benefits

A total of 9 studies evaluated the influence of PtDAs on health measures which provide benefits in terms of Quality Adjusted Life Years. Seven used the SF-36 or SF-12(68,72,72,100–103), three used the Euroqol EQ-5D(68,72,73), and one each used the menorrhagia specific utility scale,(104) and RAND-36.(77) However, despite using scales that can provide health utility values in order to generate QALYs, only four studies actually reported these. For example, in the cost-effectiveness analysis, Kennedy and colleagues measured the EQ-5D at baseline, 6, 12 and 24 months. This was used to determine that the PtDA with interview provided 0.009 more QALYs in comparison to

usual care. The amount of money spent to provide these benefits (in this case a £779 saving) can be compared to other interventions and treatments to discern the value of care.

In the studies that did not report QALYs, two studies found significant improvements in the PtDA group on total scores, one in patients with benign prostatic hyperplasia(100), and the other in women with menorrhagia,(104) while two others found significant improvements on subscales.(68,77) It is therefore unclear whether these would translate to QALY gains.

2.4 Discussion

Informing patients about their health and their choices is the future of healthcare, and as such, PtDAs should be a central component of healthcare reforms. However, economic realities suggest that potential investments in PtDAs need to be weighed against competing technologies and interventions. In order to make funding decisions, policy makers require rigorous economic evidence. Our review finds evidence that there will be upfront costs associated with administering and delivering PtDAs, but unclear evidence on whether these costs provide good value for money. The vast majority of evidence relating PtDAs to short-term costs has focused on situations where patients appear to be choosing less expensive options, primarily in the area of elective surgery. In these cases, PtDAs may be reducing the use of unwanted services and so

may be cost-effective. However, the evidence is sparse and it is unclear whether implementing PtDAs in contexts where beneficial services are known to be underutilized will be cost-effective. Consequently, it would seem the cost-effectiveness of PtDAs will be context specific and need to be evaluated on a case by case basis. Further research and study on determining the most efficient way to deliver PtDA by formally evaluating all the costs involved with implementation will be important for all evaluations.(48)

Based on current evidence we believe it is inappropriate to promote PtDAs as a means of realizing cost savings. The appropriate evaluation of PtDAs requires careful consideration of both costs and benefits. There are a number of reasons for caution. Past studies have significant methodological limitations, including not comprehensively capturing costs and benefits to the patient, and using an insufficient follow-up period for analysis. Importantly, many studies have reported evidence of PtDAs decreasing healthcare costs without consideration of the benefits. Further, the economic evidence from PtDA trails has largely focused on treatment decisions where overuse may be prevalent; conclusions could be different in cases where PtDAs increase the use of beneficial services.

When costs are considered over an appropriate time frame, it is entirely possible that using a PtDA may increase expenditures; this may warrant investment if there is sufficient benefit. PtDA trials have placed a greater focus on outcomes such as

treatment choice, satisfaction, and decisional conflict, rather than costs, treatment adherence and quality of life. Decision making in many healthcare systems relies on a measure of quality, and policy-makers will consider paying more for an intervention like a PtDA if it provides a degree of benefit. It should be emphasized that when evidence of quality is not available, it cannot be assumed that costs reflect quality of care. Reduced healthcare expenditures may indicate better or equivalent quality of life because individuals are not seeking as much care, but this will not always be the case. While the majority economic evidence related to PtDAs has come from randomized trials, other study designs should be considered. Gathering trial-based economic evidence could be facilitated by small-scale PtDA implementation using stepped-wedge or cluster-randomized designs. Incorporating the routine measurement of PROs before and after treatment, as has recently been done in the UK,(105) alongside careful measurement of resource utilization, would enable policy makers to evaluate the impact of PtDAs prior to large scale implementation.

In PtDA trials, a conflict can arise when measuring benefit since patients may legitimately choose options that are less clinically effective than alternatives. For example, a patient may choose best supportive care rather than life extending but toxic chemotherapy.(106) The conflict stems from the fact that societal preferences are used to determine benefit in economic analysis, but individuals or groups within the population may have preference that are dramatically different from those of

society.(107) The premise of SDM is that this choice, despite being at odds with societal preferences or the opinion of clinicians, is the right one. The problem is that the benefit of choosing a treatment that matches the patients informed values and preferences is not easily measured or valued in a way that can be considered by policy-makers.

The use of PtDAs in contexts where individuals need to decide between therapies that require self-management for chronic diseases has been poorly researched to date. For conditions where treatment requires a degree of self-management, adherence to treatment has recurring cost and health consequences, and it has been proposed that when patients choose an option that is more congruent with their informed values and preferences, they may be more adherent.(45) A greater focus should be placed on this area given the growing prevalence of chronic conditions, and the fact that they are significant cost drivers in the healthcare system.

With over 500 PtDAs developed to date, a number that will likely grow over time, the question becomes: where to begin? This review suggests that policy makers currently have insufficient evidence on where to prioritize investments to PtDAs since it is unknown which offer the prospect of greatest value for money. PtDAs have an important role in the future, and could save costs in certain treatment decisions, and in certain contexts. Proponents of PtDAs should focus on developing rigorous economic evidence to inform policy decision making and determine how best to design and deliver PtDAs in contexts that are valued by patients and providers.

Table 1. Characteristics and findings of studies included in systematic review

First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
Arterburn, 2012 (75)	Osteoarthritis patients (hip and knee surgery), PtDA sent to patients' home	PtDA consists of DVD and booklet, historical control (no PTDA)	Pre-post observational N (hip)= 820/968), N (knee) = (3,510, 4,217)	Short: 6 months	Cost: Significant reduction in costs ($13,489 vs $16,558 and $8,041 vs $10,400 for hip and knee respectively) Administration costs not reported.
Barry, 1997 (100)	Patients with benign prostatic hyperplasia, urologic practices	Personalized video, audio, and computer graphics, and control (brochure)	RCT N=227 (104 PtDA, 123 control)	Short: 12 months	QoL: SF-36, significant improvement in PtDA group vs control
Berstein, 1998 (101)	Patients referred for coronary angiography, hospital	Videotape PtDA and, control (usual care: counselling)	RCT N=217 (112 decision aid, 105 control)	Short: 3 months	QoL: MOS Short-form 12 (SF-12), non-significant difference.
Deschamps, 2004 (84)	HRT for peri- and post-menopausal women, study clinic/home	Take home PtDA vs. pharmacist consultation	RCT N= 128 (61 decision aid, 67 pharmacist) Completed base-line survey N=105 (56 PtDA, 49 pharmacist)	Short: 12 months	Adherence: based on self-report, non-significant difference
Edwards, 2004 (85)	Atrial fibrillation, prostatism, menorrhagia, menopausal symptoms	Training skills workshop for the physician in 1) SDM or 2) risk communication materials, both were done in routine	Cluster-RCT N=747 (391 PtDA, 356 usual care)	Short: 6 months	Adherence: Based on self-reported intention to adhere to treatment, significantly higher following research

First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
		clinic time and research clinic (longer consultation)			clinics (more time for appointment).
Emmett, 2005(98)	Newly diagnosed hypertensive patients, GP office	3 PtDA: 1) decision analysis, 2) information video plus leaflet, 3) decision analysis + information video plus leaflet, and control	RCT N= 216 (51 decision analysis + video/leaflet, 52 decision analysis, 55 video/leaflet, 59 control)	Long: 36 months	Adherence: based on self-report, non-significant difference
Hamann, 2007 (99)	Psychiatric inpatients with diagnosis of schizophrenia, state hospital	16-page PtDA followed by planned talk with physician (within 24 h), and control (treatment as usual)	RCT N=86 at 6-months (39 PtDA, 47 control)	Long: 18 months	Adherence: three sources, self-report, physicians' estimates, plasma levels, non-significant difference
Hollinghurst, 2010 (22)	Pregnant women, researcher home visit	Two intervention arms: PtDA (usual care + information ; usual care + decision analysis program), and control (usual care)	RCT N= 742	Short: 10 months	Cost: No significant difference between strategies. Costs related to antenatal visits were included, but none related to administration of the decision aid.
Jones, 2009 (86)	Type 2 diabetes patients, academic medical center	PtDA, and control (cholesterol pamphlet); both delivered by physician either during visit or prior to visit	RCT N=98 (52 decision aid, 46 control)	Short: 3 months	Adherence: based on self-report, non-significant difference.
Jones, 2001 (71)	Individuals with schizophrenia, resource center	Three intervention arms: Computer information, community psychiatric nurse, combination of (1) and (2)	RCT N=112 (56 computer, 28 nurse, 28 combination)	Short: 3 months	Cost: No significant difference between strategies. Costs of administration were included.

First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
Kennedy, 2002 & 2003 (14,15)	Patients with menorrhagia, at-home prior to consultation	Two intervention arms: PtDA, PtDA + interview, and control	RCT N=894 (296 PtDA, 300 PtDA + Interview, 298 control)	Long: 12 and 24 months	QoL: EQ-5D, no significant differences in EQ-5D at 12-months Costs: PtDA + Interview group had lower mean costs than control and PtDA alone group ($1,566 vs. $2,751 and $2,026, respectively). Costs of administration were not included.
Krones, 2008 (87)	Cardiovascular disease risk, GP office	Physician continuing medical education sessions (x 2) with booklet, risk calculator and patient summary sheets, and control	Cluster-RCT N=932 (466 in both arms)	Short: 6 months	Adherence: comparison of mean difference in cardiovascular disease score, non-significant difference
Loh, 2007 (88)	Newly diagnosed depressed patients, primary care	PtDA and GP training, and control (usual care)	Cluster-RCT N=405 (263 PtDA, 142 control at baseline; 191 PtDA, 96 control, at follow-up)	Short: 2 months	Adherence: self-reported and physician assessment, non-significant difference
Man-Son-Hing, 1999 (31)	Antithrombotic therapy for stroke prevention, clinic	PtDA (29 page booklet, worksheet and audiobook, and control (usual care)	RCT N=287 (139 PtDA, 148 usual care)	Short: 6 months	Adherence: based on self-report, non-significant difference
Mann, 2010 (90)	Statins for cardiovascular disease risk, primary care	PtDA (Statin choice tool) led by provider, control (cholesterol pamphlet)	RCT N=150 (80 PtDA, 70 control)	Short: 6 months	Adherence: based on self-report, non-significant difference

First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
McCafferty, 2010 (48)	Women with borderline abnormal cervical smears, clinics	PtDA, repeat smear at 6 months, DNA testing	RCT N=314 (104 PtDA, 104 DNA testing, 106 repeat smear at 6- months)	Short: 12 months	QoL: SF-36, non-significant difference
Montori, 2011 (91)	Post-menopausal women with low bone mineral density score, primary care	PtDA, and control (pamphlet on osteoporosis)	RCT N=100 (52 PtDA, 48 usual care)	Short: 6 months	Adherence: Patient self-report (did not miss dose), pharmacy prescription profile, non-significant except for >80% of days covered (100% decision aid vs. 74% usual care)
Morgan, 2000 (103)	Ischemic heart disease, hospital	Interactive videodisc PtDA, Control	RCT N=240 (120 PtDA, 120 control)	Short: 6 months	QoL: SF-36, non-significant difference
Mullan, 2009 (92)	Antihyperglycemic therapy for diabetic patients, primary care	PtDA (and physician training), and control (usual care)	RCT N=85 (48 PtDA, 37 usual care)	Short: 6 months	Adherence: Patient self-report , Pharmacy records, HbA_{1c}, non-significant difference
Murray, 2001a (72)	Patients with benign prostatic hypertrophy, GP office	PtDA (interactive multimedia program with booklet and printed summary), and control	RCT N=112 (57 PtDA, 52 control)	Short: 9 months	QoL: SF-36, EQ-5D, non-significant differences Cost: Significantly higher cost in intervention group due to intervention (£594.10 vs. £188.80)
Murray, 2001b (73)	Women considering hormone replacement therapy, GP office	PtDA (interactive multimedia program with booklet and printed summary, and control	RCT N=205 (103 PtDA, 102 control)	Short: 9 months	QoL: SF-36, EQ-5D, non-significant differences. Cost: Significantly higher cost in intervention group

First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
					due to intervention (£306.50 vs. £90.90)
Oakley, 2006 (93)	Postmenopausal women prescribed bisphosphonate medication for osteoporotic fracture, workshop	Information booklet, audiocassette and worksheet, and control	RCT N=33 (16 PtDA, 17 control)	Short: 4 months	Adherence: self-reported and pharmacy records, non-significant difference
Protheroe, 2007 (104)	Menorrhagia, GP clinic	Computerized PtDA, Control (written information)	RCT N=146 (74 PtDA, 72 control)	Short: 6 months	QoL: Menorrhagia Specific Utility Scale, significantly higher in the decision aid group
Rothert, 1997 (94)	Hormone replacement therapy in menopausal women, University	3 PtDAs of varying intensities (brochure, lecture/discussion, active decision support)	RCT N=248	Short: 12 months	Adherence: self-report and pharmacy records, non-significant difference.
Simon, 2012 (95)	Depression or lower back pain, via internet	PtDA (electronic individually tailored), and control (static patient information)	RCT N=2480 (1,269 PtDA, 1,211 control)	Short: 3 months	Adherence: based on self-report, non-significant difference
van Peperstraten, 2010 (20)	In vitro fertilization, clinic	PtDA + nurse support + reimbursement offer, and control (standard care for in vitro)	RCT N=308 (152 PtDA, 156 control)	Short: 12 months	Cost: Significantly lower costs in the decision aid group (€170 less per couple). Cost of administration and delivery were included.
van Steenkiste, 2008 (96)	Cardiovascular risk management, GP office	Decision support tool for physicians trained on CVD guidelines, and educational materials on paper (control)	Cluster-RCT N=490 (276 PtDA, 214 control)	Short: 6months	Adherence: Self-reported physical activity, significant improvement in the decision aid group
Vuorma, 2003 & 2004 (23,55)	Gynecology patients, clinic	PtDA focusing on menorrhagia treatment options mailed to patients	RCT	Short: 12 months	QoL: RAND-36, significant on one of 9 dimensions.

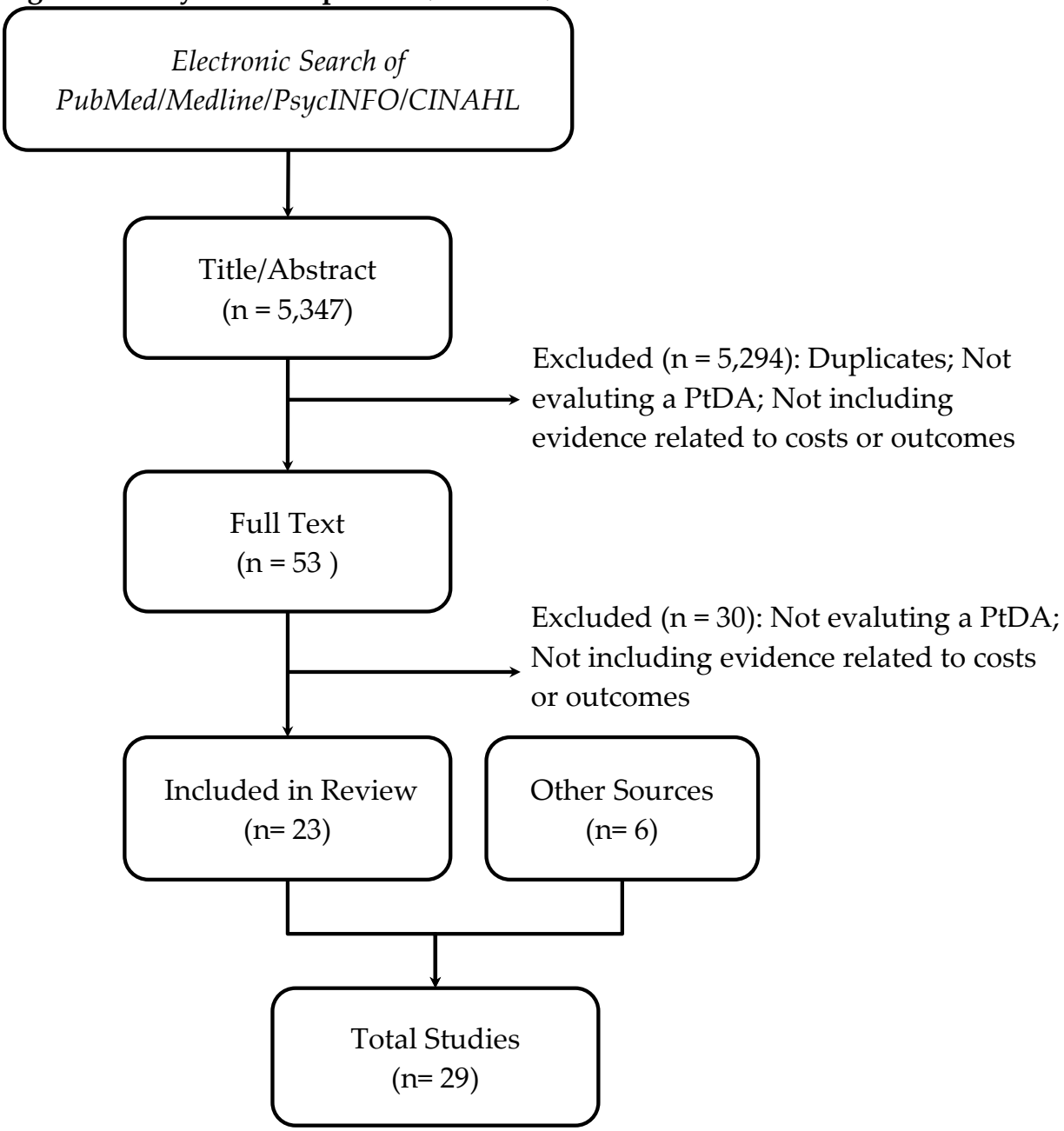
First Author, Year	Participants, Setting	DA/Control	Design and sample size	Time horizon	Outcome
		prior to treatment , Control (usual care)	N=363 (184 PtDA, 179 control)		Cost: No significant difference (€4,607 decision aid vs. €5,164 usual care). Travel costs were included but administration costs related to delivery of the decision aid were not.
Weymiller, 2007 (97)	Statins for cardiovascular disease risk, clinic	PtDA (Statin choice), and control (cholesterol pamphlet)	Cluster-RCT N=98 (51 PtDA, 46 control)	Short: 3 months	Adherence: Patient self-report: having missed one or more doses in the last week, significant increase in the decision aid group

Table 2. Conceptual methods of how patient decision aids could influence health care costs

Method	Potential to increase Costs	Potential to Decrease Costs
Administration of the PtDA	*Additional staff time, and resources (printing, scheduling etc.) to identify eligible patients, deliver and explain the PtDA, and potential increase in consultation time when discussion options.* Evidence: Most PtDA studies fail to properly measure or account for all these costs. Two trials that comprehensively accounted for costs found that administration and delivery of the PtDA cost £220-280 per patient, which was the driver of higher costs in the intervention arm.(72,73) The Cochrane review found that the median change in consultation time across 9 studies was a 2.5 minute increase.(31)	*The use of a PtDA may reduce time needed by a health professional explaining to patient.* Evidence: There is no direct evidence of PtDAs decreasing these costs. One study found an 8-minute decrease (90 minutes vs. 82 minutes) in consultation time for women referred to genetic counseling for a family or personal history of breast cancer.(108) It is unclear whether these savings would have been offset by other administration costs.
Short-term costs	*Patient might prefer a more expensive option.* Evidence: Very few studies have evaluated contexts where patients choose a more expensive option. One example is a randomized trial where 41% of individuals who received a decision aid underwent colon cancer screening, compared to 23% in the control group.(82)	*Patient might prefer a less expensive option.* Evidence: The majority of studies have focused on contexts where individuals are choosing less invasive, and thus less costly, treatment options. For example, the introduction of a PtDA for patients with osteoarthritis considering joint replacement resulted in a 12-21% decrease in health care costs over six months.(75)
Long-term costs	*Patient may prefer delaying a surgery but that leads to a more complex future surgery that costs more in the long-term. Adherence to some medications may increase costs if prescriptions were not previously being filled.* Evidence: There is no evidence of long-term costs increasing as a result of a PtDA. While no study has followed up a PtDA trial long enough, there is evidence to suggest that delaying surgery can lead to a higher rate of complications at a later date.(109)	*Patient may find it unnecessary to have surgery at all. A patient might be more adherent to option they prefer. This may lead to reduced other costs related to non-adherence* Evidence: There is very limited evidence of patients choosing options with lower costs or resource use over the long-term. The longest follow up to date is 2 years following a decision aid for menorrhagia. It found that costs were reduced in the decision aid group, driven by a decrease in hysterectomy rates and inpatients and

Method	Potential to increase Costs	Potential to Decrease Costs
		outpatient costs(68). The only other randomized trial to show cost-savings was in vitro fertilization, where a decision aid in combination with the support of a specialized nurse and a reimbursement incentive resulted in fewer twin pregnancies and associated obstetrical costs. In all, the decision aid arm saved approximately €170 per couple.(74) The most recent PtDA trial to track adherence found an improvement (100% vs. 74%), as measured by pharmacy records with prescriptions to cover >80% of days.(91)

Figure 2. Study selection process (PRISMA)



3 Exploring the cost-effectiveness of patient decision aids: a case-study in adults with obstructive sleep apnea

3.1 Introduction

It is increasingly being recognized that for many treatment decisions, identifying patient preferences is as important as a medical diagnosis.(6) Treatment options have different profiles of risks, benefits and side-effects, and the right treatment depends not only on the patients' individual clinical characteristics, but their individual preferences too.(110) For many treatment decisions, doctors have a poor understanding of their patients' preferences.(6) This may result in patients receiving treatments that they choose not to use, or are unnecessary altogether.

Shared decision making (SDM) facilitated with patient decision aids (PtDAs) appears to be the most promising approach to incorporate patient preferences into health-care decision making.(111) PtDAs are tools that make explicit that a decision is being made, provide information about competing treatment options, including benefits and harms, and elicit patient preferences and values.(112) They have been shown to improve knowledge, increase involvement in decision making, and result in better patient-doctor communication.(113,114) Consequently PtDAs hold the potential to improve treatment adherence since the patient is more likely to receive a treatment they prefer.(45) However, there has been limited study of whether PtDAs can improve

treatment adherence,(91,96,97,112) and it is often viewed as a secondary outcome rather than the focus of the analysis.

While PtDAs can potentially improve decisions and save costs through reducing unnecessary treatment, in many cases delivering a PtDA requires an investment by requiring training for clinicians or staff, additional time to identify and distribute the PtDA to eligible patients, or by lengthening the consultation between the patient and provider.(112) Additionally, some PtDAs could lead to patients choosing more expensive treatments.(115) There is strong evidence that PtDAs improve patient knowledge and result in more value-congruent decisions,(112) however the economic argument supporting their use is lacking.

Here we use a case study of treatment decisions for patients with Obstructive Sleep Apnea (OSA) to explore the cost-effectiveness of delivering a PtDA. We use a decision analytic model to explore the consequences of delivering a PtDA for patients with OSA and determine what level of treatment adherence would have to be reached in order for the PtDA to be cost-effective. We use the results to explore what future clinical studies should consider in this area.

3.1.1 Case study – obstructive sleep apnea

OSA is a common respiratory sleep disorder, where sufferers experience breathing pauses or 'apneas' during the night, resulting in an increased risk of motor

vehicle crashes, cardiovascular events, and premature death.(116–118) Evidence on the potential benefits of a PtDA in OSA comes from a meta-analysis which found two primary treatment options were clinically effective in treating patients with moderate OSA: a continuous positive airway pressure (CPAP) machine which pushes a stream of air through a mask into patient's noses or mouth to keep their throat and airway open, and Mandibular Advancement Splints (MAS), a form of dental appliance.(119) These therapies differ in efficacy, cost, comfort and side effects. The review concluded that "based on direct and indirect comparisons, CPAP appeared to be more effective than MAS. However, given the issues with noncompliance with CPAP, the decision as to whether to use CPAP or MAS will likely depend on patient preferences."(119)

While CPAP is considered more *clinically effective* in improving certain sleep parameters, such as the number of apneas individuals experience each night,(120) this is only true when used appropriately. Patients judge the effectiveness of therapy on a number of factors, including clinical effectiveness, convenience, cost and so forth. This, in turn, influences whether or not a patient is adherent to therapy. When a patient chooses not to use a treatment, he or she has determined that this is more effective, from their perspective, than using treatment. This could be because the negative aspects of treatment (inconvenient, costly) outweigh the clinical effectiveness.

Non-adherence to CPAP is a serious problem, and interventions to improve treatment adherence have shown little impact.(119) Cross-over trials indicate that about

half of OSA patients prefer MAS to CPAP.(81) Further, despite the lower efficacy of MAS in reducing apneas, the two therapies have shown similar improvement in symptoms and cardiovascular markers, which has been hypothetically related to a higher adherence rate to MAS.(81,121,122) Despite this, without a means for doctors to identify preferences, nearly all patients receive CPAP. This is considered to be a major reason for the high rates (up to 50%) of patients that are non-adherent to CPAP in the first year.(123,124) While in theory the best strategy would be to provide all patients with the most cost-effective treatment, CPAP, and offer MAS to those that are non-adherent, this is not always feasible. The two main reasons for this are cost and loss to follow-up. While many manufacturers offer a free trial period for the CPAP machine, patients often purchase the machine prior to becoming non-adherent, and if not, there are still substantial costs for consumables, such as masks and tubing. After such expenditures, individuals may be hesitant to pay over $2,000 out-of-pocket for a MAS. In addition, many patients who become non-adherent to CPAP do not return to seek alternative treatments. This may be because patients are unaware that alternative treatments such as MAS exist. Thus, the development of a PtDA in OSA is justified, both as a means to ensure informed patients are offered the treatment they prefer in the first place, and to notify them that other treatment options exist should their initial choice not be acceptable to them. It is currently unknown how a PtDA will affect actual decisions and subsequent treatment adherence, but a trial is being planned.

3.2 Methods

3.2.1 Overview

We developed a decision analytic model that simulates the natural course of obstructive sleep apnea in patients with moderate OSA [>15-30 events per hour] who followed conventional practice as outlined by Canadian guidelines.(125) A previous state transition model(126) was further developed for the purpose of this analysis.(127) Briefly, the model evaluates the impact of different treatment strategies (no treatment, CPAP and MAS) on annual risks of motor vehicle crashes (MVC), cardiovascular (CV) events such as myocardial infarction (MI) and stroke, costs, quality of life and mortality over a 5-year period (the life expectancy of a CPAP machine). This time horizon was chosen because we believe it gives an accurate representation of the incremental difference in costs and benefits between the two strategies. The cycle length of the model is one year, and a half-cycle correction was applied. The model structure is illustrated in Figure 3. The objective of modifying this model was to estimate the incremental costs and benefits (in terms of Quality Adjusted Life Years (QALYs)) of delivering a PtDA from a Canadian societal perspective. The PtDA was assumed to: 1) change the proportion of patients receiving each treatment strategy (including the proportion of patients that choose a treatment rather than no treatment); 2) change the rate of adherence to each treatment; 3) incur a cost for its delivery.

3.2.2 Strategies and embedded treatment options

Current strategy: Physicians typically select CPAP as the primary treatment since it is the most clinically effective option in reducing the number of apneas per night.(126) For patients who become adherent users of CPAP, this is the most appropriate course of action. However, approximately 37% of patients are non-adherent in the first year because the discomfort, noise and embarrassment of CPAP outweighs the potential benefits.(123) Untreated OSA is associated with increased risks of strokes,(116) myocardial infarction,(118) motor vehicle crashes,(117) reduced work performance and increased occupational injuries.(128)

PtDA strategy: We hypothesize that a PtDA could be delivered within the OSA diagnosis and treatment pathway. Currently, patients receive information about CPAP therapy, and we propose substituting this information with a PtDA after they have been diagnosed with OSA, but prior to their consultation about treatment. We assumed delivering a PtDA would require an additional consultation to deliberate and reach consensus on the course of action, and change the proportion of patients receiving CPAP and MAS. This is based on evidence from a randomized cross-over trial where all patients received CPAP and MAS, and afterwards 51% of patients said they would prefer to use MAS, 23.1% CPAP, 21.3% either, and 4.6% neither.(81) It was also assumed that delivering a PtDA would decrease treatment non-adherence to treatment as patients are offered the modality they prefer.

3.2.3 Treatment non-adherence

While there has been limited research directly comparing rates of non-adherence to CPAP and MAS,(119) indirect comparisons suggest CPAP has higher non-adherence rates in the first year in moderate OSA patients (37% vs. 23%),(123,129) with both having a gradual decline in adherence over the next 5 years.(130) By allowing informed patients to choose their treatment, it is expected that treatment non-adherence in the PtDA strategy for both patients that receive CPAP and MAS would decrease. However, the magnitude of this change is unknown. In the absence of empirical evidence we considered a distribution that incorporates a plausible range of improvement. The basecase analysis explored a 20% reduction in treatment non-adherence. Additional scenarios were also considered, ranging from a 0% reduction in treatment non-adherence, to a 50% reduction.

3.2.4 Key clinical and environmental outcomes

For patients who adhere to treatment, the annual costs and benefits were estimated in terms of the reduction in the rate of motor vehicle crashes (MVC) and cardiovascular (CV) events, such as myocardial infarction (MI) and stroke.(118,131) These were modeled through the influence of CPAP and MAS on surrogate outcomes (apneas per night) based on a previous meta-analysis.(126) Apneas per night are correlated with subjective and objective measures of the severity of OSA.(132) The

model assumed that patients fully adherent to CPAP would have an incidence similar to that of the general population (as suggested by previous studies).(118,133) Patients adherent to MAS were assumed to have some additional risk of events, though this was varied in a scenario analysis to reflect emerging evidence that MAS is equally effective in reducing the risk of cardiovascular disease.(134) Baseline risks of MVC and CV events were based on national data (see Table 5). Those non-adherent to treatment were assumed to experience the same rate of adverse events as those choosing 'no treatment'. As treatment non-adherence decreased in the PtDA arm, a greater proportion of individuals were undergoing treatment and thus the rate of adverse events decreased linearly.

3.2.5 Costs and quality-of-life

All costs are in 2010 Canadian dollars. We assumed that implementing the PtDA would require an additional follow-up visit ($43.09),(135) during which questions on the options and discussion on what aspects matter most can occur. This cost was varied from $0 to two billings ($86.18) in a scenario analysis to reflect the opposing evidence including some studies that show a PtDA may not actually increase physician time at all.(112) Both MAS and CPAP costs vary by manufacturer. We used the broad range of costs available to patients in Canada. Since CPAP is less expensive in jurisdictions where it is covered by insurance or provincial government plans, we explored smaller

costs in the scenario analysis. It is hypothesized that OSA is associated with losses to productivity; however there are no data to estimate this effect so it was not included in our analysis.

The effectiveness outcome was the QALY where various health states were adjusted for the quality of life (health utility) of health states multiplied by their duration.(136) Health utilities for adherent and non-adherent OSA by treatment (Table 3), along with those associated with each event (MVC, stroke, MI) were included (see Tables 5 and 6). Costs and QALYs were discounted at 5% per the Canadian Agency for Drugs and Technologies in Health guidelines.(137)

3.2.6 Analysis

We calculated the incremental cost-effectiveness ratios (ICERs) of the PtDA strategy vs. current practice. To reflect uncertainty in the evidence, we used a probabilistic analysis to calculate expected costs and QALYs. Probability distributions were assigned to parameters using evidence and expert opinion.(138) The probability that each strategy represents the most cost-effective use of resources was reported given currently available evidence.

3.2.7 Value of information analysis

To inform future research priorities, we performed Value of Information analysis. The expected value of perfect information (EVPI) was calculated to quantify the value of eliminating all uncertainty through additional (perfect) research.(139) Although, in practice, research cannot be undertaken to eliminate all uncertainty, EVPI provides a broad indication of the extent of the remaining uncertainty in the decision about the cost-effectiveness of the PtDA. Furthermore, the EVPI can be compared with the potential cost of additional research to indicate whether there is value in further research to reduce uncertainty of parameters. We further calculated the expected value of partial perfect information (EVPPI) to identify the value of resolving uncertainty around individual parameters or a set of related parameters.(140)

One-way sensitivity analyses were undertaken to explore the individual influence of key parameters such as the cost of the PtDA, the proportion of individuals' switching from CPAP to MAS treatment, the influence of the PtDA on treatment choice, the relative efficacy of MAS compared to CPAP, and the cost of the CPAP machine on the cost-effectiveness of the PtDA strategy.

3.3 Results

3.3.1 Primary results

In the basecase which considered a 20% reduction in treatment non-adherence (corresponding to an increase in adherence from 63% to 70% for CPAP, and from 77% to 82% for MAS in year 1), the PtDA strategy was estimated to provide marginal benefits (QALY of 3.324 vs 3.322) at an incremental cost of $84 ($4,419 vs $4,335) providing an ICER of $62,414/QALY. If non-adherence to treatment was unchanged when using a PtDA, the strategy was dominated since more patients use MAS (which is less clinically effective) at baseline (total QALYs 3.346 vs. 3.351 for PtDA and no PtDA, respectively) and incur higher costs through PtDA delivery ($4,398 vs. $4,343). Decreases in treatment non-adherence lead to increases in both incremental costs and QALY in the PtDA strategy. If treatment non-adherence is decreased by 30% (corresponding to an increase in adherence from 63% to 74% for CPAP, and from 77% to 84% for MAS, in year 1), the ICER was $20,565/QALY (incremental costs and QALYs of $98, 0.005, respectively).

However, as depicted in the cost-effectiveness scatter plot (Figure 4), and cost-effectiveness acceptability curve (CEAC, Figure 5), there was considerable uncertainty in this estimate. The probability that the PtDA strategy was the most cost-effective ranges from 27% to 52% between thresholds ranging from $0 to $100,000/QALY. A key source of parameter uncertainty is the reduction in non-adherence to treatment in the

PtDA strategy and the cost of delivering the decision aid. Figure 6 shows the impact of these two parameters on the ICER.

Other influential parameter uncertainties were explored in Table 4. The largest effect on the ICER came from varying the cost of the CPAP machine, ranging from $230,184 per QALY when the cost was $750 (half of the base-case), to the PtDA strategy dominating the alternative when the cost was increased to $2,000.

3.3.2 Value of information analysis

The EVPI at the $50,000 per QALY threshold was $91 per person-year. The EVPPI analysis suggests parameters contributing the most to this uncertainty were the adherence to chosen treatment in the PtDA strategy ($56), and treatment choice in the both the PtDA strategy ($39) and conventional strategy ($34). Since nearly 1 million Canadians have OSA, and given it is a chronic disease requiring ongoing treatment, the population EVPI and EVPPIs are in the $100's of millions.

3.4 Discussion

While PtDA are important tools for improving patient-centered care, as health technologies, decisions about wide-scale implementation will require evidence of both clinical and economic benefits. This study finds that delivering a PtDA for OSA can be a cost-effective use of resources, provided that it reduces treatment non-adherence.

Further, we find that a trial of a PtDA in this patient group that measures the proportion that choose each treatment option, and their subsequent adherence to treatment, would be economically justified (since this trial would most likely cost less than the population EVPI). The results are subject to considerable uncertainties for other parameters, particularly with respect to the cost of the PtDA and treatment devices. Importantly, the actual cost of delivering the PtDA - something that is not often measured in trials - is a sensitive parameter to the cost-effectiveness results.(112) Once this trial has been conducted, we propose this analysis be updated with the new evidence for key parameters such as treatment adherence, proportion choosing treatment and the cost of delivering the PtDA so that the economic argument for widespread implementation can be considered.

Previous studies have considered the economics of PtDAs, but primarily alongside clinical trials. For instance, Kennedy et al. found that a combination PtDA and interview for patient with menorrhagia had lower costs and higher QALYs than the control.(69) Two separate randomized trials evaluating PtDAs for benign prostatic hypertrophy and hormone replacement therapy found no significant differences in health outcomes between the two groups, but the PtDA strategy incurred greater costs through the use of video equipment.(72,73) Studies of PtDAs that have shown reductions in the proportion of hip and knee replacements(75) have been used to promote the economic benefits of PtDAs.(3) However, such conclusions are limited by

the lack of understanding of the consequences of a PtDA on all downstream costs and benefits, over a sufficiently long time horizon.(66) Gage et al used a model to consider incorporating patient preferences into decisions on antithrombotic therapy for nonvalvular atrial fibrillation(141). While not specifically a PtDA, it is an example of how decision analytic models can be used to anticipate the cost-effectiveness of such interventions. Models enable the combination of different sources of evidence in appropriate time-horizons to examine the impact of hypothetical scenarios on long-term costs and benefits. As decision analytic models are now routinely used for economic evaluation of healthcare interventions, we expect similar models will be more widely used in the evaluation of PtDAs. The cost-effectiveness of PtDAs for other conditions will depend on their specific contexts, but we have demonstrated a framework for how this can be determined.

In many ways, economic evaluations of PtDAs are no different to evaluations of other health care tests and technologies, but one unique issue warrants further attention. This is in addressing the conflict that arises when the goals of economic evaluation and patient preferences are not aligned. For example, patients using a PtDA may legitimately choose a treatment option that is less clinically effective – examples include cancer patients choosing palliation over chemotherapy, or OSA patients choosing MAS over CPAP. From a patient preference perspective, this is the most effective option *to the patient.* However, this is at odds with current economic evaluation

methods which use societal weights for health states that would have assigned fewer QALYs for the less clinically effective treatment option. This conflict stems from current QALY measurement techniques that fail to capture some of the known benefits of PtDAs, including the satisfaction a patient might get from receiving the option that is most congruent with their values and preference. New techniques such as discrete choice experiments provide an avenue for valuing these benefits in the future. However, until then we must assume that current evaluation techniques are underestimating the benefit of PtDAs.

A number of limitations merit consideration. The model of OSA adapted for this analysis relies on surrogate measures for CVD outcomes. This mainly reflects lack of direct empirical evidence relating the severity of OSA and effect of treatment on outcomes. Our analysis was completed based on the age distribution from our local centre; the cost-effectiveness could change depending upon the age structure of the population and other associated comorbidities (such as diabetes and cancer) that we have not considered. In addition, we have relied on expert opinion for a number of the parameters, but have attempted to address this by using appropriate probability distributions to describe the associated uncertainty. However, by modeling uncertainty around individual parameters, we have ignored their potential correlation. This could lead to either over- or under-estimates of uncertainty in our results, but the direction of this is unclear. We have modeled the costs of devices based on our local (British

Columbia) context, where individuals pay out-of-pocket or through extended health insurance. As identified in the scenario analysis, these costs have a substantial impact on the ICER. As such, the results may not be applicable to regions where the costs of one or both of the treatments are dramatically different. Finally, we did not consider some possible externalities with the implementation of PtDAs which might influence which physicians patients choose to visit, and how manufacturers change the price of treatments.

In conclusion, we have shown that in OSA a PtDA may be cost-effective if it decreases treatment non-adherence. A trial of a PtDA in this patient group is justified in order to determine its impact. A number of important questions around the appropriateness of benefit measurement for PtDA trials have been highlighted, and we conclude that current techniques underestimate benefits. Future studies should consider methods to better capture the benefits of PtDAs.

Table 3. Key parameters for the decision-analytic model

Parameter	Mean	Percentiles		Distribution and Source	
		2.5%	97.5%		
Proportion Choosing Treatment†					
Decision Aid					
CPAP	42.5%	31.7%	53.2%	Dirichlet	(81)
MAS	52.5%	42.6%	62.2%		
No Treatment	5.0%	1.6%	10.1%		
Non-Decision Aid					
CPAP	79.1%	69.8%	87.1%	Dirichlet	*
MAS	5.9%	2.0%	11.6%		
No Treatment	15.1%	8.9%	22.5%		
Switching from CPAP to MAS‡	30.0%	10.7%	49.5%	Normal	*
Relative Efficacy of MAS	70.4%	46.6%	97.2%	Beta	(142)
Treatment non adherence §,‖					
CPAP					
Year 1	29.3%	20.7%	36.3%	Normal	(123)
Year 3	39.4%	24.8%	52.8%		
Year 5	43.2%	21.6%	63.5%		
MAS					
Year 1	18.1%	12.3%	23.2%	Normal	(129,130)
Year 3	29.9%	17.0%	42.0%		
Year 5	33.0%	13.4%	51.7%		
% Reduction in treatment non-adherence in PtDA Strategy(basecase)	20.0%	4.2%	40.3%	Beta	*
Utilities					
OSA (no treatment)	0.730	0.350	0.968	Beta	(143)
Incremental CPAP	0.039	0.001	0.155	Beta	(143,144)
Incremental MAS	0.028	0.001	0.107	Beta	(143,144)
Costs					
Decision Aid	$43	$35	$51	Normal	(135)
CPAP					
Machine	$1,498	$1,125	$1,875	Normal	*
Initial Visit	$43	$35	$51	Normal	(135)
Follow-up visit (annual)	$166	$136	$197	Normal	(135)
Recurrent costs (annual)	$351	$285	$418	Normal	*
MAS					
Appliance	$2,400	$2,103	$2,704	Normal	*

Parameter	Mean	Percentiles		Distribution and Source
		2.5%	97.5%	
Follow-up visit (biannual)	$166	$136	$197	Normal (135)

CI = Confidence Interval; CPAP = continuous positive airway pressure; MAS = mandibular advancement splint; OSA = obstructive sleep apnea

* Expert opinion, we elicited min, max and median values from our clinical experts and fit distributions following guidance in O'Hagan et al.

† The proportion of people choosing no treatment decreased when using a decision aid. This is supported by trials finding that decision aids are able to decrease the proportion of people who do not make a decision.

‡ Switching was assumed to occur only from CPAP from MAS. Expert opinion suggests that switching from MAS to CPAP is extremely rare.

§Adherence curves were developed using a polynomial regression line to approximate the adherence values reported by the above sources. With respect to CPAP adherence specifically, this study was chosen over alternatives because it best approximates real-world conditions.

|| Improvements in adherence were modeled as percentage decreases in non-adherence. This was done for two reasons, first it assumes that as baseline adherence improves it becomes more difficult to improve adherence, an assumption that has face validity. Secondly it avoids a ceiling effect, where adherence could go over 100%.

Table 4. Probabilistic results from scenario analysis

	PtDA		No PtDA		ICER
	Cost	QALY	Cost	QALY	
Base Case	$ 4,419	3.32353	$ 4,335	3.32218	$ 62,414
Sensitivity Analysis					
Discount 3%	$ 4,529	3.44621	$ 4,453	3.44435	$ 40,799
Discount 0%	$ 4,697	3.64326	$ 4,639	3.64082	$ 23,513
Switching CPAP to MAS = 10%	$ 4,429	3.34179	$ 4,350	3.34156	$ 348,637
Switching CPAP to MAS = 50%	$ 4,411	3.32706	$ 4,326	3.32439	$ 32,020
Proportion choosing treatment - current practice*	$ 4,451	3.35191	$ 4,344	3.34395	$ 13,512
Proportion choosing no Tx - 15% in both arms	$ 4,205	3.32635	$ 4,346	3.33466	$ 17,020
Efficacy of MAS = CPAP	$ 4,312	3.34734	$ 4,308	3.34500	$ 2,004
Efficacy of MAS = 0.5 * CPAP	$ 4,504	3.34196	$ 4,367	3.34121	$ 182,413
Cost of PtDA = 0	$ 4,378	3.33760	$ 4,339	3.33624	$ 28,933
Cost of PtDA = 2 * Initial Visit	$ 4,460	3.33760	$ 4,333	3.33624	$ 93,204
Cost of CPAP = 750	$ 4,122	3.33760	$ 3,810	3.33624	$ 230,184
Cost of CPAP = 2,000	$ 4,633	3.33760	$ 4,700	3.33624	Dominated
Cost of MAS = 1,200	$ 3,769	3.33760	$ 4,167	3.33624	Dominated
MVC Effect Removed	$ 4,104	3.32030	$ 4,012	3.31882	$ 62,162

QALY= quality adjusted life year; ICER = incremental cost-effectiveness ratio; MAS = mandibular advancement splint; CPAP = continuous positive airway pressure

*This sensitivity analysis applies the distribution of patients choosing treatment (CPAP – 80%, MAS – 5%, no treatment – 15%) from current practice, to the decision aid arm.

Table 5. Additional model parameters

Parameter	Value		Source	Parameter	Value	Source
	Male	**Female**				
Age Range (%)			Sleep disorders program, Vancouver general hospital, Vancouver, BC, Canada	Male/Female Prevalence Ratio	3.3:1	(145)
25-34	11.0	8.4		Proportion of severe stroke/total strokes	0.27	(143)
35-44	26.7	16.9		MVC by Severity		
45-54	34.6	44.6		PDO	82.50%	(146)
55-64	27.7	30.1		MAIS 0	5.70%	
Annual probability of MVC in general population			(147)	MAIS 1	10.40%	
25-34	4.9%	3.5%		MAIS 2	1.00%	
35-44	3.8%	2.8%		MAIS 3	0.30%	
45-54	3.7%	2.3%		MAIS 4	0.08%	
55-64	2.7%	1.8%		MAIS 5	0.02%	
Incidence of MI (per 100,000) in general population			(148)	Fatal	0.01%	
25-34	170	10		Adverse Events		
35-44	570	120		RR of MVC	1.90	(117)
45-54	1170	480		RR of MI	1.53	(118)
55-64	1950	920		RR of Stroke	1.53	(118)
Incidence of stroke (per 100,000) in general population			(149)	Cost of MVC		
25-34	14	17		PDO	$1,424	(146)
35-44	47	45		MAIS 0	$980	
45-54	161	132		MAIS 1	$7,962	
55-64	469	153		MAIS 2	$42,900	

Parameter	Value		Source	Parameter	Value	Source
Proportion of fatal MI/total MI	9.1%	14.8%	(150)	MAIS 3	$123,679	
Proportion of fatal stroke/total stroke	14.0%	20.0%	(151)	MAIS 4	$291,533	
				MAIS 5	$694,959	
				Fatal	$240,432	

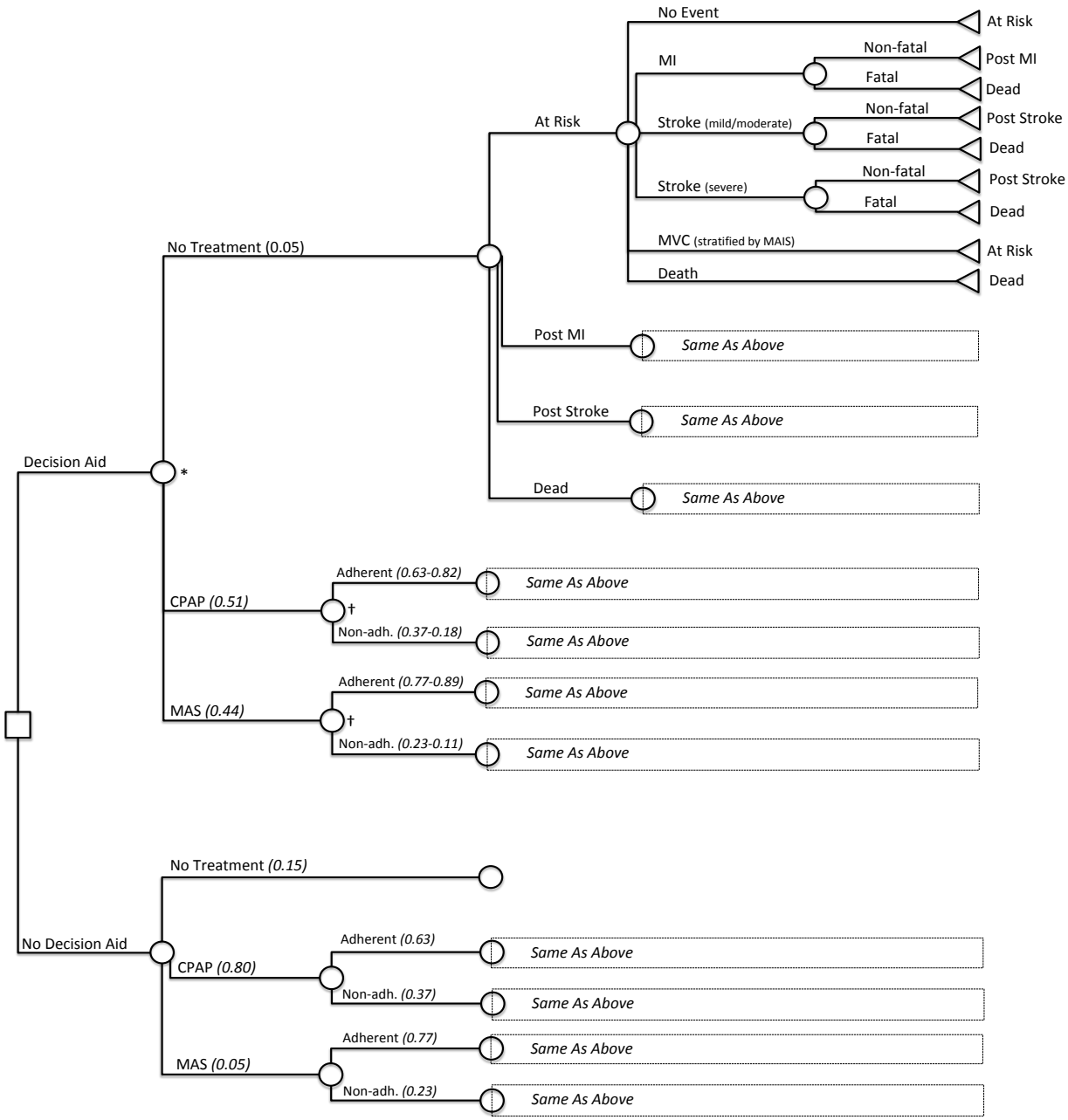
MVC = motor vehicle crash; MI = myocardial infarction; PDO = property damage only; MAIS = maximum abbreviated injury scale

Table 6. Additional model distributions

Parameter	Mean	95% CI		Distribution and Source
		Low	High	
Utilities				
MI				
First 6 Months	0.570	0.166	0.917	Beta (152)
After 6 Months	0.733	0.277	0.988	Beta (152)
Stroke				
First 6 Months	0.310	0.240	0.386	Beta (153)
After 6 Months	0.620	0.554	0.684	Beta (154)
Costs				
Stroke				
Initial (Fatal)	$17,040	$ 13,798	$ 20,281	Normal (155)
Initial (Non-fatal)	$35,566	$ 28,815	$ 42,230	Normal (155)
Annual	$13,195	$ 10,636	$ 15,652	Normal (155)
MI				
Initial (Fatal)	$7,453	$ 6,021	$ 8,906	Normal (155)
Initial (Non-fatal)	$3,318	$ 9,338	$ 13,670	Normal (156)
Annual	$3,318	$ 2,691	$ 3,953	Normal (155)

CI = confidence interval; MI = myocardial infarction

Figure 3. Diagrammatic representation of the decision analytic model



MI= myocardial infarction; MVC = motorvehicle crash; MAIS = maximumabbreviated injury scale; CPAP = continuous positive airway pressure; MAZ = mandibular advancement splint; Non-adh. = non-adherent

* Arrows eminating from this node refer to changes in the proportion of individuals choosing each option relative to the *No Decision Aid* arm. The No Decision Aid arm was modelled according to current practice, with 80%, 5%, and 15% of individuals receiving CPAP, MAS, and No Treatment, respectively. By comparison, in the Decision Aid arm, 44%, 51%, and 5% of individuals receive CPAP, MAS, and No Treatment, respectively.
† Arrows eminating from these nodes refer to changes in the adherence rates for individuals relative to the *No Decision Aid* arm. All adherence and non-adherence values are at year 1. Adherence in the *No Decision Aid* arm was modelled as 63% for CPAP and 77% for MAS at one-year.

Figure 4. Cost-effectiveness scatter plot

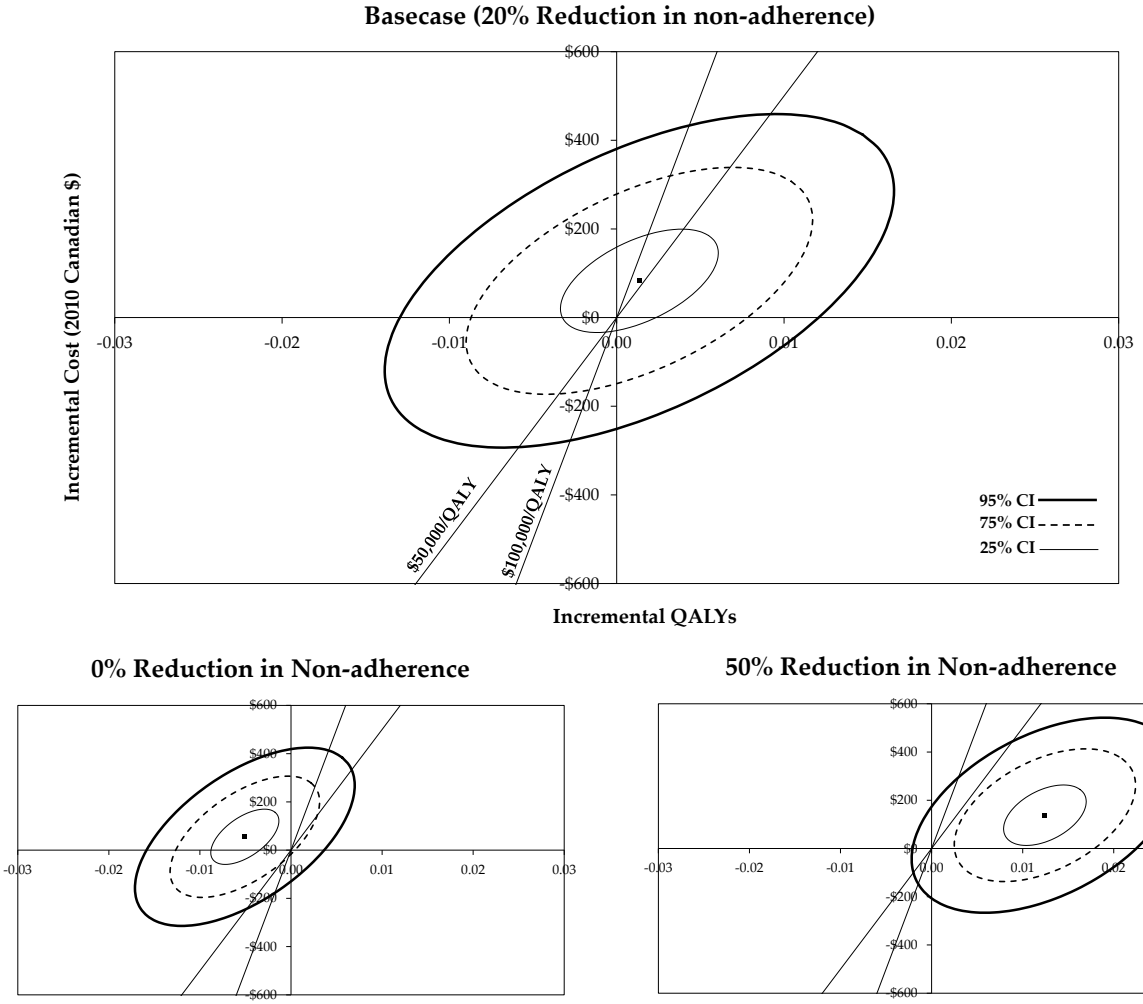
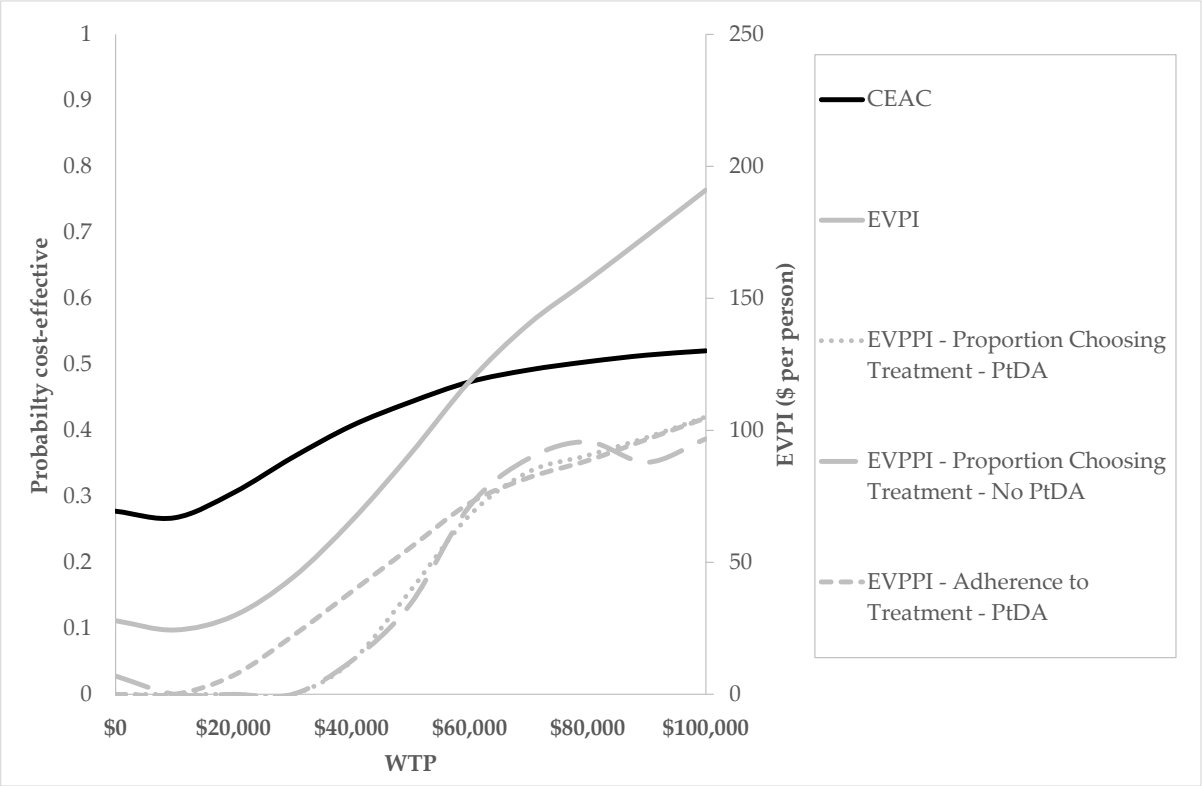
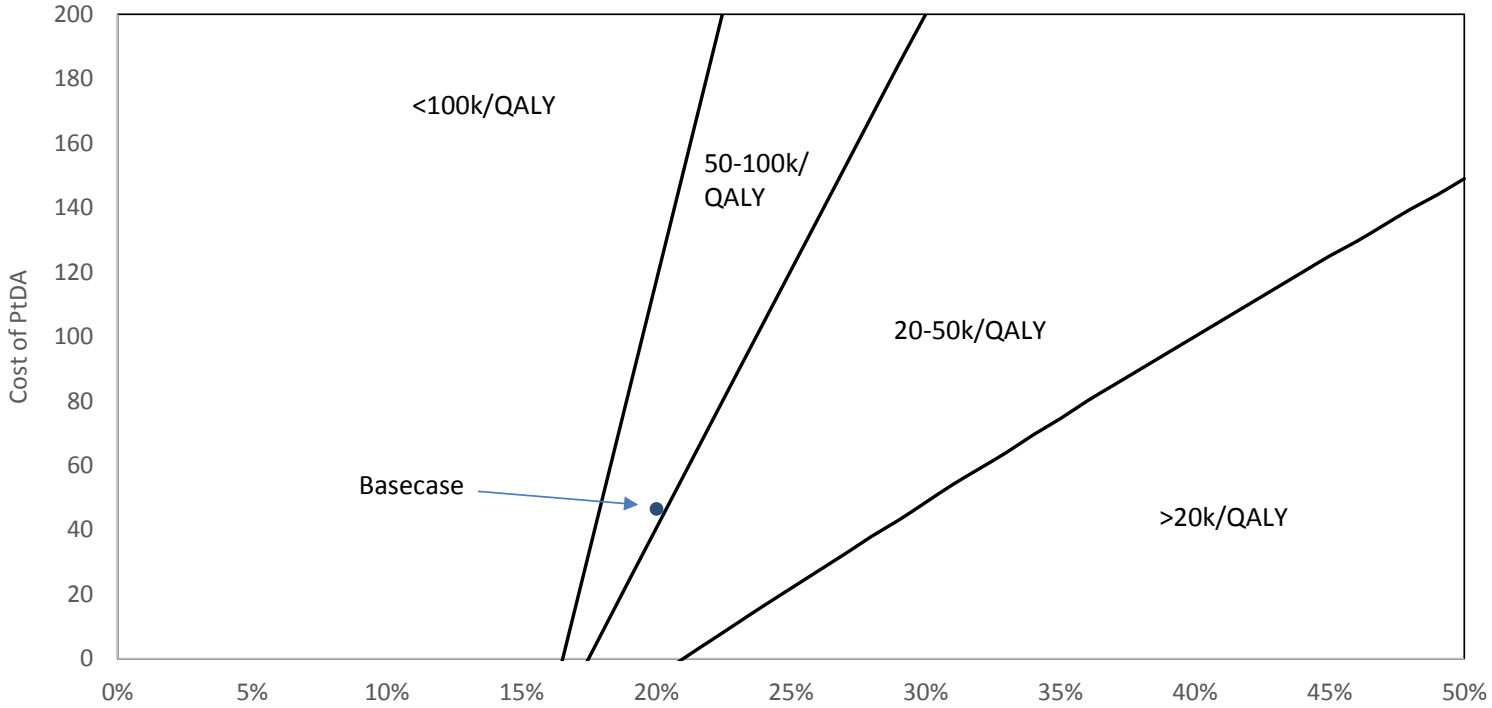


Figure 5. Cost-effectiveness acceptability curve and expected value of information



WTP = Willingness to pay for an additional QALY
CEAC = Cost-effectiveness acceptability curve
EVPI = Expected value of perfect information
EVPPI = Expected value of partial perfect information

Figure 6. Influence of cost of PtDA and adherence to treatment on incremental cost-effectiveness ratio



Corresponding 1 year adherence rates for:											
CPAP	63%	64%	67%	69%	70%	72%	74%	76%	78%	80%	82%
MAD	77%	78%	79%	80%	82%	83%	84%	85%	86%	87%	89%

4 Development of a patient decision aid prototype for adults with obstructive sleep apnea

4.1 Introduction

Obstructive sleep apnea (OSA) is a condition characterized by repeated episodes of partial or complete airway collapse during sleep which results in apneas and hypopnea.(131) Three percent of Canadians over 18 years of age report having physician diagnosed sleep apnea, though the true burden is much higher, as an estimated 70-90% of individuals with OSA are undiagnosed.(116,131,157) Risk factors for OSA include being male, over 40 years of age, overweight or obese, some ethnic groups, some genetic factors, craniofacial or upper airway abnormalities, being a smoker, consuming alcohol regularly, nighttime nasal congestion, and post menopause.(116,131) Individuals with OSA experience daytime sleepiness and decreased cognitive function which results in an increased risk of motor vehicle crashes and job-related accidents.(158,159) OSA is also associated with a significantly increased rate of cardiovascular and cerebrovascular disease, and all-cause mortality.(116,158)

Continuous positive airway pressure (CPAP), along with conservative management which includes weight loss and reduced alcohol intake, is considered the gold-standard treatment for OSA.(160) Despite being highly effective at reducing the rate of apneas and adverse outcomes, due to multiple factors including inconvenience of use, adherence to CPAP is poor. Estimates of adherence range from 28% to 83%

depending on how it is defined, severity of OSA and timing of when adherence is measured.(161) In recent years mandibular advancement splints (MAS) have emerged as a viable alternative to CPAP. These are forms of dental appliances which are more convenient to use than CPAP. While they are considered less effective at reducing OSA parameters on polysomnography, this has not translated into worse health outcomes in clinical practice.(120) This finding may be explained by greater nightly use of MAS, as many patients prefer MAS to CPAP. A recent cross-over trial with 108 newly diagnosed sleep apnea patients found that twice as many patients preferred MAS to CPAP.(81) Given the serious personal and societal consequences associated with untreated sleep apnea, ensuring that each patient has access to a treatment that he or she will use is of paramount importance. Social, economic, clinical, and personal factors all influence an individual's decision to adhere to treatment.(124) CPAP and MAS, despite treating the same condition, are fundamentally different therapies that have unique treatment characteristics. As such, the best treatment for each patient will depend upon his or her informed values and preferences.(119)

There is a growing desire within all areas of medicine to inform patients of their choices and enhance the patient-centeredness of care.(162) During the clinical encounter, this can be realized through shared decision making (SDM) between the patient and physician. One way of promoting SDM is through the use of patient decision aids (PtDAs), tools that present information about competing treatment

options, while also helping the patient deliberate and clarify their values and communicate these to their healthcare professional. Studies have shown that PtDAs are able to increase knowledge about the treatments, result in patients having more realistic expectations about the benefits and harms, and reduce decisional conflict compared to usual care.(112) PtDAs also have the potential to improve treatment adherence, as patients are more likely to receive a treatment they prefer(45), however, the evidence supporting this is lacking.(112)

This study describes the development of a PtDA prototype for adults with OSA that aims to help newly diagnosed individuals make an informed treatment decision.

4.2 Methods

Development of the OSA PtDA was guided by the International Patient Decision Aid Standards.(163,164) As outlined by Coulter et al. (see Figure 7), development of PtDAs is a multistage profess and involves the following stages: 1) scope and purpose, 2) design and evidence synthesis, 3) prototype development, 4) alpha (usability) testing, 5) beta (usability) testing. This study focuses on stages 1 through 3, describing the development of an OSA PtDA prototype.

4.2.1 Scope and purpose

To guide the development of the PtDA a multidisciplinary team of health researchers, animators, clinicians, and patients was assembled. The target population was newly diagnosed OSA patients who were considering first-line treatment options. The PtDA was designed to be completed by patients at home following OSA diagnosis by polysomnography and focus on first-line treatment options.

4.2.2 Design and evidence synthesis

4.2.2.1 Assessing patient and clinicians' views on decisional needs

Patients' views on decisional needs and key factors influencing their decision-making were previously elicited through focus groups, the results of which are published elsewhere.(124) Two clinicians, a respirologist (Dr. Najib Ayas) and a dentist (Dr. Fernanda Almeida) were consulted to elicit their views on the decisional needs of individuals presenting in their practice. Given the scope of focusing on first-line treatments, the steering group identified two primary treatment options: 1) continuous positive airway pressure (CPAP), and 2) dental appliances. Notably surgery is not chosen because is not recognized as a first-line therapy.(165) Given the high rates of non-adherence to treatment, it was also decided that 'Not using a treatment' would be presented as a third option. This allows for patients to see how the potential benefits,

harms and costs differ if they either choose neither treatment, or choose a treatment but do not adhere.

4.2.2.2 Theoretical framework

A number of relevant prescriptive and descriptive decision-making theories have informed the use and development of PtDAs, including the Behavioural Decision Framework, Conflict Model, Differentiation and Consolidation, Fuzzy Trace Theory, Image Theory, Parallel Constraint Satisfaction, the Search for Dominance, and multi-attribute utility theory.(166) Feldman-Stewart et al., note that while all have distinct properties, all models suggest that in order for individuals to make an informed decision, individuals need a developed knowledge based about the decision problem, and to establish their preferences for aspects of the different options, or the options as a whole.(166) The development and design of the OSA PtDA was guided by the practical decision making framework developed by Howard et al. (167) It encompasses a number of the above theories, and involves eliciting preferences and trade-offs between multiple competing attributes, such as effectiveness, side-effects, and costs. (168) This framework has previously been used in decision aid development,(169) and outlines 7 elements that constitute a high quality decision-making process. These include: 1) appropriate framing; 2) quality information/evidence; 3) creative alternatives; 4) clarifying values; 5)

integrating and evaluating alternatives with logic; 6) balance of basis; 7) a commitment to action.(167)

4.2.2.3 Determining format and distribution plan

Clinical evidence is constantly evolving. IPDAS standards state that the information in PtDAs should be comprehensive, critically appraised, and up-to-date.(170) Incorporating emerging evidence into paper- and video-based PtDAs can be challenging and expensive. For these reasons, the steering group decided the OSA PtDA would use an electronic platform that could be updated quickly, and at minimal cost. The dynamic computer interactive decision application (DCIDA) platform was chosen. It is a web-based platform (www.dcida.ubc.ca) that enhances decision-making using the principles of behavioural economics and allows the PtDA to be individualized based on demographic or clinical characteristics. DCIDA includes 8 stages.

Stages one through three constitute the *Introduction*, which explains the importance of the decision facing the patient, explicitly states the decision that needs to be made, outlines general features of the different options, and asks some basic demographic and clinical questions. Stages 4 and 5 constitute the V*alue Clarification* component, and ask users to consider a number of attributes that differ between the options. Based on a brief description, individuals are asked to choose the four attributes that they felt were the most important, and then rank these based on their relative

importance. Following this, they were provided with additional information about how the treatments differed on each of their attributes, and their preferences were elicited on scale ranging from poor to excellent. The results from this component were used to tailor how the information is presented in the coming stages. Stages 6 and 7 form the *Information* component, and present individualized information about each of the different options in a table format, with attributes ranked based on the values clarification exercise. Attributes chosen as the most important appear at the top, and are sized based on their relative importance. Based on the values clarification exercise, a default option is selected. This is targeted at overcoming the status quo bias which recognizes that individuals are more likely to stick with the default choice (status quo) than change this option.(171) This is followed by a brief quiz to test the users' knowledge. The final stage presents a one page summary of all results that can be printed out or emailed to the user or his or her clinician.

4.2.2.4 Review and evidence synthesis

Decisions regarding which treatment attributes to include were guided by a previous qualitative study on the treatment experiences and preferences of individuals with OSA,(124) and input from clinical experts. Seven attributes were included in the PtDA, including: 1) impact on cardiovascular disease; 2) side-effects; 3) daytime sleepiness; 4) snoring/apneas; 5) embarrassment/noise; 6) convenience/transport; 7) cost.

The latest evidence for each of the attributes was taken from the published literature, specifically a recently conducted systematic review by the Agency for Healthcare Quality (ARHQ) in the US and the complementary patient education booklet,(119,172) and content was developed using the most recent literature in risk communication.(173)

4.2.3 Prototype development

Following approval from the steering committee, an OSA PtDA prototype was developed. Weekly meetings were held between researchers and software designers to identify and resolve any issues that emerged. It also gave the team the opportunity to voice concerns and deliberate on potential solutions. Over the course of these meetings, a number of features were added to the DCIDA platform. Many were targeted at minimizing the amount of content shown to users initially, but allowing users to access this 'additional' information if desired. Two such features are text accordions that allow for text to be shown or hidden, and pop-up risk graphics or videos.

It is recognized that individuals process information through different media, so we made a point of including a variety of visual aids to help illustrate important concepts. This included a short animated video that describes the value of using a PtDA. We used an animated whiteboard style which is simple, inexpensive, and visually appealing. The video was developed in Sparkol software (North Somerset, UK) which allows the user to develop videos from static images. In addition, animated

drawings were used throughout the decision aid to help illustrate key points and to provide a cohesive feel to the visual content. The video development process was iterative and involved script writing, storyboarding and discussing image transitions. The images and voice-over were then loaded into the software and produced into the final video.

4.2.3.1 Prototype usability testing

This study was approved by the Behavioural Research Ethics Board at the University of British Columbia. The main objective of this study is to assess usability issues in the PtDA. Usability issues were defined as: when a participant was not able to advance to the next step due to design or programming error, or when a participant was distracted by a particular design or content of the online tool.(174) Usability on each of these points was assessed as follows:

1.) by determining if participants were unable to advance during the PtDA through evaluating the database and by reviewing user comments from each page, and
2.) through content analysis of open-ended user feedback

4.2.3.2 Participants

The objective was to obtain feedback from persons who would be similar to those patients that would use the PtDA. OSA sufferers who already had a diagnosis and

were being treated would bring pre-existing knowledge of the disease and treatments, and so it was chosen not to focus on this group. The ideal group would be patients who have just been diagnosed with OSA and are considering their treatment options. However, it was decided it was not appropriate to use the untested tool in this patient group at this stage, and rather use this patient group in beta testing. Instead it was decided to recruit persons with suspected OSA, but who did not have a diagnosis and were treatment naive. For this, an online survey recruiting respondents from Amazon's Mechanical Turk (MTurk) was used. MTurk is "a web-based platform for recruiting and paying subjects to perform tasks."(175) Participants have been shown to be more demographically diverse than standard internet panels or samples of college students, and "…data obtained are at least as reliable as those obtained via traditional methods."(176) The survey was advertised as an online decision tool for individuals who are over 50, experience daytime sleepiness, and snore when they sleep. Eligible individuals were those from North America, with over 1000 'human intelligence tasks' completed (meaning that they were experience MTurk workers) and an approval rating of 97% or greater (suggesting they have demonstrated an ability to complete tasks per instructions).

In the interest of recruiting those in the target audience of the PtDA, namely diagnosed individuals with OSA, only the responses of those at 'high-risk for OSA' based on the STOP-bang OSA screening questionnaire were retained(see Appendix

A).(177) Further, those taking insufficient time to appropriately complete the introductory survey (45 seconds), PtDA (5 minutes) or concluding survey (60 seconds) were also excluded. These minimum times were determined by having a group of individuals completing each section correctly, and as fast as possible. Multiple entries from the same IP address, and those using an internet proxy which did not allow for the surveys to be linked to the PtDA were also excluded.

4.2.3.3 Survey Design

After providing written informed consent, patients were provided with a URL link and unique identifier that could be used to login. Following completion of the PtDA (Appendix B and C), participants were linked to the next page of the survey, which included two scales to assess usability (see Appendix D). The first was a modified version of the patient acceptance of decision aid scale which assesses the navigation, format, length, and usefulness of the decision aid.(178) The second was the System Usability Scale (SUS), which is a ten-item scale where items are scored based on level of agreement(179). The scale is scored from 0-100, with a higher score indicating greater usability. Lastly, participants were asked to assess the strengths and weakness of the PtDA in an open-ended format across five categories; information, navigation, visual design, visual tools, and items they would change or keep the same.

4.2.3.4 Data Analysis

Participant characteristics were summarized using descriptive statistics. The patient acceptance of decision aid scale and SUS scale were used as a descriptive analysis of the PtDA. Open-ended questions were analyzed with a conventional content analysis. This involved two researchers reading all data repeatedly, followed by the development of codes that were reflective of key thoughts. (29) These codes were then sorted into positive and negative categories. Content analysis was performed by two researchers (LT and SM), and codes were represented with the number of times the code appeared and participant quotes. Any discrepancies in coding or counts were resolved through discussion. Statistical analysis was completed using R Version 3.0.2 by the R Foundation for Statistical Computing (Vienna, Austria).

4.3 Results

A total of 150 persons began the survey. In the end, 80 were retained for analysis, excluding respondents at low-risk for OSA (n=30), taking insufficient time to complete the PtDA and/or surveys (n=55), or having a duplicate IP address or using an internet proxy (n=12). There was no missing data, as online surveys require each question to be completed before advancing, thus all results are based on a sample size of 80 participants. Baseline characteristics of the study population are described in Table 7. The mean age was 54 years (SD = 8.9), and 60% of the sample was male, 78% were

university educated, and 64% were employed full-time. Participants took an average of 13.7 minutes (SD = 9.6) to complete the PtDA. In total, 39 participants chose CPAP, 25 chose dental appliances, and 16 chose no treatment.

Table 8 presents the results from the patient acceptance of decision aid scale. The vast majority of individuals thought the PtDA was useful in making a decision (n=77, 96%) and would recommend it to others (n=77, 96%). The majority felt the length (n=71, 89%), and amount of information presented (n=68, 89%) was 'just right', and that 'everything' or 'most' things (n=72, 90%) were presented with clarity. Notably, a third of participants felt the PtDA was slanted towards CPAP. Table 9 describes the results from the SUS, with the PtDA scoring a 78.22 (SD = 15.13).

4.3.1 Prototype usability testing

Firstly, we first sought to determine if there were any usability issues in the OSA PtDA. The first aspect of usability was whether a participant was not able to advance to the next step due to design or programming error. A total of 8 individuals abandoned the survey. Analysis of both the survey and PtDA databases found no specific pattern emerging with respect to where this occurred. Further, only one participant mentioned difficulty in advancing through the PtDA, experiencing 'endless loops' during the rating task. Secondly, we aimed to identify any design or content aspects that distracted

participants. This was done through 5 open-ended questions across the following domains: information, navigation, visual design, and visual tools (see table 10).

4.3.1.1 Information

The majority of participants spoke positively of the information provided in the decision aid, stating that the content was easy to understand and they would not change anything. As one participant responded, "I wouldn't change anything, it was easy to use, and read – I think it's a winner to help people like me." Many respondents made comments regarding the length of the content, with suggestions to reduce the length of the content on the introduction screen and reduce the amount of information provided on the options' risks and benefits. Many noted that there was too much information for their preference ("I would perhaps summarize the information a little more concisely"). Conversely, some requested more information on the options, including the costs of treatments, on CPAP in general, on weight loss, and on the use of the dental appliance with dentures. Some commented on the literacy level of the content, requesting "less long words" and more diagrams and visual aids to communicate information.

4.3.1.2 Navigation

On balance, participants found the decision aid easy to navigate. One participant responded, "Maybe because I am computer literate and use system like this all the time, I found the whole system and interface easy and natural – I wouldn't change anything." A number of participants enjoyed the summary screen where they could select their preferred treatment option, in addition to the "large" buttons. A small number of participants experienced challenges with the ratings task, with one participant noting "the hardest part was rating importance, I got into endless loops."

4.3.1.3 Visual design

Most participants felt positively about the visual design of the PtDA. Participants noted that it was "visually appealing," and that the "visual presentation was good." Others noted that they "loved the colors," and that the "color scheme was nice and unobtrusive." Conversely, a number of participants desired more color, with one noting that "some of the plain, black and white pages looked too unprofessional." Others expressed concern over the color scheme, noting that "some of the colors clashed with each other." Lastly, a few participants felt they could benefit from larger text, while others felt it was easy to see.

4.3.1.4 Visual tools

Most participants found the visual tools were "easy to use" and "added to the experience." Some suggested that they would like more visual tools, and that existing ones could be improved. One participant noted, "I think better pictures and diagrams could be used. The sketches were a bit basic and dull." A number of participants indicated that they did not notice the videos, with one suggesting, "I didn't notice any of the videos so I guess making those more obvious may be good." One participant made a similar content about the risk graphics, "I did not know they would come up until I pressed on the group of people. You might want to tell people about that because they might not know. I just pressed on it to see what would happen and was surprised to see the data."

4.4 Discussion

The goal of this study was to describe the development and preliminary usability testing of an online PtDA for individuals with OSA. The results from the patient acceptance of decision aid scale suggest that the vast majority of participants found the decision aid acceptable, though 33% believed it was slanted towards CPAP. The total SUS score was 78.22 (SD = 15.13). There is no universally accepted standard for what constitutes a 'good' SUS score, however, Bangor et al evaluated 2,323 surveys over 206 studies and concluded that a score of 70 was considered 'passable', with 'better'

products scoring in the high 70's or 80's, and 'truly superior' products scoring 90 or above.(181) Notably, internet web-based applications scored significantly lower than other interfaces (graphical user interface for OS-based devices, voice response systems) with a mean score of 68.05 (SD = 21.56). Based on this, the OSA PtDA is considered a good product with respect to usability.

By evaluating data from the survey and PtDA database, we found no examples where participants were unable to advance to the next step. Through analysis of open-ended survey responses, we found that in general, the information provided was easy to understand and the PtDA was easy to navigate. Further, participants had overwhelmingly positive feedback on the visual design and visual tools. A number of potential improvements were identified. Participants suggested that, in general, the amount of text could be reduced, with additional information provided through the use of visual aids, such as photos, graphics and diagrams.

The study team expressed concerns during prototype development over the amount of content in the PtDA. Despite efforts to reduce the amount of text shown to users through the implementation of accordions, a number of participants felt there was still too much information, that the text should be broken up with graphics, or that the content could be spread out over a number of pages. The two pages of the PtDA that were singled out by participants were the introduction and the ratings component of the values clarification section. In response, we have reduced the amount of text that

appears in the accordions, and decreased the number of accordions that are open by default. In the values clarification section, the study team is reconceptualising the ratings section, and attempting to integrate it within the summary table. This avoids redundancy in the information provided and streamlines the ratings task.

A third of participants felt the PtDA was slanted towards CPAP. How information is presented or 'framed' is known to have a significant impact on how individuals make decisions. Tversky and Kahneman define framing as being "controlled by the manner in which the choice problem is presented as well as norms, habits, and expectancies of the decision maker."(182) Framing information is unavoidable,(183) and given that CPAP is the more clinically effective of the two options, we believe that encouraging people to try CPAP before dental appliances is preferred to the alternative.

There are a number of limitations that warrant consideration. In order to recruit patients in our target audience (i.e., OSA patients who are treatment naïve) we used a screening tool for OSA and included those who were 'at high risk for OSA.' In a population of Canadian patients undergoing elective surgery, this screening tool has been shown to have a sensitivity and specificity of 84.1% and 40.3%, respectively, and a positive and negative predictive value of 75.3% and 54.0% respectively.(184) Based on these values, a majority of patients in our sample will have OSA, however it likely a number of individuals will not.

It has been shown that MTurk respondents 'cheat,'(185) and thus we put in a number of checks to ensure our population was at high risk for OSA, and that we received legitimate survey responses. These controls included eliminating responses with the same IP address, which may have indicated the same person completing the survey twice, and those where the IP address could not be linked. In the latter case the user may have been using a proxy server to appear in the US, when in fact they were not. Lastly, we eliminated those who completed any aspect of the survey or PtDA in a time that would indicate they were simply clicking through. While these likely eliminated a number of individuals who were not providing valid and thoughtful responses, some response bias may remain.

Our sample was highly educated, with 78% having completed some post-secondary education, and with a high-level of self-rated computer skills (76% rated excellent or very good). The OSA PtDA was designed as an online tool, but it is unclear how usability would be in a sample with less computer literacy. Given its online platform, it is only available to those that have access to the internet, and thus may increase socioeconomic health disparities.(186,187) Others have argued the opposite, that given the increasing availability of the internet and connected devices, online tools may help to reduce such disparities.(188) An additional limitation is that the PtDA is only available in English, and may not be sensitive to cultural differences. Lastly, only one of the eighty participants used a tablet, so our conclusions are limited to those who

used a desktop or laptop computer. As such, we cannot make inferences about usability on devices with lower resolution or touch screens.

4.4.1 Conclusion and next steps

In conclusion, we have described the development of an OSA PtDA prototype. We found no evidence that users became stuck or experienced errors in the PtDA during usability testing, and the majority of users found it easy to use, said that it helped them make a decision, and would recommend it for others. Feedback has been used to improve the OSA PtDA prototype. It will now undergo alpha and beta usability testing in patients at the Vancouver sleep disorders clinic at the University of British Columbia and clinicians, including Dr. Najib Ayas and Dr. Fernanda Almeida. After finalization of the OSA PtDA, it will be evaluated in a randomized trial to determine its' impact on patient choice, adherence to treatment, consultation time with physicians, and cost-effectiveness.

Table 7. Baseline characteristics of participants (n=80) for usability testing

	N	%
Gender		
Male	48	60%
Female	32	40%
Education		
Elementary school	0	0%
High school	18	23%
Post-secondary	62	78%
Annual income		
<20k	18	23%
20-40k	27	34%
40-60k	16	20%
60k+	25	31%
Work Status		
Full-time employed	51	64%
Part-time employed	13	16%
Retired	6	8%
Other	10	13%
Health Insurance		
Private/employer	47	59%
Government	15	19%
None	15	19%
Other	3	4%
Self-rated health		
Excellent	10	13%
Very Good	12	15%
Good	27	34%
Fair	4	5%
Poor	27	34%
Self-rated computer skills		
Excellent	32	40%
Very Good	29	36%
Good	14	18%
Fair	5	6%
Poor	0	0%
Device		
Desktop computer	42	53%
Laptop computer	37	46%
Tablet	1	1%
Stop-BANG Score		
3	22	28%
4	21	26%
5	13	16%
6	16	20%
7	7	9%
8	1	1%

Table 8. Patient acceptance of decision aid scale

	Yes (%)	No (%)		
Was the decision aid useful in making a decision?	77 (96%)	3 (4%)		
Would you recommend the decision aid to others?	77 (96%)	3 (4%)		
Was there enough information to decide between the options?	75 (94%)	5 (6%)		
Was it easy to understand your risk of serious medical conditions and potential benefits?	77 (96%)	3 (4%)		
Length of decision aid	Too long, 7 (9%)	Just right, 71 (89%)	Too short, 2 (3%)	
Amount of information	Too much, 6 (8%)	Just right, 68 (85%)	Too little, 6 (8%)	
Were things presented clearly?	Few, 0 (0%)	Some, 8 (10%)	Most, 36 (45%)	Everything, 36 (45%)
Was the decision aid balanced? (Slanted towards one option)	Not slanted, 53 (66%)	Slanted to CPAP, 26 (33%)	Slanted to DA, 1 (1%)	

Table 9. Modified systems usability scale (SUS)

(1=strongly disagree to 5=strongly agree)

	Mean (SD)
I liked using the decision aid as a tool for making an informed decision about treatments for OSA	3.19 (0.78)
I found the decision aid unnecessarily complex	0.90(0.83)
I thought the decision aid was easy to use	3.21(0.92)
I think I would need the support of a technical person to be able to use the decision aid	0.76(1.02)
I found the content and navigation in the decision aid was well integrated	3.08(0.83)
I thought there was too much inconsistency between the design and navigation of the decision aid	1.03(1.01)
I would imagine that most patients with OSA would learn to use the decision aid very quickly	2.98(0.81)
I found the decision aid very cumbersome to use	0.93(1.02)
I would be very confident using the decision aid	3.18(0.75)
I would need to learn a lot of things about using computers before I could get going with the decision aid	0.73(1.00)
Total SUS* score (out of 100)	**78.22(15.13)**

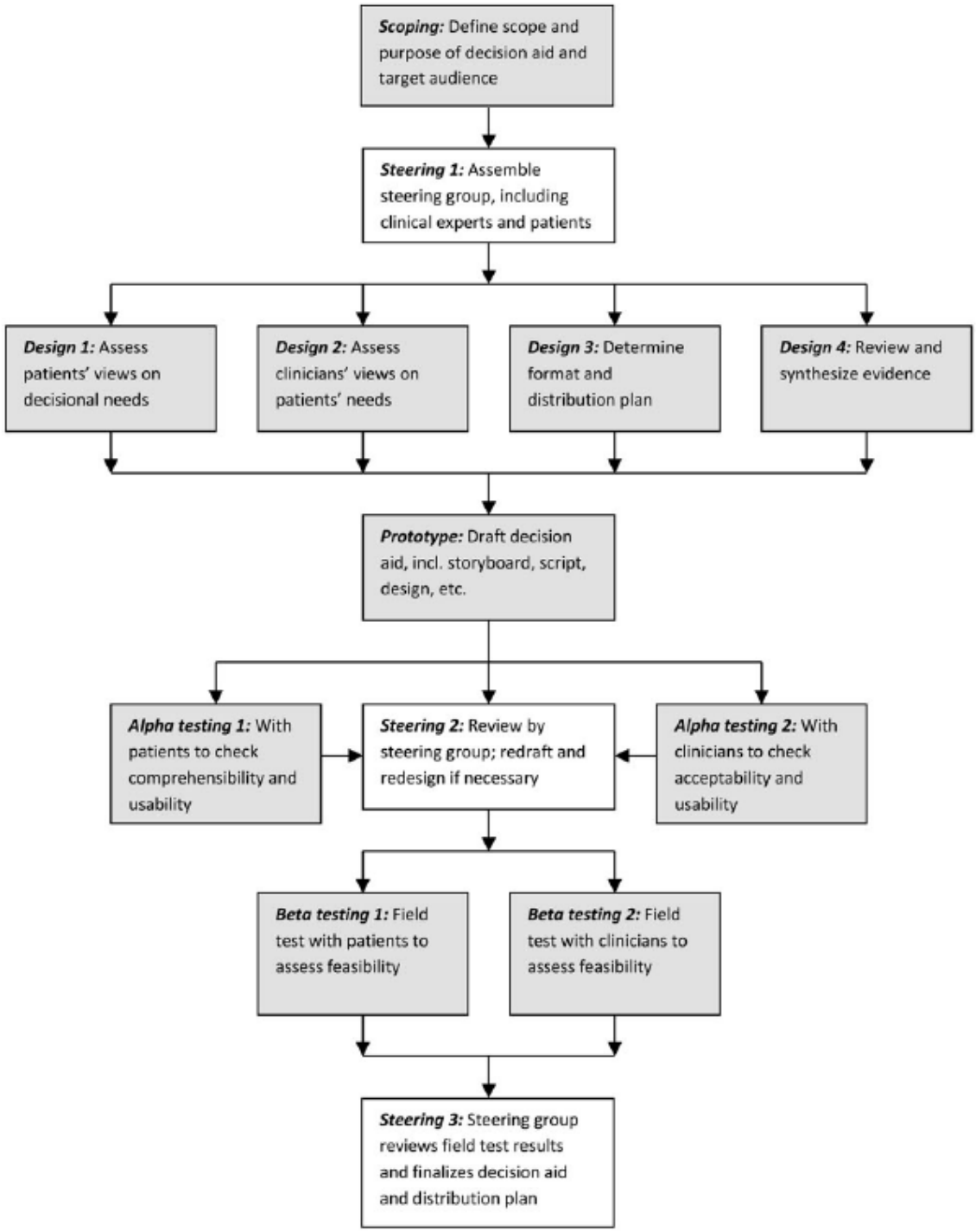
*scores of the 10 items were transformed into a summary score ranging from 0 to 100, with 100 being more user-friendly

Table 10. Open-ended feedback on the OSA PtDA

Category	Selected Quotes
Information	
Positive	"I wouldn't change anything, it was easy to use, and read – I think it's a winner to help people like me."
Easy to understand (n=16)	
Negative	
More information	"The information seemed sufficient, but why the dental appliance was so expensive, even more so than CPAP, is a mystery and could have been explained."
General (n=3)	"I would mention more about a person losing weight to change their sleep apnea condition."
CPAP/DA(n=3)	"It would be helpful if there wasn't so much info on each page."
Weight loss/Ref(n=2)	"In the risks section there was a bit too much information. Most people my age know about these risks and don't need them to be outlined further."
Less information	"Make it a bit easier to understand and not so technical."
General (n=3)	
Risk (n=1)	
Literacy (n=2)	
Navigation	
Positive	"Maybe because I am computer literate and use system like this all the time, I found the whole system and interface easy and natural – I wouldn't change anything."
Easy to navigate (n=36)	"The parts that were easiest were when an option was presented and you just had to click on the option wanted. It made the site feel intuitive and I was able to pay attention to the information more."
Summary table (n=3) Buttons(n=3)	
Sliders (n=1)	
Negative	
Ratings (n=2)	"The hardest part was rating importance, I got into endless loops."
Tables/Graph/Slider (n=3)	"The slider portion was a bit tricky but manageable."
Visual Design	
Positive	"I liked the crisp, white design. It was simple and sleek while still being very effective. The look of the guide is perfect."
General (n=20)	"I liked the size of the font, the color was easy to see."
Text (n=2)	
Negative	"Some of the plain, black and white pages looked too unprofessional…I might want the colored aspects on the other pages to be implemented…if possible."
More color (n=6)	"The color scheme was weak."
Change color scheme (n=4)	"I liked everything, but I wish there were bigger text to read."
Larger (n=3)	"I would change the red color at the top of the page."
Dislike red (n=2)	
Visual Tools	
Positive (n=20)	"The visual tools were very easy to use; they're extremely intuitive."
Negative	

Category	Selected Quotes
Pictures	
More (n=2)	"I liked everything, but would like more pictures."
Dislike cartoons (n=2)	"The sketches were a bit basic and dull."
Real pictures (n=2)	"I would use real world pictures rather than black and white graphics."
Accessibility	"I didn't notice any of the videos so I guess making those more obvious may be good."
Video (n=2)	"I did not know they would come up until I pressed on the group of people. You might want to tell people
Risk graphics (n=1)	about that because they might not know. I just pressed on it to see what would happen and was surprised to see the data."

Figure 7. IPDAS systematic PtDA development process (163)



5 Summary

The goals of this thesis were to explore the potential economic impact of PtDAs. I began by systematically reviewing the economic evidence of PtDA trials. This found that there is some evidence to suggest that PtDAs can reduce the rate of elective surgeries and associated costs in the short-term; however this evidence is fraught with methodological issues, such as an inadequate follow-up period and a lack of consideration of the impact on benefits, such as health outcomes. As modelling is a method that can be used to investigate the potential costs and outcomes of health technologies over a long time frame, I next developed a cost-effectiveness model of a PtDA for adults with moderate OSA. This serves as an example for how policy makers can determine the contexts where PtDA are both effective and cost-effective. The analysis found that a PtDA for OSA could be cost-effective provided that it increased adherence to treatment. However, the model was limited by considerable parameter uncertainty. Based on a value of information analysis, a trial of a PtDA in OSA was found to be economically justified. Lastly, I developed and preliminary usability tested an OSA PtDA with patients and clinicians that can be used in this trial.

5.1 Policy implications

It is clear that policy-makers lack sufficient economic evidence to make informed decisions about whether and where to invest in PtDAs. This could be a factor

contributing to the slow implementation of PtDAs, as health system policy-makers may be more likely to prioritize investments in interventions with a more rigorous economic evidence base. Using OSA as a case study, this work demonstrates a framework that can be used to evaluate the potential cost-effectiveness of a PtDAs for different treatments. This evaluation contributes to the sparse economic evidence of PtDAs and suggests that modelling, while requiring an up-front investment, could be a valuable avenue to evaluate the potential cost-effectiveness of PtDAs prior to their development, and/or overcome the short follow-up time frame of trial-based evaluations.

It stands to reason that the prospect of saving costs is a significant driver of PtDA development and implementation. This thesis argues that the evaluation of PtDAs should move beyond simply considering costs and simultaneously consider the benefits (or lack thereof) they afford. From a research perspective, the development of a PtDA is a costly and time-consuming process. PtDA development is often driven by perceived need by patients and physicians, with less consideration of the economic impacts and their potential of providing 'value for money.' It seems prudent that in this context, opportunity costs should be considered and investments in PtDAs, both in research and in practice, should be prioritized to those that offer the greatest potential value.

5.1.1 Addressing the conflict between the health system and individuals

Through the work on OSA, a number of important considerations were identified about the conflict between system-level objectives of cost-effectiveness analysis and clinical decisions made between the patient and their provider. As stated by Brazier, "the case for incorporating patient preferences into clinical decision making rests on the premise that this will lead to improved satisfaction and better health outcomes."(107) Through a systematic literature search, very little evidence was found to support that PtDAs improve health outcomes – though there is evidence that satisfaction is improved.(31)

One criticism of the current economic evaluation paradigm is that it relies on societal valuations of benefits. Observers note that this fails "…to take into account important inter-individual differences that might affect the value of a particular intervention. The choice that maximizes the population's health … is not always the same as the best choice for a specific individual."(189) Performing cost-effectiveness in population subgroups or even in individuals with significantly different preferences than the general population have also been proposed as methods that may help to "reflect the heterogeneity of individuals' preferences."(190) This might be most important when a subgroup possesses dramatically different preferences. Conversely, sometimes even small differences in benefit can have a dramatic impact on the cost-effectiveness, as demonstrated by the OSA cost-effectiveness model.

It is apparent that the QALY framework, currently used across the world to inform resource allocation decisions, does not adequately capture the potential benefits of PtDAs related to patients improved satisfaction with the decision-making process, and some aspects of health outcomes, such as reduced anxiety. While in theory general measures of health-related quality-of-life (HRQoL) should capture these benefits, in reality this is unlikely since most preference based measures such as the Euroqol-5D are necessarily short, and therefore can be insensitive to these changes. One potential solution is to measure these benefits and incorporate them as an add-on to current preference-based tools.

In healthcare systems that face resource constraints, there may be limitations in treatments available. Effective treatments may be denied on the basis of cost, which may create a conflict between the patient's preferred treatment and those covered by the payer.(190) This could mean that the most effective treatment is denied based on exorbitant cost, or that treatments that are less cost-effective than current practice are not covered, despite being preferred by a subgroup of the population. This is the case in Ontario, where CPAP is currently covered while dental appliances are not. In British Columbia neither treatment is covered by public agencies. Given the preference-sensitive nature of the decision between treatment options for patients with OSA, payers need to be cognisant of how coverage decisions may impact patient choice. In Ontario, for example, it is conceivable and perhaps likely that a considerable number of

informed OSA patients would choose CPAP over dental appliances on the basis of out-of-pocket costs. Encouraging the use of CPAP as a first-line treatment is defensible given it is the more clinically effective of the two options at reducing the rate of apneas and other objective parameters as measured by polysomnography.(120) However, this established view of CPAP as the superior treatment due to its clinical effectiveness masks the importance of adherence which can be influenced by a variety of factors, such as convenience and cost. Policy makers should recognize that patients judge effectiveness on more than just clinical effectiveness – which can influence which treatment they prefer and whether or not they adhere to a treatment. From a policy perspective, the goal should be top ensure that patients as possible are using a therapy. In a jurisdiction like Ontario where CPAP is covered, this could be accomplished through a policy whereby those that fail CPAP therapy would be eligible for a dental appliance covered by the government. It has been suggested that providing patients with information about the cost the options, even those covered by the public system, could allow patients to consider the cost-effectiveness of treatments when making their decisions. However it is unclear if individuals are willing to act altruistically to achieve the social goals of efficiency and equity.(190)

5.1.2 Engaging physicians in PtDA implementation

Physicians are key players in the successful implementation of PtDAs. In Washington State, a pilot project was undertaken which sought in integrate SDM into clinical practice through the use of PtDAs. One of the participating groups, Group Health, covers over 600,000 individuals in Washington and Idaho States, implemented PtDAs for a variety of elective surgeries. This experience highlighted important considerations around PtDA implementation and physician incentives. The physicians at Group Health were salaried rather than fee-for-service. Much of the demand for elective surgeries is believed to be driven by physicians – a concept known as 'supplier induced demand'.(191) When physicians are salaried they do not have the same incentive to provide services, and are less likely to object to the use of PtDAs. The pilot project was considered successful, however "only one-third of patients identified as having hip or knee osteoarthritis received a decision aid."(75) This highlights the fact that strategies to increase PtDA usage, especially in Canada where physicians are fee-for-service and have incentive to provide services, will need to be multifaceted and require physician buy-in.(192)

In certain contexts it may be worth reimbursing physicians to use PtDAs to overcome potential financial losses. There are other incentives that could be used to support the use of PtDAs. In Washington State, policy makers have embedded the use of PtDAs and SDM into law as evidence of informed consent.(193,194) Given the highly

litigious nature of the United States and the country's healthcare system in particular, protection against litigation could serve as a significant incentive for physicians to use PtDAs. It remains to be seen if such policy could be applied in less litigious jurisdictions, such as Canada; however, it is one option that policy makers can consider. Payers could also require that physicians use and document, through a signature by both themselves and the patient, the use of PtDAs for specific health decisions as a prerequisite for funding. This has on part been implemented in British Columbia for the special authorization of biologic agents for rheumatoid arthritis, where physicians must document use and failure of first-line treatments to BC pharmacare before a biologic can be prescribed. By incorporating a PtDA into this process, it could help overcome the overuse of biologics in patients who prefer less toxic and cheaper alternatives.

5.2 Questions for future research

This research has raised a number of questions that warrant future investigation and which I plan to pursue through my doctoral program of research. They are as follows:

1) How can PtDAs best be evaluated through the use of trial- and model-based techniques?
    a. How does incorporating evidence from the OSA PtDA trial impact the cost-effectiveness?

b. Is a PtDA for adults with hip and knee osteoarthritis cost-effective?

2) Do patients value the SDM process?
    a. Can this value be incorporated into QALY measures to inform reimbursement decisions?
    b. How does incorporating this value into the economic analysis impact the cost-effectiveness results?
3) How can physicians be incentivized to employ shared decision-making in preference sensitive situations?

First, the OSA PtDA will be further refined through interviews with individuals at the Vancouver sleep clinic and expert clinicians (beta testing). When completed, it will be evaluated in a recently CIHR funded three-arm trial. In this, individuals will be randomized to either 1) CPAP, 2) dental appliance, or 3) the PtDA where they will choose their preferred treatment. This trial will provide valuable information on the benefit of the PtDA, but will also provide important evidence for its cost-effectiveness. Notably, it will reduce the uncertainty in a number of key parameters, such as the cost of implementation (we will measure the time the physician spends with the patient in each arm), the proportion of informed patients choosing specific treatments, and the subsequent impact on adherence. Following the completion of this trial, I will then

complete an economic evaluation based on the trial data which will allow for comparison with our modelling results.

Concurrently, I will be performing a trial-based economic evaluation of a trial where a PtDA was used for end-stage hip and knee osteoarthritis. The trial is completed and followed approximately 350 patients for 2-years after using a PtDA. In Chapter 2, it was noted that the longest any economic evidence has been collected in a PtDA trial is 2-years, and that longer term evidence is required in order to fully understand the economic impact of PtDAs. I plan on extending this analysis beyond the 2-year follow-up period in one of two ways. Ideally, I will be able to link trial data with administrative data that will provide a total follow-up of approximately 4 years. If this linkage is not possible, I will extend the trial data with a Markov cohort decision-analytic model with a lifetime horizon. Regardless of the method, this analysis will add to the sparse body of economic evidence for PtDAs.

Finally, it has also been noted that current economic evaluations of PtDAs likely underestimate the benefits they provide. During my PhD, I propose to also investigate how much patients value the SDM process, with the aim of incorporating this value within QALY measurement. My preliminary plan is to focus on OSA and hip and knee replacement decisions, with the purpose of investigating how much individuals value their physician engaging in SDM when making a treatment decision. In order to do so, I will develop a survey that will ask participants to imagine having been diagnosed with

the disease, and needing them to decide between conservative management with pain medication and exercise or having knee replacement surgery. In order to make their decision, they will be asked to trade-off a longer waiting time for a surgeon/physician that puts greater effort in SDM. SDM descriptions will be based on descriptions from the collaboRATE scale,(195) conceptualized as the physician putting effort into explaining the patient's health issues, listening to the things that matter to the patient, and including what matters most to the patient in choosing what to do next. The objectives are to determine: 1) the maximum amount of time patients are willing to wait for a consultation with a surgeon that engages in SDM (greater or less than a surgeon that does not), and 2) how this willingness to wait is influenced by the pain they are experiencing and the probability that SDM will influence their treatment choice. I hypothesize that individuals will be willing to wait longer for a consultation with a physician who engages in SDM, and that individuals will be willing to wait longer if the pain they are experiencing in their knee is less severe. I plan to refine the survey and incorporate any addition benefit as an add-on to EQ-5D. If we are able to do so, I will compare the cost-effectiveness of the hip and knee osteoarthritis PtDA with, and without, this additional benefit.

The success of implementing PtDAs into clinical practice hinges on support from practicing physicians. I also plan to investigate how we can incentivize physicians to use PtDAs. This may involve comparing different types of incentives, such as protection

from litigation or additional payments. Such work would consider how physicians' desire to use PtDAs is influenced by these incentives and other benefits, such as shorter consultation times, improved health system efficiency, improved patient adherence to treatment, and improved health outcomes. Findings from this research could help determine what evidence or policies would encourage the use of PtDAs by physicians.

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Appendix A: STOP-bang OSA screening tool

1. Do you snore loudly (louder than talking or loud enough to be heard through closed doors)?
2. Do you often feel tired, fatigued, or sleepy during daytime?
3. Has anyone observed you stop breathing during your sleep?
4. Do you have or are you being treated for high blood pressure?
5. BMI more than 35 kg/m^2?
6. Age over 50 yr old?
7. Neck circumference greater than 40 cm?
8. Gender male?

High risk of OSA: answering yes to three or more items
Low risk of OSA: answering yes to less than three items

Appendix B: OSA PtDA prototype content

Intro page

Welcome to the sleep apnea decision aid

There are 7 simple steps in this tool that will start you on the path to making an informed treatment decision

Introduction

Who is this decision aid for?
This decision aid is designed for adults recently diagnosed with obstructive sleep apnea (OSA). It provides information about OSA and treatment options.
This is NOT for:

- people who have **not** been diagnosed with OSA by their doctor
- children (under 18 years of age)
- people who have been diagnosed with *central sleep apnea*.

Why is this decision important?
Deciding between treatments for OSA is challenging, because there is no "best" treatment option.
The options are very different and deciding what is best for you means making trade-offs between things like effectiveness, side effects, convenience, and cost.
It is hard for doctors to select a treatment for you, because they don't always know what is important to you.
This tool will help you review your options and think about what you prefer, so you can make an informed decision with your healthcare provider.

What treatment options does this decision aid discuss?
This decision aid covers the 2 most popular, well-researched treatments for OSA:
1.) Continuous positive airway pressure (CPAP)
2.) Dental appliance

What is obstructive sleep apnea?
Obstructive sleep apnea (OSA) is a chronic (i.e., ongoing) sleep disorder. People with OSA experience stops, "pauses," or shallow breathing when they sleep. An estimated 365,000 Canadians have OSA. Many more people may have OSA and do not know it because they have not been tested.

Who is affected by OSA?

- OSA is very common. About 4% of men and 2% of women have been diagnosed with OSA.
- OSA affects people of all ages, but it is more common in those over 40 years of age and those who are overweight or obese.
- Other risk factors for OSA include smoking and alcohol consumption, nighttime nasal congestion, menopause, and genetic factors.

What happens during OSA?

- People with OSA experience pauses in their breathing (called "apneas") or shallow breathing (called "hypopnea")
- Both apneas and hypopneas result in low levels of oxygen in the blood.
- In order to be diagnosed with OSA, people must experience at least 5 pauses per hour.
- People with more severe OSA may experience pauses 30 to 60 times per hour.
- Often people with OSA start breathing again with a loud snort or choking sound.

How is OSA classified?

OSA can be mild, moderate, or severe, depending on:

- The number of times an hour a person pauses their breathing or has lower airflow.
- The oxygen level in a person's blood.
- How sleepy a person feels during the day.

Can losing weight cure OSA?

- Regardless of the choice you make about treatment, your doctor may recommend losing weight. Some overweight/obese people may reduce or cure their OSA by losing weight.

How serious is OSA?

Untreated OSA may:

- Cause poor sleep quality, leading to daytime sleepiness and an increased risk of accidents at work or when driving.
- Increase the risk of serious health problems, including heart attacks, strokes, diabetes, and even death.

Where does this information come from?
Researchers from the University of British Columbia have reviewed the evidence and asked patients with OSA what they think about treatments. The researchers have not been funded by the makers of any OSA treatments and they are impartial about which treatment you use.

How does my doctor know if I have OSA?

- Your doctor may ask you questions about whether you have symptoms of OSA.
- Your doctor or health care provider may ask you to do a "sleep study." This is an overnight stay at a special clinic where trained professionals watch your breathing, heart rate, and other vital signs while you sleep.
- Your doctor may use a home monitor to check how often you pause or stop breathing or have less airflow when you sleep.

Disclaimer

- The information in this decision aid does not replace the advice of a doctor. The University of British Columbia and the researchers who developed this decision aid disclaim any warranty or liability for your use of this information.

About Me

In order to provide individualized information, please answer the following questions about yourself.

1. How severe is your OSA?
   ○ Mild ○ Moderate ○ Severe ○ Don't know

2. Are you over 18 years of age? (This guide does not cover treatment for children)
   ○ Yes ○ No

3. Are you over 65 years of age?
   ○ Yes ○ No

4. What is your sex?
   ○ Male ○ Female

5. Your body mass index (BMI) suggests you are:
   ○ Normal weight ○ Overweight ○ Obese ○ Don't Know

My Options

The next pages will help you to consider which OSA treatment you prefer. These options treat your OSA by keeping your airway open while you sleep.

Continuous positive airway pressure (CPAP) machine

- A CPAP machine pushes a stream of air through a mask you wear while you sleep. The air flows through the mask into your nose or mouth to keep your throat and airway open.
- There are many kinds of CPAP machines and masks. Some masks fit over your nose and others cover both your nose and mouth.
- CPAP is the most common and most researched treatment for OSA. It is usually the first treatment that a doctor will suggest for OSA.
- *In order to be effective, CPAP needs to be used every time you sleep. Most people need to continue using CPAP for their entire lives.*

Dental Appliance

- A dental appliance is a mouthpiece you wear while sleeping. The mouthpiece keeps your jaw forward and your airway open. Some devices also hold your tongue in place.
- There are many types of dental appliances. Most are made of hard plastic that covers your upper and lower teeth. These devices are fitted and sold by a dentist or orthodontist, who makes a mold of your mouth to create a customized device.
- *In order to be effective, a dental appliance needs to be used every time you sleep. Most people need to continue using a dental appliance for their entire lives.*

Not Using a Treatment

- You may choose not to use a treatment. This could involve not purchasing a treatment or purchasing one but not using it regularly.
- Losing weight may also help reduce your OSA symptoms.
- In the first year after being diagnosed with OSA, as many as 40% of patients do not use a treatment or do not use it properly.
- **Not using a treatment may be more convenient and less costly in the short term, but it would increase your risk of serious long-term medical conditions including heart attack, stroke, diabetes, cancer, and death.**

Other Treatments
Surgery

- Some people with OSA may have surgery to remove tissue from the back of the throat. This makes the airway wider.
- All surgeries come with risks, including bleeding and infection, nerve damage, and even death. After surgery, some people experience difficulty swallowing, change in speech or voice, or narrowing of their airway.
- There is not enough research to compare surgery to the other treatments for OSA.
- Researchers believe that surgery can improve OSA, but they do not know how well and who might benefit from it most.

Other Options

- There are other treatments and products that claim they can stop snoring or sleep apnea. There is not enough research to know if or how well these work.
- You may see ads for mouthpieces that claim to help stop snoring or sleep apnea. These devices are purchased directly from companies and do not need fitting. These are not the same as the dental appliances studied by researchers. Always check with your doctor or dentist before ordering a mouthpiece.

My Values

There are many issues to consider when deciding between treatment options for OSA. Things that are important to some people are not important to others.
Please carefully review each of the issues below and pick the **FOUR** that are most important to you. You can return to this page to change your choices at a later point.

1. **Cardiovascular disease**: People with sleep apnea are at increased risk of cardiovascular disease, including high blood pressure, heart attack, and stroke. Without appropriate treatment, your risk of heart attack and stroke is about 2 times higher than with treatment.
2. **Side Effects**: OSA treatments have different side effects, which range from mild (dry throat, blocked up nose, increased awakenings during the night) to more severe (pain, shifting bite, anxiety and claustrophobia).
3. **Daytime Sleepiness**: You may start dozing off in situations when you are reading, watching television, sitting quietly, lying down, talking to someone, or driving. With appropriate treatment your daytime sleepiness will be reduced.
4. **Snoring/Apneas**: Apneas - incidents where you stop breathing during sleep - can contribute to snoring or gasping. You may or may not be aware that apneas are happening through the night. When used properly, treatments for OSA can completely reduce apneas in 40-75% of people.

5. **Embarrassment/Noise**: Treatments differ in how they look when worn, and the noise they make. Some people report being embarrassed when wearing their treatment in front of other people. Treatments need to be used every night in order to be effective, but embarrassment or noise may influence how often you use yours.
6. **Convenience/Transportability**: Treatments may be as large as a backpack or as small as a mouth guard. This may influence your desire to take your treatment with you if you travel.
7. **Cost**: Treatments have different up-front costs and recurring costs. Up-front costs range from $1,000 to $2,500 depending on the treatment. The amount you pay out-of-pocket depends on the type of health insurance you have.

Consider further what matters most to you. Is each of these issues equally important or does one issue matter much more than the others?

Move the sliders to show the relative importance of these four issues and press 'Compare Options' when you are finished.

Compare Options

Below is information on the issues you selected.

Spend some time thinking about the information for each issue and then rate each issue with the buttons. You may explore additional issues by selecting 'Additional Features' at the bottom.

Cardiovascular disease: Treatments for OSA can potentially reduce your risk of cardiovascular disease, including high blood pressure, heart attacks, and stroke. When using no treatment, about 2 out of 100 (2%) people like you will experience a heart attack or stroke over the next year.

- CPAP

  When using CPAP, about 1 in 100 (1%) fewer people like you (1 in 100 total) will experience a heart attack or stroke over the next year compared to no treatment. This is the same as healthy people without OSA.

- Dental Appliance

  When using a dental appliance, about 1 in 100 (1%) fewer people like you (1 in 100 total) will experience a heart attack or stroke over the next year compared to no treatment. This is the same as healthy people without OSA.

Side Effects: Treatments for OSA have side effects. Most are minor but they may influence how often you use treatment.

CPAP: The most common side effects of CPAP include: dry nose, mouth, or throat, nosebleeds, sore gums or lips, chest discomfort, and/or feeling trapped or claustrophobic.

MAD: The most common side effects of dental appliance include mouth dryness, teeth pain, excessive salivation, jaw pain, and permanent changes in the bite. Many people who use a dental appliance have to do jaw stretches in the morning or see a dentist regularly to check for teeth problems.

Daytime Sleepiness: Treatments for OSA decrease your daytime sleepiness and fatigue, which decreases your risk of motor vehicle crashes and work accidents. When using no treatment your daytime sleepiness will remain the same.

- CPAP

  When using CPAP, your daytime sleepiness will be reduced compared to no treatment.

- Dental Appliance

  When using a dental appliance, your daytime sleepiness will be reduced compared to no treatment.

Snoring/Apneas: Treatments for OSA decrease the number of times you stop breathing while you sleep (apneas), and can potentially eliminate or reduce snoring. When using no treatment, people like you experience between 15 and 30 apneas per hour, on average.

- CPAP

  When using CPAP, about 75 in 100 (75%) of people like you experience a complete treatment response (less than 5 apneas per hour).

- Dental appliance

  When using a dental appliance, about 40 in 100 (40%) of people like you experience a complete treatment response (less than 5 apneas per hour).

Embarrassment/Noise: Treatments differ in how they look when worn and the noise they make. Some people report being embarrassed when wearing their treatment in

front of other people. Treatments need to be used every night in order to be effective, but embarrassment or noise may influence how often you use yours.

- CPAP: CPAP can be noisy and may disturb you and/or your bed partner's sleep. It involves wearing a face mask connected to a long breathing tube that can cause embarrassment in some people.
- Dental appliance: A dental appliance does not make any noise. It is placed in the mouth like a sports mouth guard.

Convenience/Transport: Treatments differ in size and how difficult they are to clean. This may influence your desire to use them or take them with you when you travel.

- CPAP: CPAP machines can be quite large and difficult to pack for travelling. They require a power source to operate, and some require a source of fresh water to fill the humidifier.
- Dental appliance: A dental appliance is small and easy to pack for travelling. It may require warm water to clean it before and after use.

Cost: Treatments have different up-front costs and recurring costs. Importantly, the amount you pay out-of-pocket depends on the type of health insurance you have. Using no treatment will not cost you any money.

- CPAP:
    - The average retail cost (before insurance payment) of most CPAP machines is between $750 and $2,000. A machine will last on average 5 years.
    - The average cost of CPAP supplies (mask, tubes, and filters) is between $300 and $800 per year. You will have to buy new supplies every year.
- MAD:
    - The average retail cost (before insurance payment) of a dental appliance is between $2,000 and $2,500. Your custom device will last on average 3 years.
    - Using a dental appliance may require that you visit a dentist more often to check for teeth and jaw problems. Dental visits may be covered by your health plan.

8. My Choice

Below is a summary of your options. Please compare them and select the one that you feel is the best fit for you. We have highlighted in pink the option that we think best matches your profile, but only you will know which treatment is best.

Review

Please answer the following questions to the best of your ability. Your answers will tell us how well we presented the information, how you feel about your choice, and how much this decision aid has improved your knowledge. Your personalized summary will be provided on the next page.

Knowledge Questions

Which of the following is NOT a risk factor for OSA?

a. Obesity
b. Older Age (> 40)
c. Alcohol Consumption
d. None of the above

Untreated OSA increases your risk for which serious medical condition?

a. Heart attack and stroke
b. Cancer
c. Diabetes
d. Death
e. All of the above

CPAP stands for:

a. Continues to push air past your nose
b. Close passages and pressures
c. Continuous positive airway pressure
d. Central pauses and pressures

Treatments work best when used:

a. Every night
b. Every second night
c. Weekdays only
d. Weekends only

Other Questions

Do you feel you know the benefits, risks, and side effects of each treatment?

- Yes
- No

Are you clear about which aspects of the different options are most important to you (benefits, side effects, convenience, etc.)?

- Yes
- No

Do you feel that you have enough information and advice to make a choice?

- Yes
- No

Do you feel sure about what option is best for you?

- Yes
- No

Summary and Next Steps

You have completed the decision aid! We notice that you have chosen <treatment> as your preferred treatment. We would like to encourage you to try CPAP because the evidence suggests that it is more effective at reducing your long-term risk of serious medical conditions. Many places offer a free one-month trial so you can determine if CPAP will work for you without purchasing a machine. If you don't like CPAP, we would then encourage you to talk to your doctor about a dental appliance. Regardless of what you choose to do, we hope that you have more knowledge about the options available to you.

Appendix C: OSA PtDA prototype screenshots

OBSTRUCTIVE SLEEP APNEA DECISION AID

1. ABOUT ME | 2. MY OPTIONS | 3. MY VALUES | 4. COMPARE OPTIONS | 5. MY CHOICE | 6. NEXT STEPS | 7. SUMMARY

Introduction

▾ Who is this for?

This decision aid is designed for adults recently diagnosed with obstructive sleep apnea (OSA). It provides information about OSA and treatment options.

This is NOT for:

- people who have **not** been diagnosed with OSA by their doctor
- children (under 18 years of age)
- people who have been diagnosed with *central sleep apnea*.

▾ Why is this decision important?

- Deciding between treatments for OSA is challenging, because there is no "best" treatment option.
- The options are very different and deciding what is best for you means making trade-offs between things like effectiveness, side effects, convenience, and cost.
- It is hard for doctors to select a treatment for you, because they don't always know what is important to you.
- This tool will help you review your options and think about what you prefer, so you can make an informed decision with your healthcare provider.

Watch Video

▾ What treatment options does this decision aid discuss?

This decision aid covers the 2 most popular, well-researched treatments for OSA:

1. Continuous positive airway pressure (CPAP)

OBSTRUCTIVE SLEEP APNEA DECISION AID

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The next pages will help you to consider which OSA treatment you prefer. These options treat your OSA by keeping your airway open while you sleep.

OPTIONS

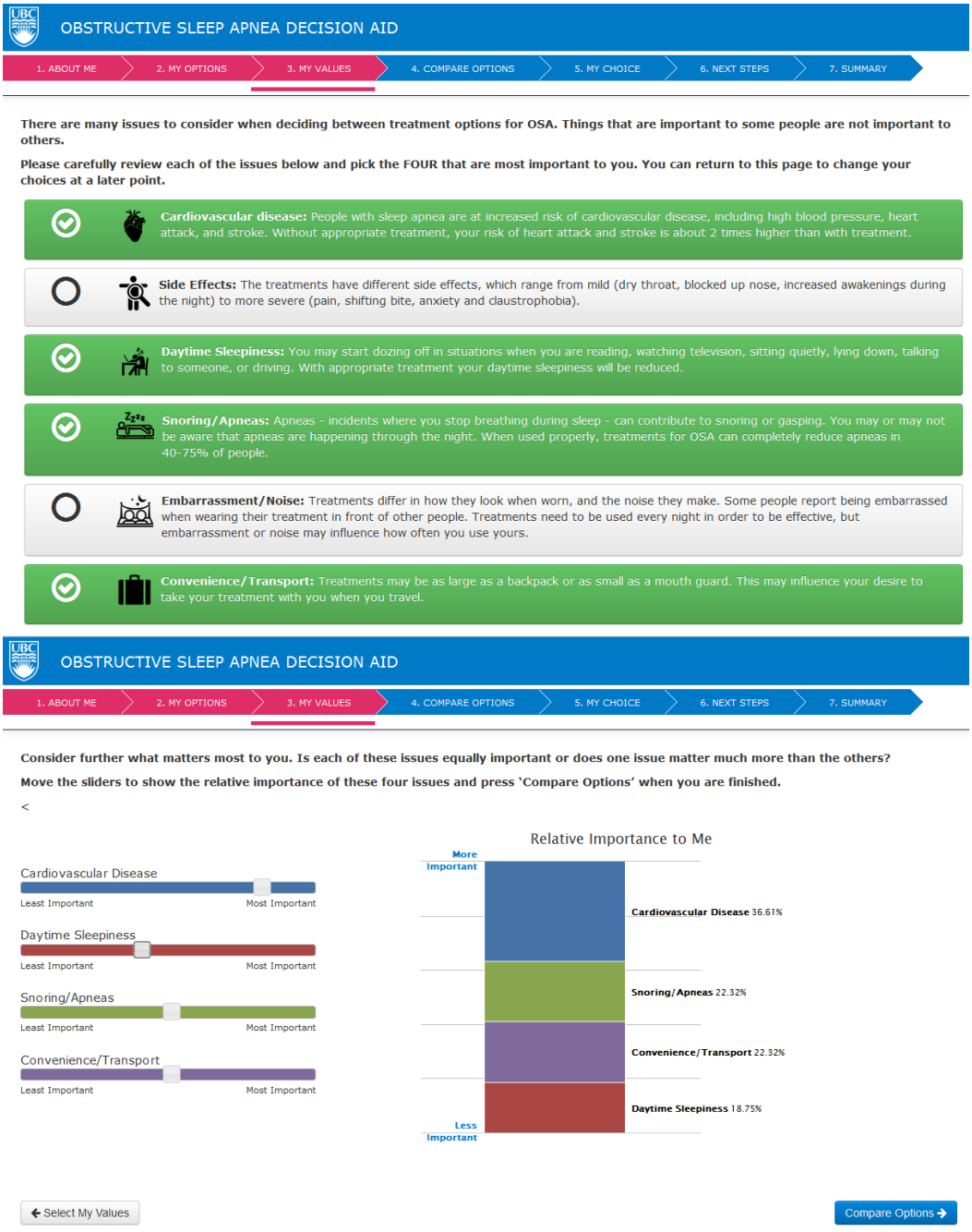
- Continuous Positive Airway Pressure (CPAP)
- Dental Appliance
- Not Using a Treatment

Continuous Positive Airway Pressure (CPAP)

- A CPAP machine pushes a stream of air through a mask you wear while you sleep. The air flows through the mask into your nose or mouth to keep your throat and airway open.
- There are many kinds of CPAP machines and masks. Some masks fit over your nose and others cover both your nose and mouth.
- CPAP is the most common and most researched treatment for OSA. It is usually the first treatment that a doctor will suggest for OSA.
- *In order to be effective, CPAP needs to be used every time you sleep. Most people need to continue using CPAP for their entire lives.*

Dental Appliance

- A dental appliance is a mouthpiece you wear while sleeping. The mouthpiece keeps your jaw forward and your airway open. Some devices also hold your tongue in place.
- There are many types of dental appliances. Most are made of hard plastic that covers your upper and lower teeth. These devices are fitted and sold by a dentist or orthodontist, who makes a mold of your mouth to create a customized device.
- *In order to be effective, a dental appliance needs to be used every time you sleep. Most people need to continue using a dental appliance for their entire lives.*


OBSTRUCTIVE SLEEP APNEA DECISION AID
1. ABOUT ME
2. MY OPTIONS
3. MY VALUES
4. COMPARE OPTIONS
5. MY CHOICE
6. NEXT STEPS
7. SUMMARY
There are many issues to consider when deciding between treatment options for OSA. Things that are important to some people are not important to others.
Please carefully review each of the issues below and pick the FOUR that are most important to you. You can return to this page to change your choices at a later point.
Cardiovascular disease: People with sleep apnea are at increased risk of cardiovascular disease, including high blood pressure, heart attack, and stroke. Without appropriate treatment, your risk of heart attack and stroke is about 2 times higher than with treatment.
Side Effects: The treatments have different side effects, which range from mild (dry throat, blocked up nose, increased awakenings during the night) to more severe (pain, shifting bite, anxiety and claustrophobia).
Daytime Sleepiness: You may start dozing off in situations when you are reading, watching television, sitting quietly, lying down, talking to someone, or driving. With appropriate treatment your daytime sleepiness will be reduced.
Snoring/Apneas: Apneas - incidents where you stop breathing during sleep - can contribute to snoring or gasping. You may or may not be aware that apneas are happening through the night. When used properly, treatments for OSA can completely reduce apneas in 40-75% of people.
Embarrassment/Noise: Treatments differ in how they look when worn, and the noise they make. Some people report being embarrassed when wearing their treatment in front of other people. Treatments need to be used every night in order to be effective, but embarrassment or noise may influence how often you use yours.
Convenience/Transport: Treatments may be as large as a backpack or as small as a mouth guard. This may influence your desire to take your treatment with you when you travel.
OBSTRUCTIVE SLEEP APNEA DECISION AID
1. ABOUT ME
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6. NEXT STEPS
7. SUMMARY
Consider further what matters most to you. Is each of these issues equally important or does one issue matter much more than the others?
Move the sliders to show the relative importance of these four issues and press 'Compare Options' when you are finished.
Cardiovascular Disease
Least Important
Most Important
Daytime Sleepiness
Least Important
Most Important
Snoring/Apneas
Least Important
Most Important
Convenience/Transport
Least Important
Most Important
Relative Importance to Me
More Important
Less Important
Cardiovascular Disease 36.61%
Snoring/Apneas 22.32%
Convenience/Transport 22.32%
Daytime Sleepiness 18.75%
Select My Values
Compare Options

Appendix D: Usability questionnaire

Patient acceptance of decision aid

- Was the decision aid useful in making a decision?
  - Yes, No
- Would you recommend the decision aid to others?
  - Yes, No
- Was there enough information to decide between the options?
  - Yes, No
- Was it easy to understand your risk of serious medical conditions?
  - Yes, No
- Length of decision aid:
  - Too long, Just right, Too short
- Amount of information:
  - Too much, Just right, Too little
- Clarity: were things presented clearly?
  - Few, Some, Most, Everything
- Was the decision aid balanced? (Slanted toward one option?)
  - Not Slanted, Slanted towards CPAP, Slanted towards dental appliance

Modified system usability scale

(1 = Strongly disagree; 5 = Strongly agree)

1. I liked using the decision aid as a tool for making an informed decision about treatments for OSA/knee pain
2. I found the decision aid unnecessarily complex
3. I thought the decision aid was easy to use
4. I think I would need the support of a technical person to be able to use the decision aid
5. I found the content and navigation in the decision aid was well integrated
6. I thought there was too much inconsistency between the design and navigation of the decision aid
7. I would imagine that most patients with OSA/knee pain would learn to use the decision aid very quickly
8. I found the decision aid very cumbersome to use
9. I would be very confident using the decision aid
10. I would need to learn a lot of things about using computers before I could get going with the decision aid

Prompting Questions (open-ended response)

1. What would you change about the information provided in this decision aid? What information was easy or hard to understand?
2. What parts of the decision aid were easy to navigate? What parts were hard to use?
3. What did you like or dislike about the visual design of the decision aid? Do you have feedback on the colour scheme, text, or overall look of the decision aid?
4. What are your thoughts on the visual tools used in the decision aid? Do you have any feedback on the videos, pictures, or diagrams provided?
5. What is one thing you would change about this decision aid? What is one thing you would keep the same?