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Cost-effectiveness of Telemedicine-directed Specialized vs Standard Care for Patients With Inflammatory Bowel Diseases in a Randomized Trial

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BACKGROUND & AIMS: Telemedicine can be used to monitor determinants and outcomes of patients with chronic diseases, possibly increasing the quality and value of care. Telemedicine was found to reduce outpatient visits and hospital admissions for patients with inflammatory bowel diseases (IBD). We performed a full economic evaluation of telemedicine interventions in patients with IBD, comparing the cost-utility of telemedicine vs standard care.

METHODS: We performed a randomized trial of 909 patients with IBD at 2 academic and 2 non-academic hospitals in The Netherlands. Patients were randomly assigned to groups that received telemedicine (myIBDcoach; n = 465) or standard outpatient care (n = 444) and followed for 12 months. Costs were measured from a societal perspective. Direct healthcare costs were based on actual resource use. Indirect costs comprised self-reported hours sick leave from work, intervention costs (annual license fee of €40 per patient [$45]), and utility costs (assessed using EQ5D). Cost-utility and uncertainty were estimated using the non-parametric bootstrapping method.

RESULTS: Telemedicine resulted in lower mean annual costs of €547/patient [$612] (95% CI, €1029–2143 [$1150-2393]; mean costs of €9481 [$10,587] for standard care and €8924 [$9965] for telemedicine) without changing quality adjusted life years. At the Dutch threshold of €80,000 [$89,335] per quality adjusted life year, the intervention had increased incremental cost-effectiveness over standard care in 83% of replications and an incremental net monetary benefit of €707/patient [$790] (95% CI, €1241–2544 [$1386-2841]).

CONCLUSIONS: Telemedicine with myIBDcoach is cost saving and has a high probability of being cost effective for patients with IBD. This self-management tool enables continuous registration of quality indicators and (patient-reported) outcomes and might help reorganize IBD care toward value-based healthcare. ClinicalTrials.gov no: NCT02173002.

Keywords: Smartphone App; Home Care; QALY; Efficiency.

Abbreviations used in this paper: CD, Crohn's disease; CI, confidence interval; EQ-5D, EuroQoL 5-dimensions; IBD, inflammatory bowel disease; iCUR, incremental cost utility ratio; iNMB, incremental net monetary benefit; PROM, patient-reported outcome measure; QALY, quality-adjusted life year; UC, ulcerative colitis; WPAI, work productivity and activity impairment; WTP, willingness to pay.

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See editorial on page 1688; also see related article on page 1882.

Improving quality of care is a continuous task for every healthcare professional and institution and is especially challenging when increasing healthcare expenditures force professionals to constrain costs. During recent years, value-based healthcare has been considered a way forward to control costs while improving quality by emphasizing the value for patients as the common goal of all stakeholders (patient, healthcare professional, insurance company, and society).[1] In this framework, regular measurement of clinical and patient-reported health outcomes is necessary. In a traditional healthcare organization frequent monitoring of these outcomes is challenging because of time constraints and a further increase of the administrative burden. Telemedicine enables regular collection of health outcomes without disrupting the clinical workflow.[2,3] However, no data are available regarding cost-effectiveness of telemedicine.

The inflammatory bowel diseases (IBDs), Crohn's disease (CD) and ulcerative colitis (UC), are debilitating, chronically relapsing, inflammatory disorders primarily affecting the gastrointestinal tract. Because IBD is typically diagnosed in young people during the most productive years of their lives[4] and requires life-long medical therapy and monitoring, the disease has a significant impact on patients' quality of life. It constitutes a high economic burden on society.[5,6] Cost-of-illness studies report total annual healthcare costs in Western European countries ranging from €6338–€8004 for CD and €3600–€4819 for UC patients.[6–8] Today, the direct healthcare costs for IBD are mainly driven by medication costs, hospitalizations, and outpatient clinic visits, whereas diagnostic procedures and surgery account for a relatively small portion.[6–8] In addition, productivity losses resulting from sick leave and work disability account for up to 50% of the total costs of illness in IBD.[7]

IBD care is traditionally organized by standard scheduled outpatient visits, with a frequency predominantly based on medical treatment but independent of the occurrence of unpredictable flares.[9,10] This might result in an inefficient and potentially costly way of medical resource use and patients' time. In addition, treatment strategies for IBD are shifting from mere management of symptoms toward tight and personalized control of intestinal inflammation and timely intervention if inflammation (re-)occurs to prevent structural bowel damage and eventually improve long-term disease outcome.[9–11] Together with the rising incidence of IBD, this results in overburdened IBD clinics and escalating costs.[12,13]

Implementation of telemedicine for IBD can improve efficiency of care, enable tight disease monitoring and registration of patient-reported outcome measures (PROMs), and answers to patients' demands for more participation in disease management. A pragmatic randomized multicenter trial with the telemedicine tool myIBDcoach showed that personalized telemonitoring reduced the number of outpatient visits by 36% and the number of hospitalizations by 50%, while increasing adherence to medication when compared with standard care.[14]

Although this trial convincingly showed that the telemedicine tool reduced healthcare utilization, broad implementation is currently hindered by some barriers of which reimbursement is considered to be the key determinant.[3] Therefore, research focusing on economic evaluation of telemedicine interventions is warranted to aid decision makers in estimating the value of healthcare reorganization toward reimbursement of telemedicine.[2] Therefore, the objective of this study was to calculate, from a societal perspective, the cost-effectiveness of telemedicine with myIBDcoach compared with standard care for IBD patients.

What You Need to Know

Background

Telemedicine can be used to monitor patients with chronic diseases, possibly increasing the quality and value of care for patients with inflammatory bowel diseases (IBD).

Findings

Telemedicine with myIBDcoach was cost saving and appeared to be cost effective for patients with IBD.

Implications for patient care

This self-management tool enables continuous registration of quality indicators and outcomes and might increase value-based healthcare for patients with IBD.

Methods

Study Design and Participants

The telemedicine tool myIBDcoach monitors disease activity and other disease-modifying factors such as medication adherence, nutrition, and psychosocial factors every 3 months. Furthermore, the system includes intensified monitoring modules (weekly in case of flare), outpatient visit modules (to prepare for an outpatient visit), e-learning modules, a personal care plan, and a back office (administrator page) used by the healthcare professional, ie, gastroenterologist or nurse. When parameters recorded by the monitoring modules exceed a predefined threshold, an alert is created in the back office of each local hospital, and the patient is contacted for further evaluation within 2 working days. At any time, patients can communicate easily with their healthcare professional by sending a message to the back office.[15] The myIBDcoach trial was carried out in 2 academic and 2 nonacademic hospitals in the Netherlands, each caring for 1500–2000 IBD patients, with 1 or 2 dedicated nurses, as well as an e-mail and telephone consultation structure for patients to contact the IBD team. IBD patients who were between 18 and 75 years of age were eligible for inclusion when they had no history of an ileoanal pouch or ileorectal anastomosis. In addition, those with a hospital admission within 2 weeks before inclusion were excluded. Patients were randomized in a 1:1 ratio to care via myIBDcoach (intervention group) or standard care (control group) and were followed for 12 months. At baseline and after 12 months, all participants received a paper questionnaire regarding health-related quality of life and work productivity. At 12 months an additional questionnaire on healthcare utilization (number of e-mail and phone consultations) was sent to the participants. This cost-effectiveness analysis was carried out from a societal perspective following the recommendation of Sanders et al.[16] The study has been approved by the Medical Ethics Committee of the Maastricht University Medical Centre+, and all subjects gave written informed consent before participation. This approval was applicable to all participating centers. All authors had access to the study data and reviewed and approved the final manuscript. This trial was registered at ClinicalTrials.gov (NCT02173002).

Clinical and Demographic Data

Demographic and clinical data, ie, patient characteristics, disease phenotype, disease duration, and baseline disease activity, were obtained from patients' electronic medical records.

Direct Healthcare Costs

Information on IBD-related healthcare utilization during the 12-month study period, ie, outpatient service, inpatient service, medication use, diagnostic procedures, and surgery, was collected from patients' electronic medical records and hospital information systems. Outpatient services included the number of visits to a gastroenterologist or IBD nurse, scheduled phone consultations, and visits to the emergency department. Inpatient service was defined as the number of days hospitalized. Medication use for IBD was calculated by using the mean number of daily dosages per type of drug used. Diagnostic procedures included the number and type of endoscopies and radiologic procedures. Furthermore, the number and type of surgeries were retrieved. Information regarding the number of additional unscheduled phone consultations and e-mail contacts with the IBD nurse was obtained from the 12-month follow-up questionnaire. For each of the abovementioned resource categories, the annual costs per patient were calculated by multiplying units of resource use by their unit costs.[17] For most healthcare resources, unit costs were available from the Dutch Guideline for Economic Evaluations in Healthcare and the Healthcare Insurance Board (Supplementary Table 1).[18–20] Unscheduled phone consultations and e-mail contacts with the nurses were charged as 15 minutes of their hourly wage.[17] Medication costs were calculated on the basis of the individual dosage and their dose prices (Supplementary Table 1).[17,20,21] Non-healthcare costs, ie, transport and parking costs related to an outpatient visit, were calculated by using standardized prices.[17,20] Finally, for patients using myIBDcoach, an estimated annual license fee of €40 was added to their total direct healthcare costs. The total annual direct (healthcare and non-healthcare) costs per patient equaled the sum of all these cost categories.

Indirect Healthcare Costs

To estimate the indirect healthcare costs, only productivity loss because of sick leave for those with a paid job were included. A time horizon of 1 year would not provide reliable information on the effect of the intervention on work disability. Self-reported hours of absence from work because of health problems were measured using the Work Productivity and Activity Impairment questionnaire (WPAI).[22] Hours absent per week were extrapolated for the trial duration and multiplied by the average hourly Dutch wage retrieved from the Dutch Guideline for Economic Evaluations in Healthcare.[19]

Health Utility

Changes in value for health or effectiveness during the 12-month study period were expressed as quality-adjusted life years (QALYs) gained and captured by using the EuroQoL 5-dimensions (EQ-5D),[23] filled out by all participants at baseline and at 12-month follow-up. The EQ-5D is an instrument assessing 5 health-related quality of life dimensions: mobility, self-care, daily activities, pain/discomfort, and depression/anxiety. On the basis of preferences elicited from a general Dutch population,[24] the EQ-5D scores were converted into utilities. The change in utility at 12 months compared with baseline equals the change in QALYs.

Statistical Analyses

Incremental costs, utilities, and cost-utility were assessed by the nonparametric bootstrapping method as recommended by the International Society for Pharmacoeconomics and Outcomes Research for cost-effectiveness analysis alongside trials.[25] Between 3% (data derived from electronical medical records) and 26% (data derived from questionnaires) of observations contained missing values for costs or utilities. Preceding the bootstrap analyses, where 5000 bootstrap replications were generated, these were replaced with estimates using stochastic imputation. The variables medical center (4 centers), subtypes of IBD (CD or UC), treatment (no medication or mesalazine, immunosuppressive drugs, or biological therapy), age (numeric), sex (male or female), disease duration (numeric), disease activity at baseline (remission or active), smoking (nonsmoker, active smoker, or ex-smoker), and educational level (5 levels) were used as predictors in the stochastic imputation model. All randomized patients were included in the analyses (intention-to-treat population). The results of the bootstrap iterations were presented in an incremental cost-utility ratio (iCUR) plane, an incremental net monetary benefit (iNMB) curve, and a cost-effectiveness acceptability curve, representing the probability the intervention is cost-effective. The iNMB was calculated as iNMB $= \lambda * \Delta$QALY – Δcost, where λ is the decision maker's maximum willingness to pay (WTP) for a QALY. An iNMB above 0 indicates the intervention generates monetary benefits at the chosen λ. In the Netherlands, a WTP threshold of €80,000 per QALY is used. Because many bootstrapped iCUR replications indicated cost savings but minor loss in QALY, we furthermore used a willingness-to-accept threshold of €100,000 savings per QALY lost. A two-sided P value $\leq$.05 was defined as statistically significant. All statistical analyses were performed by using SPSS version 22.0 (IBM, Chicago, IL).

Results

In total, 909 patients were included in the myIBDcoach study, of whom 465 were randomized to the intervention group and 444 to the control group. The baseline characteristics of the 2 study groups were similar and were representative for the general IBD population in the Netherlands with regard to demographics and disease activity (Table 1).[26] At the end of the 12-month study period, 27 patients (6%) in the intervention group and 1 patient (0.02%) in the control group were lost to follow-up. At baseline, 382 patients (82%) in the intervention group and 369 (83%) in the control group completed the paper questionnaires on healthcare utilization, health-related quality of life, and work productivity. At 12 months, these questionnaires were completed by 340 patients (73%) in the intervention group and 331 patients (75%) in the control group.

Direct and Indirect Annual Costs and Quality-Adjusted Life Years

The mean annual direct costs in the intervention group were €7048 per patient compared with €7423 per patient in the control group, whereas the mean indirect costs were €1886 in the intervention group compared with €2058 in the control group. This resulted in a mean annual cost saving of €547 per patient (95% confidence interval [CI], €–1029 to €2143) favoring telemedicine. Table 2 shows averages of the main annual direct cost categories, sick leave costs, and QALYs per year in the 2 treatment strategies. Within the direct healthcare costs outpatient visits and drug costs were the main cost savers, responsible for an annual saving of €104 (95% CI, €79–€129) and €216 (95% CI, €–776 to €1209), respectively. Patients in the intervention group showed a mean gain in QALYs of 0.002 (95% CI, –0.022 to 0.018).

Incremental Cost-Effectiveness and Uncertainty

The scatter plot of the iCURs resulting from the 5000 bootstrapped replications for the imputed data set from a societal perspective is provided in Figure 1 (for the cost-effectiveness planes from a healthcare perspective, see Supplementary Figures 1 and 2). Using a WTP threshold of €80,000 and a willingness-to-accept threshold of €100,000, the intervention was cost-effective in 83% of all replications. In only 6% of replications the intervention was accompanied by higher cost and lower health utility. At the Dutch cost-effectiveness threshold of €80,000 per QALY, the intervention has an iNMB of €707/patient (95% CI, –1241 to 2544) per year (Figure 2). Figure 3 shows the uncertainty analyses of the iCUR and iNMB. MyIBDcoach dominates usual care even at a WTP threshold of €0 per QALY, some uncertainty remains, and there is no threshold at which myIBDcoach is 100% certain cost-effective.

Discussion

In this cost-utility analysis regarding data from a pragmatic, randomized controlled trial, we showed that telemedicine with myIBDcoach has a high probability of being cost-effective compared with standard care for IBD patients. Positive effects of telemedicine on IBD outcome have extensively been demonstrated. However, broad implementation was currently hindered as a result of lacking evidence on cost-effectiveness. This was a complete economic evaluation of a telemedicine intervention versus standard care for IBD patients.

Telemedicine with myIBDcoach resulted in lower mean annual costs and a small mean gain in QALYs. At the Dutch threshold of €80,000 per QALY, the iNMB was €707 per patient per year. Cost benefits of telemedicine in IBD have been evaluated in 2 previous studies. The first trial, in which 333 patients in Denmark with mild-to-moderate UC treated with mesalazine were randomized to care via a telemedicine tool or standard care and followed for 12 months, showed that tight monitoring of disease activity and personalized treatment strategies resulted in cost savings of €189 per patient per year, mostly because of fewer outpatient visits.[27] In addition, improvement in

Table 1. Patient Characteristics

	MyIBDcoach (n = 465)	Standard care (n = 444)
Center, N		
Maastricht University Medical Centre	133	131
Leiden University Medical Centre	144	152
Zuyderland Medical Centre Sittard	117	102
St Antonius Hospital Nieuwegein	71	59
Gender, N (%)		
Male	194 (41.7)	180 (40.5)
Female	271 (58.3)	264 (59.5)
Age at diagnosis, y, mean (SD)	30.7 (13.5)	30.4 (13.6)
Age at inclusion, y, mean (SD)	44.0 (14.1)	44.1 (14.2)
Disease duration (y), mean (SD)	12.8 (10.4)	13.1 (10.8)
Phenotype, N (%)		
Crohn's disease	282 (60.6)	262 (59.0)
Ileal	87/282 (30.9)	68/262 (26.0)
Colonic	67/282 (23.8)	63/262 (24.0)
Ileocolonic	128/282 (45.4)	131/262 (50.0)
Upper gastrointestinal modifier	34/282 (12.1)	26/262 (9.9)
Nonpenetrating, nonstricturing	169/282 (59.9)	152/262 (58.0)
Stricturing	76/282 (27.0)	70/262 (26.7)
Penetrating	37/282 (13.1)	40/262 (15.3)
Perianal disease modifier	67/282 (23.8)	62/262 (23.7)
Ulcerative colitis	183 (39.4)	182 (41.0)
Proctitis	26/183 (14.2)	27/182 (14.8)
Left-sided	81/183 (44.3)	70/182 (38.5)
Pancolitis	76/183 (41.5)	85/182 (46.7)
Treatment, N (%)		
No medication/mesalazine	173/465 (37.2)	147/444 (33.1)
Immunosuppressives	122/465 (26.2)	131/444 (29.5)
Biologicals	170/465 (36.6)	166/444 (37.4)
Baseline disease activity, N (%)		
Remission	394/465 (84.7)	380/444 (85.6)
Active disease	71/465 (15.3)	64/444 (14.4)
Employment at baseline, N (%)	241/384 (62.7)	235/372 (63.2)
Work-productivity (WPAI; mean hours of productivity loss per study year, (SD))	54.3 (232.6)	59.1 (218.4)
Utility (EQ-5D; mean score, SD)	0.83 (0.15)	0.81 (0.17)

EQ-5D, EuroQoL 5-dimensions; SD, standard deviation; WPAI, work productivity and activity impairment.

disease-specific quality of life was found. Because quality of life was not measured by the EQ-5D, cost per QALY could not be calculated. Regarding indirect healthcare costs, no statistically significant difference in number of workdays lost through illness was observed between the 2 groups. The second study by the same group, a 12-month observational study in 92 CD patients (23 web-based care), found lower direct costs of €699 per patient per year when infliximab treatment was personally scheduled by web administration, as compared with standard scheduled infliximab administration in the outpatient clinic.[28] However, this web-based strategy did not show an improvement in quality of life or a change in time missed from work because of CD.[29] In line with our results, both studies showed that individualization of disease management enabled by telemedicine was accompanied by a reduction of direct healthcare costs due to more efficient use of resources. In addition, telemedicine can contribute to earlier identification of patients with insufficiently controlled disease who tend to be undertreated in current practice. Mounting evidence underscores that tight control of disease activity and optimization of medication adherence prevent chronic uncontrolled mucosal inflammation, subsequent irreversible bowel damage, surgeries, and hospitalizations.[30–32] By implementing such a strategy with telemedicine, flares can be detected earlier, and the delay to treatment can be minimized. The abovementioned UC study indeed showed that implementation of guided self-management with personalized action plans by which patients could immediately self-

Table 2. Mean Costs (in Euro) and QALY per Patient in the 2 Groups During the Study Period

	Intervention group (SD)	Control group (SD)	Incremental (95% CI)
Direct healthcare costs	7048 (8458)	7423 (8522)	375 (–742 to 1501)
Non-drug healthcare costs	762 (2412)	960 (2386)	198 (–157 to 554)
Drug costs	6244 (7487)	6460 (7757)	216 (–776 to 1209)
Out-of-pocket costs	2 (1.9)	3 (2.2)	1 (0.8–1.3)
IBD-coach related	40	0	+40
Indirect healthcare costs	1886 (8084)	2058 (7587)	172 (–848 to 1192)
Total costs	8934 (12,256)	9481 (11,757)	547 (–1029 to 2143)
QALY gains	0.034 (0.150)	0.032 (0.162)	0.0020 (–0.022 to 0.018)

CI, confidence interval; IBD, inflammatory bowel disease; QALY, quality-adjusted life year; SD, standard deviation.

initiate treatment in case of symptoms shortened flare duration.[27] The combination of close monitoring and immediate intervention in case of a relapse might also have contributed to the reduction in hospitalizations in the myIBDcoach study.[14]

The cost savings in our study were mainly due to a reduction in outpatient visits and medication costs. We speculate that the latter might have been due to a reduction in flare duration and thereby a shortened therapy window. In addition, timely optimization of conventional therapy might prevent escalation to more expensive biologicals. Currently, several trials assess the possibilities for de-escalation of biological treatment in patients with sustained remission to improve safety and reduce healthcare costs. Proposed strategies include reducing dosage, increasing dosage intervals, or discontinuation of biological therapy.[33,34] Telemedicine can add value to these strategies by closely monitoring disease activity and immediate re-escalation when indicated. MyIBDcoach can further contribute to healthcare cost reduction because it systematically registers PROMs and quality indicators and thereby enables benchmarking and evaluation of practice variations. Our study showed that use of telemedicine only resulted in a small nonsignificant mean gain in quality of life, probably because of the fairly short follow-up of 12 months and the high baseline scores on quality of life in both the telemedicine and standard care groups. We expect that

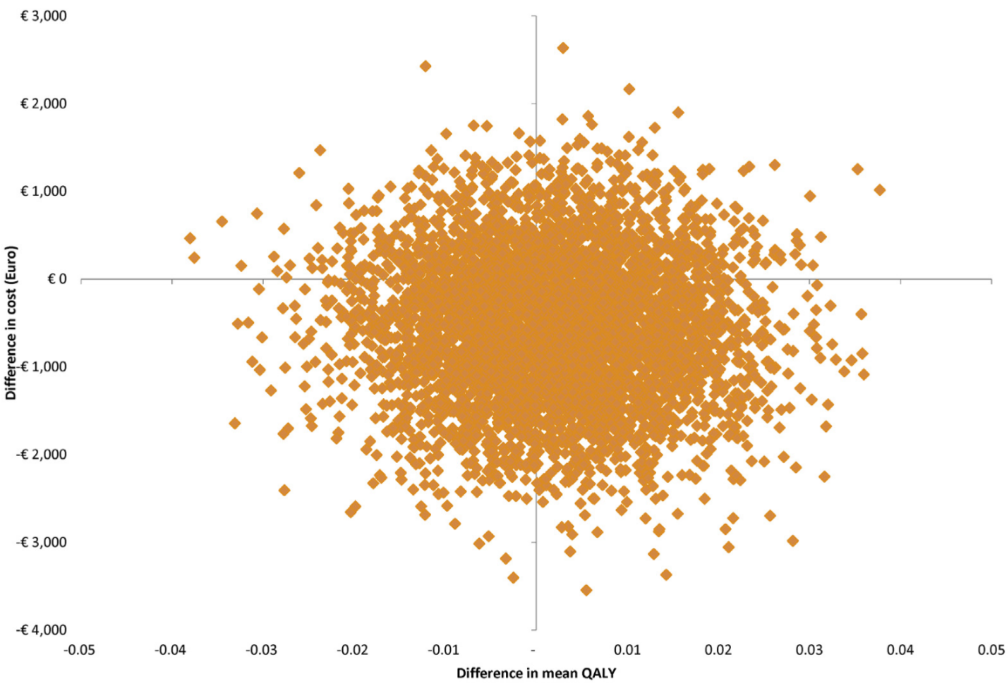

Figure 1. Scatter plot of incremental mean cost against incremental mean quality-adjusted life years (QALY) of telemedicine compared with standard care from among 5000 bootstrap samples, representing the uncertainty surrounding the incremental cost-effectiveness ratio. The percentage of joint density occupying each quadrant indicates the likelihood that cost-effectiveness lies in that quadrant. Southeast: more effective and less costly (ie, dominant); Northeast: more effective and more costly; Northwest: less effective and less costly (ie, inferior); Southwest: less effective and less costly. For SW and NE quadrants, cost-effectiveness depends on the threshold applied.

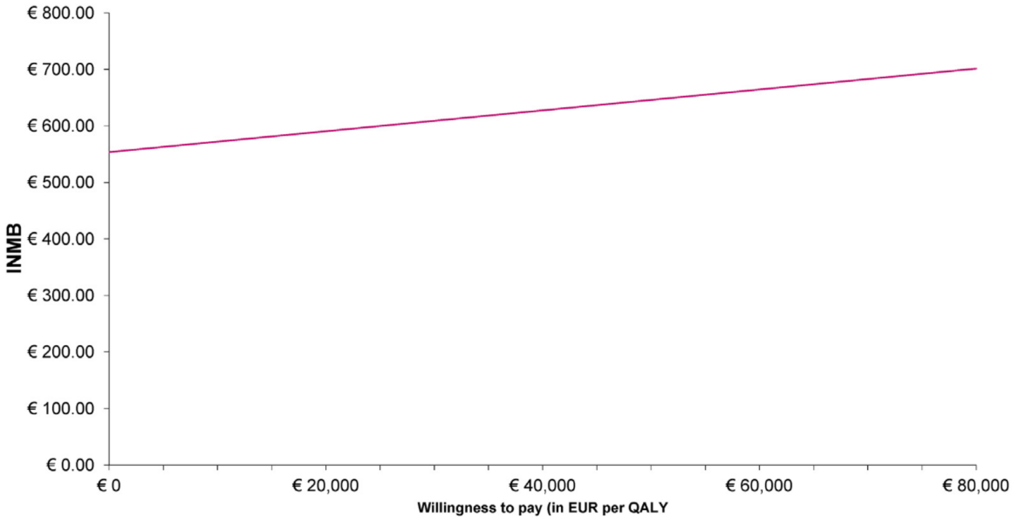

Figure 2. Incremental net monetary benefit (iNMB) curve for telemedicine versus standard care. QALY, quality-adjusted life year.

additional interventions based on aberrant patient-reported values, such as smoking, an unfavorable nutritional status, or medication nonadherence, might contribute to better disease outcome and quality of life on the long-term. Although this study showed that use of telemedicine has a high chance of being cost-effective for all subtypes of IBD patients, there still remains some uncertainty. Future research should focus on identifying subgroups that benefit most from the intervention and whether the intervention is safe and effective in patients with severe disease activity at baseline.

The main strength of this economic evaluation was that the data were obtained from a trial with a pragmatic randomized design; thus it was based on common daily practice. A large unselected heterogeneous group of patients, clinicians, and clinical practices were included to maximize the generalizability of the results to everyday practice. A possible limitation of this study was the lack of detailed information regarding laboratory tests because they were not systematically reported in medical records. However, these tests were previously found to account for a small proportion of the total costs and are therefore unlikely to have major impact on the reported results. Furthermore, because laboratory tests are regularly linked to outpatient visits and hospital admissions, the total costs related to these tests are probably lower in the telemedicine group. In addition, some parameters were collected by questionnaires, resulting in possible selection bias. Yet, we found no statistically significant differences in gender, age, phenotype, disease severity, or educational level between patients who returned their questionnaires and those who did not. We calculated the indirect healthcare costs using the self-reported hours of sick leave from work during the past 7 days obtained from the WPAI and extrapolated these data for 1 year. This method could have underestimated or overestimated the total amount of indirect healthcare costs. However, in view of the large sample size and under the assumption that weekly sickness absence would occur at a constant rate throughout the observation period in each treatment group, the indirect costs estimated can be considered accurate. In addition, we might have slightly overestimated the total net monetary benefit because we calculated a plain annual license fee

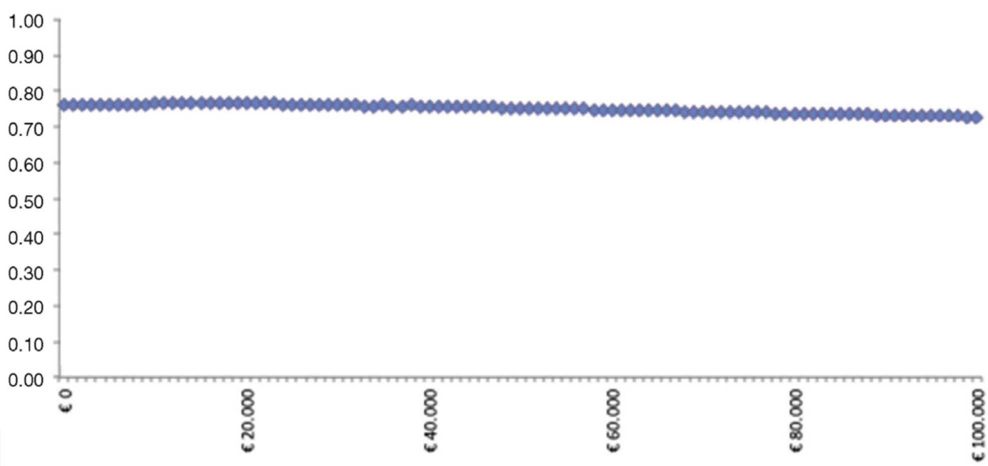

Figure 3. Cost-effectiveness acceptability curve (CEAC) for telemedicine versus standard care.

of €40 per patient using telemedicine. These costs did not cover initial implementation costs and additional costs related to any potential healthcare reorganization (eg, extra hours of work for an IBD nurse). However, we experienced that monitoring of the back office of myIBDcoach was limited to an hour daily (not all monitoring sessions resulted in alerts that require action), and we expect that the additional actions related to myIBDcoach performed by the IBD nurses are at least partly compensated by the information and education function of the e-learning modules, which is normally a face-to-face task of the IBD nurse. Last, the fairly short follow-up period can also be considered a limitation of the study because clinicians and patients require time to adapt to an altered clinical workflow. Therefore, trials with longer follow-up periods are required to determine whether myIBDcoach can control costs and improve disease outcomes on the long-term.

Telemedicine with myIBDcoach was cost-saving and has a high probability of being cost-effective. Implementation of telemedicine was shown to improve health outcomes and was accompanied by a reduction of healthcare costs without a decline in quality of life. Incremental costs of telemedicine are relatively low, and the chance of telemedicine increasing costs without any health benefit is low. Therefore, telemedicine may be a promising tool in restructuring IBD care toward more personalized, efficient, and value-based healthcare.

Supplementary Data

Note: to access the supplementary materials accompanying this article, visit the online version of *Clinical Gastroenterology and Hepatology* at www.cghjournal.org, and at https://doi.org/10.1016/j.cgh.2020.04.038.

References

1. Huber M, et al. How should we define health? BMJ 2011; 343:d4163.
2. Jaboli F, et al. Telehealth in inflammatory bowel disease: every patient may need a coach! Gastroenterology 2018; 154:1196–1198.
3. Tuckson RV, et al. Telehealth. N Engl J Med 2017; 377:1585–1592.
4. Johnston RD, et al. What is the peak age for onset of IBD? Inflamm Bowel Dis 2008;14(Suppl 2):S4–S5.
5. Bernklev T, et al. Health-related quality of life in patients with inflammatory bowel disease measured with the short form-36: psychometric assessments and a comparison with general population norms. Inflamm Bowel Dis 2005;11:909–918.
6. van der Valk ME, et al. Healthcare costs of inflammatory bowel disease have shifted from hospitalisation and surgery towards anti-TNFalpha therapy: results from the COIN study. Gut 2014; 63:72–79.
7. van der Valk ME, et al. Evolution of costs of inflammatory bowel disease over two years of follow-up. PloS One 2016;11: e0142481.
8. Bassi A, et al. Cost of illness of inflammatory bowel disease in the UK: a single centre retrospective study. Gut 2004; 53:1471–1478.
9. Dignass A, et al. Second European evidence-based consensus on the diagnosis and management of ulcerative colitis part 2: current management. J Crohns Colitis 2012; 6:991–1030.
10. Dignass A, et al. The second European evidence-based consensus on the diagnosis and management of Crohn's disease: current management. J Crohns Colitis 2010;4:28–62.
11. Colombel JF, et al. Management strategies to improve outcomes of patients with inflammatory bowel diseases. Gastroenterology 2017;152:351–361.
12. Molodecky NA, et al. Increasing incidence and prevalence of the inflammatory bowel diseases with time, based on systematic review. Gastroenterology 2012;142:46–54 e42, quiz e30.
13. van den Heuvel TRA, et al. A 20 year temporal change analysis in incidence, presenting phenotype and mortality in the Dutch IBDSL cohort: can diagnostic factors explain the increase in IBD incidence? J Crohns Colitis 2017;11:1169–1179.
14. de Jong MJ, et al. Telemedicine for management of inflammatory bowel disease (myIBDcoach): a pragmatic, multicentre, randomised controlled trial. Lancet 2017; 390:959–968.
15. de Jong MJ, et al. Development and feasibility study of a telemedicine tool for all patients with IBD: myIBDcoach. Inflamm Bowel Dis 2017;23:485–493.
16. Sanders GD, et al. Recommendations for conduct, methodological practices, and reporting of cost-effectiveness analyses: second panel on cost-effectiveness in health and medicine. JAMA 2016;316:1093–1103.
17. Health Insurance Board. 2018. Available at: www.medicijnkosten.nl. Accessed December 3, 2018.
18. National Health Care Institute Netherlands. Guideline for conducting economic health care evaluations. 29-02-2016.
19. Hakkaart-van Roijen, van der Linden, Boumans, et al. Methodology of economic evaluations and reference prices for economic evaluations in healthcare. 29-02-2016.
20. Dutch healthcare authority. 2018. Available at: www.nza.nl. Accessed December 3, 2018.
21. Farmacotherapeutisch Kompas. 2018. Available at: http://farmacotherapeutischkompas.nl. Accessed December 3, 2018.
22. Reilly MC, et al. The validity and reproducibility of a work productivity and activity impairment instrument. PharmacoEconomics 1993;4:353–365.
23. EuroQol G. EuroQol: a new facility for the measurement of health-related quality of life. Health Policy 1990; 16:199–208.
24. Lamers LM, et al. The Dutch tariff: results and arguments for an effective design for national EQ-5D valuation studies. Health Economics 2006;15:1121–1132.
25. Ramsey S, et al. Good research practices for cost-effectiveness analysis alongside clinical trials: the ISPOR RCT-CEA Task Force report. Value in Health 2005;8:521–533.
26. van den Heuvel TR, et al. Cohort profile: the inflammatory bowel disease South Limburg cohort (IBDSL). International Journal of Epidemiology 2015;1–9.
27. Elkjaer M, et al. E-health empowers patients with ulcerative colitis: a randomised controlled trial of the web-guided 'Constant-care' approach. Gut 2010;59:1652–1661.

28. Pedersen N, et al. eHealth: individualisation of infliximab treatment and disease course via a self-managed web-based solution in Crohn's disease. Aliment Pharmacol Ther 2012;36:840–849.
29. Cross RK, et al. Randomized, controlled trial of home telemanagement in patients with ulcerative colitis (UC HAT). Inflamm Bowel Dis 2012;18:1018–1025.
30. Pedersen N, et al. eHealth: individualization of mesalazine treatment through a self-managed web-based solution in mild-to-moderate ulcerative colitis. Inflamm Bowel Dis 2014;20:2276–2285.
31. Lakatos PL. Prevalence, predictors, and clinical consequences of medical adherence in IBD: how to improve it? World J Gastroenterol 2009;15:4234–4249.
32. Colombel JF, et al. Effect of tight control management on Crohn's disease (CALM): a multicentre, randomised, controlled phase 3 trial. Lancet 2018;390:2779–2789.
33. Peyrin-Biroulet L, et al. Stopping infliximab in Crohn's disease: still an ongoing STORI. Inflamm Bowel Dis 2012;18:2201–2202.
34. Reenaers C, et al. Outcomes 7 years after infliximab withdrawal for patients with Crohn's disease in sustained remission. Clin Gastroenterol Hepatol 2018;16:234–243 e2.

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