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Title: Economic evaluation strategies in telehealth: Obtaining a more holistic valuation of telehealth interventions

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Abstract

Telehealth is an emerging area of medical research. Its translation from conception, to research, and into practice requires tailored research and economic evaluation methods. Due to their nature telehealth interventions exhibit a number of extra-clinical benefits that are relevant when valuing their costs and outcomes. By incorporating methods to measure societal values such as patient preference and willingness-to-pay, a more holistic value can be placed on the extra-clinical outcomes associated with telehealth and evaluations can represent new interventions more effectively. Cost-benefit analysis is a method by which relevant costs and outcomes in telehealth can be succinctly valued and compared. When health economic methods are conducted using holistic approaches such as cost-benefit analysis they can facilitate the translation of telehealth research into policy and practice.

Introduction

Economic evaluations inform decisions regarding the models for health care provision, including the design and delivery of new services such as telehealth interventions. Tailoring analysis methods appropriately to the intervention and context being studied is vital in order to produce findings that have generalisability outside of the research environment ¹. Economic analysis requires the assessment of costs and benefits associated with different services, enabling existing services to be compared with new interventions not just in terms of clinical efficacy (as in clinical research) but in terms of

costs and therefore value for money. Understanding and applying economic methods in health research is vital to promote the efficiency and sustainability of healthcare – one of the core goals that telehealth aims to achieve.

The expected growth of telehealth solution adoption is higher than the observed growth. REF Consequently, the question of the efficiency of telehealth as a solution for the delivery of healthcare is contentious. The lack of a clear answer on the efficiency of telehealth services may be due to a number of factors, including a lack of economic evaluations undertaken for telehealth interventions, the quality of existing studies, and the difficulty in measuring and valuing meaningful outcomes from telehealth interventions. REF This latter issue can be addressed by careful consideration of the outcomes of importance and the design of the economic evaluation, as we will discuss shortly.

When undertaking an economic analysis of a new telehealth intervention an understanding of the relevant service outcomes that might change as a consequence of the intervention and be meaningful from the perspective of the relevant payer and beneficiary is necessary to appropriately assess the value. However, the many possible societal benefits of telehealth extend beyond the health benefits that are captured in conventional approaches to economic evaluation and which have generally been

measured and valued in healthcare studies to date. For example, improved equity of access for isolated populations, access to specialist opinion, and reduced travel requirements all feature as benefits in telehealth and may not be captured by conventional approaches ².

In this paper, we discuss the potential limitations of using conventional measures of health outcome in the economic evaluation of telehealth interventions. We highlight the potential for willingness to pay to overcome some of these limitations within a cost-benefit evaluative framework, as well as some of the methods and challenges for achieving this. Finally, we outline some of the related key challenges still posed for the economic evaluation of telehealth. We aim to promote a debate amongst telehealth researchers and practitioners around the most appropriate and holistic frameworks for evaluating their services, in order to extend the body of evidence that exists to support decision-making in this area.

The most common form of economic evaluation published in telehealth is cost-minimisation analysis (CMA) ^{1,2}. CMA only compares costs; however, to perform a robust CMA it is expected that the outcomes of the two interventions have been proven equivalent (or at a minimum, non-inferior). If the economic analysis does not address the potential difference in outcomes between the comparators, then it is a simple cost-

analysis (CA). Other analyses include cost-effectiveness analysis (CEA), cost-utility analysis (CUA), and cost-benefit analysis (CBA), all of which compare both the intervention's health benefits (measured by units of clinical effectiveness, utility or monetary value) and costs with the health benefits and costs of a comparator. Another arguably underutilised method of analysis is a cost consequence analysis (CCA), which presents costs and outcome measures for interventions side by side in a disaggregated format. CCA is proposed to provide decision makers with all available information in a non-preferential format to enable them to form their own judgement regarding the relative importance of the costs and outcomes presented.

Outcomes in telehealth

The “gold standard” method in the economic evaluation of health care is generally considered to be CUA³. CUA usually employs the quality-adjusted life year (QALY) as the utility outcome metric. This captures the value of gains in health-related quality of life. However, telehealth interventions frequently aim to provide greater efficiency, convenience and access for patients. It may often be unrealistic to anticipate a measurable improvement in *health-related* quality of life. It may however be reasonable to anticipate that the gains in convenience and access lead to gains in global quality of life and overall utility. From a welfarist point of view, the theoretical underpinnings of economics, these gains contribute to overall utility and should therefore be valued.

The non-health or process-related outcomes associated with telehealth would be more readily captured and valued within a cost-benefit evaluation framework, which places a monetary value on the outcomes of healthcare as well as on the costs. However, cost-benefit analysis (CBA) has been seldom applied in health more widely or in telehealth specifically, due to its increased complexity and requirement for extra data such as willingness-to-pay (WTP) values, which are not routinely collected in randomised controlled trials. In fact it has only been during the last decade that economic analysis has been routinely performed for telehealth interventions⁴. Nevertheless, telehealth above many other health service foci would benefit from a more holistic cost-benefit evaluative approach, and this is becoming feasible with rapid methodological developments in the estimation of societal willingness to pay, which we discuss below.

As well as CBA, cost-consequence analysis (CCA) also provides a framework by which holistic results on costs and benefits can be presented in a cumulative format for decision-makers to consider. Whilst CCA may be comparatively easy for decision-makers to understand, the main disadvantage of CCA is that the disaggregated format of the costs and outcomes rely entirely on decision-maker judgement as to what constitutes value for money in each situation.REF This compares to the more aggregated format of CBA where a systematic decision criterion of acceptable cost-

effectiveness if the net monetary benefit (that is incremental benefits of an intervention, less its incremental costs, expressed as a dollar value) exceeds zero can be adopted.

In telehealth research, just as in any other area of health, it is important that the most appropriate outcome is selected to best represent the health benefits associated with the intervention being tested. In general outcome measures are selected due to a combination of their ability to represent health benefits, ease of measurement and convenience. Telehealth economic analysis should incorporate best practice guidelines, comparative gold-standard therapy and validated frameworks where possible. CBA provides a framework for this ^{2,5}. It combines the relevant clinical outcomes and telehealth specific outcomes (such as patient or practitioner travel or satisfaction, cost changes for all stakeholders, and willingness-to-pay) into one succinct economic analysis ^{2,5,6}. This approach values not only the health outcomes (as in CEA or CUA when using QALYs), but also the preferences of the patient to use the intervention being proposed ^{1,7}. It also provides a framework by which holistic results can be presented in a cumulative format compared to the disaggregated format of CCA that relies more strongly on judgement. CBA is a method for streamlining health outcome, cost information, and societal values in order to increase the ease with which interventions can be compared.

In considering outcomes, careful selection of the economic analysis perspective impacts the outcomes that are relevant, especially in telehealth. If an economic analysis is conducted from the perspective of the government then extra-clinical costs such as patient-funded travel or loss of productivity are not taken into consideration, while reimbursed time for a consultant or technology resources may be ². A broader societal perspective would allow these wider costs to be included in the evaluation. This is particularly pertinent in the case of telehealth.

Methods must be carefully selected and economic analysis well designed with outcomes considered to ensure that all of the relevant factors are captured by the analysis where possible. For example, Bini and Manahajan used multiple evidence-based tools in their method they minimised bias, increased the transparency of their study and increased the comparability of their outcomes ⁸. Their study compared two physical rehabilitation interventions using reporting guidelines (SQUIRE), validated questionnaire tools (VAS and KOOS), and standard institution care procedures ⁸⁻¹². However, applying standard validated clinical questionnaire tools may not capture all relevant information. It is therefore appropriate to use informal methods such as free-form questions to capture specific additional data relevant to a more holistic evaluation in telehealth. Bini and Manahajan asked their participants further questions in free-form style, and collected information about time and travel investments ⁸. In doing this they provided a well-

designed example for future telehealth research.

Societal values: Willingness-to-pay

Inadequacy arising from including only conventional effectiveness outcomes in telehealth evaluation raises questions about how to value interventions using a more holistic set of outcomes. One way of doing this is by incorporating Willingness to pay (WTP) into evaluations. WTP estimates the contingent value of a service by eliciting population preferences regarding a service, and placing a monetary value on the benefit associated with a service, from a societal perspective. WTP can place a value on both the non-health as well as the health-related outcomes from a service. Thus, WTP information is integral to estimating changes in societal welfare using CBA, and it is used to advise policy makers about a population's preference to pay from personal funds in order to receive a specific service. The outcomes not only provide relevant monetary estimates but can also be used as an indication of the intensity of the sample population's interest in receiving the service, and likely service demand. Using appropriate methods to collect and apply WTP within telehealth research moves one step closer to holistic outcome measurement alongside the systematic and reproducible application of decision criteria.

There are several approaches to measuring WTP in healthcare, the most relevant for

telehealth research are direct contingent valuation and discrete choice experiment (DCE) surveys¹³. Several studies have demonstrated that the elicitation method used for WTP may affect the responses and therefore the values assigned¹⁴⁻¹⁶. With this in mind, it is important that telehealth researchers use appropriate methods to accurately measure WTP and apply the results. This is especially important given the innovative and dynamic nature of telehealth interventions, because respondents may find WTP enquiries in telehealth more challenging to respond to than in alternate medical research with more familiar concepts.

Patient contingent valuation surveys can be used to measure WTP. Contingent valuation methods can use multi-choice or open-ended questions, or alternative “ping-pong” or referendum formats^{16, 17}. However, their structure needs to be specific in order to elicit a consumer response that reflects the intervention being costed. WTP can be ascertained by asking the participant to select a range from pre-stated options, or by providing a free-form question enabling them to provide an unguided answer¹⁸. When using this approach to identify WTP it is important to provide participants with specific information about what the service (and associated benefit) is that they are valuing, and to provide information about the aspects that are not included in the service. Participants must be well informed about the potential changes in not only present healthcare costs (the intervention), but future health care costs and potential impact on future gains and

productivity. For telehealth interventions immediate productivity gains are often a factor due to reduced requirements for travel and extended interruption to work.¹⁷ Stahl et al. conducted a randomised-cross design trial and asked participants to respond to a contingent valuation survey that used a 5-point Likert scale to express satisfaction, perception of quality, and WTP¹⁸. As with the DCE, it is important when ascertaining WTP via contingent valuation that participants understand the full impact of the telehealth intervention; both clinical and non-clinical outcomes^{7, 18}. This is important because the value of telehealth is often in its societal gain for participants in terms of time, access and clinical non-inferiority.

Alternatively, the Discrete Choice Experiment (DCE) approach can be used to determine WTP. Grounded in random utility theory, DCEs involve asking participants a series of trade-off questions that require them to make discrete choices^{17, 19, 20}. The trade-off design of this method allows for variation between the options to be captured and described concisely to the respondent^{10, 21}. This method allows researchers to determine the relative importance that respondents place on specific aspects of the treatment or service, as well as their overall WTP for the service^{19, 20}. This makes DCE a very attractive method within telehealth research. It enables researchers to elicit consumer preference around potential variations that could be made to the intervention itself, the delivery method, the proposed costing structure, or other relevant factors.

A well-designed DCE uses best practice guidelines, clear objectives and comparators, and knowledge of current literature. Although the DCE method is becoming popular in health care, few examples of the DCE method being applied in telehealth have been published. One relevant, though small study, Spinks et al. (2015) used a DCE to determine consumer WTP for teledermoscopy screening in an Australian population²². The authors followed the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) guidelines and developed a rigorous design^{17,22}. The comparators and their varied characteristics were devised in part from previously published surveys in the target population²². In order to use the DCE to accurately estimate WTP and uptake for telehealth initiatives, the trade-off attributes must include all of the relevant characteristics for telehealth (such as patient time, technology options, wait time for results, clinical reviewer). Spinks et al., incorporated these aspects into their DCE while the DCE section of the survey remained concise (twelve trade-offs)²². Another recently example of DCE being applied in telehealth is the work by Kaambwa et al. (2016) which surveyed a larger population (n=330) to determine consumer preference for specific telehealth characteristics and WTP for an Australian population of consumers over 65 years of age²³. The resultant WTP values provided an estimate for proposed policy changes and also indicated the consumers weighted preferences towards some of the variable aspects of the proposed telehealth interventions^{22,23}. Both Spinks et al. and

Kaambwa et al. found that given the choice many people would prefer to undertake a conventional face-to-face consultation, however telehealth becomes more appealing when the face-to-face consultation requires hours of travel away from home, time off work, and an increased wait for an appointment^{2, 22, 23}.

Unintended consequences and budget impact

Just like the possible societal benefits of telehealth that cannot always be captured by standard economic analysis methods, new interventions may potentially have unintentional consequences, which also need to be considered. These consequences can arise from unexpected uptake, which substantially increases costs (either greater or lower uptake than predicted), unexpected workload changes, or other unforeseen factors. Unintended consequences are described as such because they are generally only discovered once an intervention is offered widely and they substantially change the predicted costs and impact on the intervention budget.

Using CBA or budget impact analysis (BIA) in telehealth research along with appropriately selected sensitivity analyses can assist in identifying unintended outcomes that would not routinely appear in analyses taking a narrower perspective. As an example, Fortney et al. conducted a budget impact analysis and found that the cost-effectiveness of a telepsychiatry intervention was dependent on the severity of the

symptoms, but concluded that the intervention did not increase the work load for health professionals making it a viable service provision option ²¹. This paper captured the impact of societal factors that may not have been captured using a conventional CEA/CUA method such as health professional time ²¹.

Sensitivity analysis within telehealth economic evaluation

Economic analysis in telehealth is generally proposed to inform funding models, resource allocation decisions and policy initiatives. Sensitivity analysis is an essential part of the evaluation. Sensitivity analysis can be used to test potential uncertainties in parameters and assumptions made within the analysis. Therefore, sensitivity analysis can assist the translation of evidence-based medicine into practice by providing appropriate evidence around the uncertainty in findings - an aspect that is important for decisions. Sensitivity analysis method selection is an important concept for rigorous telehealth research ^{24, 25}. Guidelines for sensitivity analysis methods are available and should be selected based on the form of economic analysis being undertaken ^{24, 26}.

When applied to economic modelling for telehealth, sensitivity analysis can be used to vary the probabilities used to inform the model, enabling the probabilities that exerted the largest effect on the cost-effectiveness outcome to be identified. For example, Jackson et al. used this method to examine the outcomes of their economic model, a

CUA of a telehealth initiative aimed at managing retinopathy of prematurity⁵. Although sensitivity analysis needs to be appropriately applied and interpreted it does not need to be complex. Smith et al. used sensitivity analysis to determine the threshold based on workload (number of consultations) where the costs of telehealth were the same, less or greater than alternative methods of service delivery²⁷. Using this method allowed the authors to show which variables the financial viability of their service was most sensitive to²⁷. This form of sensitivity analysis is simple to understand and apply and can reveal information about the economic analysis and the intervention being investigated, along with the robustness of any decision based on the findings.

Conclusion

Performing appropriate economic analysis on telehealth is imperative to enable the translation of any proposed intervention into practice²⁸. Given the innovative nature of telehealth interventions and the dynamic nature of technology conducting an economic analysis in this area should involve the incorporation of societal values and the preferences of users, something that is possible with CBA. In order to undertake CBA researchers must include rigorous methods for eliciting WTP (such as DCE), and ensure that all relevant clinical and extra-clinical outcomes affecting costs or patient preferences are measured and valued. In assessing the effects of telehealth, CBA could be the most appropriate and challenging approach. However, future research should

seek to demonstrate that the current studies that adopt the CBA approach are more appropriate in showing the value of telehealth, with respect to alternative approaches like CEA or CUA. This could be achieved for example through a literature review of existing economic studies and/or through repeating different study designs on the same evaluation problem. Appropriately applied economic analysis and improved understanding of the economics of telehealth can positively influence policy decisions, potential investment, practice changes and uptake in the health sector.

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