Valuing a Hypothetical Cure for Rheumatoid Arthritis Using the Contingent Valuation Methodology: The Patient Perspective

BRUNO FAUTREL, ANN E. CLARKE, FRANCIS GUILLEMIN, VIVIANE ADAM, YVAN ST-PIERRE, TINA PANARITIS, PAUL R. FORTIN, HENRI A. MENARD, CAM DONALDSON, and JOHN R. PENROD

ABSTRACT. ***Objective.*** A willingness-to-pay (WTP) survey measures the value of a given intervention in money terms. We examined the WTP of Canadian patients with rheumatoid arthritis (RA) for a hypothetical cure for RA under private and public scenarios. The validity of the survey was explored by studying the association between WTP and variables thought to be associated with WTP and randomly-varied variables of the survey materials.

Methods. A telephone survey was carried out in a sample of 121 patients with RA from 5 rheumatologists affiliated with the McGill University Health Centre. In advance, patients had been sent a 4-page brochure providing a comprehensive description of the disease (including photos or no photos). The hypothetical cure for RA was presented through 2 scenarios: a private insurance implying an annual premium and a public coverage requiring additional income taxes. The survey included questions related to their WTP, socioeconomic status (ability to pay), general health, opinion about the performance of the healthcare system, and their opinion about the difficulty of the survey. For elicitation of WTP, patients were randomized to one of 3 payment cards. Mailed questionnaires concerning RA health status were also completed. A series of univariate comparisons and multivariate ordered logit regressions were carried out to examine the association of WTP and patient and study variables.

Results. Patients were willing to pay annually significantly more for the private program (mean CAD $1190) than for the public program (mean CAD $502). Annual WTP was associated with age, household income, site of care (private program), private health insurance, opinion about the performance of the public healthcare system (public program), and presence of brochure photos. The payment card did not affect WTP for either program.

Conclusion. The WTP survey was well understood and accepted by the patients with RA. Although measures of RA-specific health status (e.g., Health Assessment Questionnaire) were not found to be associated with WTP, many variables thought to be associated with WTP were found to be related in the expected directions. Since WTP for the private program was higher than that for the public program, our study design did not fully capture altruistic valuations of RA patients. Thus, our estimates represent a lower bound on patients' WTP for an RA cure. (J Rheumatol 2005;32:443–53)

Key Indexing Terms:
RHEUMATOID ARTHRITIS ECONOMICS DECISION MAKING
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Many studies have estimated the burden of rheumatoid arthritis (RA) in monetary terms[1-6]. These estimates, referred to as cost-of-illness or burden-of-illness studies, are based on the sum of the direct costs of providing medical care and the indirect costs of productivity losses, as valued by the human capital method. The published estimates of the

From the Department of Rheumatology, Hospital Pitié-Salpêtrière, Paris, France; Division of Clinical Epidemiology, Division of Clinical Immunology/Allergy, and Division of Rheumatology, McGill University Health Center, Montreal, Canada; School of Public Health, Faculty of Medicine, Nancy, France; Division of Rheumatology, University Health Network, University of Toronto, Toronto; and Department of Health Economics, University of Calgary, Calgary, Canada.

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B. Fautrel, MD, Department of Rheumatology, Hospital Pitié-Salpêtrière, Division of Clinical Epidemiology, McGill University Health Center, EA 3444 School of Public Health, Faculty of Medicine, Nancy; A.E. Clarke, MD, MSc, Division of Clinical Epidemiology, Division of Clinical Immunology/Allergy, Division of Rheumatology, McGill University Health Center; F. Guillemin, MD PhD, EA 3444 School of Public Health, Faculty of Medicine, Nancy; V. Adam, MSc; Y. St-Pierre, MSc; T. Panaritis, BSc; J.R. Penrod, PhD, Division of Clinical Epidemiology, McGill University Health Center; P.R. Fortin, MD, MPH, Division of Rheumatology, University Health Network, University of Toronto; H.A. Menard, MD, PhD, Division of Rheumatology, McGill University Health Center; C. Donaldson, PhD, Department of Health Economics, University of Calgary.

Address reprint requests to Dr. B. Fautrel, Department of Rheumatology, Hospital Pitié-Salpêtrière, 83 bd de l'Hôpital, 75013 Paris, France. E-mail: bruno.fautrel@psl.ap-hop-paris.fr

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RA burden vary widely. For instance, its annual direct costs have been estimated to be between $2400 and $7200 CAD (i.e., $2000 and $6000 US)[1-3,5-7]. Differences are even more dramatic for indirect costs, which have been valued between $1800 and $26,400 CAD (US $1500 and $22,000)[1,7-14]. These discrepancies are mainly related to the method used for the valuation of non-market productivity losses.

Such estimates of the economic burden of a disease also correspond to the benefit, in monetary terms, that would arise if a curative treatment for RA were to become available. However, several investigators have noted that human capital estimates are conceptually invalid measures of such a benefit of a given program (or of the burden of a disease)[15-17]. According to standard economic theory, the measure of the benefit to the individual in money terms is defined as "the individual's maximum willingness to pay (WTP) for the program when supplied with information complete as it can be, given the scientific knowledge at the time"[16], a theoretical result that follows directly from utility maximization subject to a budget constraint. Among other things, the human capital method implies that society is productivity-maximizing rather than utility-maximizing, and fails to account for important health consequences that influence the utility of individuals but do not translate into productivity losses (for example, pain that does not result in productivity loss). At best, human capital estimates can be seen as a lower bound on WTP[16].

For new programs or public goods, WTP must be observed through survey methods[16,17]. WTP surveys are the most common form of the contingent valuation method, the name derived from the fact that the responses are contingent on the existence of a hypothetical market. First applied to environment economics[18], this method has been adapted to health economics[15,17,19-21]. Because of its direct link to economic theory, it has been recommended over human capital methods for cost-benefit analyses in the health domain by the Canadian Coordinating Office for Health Technology Assessment[22].

To date, WTP studies have been conducted in many different fields[17,23-42]. Several of these studies focused on individuals affected by a specific condition, with the aim of incorporating a treatment or a health program into an optimal budget-allocating framework[25,29,30,40,41,43,44]. Another set of studies addressed the question of the WTP for a hypothetical treatment or program — usually addressing a complete cure of a disease — either among affected individuals only or of the general population (both affected and unaffected individuals)[23,24,32,34,43,45,46]. The elicited WTP value, in combination with reductions in direct costs of care, corresponds to the monetary valuation of the burden of a disease.

However, many factors might influence respondents' WTP, making more complex the interpretation of WTP studies. For example, it has been shown that tools used to elicit WTP may introduce bias (payment cards, open-ended questions, take-it-or-leave-it questions)[20,21,47]. The respondents' prior knowledge of the disease — whether they are affected or healthy — might influence their WTP responses, which is rarely described in published reports[48-52]. Moreover, the economic context in which the question is presented is also of importance, some respondents favoring public funding for healthcare, some private. A study in the United States compared WTP for both public and private programs[28], but such a comparison has not been carried out in a context where healthcare is universally provided in a tax-financed system.

To address these issues, we evaluated a survey approach to measuring the WTP for a complete cure for RA in a convenience sample of Canadian patients with RA. This study of RA patients was carried out as part of a larger WTP study that included a survey of unaffected individuals as well who, through altruistic motives or the desire to insure against potential eventualities, may also be willing to pay. Following the recommendations on the implementations of WTP surveys, the elicitation is based on the WTP for an ex-ante, insurance-based program, which better reflects the real context of healthcare financing than an out-of-pocket scenario[17,28,53]. Specifically, our aims were (1) to estimate by means of a WTP survey the value of a hypothetical program providing a 100% effective cure for RA in a convenience sample of Canadian RA patients; (2) to measure whether RA patients would be willing to pay more for a public program that would cover everyone than for a private insurance that would cover only themselves and their family; (3) to explore the validity of the WTP estimates by estimating their relationship with variables theoretically associated with WTP; and (4) to examine the effect of variations in the study instruments on the WTP responses.

MATERIALS AND METHODS

Sampling. The design of the study is summarized in Figure 1. The convenience sample of RA patients was identified from the files of 5 participating rheumatologists affiliated with the McGill University Health Center (MUHC) Division of Rheumatology. Patients could be drawn from the MUHC rheumatology clinic or from their private practices. The medical charts of all patients were reviewed by a research nurse to ensure RA diagnosis on the basis of the 1987 American College of Rheumatology (ACR) classification criteria. All patients then received by mail from their rheumatologist: (1) an introductory letter describing the survey; and (2) an informed consent document, to be signed and returned by mail. In cases where consent to participate was received, participants were contacted by telephone by specially trained interviewers to confirm their participation and arrange a telephone interview. Prior to the interview, participants were sent a 4-page brochure summarizing the main information about RA. A few days later, the survey questionnaire was administered during the scheduled telephone call. Finally, within the following month, patients were asked to complete 2 mailed questionnaires: the Health Assessment Questionnaire (HAQ) disability index and a questionnaire recording the healthcare system resource use and productivity losses, derived from the economic component of the HAQ[54]. The study was approved by the MUHC Research Ethics Committee.

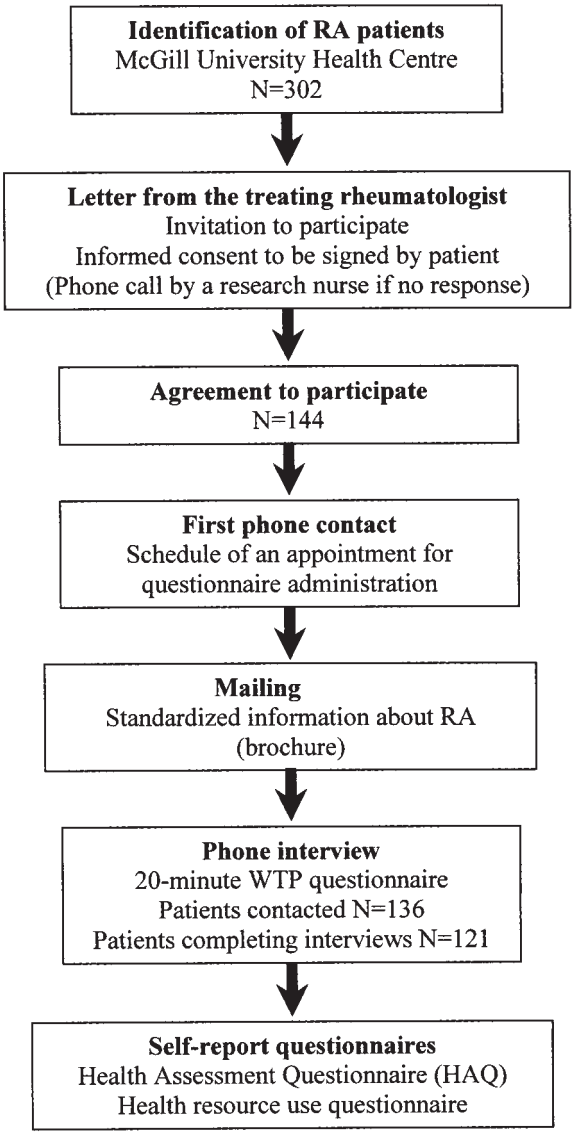

Figure 1. General design of the study.

RA information brochure. Since the knowledge of RA patients about their disease may be heterogeneous, standardized information about RA was provided in a 4-page brochure, entitled "Rheumatoid arthritis: 10 questions and 10 answers," explaining the symptoms of the disease, their impact on patients' activities of daily living, and their evolution with or without treatment. This brochure was developed by a group of 8 rheumatologists and a member of an arthritis consumer advocacy organization. To test for sensitivity of the WTP responses to the mode of presentation of the health state description, 2 brochure formats were produced and the brochure type was randomly assigned to respondents; one contained only text, as is often the case in WTP studies, and the other included pictures of joint deformities in the hands of a patient with RA in addition to the text, giving visual insight into the functional and cosmetic consequences of the disease.

WTP questions. Respondents were informed that this treatment was purely hypothetical and not based on the knowledge of new therapies either already available or in development (Appendix). As in a previous report[28], we elicited the WTP for a hypothetical cure, which would be 100% effective, under 2 different scenarios — private insurance and public insurance. In the private insurance scenario, we asked the participants to imagine that they lived in a country where there is only private health insurance, and that they had a one-time chance to purchase supplemental insurance to cover the RA cure for themselves and their family. In this case, the WTP was measured as the maximum annual premium for which they would purchase the supplemental RA insurance. Following Neumann and Johannesson[28], the one-time aspect of the offer was included to discourage "free-riding" by patients until some future period. In the public (second) scenario, the patients were asked to consider that they lived in a place such as Quebec, where medical care is provided through a tax-financed public system. The WTP was then measured as the maximum amount of additional annual taxes that they would accept in a provincial referendum concerning the program. It was explained to participants that, if the referendum did not pass, the RA treatment would not be offered in the province.

The elicitation of the WTP value was based on the payment "card" method. Each respondent was randomly allocated one of 3 cards. The 3 "cards" included overall ranges of (Canadian dollars): 0 to CAD $2000 or more; 0 to CAD $4000 or more; and 0 to CAD $8000 or more. Each card type was divided into 5 WTP ranges; after selecting a first range, respondents were asked to choose between 2 subranges within that category; finally, respondents were asked to provide a precise value. As another measure of the difference in their preference for the 2 programs, we also asked the patients whether they would prefer the private program scenario or the public program scenario.

Determinants of WTP. The putative determinants of WTP were based on a simple theoretical model of demand for health insurance, where WTP is related to ability to pay; measures of disease severity or measure of expected disease progression (expected benefit) and, in the case of the private plan, the presence of family members also affected by RA; and the "taste" or preference for health, relative to other possible goods. For the public plan, WTP may be more due to feelings of altruism, and this altruism could increase with the increasing number of persons among a respondent's family or acquaintances affected by RA.

The concept of ability to pay relevant to WTP is permanent income, the average income an individual or household expects to receive over a period of years[55]. Although permanent income cannot be observed, it is related to demographic and socioeconomic factors measured in our study, which included age, sex, educational attainment, household composition, current household income, the presence of private health insurance — which is related to generosity of employment compensation, and the location of usual RA care (hospital based public clinic or private clinic). To the extent that private health insurance reflects the choice of the individual rather than an employer, this variable may also reflect the respondent's taste or preference for health or his/her health status. Although physician care is fully reimbursed by the provincial health plan in both private and public clinics, the location of RA care may be a measure of the ability to pay for RA cure insurance, since the downtown location of the public clinic may be related to residential choices related to the respondents' ability to pay. Finally, to the extent that health status affects the ability to earn income, several measures of health status (HAQ, general health self-rated on a 5-category scale or on a 0–100 verbal rating scale, presence and number of comorbidities) and variables related to health status and RA risk (age, sex) are also in part measures of ability to pay.

In theory, holding the quality of the RA cure constant, the WTP for the public program should be higher than that for the private program, if there are important altruistic feelings. Although in both private and public WTP questions, we explained that the RA cure would be 100% effective, to help us understand possible reasons for differences in WTP responses between private and public programs, we included the following question on the respondents' assessment of the provincial healthcare system: "Thinking of the present healthcare system in the province, overall, how would you rate it?". The possible responses were: excellent, very good, fair, and poor.

To assess the capacity and the desire of respondents to participate in studies such as ours, the survey closed with the following 2 questions: (1) Did you find the questions difficult to answer? and (2) Do you think that

questions on allocation of resources in the health field are too difficult to be answered by the general population; therefore should they be left for the experts in the health service?

Statistical analysis. All analyses were conducted with Stata software, release 5 (Stat Corp., College Station, TX, USA). The patients' WTP in the context of the 2 programs was compared using a Wilcoxon nonparametric test, since WTP is typically not normally distributed, usually including a significant number of zero responses, with the positive responses being right-skewed[56]. The variation of WTP values according to sociodemographics, health parameters, organization of and perceptions about the existing healthcare system in the country, and the design and the format of the survey were studied. In a first step, a univariate analysis was conducted, and WTP estimates were compared in different subgroups by nonparametric tests: Mann-Whitney tests for dichotomous variables and Kruskal-Wallis tests for polytomous variables. Spearman correlation coefficients and correlation matrices were used to investigate the relations between health status variables and ability to pay, i.e., socioeconomic variables.

Finally, a multivariate analysis was conducted on the basis of an ordered logit (polytomous logistic) regression model. For this purpose, the dependent variables, i.e., individual WTP estimates for the private program and for the public one were categorized in 5 classes: 0 CAD, $1 to $200 CAD, $201 to $500 CAD, $501 to $1000 CAD, and more than $1000 CAD. The variables significantly associated with WTP in the univariate analysis were included in the analysis as explanatory variables. The level of significance for variables included in the model was set at 0.1. For each, the model provided regression coefficients, the antilogs of which were the odds ratios (OR) expressing the effect of the increment of 1 unit of one of the independent variables, the others remaining constant. The equation of the ordered logit model was:

$$Ln\left(\frac{P\,[y > j]}{1 - P[y > j]}\right) = \alpha_j + b_1x_1 + b_2x_2 + \ldots + b_nx_n,$$

where j is one of the thresholds, i.e., 0, $200, $500 or $1000 CAD, and a_j the intercept for each of these values so that $P_0[y > j] = e^{\alpha j}/(1 + e^{\alpha j})$.

RESULTS

Among the 302 patients contacted, 144 consented to participate in the study and complete responses were available for 121 (Table 1). All had RA and satisfied the 1987 ACR classification criteria. There were 96 women (79.3%), with a mean age of 57.4 years, and 60.3% were primarily English speaking. The majority of them had established RA, with mean disease duration of 13.1 years; it was less than or equal to 1 year for 7.7% of the patients, between 2 and 5 years for 25%, between 6 and 10 years for 20.2%, and exceeded 10 years in 47.1%. Their mean HAQ score was 1; only a few patients (13%) were severely disabled, i.e., with a HAQ score > 2. One or several other chronic conditions in addition to RA were observed in about one-third of the population (36.4%). Globally, respondents self-rated their health status as good or very good in more than half of the sample (58.7%), fair in 28.9%, and poor in 12.4%. Expressed on the 0–100 rating scale, 0 being death, 100 full health, the self-reported health rating was 63.3 on average, ranging from 2 to 95. Forty-four (36.4%) patients were cared for at the McGill University Division of Rheumatology, and the remaining 77 (63.6%) in private clinics of the participating rheumatologists. Overall, acceptance of the survey was favorable: 77.7% of respondents did not find the questions difficult, and 72.7% believed that the economic aspects of health should not be addressed only by experts.

Table 1. Main characteristics of patients with RA (n = 121).

Characteristic	N
Age, yrs	
Mean ± SD	57.4 ± 11.3
Median	57
Range	27–70
Female, n (%)	96 (79.3)
Married, n (%)	73 (61.3)
English speaking, n (%)	73 (60.3)
Medical information	
Site of care, n (%)	
Hospital	44 (36.4)
Private clinic	77 (63.6)
RA duration, yrs	
Mean ± SD	13.1 ± 11.5
Median	10
Range	1–51
Self-rated health status (0–100 verbal scale)	
Mean ± SD	63.3 ± 20.4
Median	70
Range	2–95
or	
Categorical, n (%)	
Fair or poor	50 (41.3)
Good	50 (41.3)
Very good/ excellent	21 (17.4)
HAQ	
Mean ± SD	1.0 ± 0.8
Median	1
Range	0–2.8
or	
Categorical, n (%)	
0 to < 1	48 (44.9)
1 to < 2	45 (42.1)
2 to 3	14 (13.0)
Pain (0–100 verbal scale)	
Mean ± SD	39.1 ± 23.7
Median	36
Range	0–92
Chronic condition(s) other than RA, n (%)	44 (36.4)
Mean number if present ± SD	1.4 ± 0.6
Median	1
Range	1–3
Socioeconomic status	
Personal annual income (CAD$)	
Mean ± SD	34,575 ± 26,880
Median	25,000
Annual household income (CAD$)	
Mean ± SD	48,702 ± 31,079
Median	45,000
Work status, n (%)	
Full-time paid work	26 (24.3)
Part-time paid work	10 (9.4)
Pension	38 (35.5)
Housekeeping	11 (10.3)
Volunteering	5 (4.7)
RA-related disability	15 (14.0)
Non-RA-related disability	1 (1)
Other	1 (1)

Patients' WTP for a hypothetical cure of RA. The WTP of the patients for a hypothetical cure of RA was obtained from 119 of the 121 respondents; 2 patients were unable to provide either category or precise value. Maximum WTP values were similar for both private and public programs and, as expected, there was a significant number of zero responses, with the positive responses skewed to the right (Table 2). Sixty percent of the respondents expressed a preference for public coverage of the hypothetical RA cure. However, the mean and median WTP were significantly higher for the private program than for the public one, $1190 CAD versus $502, respectively, evaluated at the mean, and $600 CAD and $200, respectively, evaluated at the median ($p < 0.00001$, Wilcoxon sign-rank test). Moreover, in the public program, respondents expressed more reluctance to pay, i.e., more zero values than in the private insurance program. Thirty-six respondents (30.3%, 95% CI 21.8%, 38.7%) declined to pay anything for the public program versus only 20 (16.8%, 95% CI 10.4%, 24.4%) who declined to pay for the private one. For the public program, the main explanation for declining payment was a refusal to pay additional taxes (60.5%); for the private program, 47.8% of those refusing to pay stated they could not afford to and 26.1% considered that such a treatment should be covered by public insurance.

Factors influencing WTP in the univariate analysis. Age and household income were the 2 sociodemographic variables associated with WTP estimates in both insurance programs (Table 3). Patients older than 60 years had a significantly lower WTP than younger ones: median WTP for the private insurance was $300 CAD in this age group compared to $800 and $1000 in the 2 others ($p = 0.0001$, Kruskal-Wallis nonparametric test), and $150 CAD for the public coverage versus $300 and $400 ($p = 0.039$, Kruskal-Wallis nonparametric test). The WTP was also significantly higher for both programs in the group with higher household income: in the 3 increasing-ordered income groups, the median WTP estimates were $300, $550, and $1000 CAD for the private program, and $100, $300, and $400 CAD for the public one ($p = 0.003$ and $p = 0.032$, respectively, Kruskal-Wallis nonparametric tests). Fifteen respondents (13%) refused to disclose any information about their incomes; although their mean WTP was higher than that of other respondents, their median was lower.

No association was found between health variables and WTP (Table 3). However, an association of WTP was also found with healthcare system variables (Table 3). The WTP for the private program was significantly higher in patients cared for in private clinics ($p = 0.039$, Mann-Whitney test) or those already in possession of private health or drug insurance ($p = 0.004$, Mann-Whitney test). This latter finding was also observed for the public program WTP ($p = 0.027$, Mann-Whitney test). However, in this program, the opinion about the existing healthcare system was also important, the WTP being lower for respondents who have a poor opinion ($p = 0.031$, Kruskal-Wallis test).

No significant trend was found between the 3 different grids and the WTP responses, ruling out a potential payment card bias (Table 3). However, in patients who had received an information brochure containing the 3 pictures of the hands of a patient with RA, the WTP in the context of the private insurance was significantly higher than that of the patients who received the text-only brochure, i.e., with no photos ($p = 0.049$, Mann-Whitney test). This trend was not observed in the context of the public coverage.

Correlations between WTP and explanatory variables. Several significant correlations were found. First, there was a positive correlation between the WTP values for the private and the public programs, with a coefficient of 0.51 ($p < 0.00001$; Table 4). As expected from the univariate analysis, the WTP for the private program was significantly positively correlated with household income and negatively with age. In addition, there was a negative correlation between age and income.

As in the univariate analysis, no correlation was found between the WTP and the health variables, except a trend for a positive correlation between the WTP for the public program and RA disease duration ($p = 0.06$). Consistent with the idea that health status is related to the ability to earn income, there was a significant positive correlation between mean household income and health status rating ($p = 0.001$) and a negative one with HAQ score ($p = 0.03$). This may explain in part why WTP was not associated with health status measures in Table 3: patients with poorer health status have less ability to pay.

Factors associated with WTP in the multivariate analysis. As shown above, several explanatory variables were highly correlated (Table 5). The multivariate analysis, based on an ordered logit regression, showed that the variables associat-

Table 2. Maximum WTP for a hypothetical cure of RA in the public and private insurance programs.

	N	Maximum WTP ($CAD) Mean	Minimum	Maximum	Median	IQR	p*
For private program	119	1190 ± 1589	0	8000	600	1000 (200;1200)	< 0.00001
For public program	119	502 ± 1057	0	8000	200	500 (0; 500)	

* Nonparametric tests: Wilcoxon sign-rank test.

Table 3. Variations of the maximum WTP for a complete cure in patients living with RA (univariate analysis).

Variables	WTP for Private Program N	Mean	Median	IQR	p*	WTP for Public Program N	Mean	Median	IQR	p*
Age group, yrs										
18–49	33	1684	800	1600 (400;2000)	0.0001	33	476	300	475 (125;600)	0.039
50–60	31	1514	1000	1500 (500;2000)		31	859	400	800 (0;800)	
More than 60	57	710	300	525 (75;600)		55	317	150	400 (0;400)	
Education										
Less than secondary	20	628	350	600 (50;650)	0.265	20	154	150	200 (0;200)	0.385
Secondary	30	1177	675	1450 (150;1600)		29	456	200	500 (0;500)	
Post-secondary	42	1304	650	800 (400;1200)		43	545	300	750 (0;750)	
University	27	1444	800	1925 (75; 2000)		27	741	350	800 (0;700)	
Household income (2001 $CAD)										
< 30,000	33	832	300	700 (100;800)	0.003	33	595	100	400 (0;400)	0.032
30–60,000	36	679	550	900 (200;1100)		36	297	300	400 (0;400)	
> 60,000	35	1841	1000	2900 (500;3400)		35	604	400	650 (150;800)	
Refusal**	15	1683	250	4000 (0;4000)		15	533	0	600 (0;600)	
Health status										
Fair or poor	49	1205	500	1000 (200;1200)	0.183	49	649	200	600 (0;400)	0.231
Good	50	1066	450	1075 (125;1200)		50	378	150	400 (0;400)	
Very good	20	1461	900	1550 (450;2000)		20	454	450	538 (63;600)	
HAQ score										
0 – ≤ 1	48	1024	600	1000 (200;1200)	0.663	48	419	300	500 (0;500)	0.702
1 – ≤ 2	44	1356	800	1850 (150;2000)		44	422	200	450 (0;450)	
2 – ≤ 3	14	1793	800	1700 (300;2000)		14	739	275	500 (0;500)	
Chronic conditions										
No	76	1033	500	925 (175;1100)	0.334	75	377	200	500 (0;500)	0.910
Yes	43	1467	800	1700 (300;2000)		44	715	200	625 (0;625)	
Site of care										
Hospital	44	1113	450	900 (0;900)	0.039	44	668	200	550 (0;550)	0.764
Private clinic	75	1235	800	1200 (300;1500)		77	405	200	500 (0;500)	
Opinion about healthcare service										
Poor	25	1407	600	1375 (125;1500)	0.983	25	235	30	200 (0;200)	0.031
Fair	39	1243	700	1050 (150;1200)		39	409	150	500 (0;500)	
Good or very good	55	1053	500	1150 (250;1400)		55	690	350	500 (100;600)	
Private health or drug insurance										
No	62	867	400	925 (75;1000)	0.004	61	457	125	400 (0;400)	0.027
Yes	57	1541	800	1600 (400;2000)		58	550	300	525 (75;600)	
Grid										
1: 0–2000	37	1260	500	1800 (200;2000)	0.680	37	221	150	350 (0;350)	0.077
2: 0–4000	41	1120	400	1100 (100;1200)		40	733	400	525 (125;650)	
3: 0–8000	41	1196	800	900 (300;1200)		42	530	200	600 (0;600)	
Picture										
No	55	802	400	600 (200;800)	0.049	56	315	188	400 (0;400)	0.083
Yes	64	1523	800	1863 (138;2000)		63	668	200	800 (0;800)	
Program preference										
Public	66	1264	550	1500 (100;1600)	0.696	66	574	200	500 (0;500)	0.738
Private	45	1204	600	950 (250;1200)		45	468	200	500 (0;500)	

* Nonparametric tests: Mann-Whitney for dichotomous variables, Kruskal-Wallis for polytomous variables. ** Refusal to provide information about income.

ed with the WTP for the private program were age (OR = 0.97), care in a public hospital (OR = 0.34), already having private health insurance (OR = 2.58), and the presence of the pictures on the information leaflet (OR = 2.01). For the public program, the only variables associated with the WTP were the opinion about the healthcare system (OR = 2.13) and already possessing private health insurance (OR = 2.39). Because the HAQ and general health status were shown to be related to household income in Table 4, in a set of alternative specifications not shown, we added these variables to the multivariate model. However, the coefficients on these variables were imprecisely estimated, so we cannot make any conclusions about their effects.

DISCUSSION

Our study provides an estimation of RA patients' WTP for a hypothetical complete cure of their disease. Our results represent a partial estimate of society's WTP for an RA cure;

Table 4. Correlation matrix of maximum WTP with socioeconomic and health variables.

	WTP for Private Program	WTP for Public Program	Mean Income	Mean Age	Health Status[1]	HAQ	Pain on VAS	RA Duration
WTP private program	1							
WTP public program	0.51¶	1						
Mean income	0.42¶	0.30***	1					
Mean age	–0.36†	–0.22**	–0.38†	1				
Health status	0.01	0.08	0.37†	–0.12	1			
HAQ	0.03	–0.03	–0.23**	0.01	–0.50¶	1		
Pain on VAS	–0.06	–0.07	–0.20*	0.11	–0.43¶	0.67¶	1	
RA duration	0.02	0.17*	–0.16	0.17*	–0.32†	0.31***	0.27**	1

[1] Health status has been estimated on a 0–100 verbal scale (0 worse – 100 better). * $p < 0.1$, ** $p < 0.05$, *** $p < 0.01$, † $p < 0.001$, ¶ $p < 0.00001$.

Table 5. Multivariate analysis of the factors influencing the maximum WTP for a complete cure in patients living with RA.

	Private Program		Public Program	
	OR (95% CI)	p	OR (95% CI)	p
Age*	0.97 (0.931;1.000)	0.05	0.99 (0.958;1.026)	0.62
Household income*	1.00 (1.000;1.000)	0.20	1.00 (1.000;1.000)	0.23
Care in a public hospital	0.34 (0.153;0.754)	0.01	0.86 (0.395;1.852)	0.69
Opinion about healthcare system	1.03 (0.643;1.657)	0.90	2.13 (1.316;3.448)	0.01
Private drug insurance	2.58 (1.089;6.129)	0.03	2.39 (1.014;5.649)	0.05
Picture	2.01 (0.937;4.331)	0.07	1.70 (0.809;3.559)	0.16
Intercept–Predicted P_0 (y > j)				
j = 0	0.04		2.70	
j = 200	0.09		10.89	
j = 500	0.36		42.68	
j = 1000	0.98		124.03	

* Age and household income have been treated as a continuous variable.

another part of the study will aim to incorporate the WTP for the hypothetical cure of the general population, who may be willing to pay through their desire to insure against the possibility of development of RA at a future date or because of altruistic motivations. Since our results from patients (roughly 1% of the population) are not comprehensive estimates of society's WTP, it is not possible to directly compare our results to those from human capital studies. In contrast to WTP studies directly focusing on the valuation of improvements of specific symptoms, we chose the option of a holistic scenario associated with the multidimensional change in health tied to an RA cure, as recommended by health economists[17,20]. In addition, in the preamble to the WTP questions, we reminded the subjects to place the payment associated with the RA programs in the context of their regular expenses, in order to elicit more realistic answers. Thus, we did not receive any unrealistic answers, and the percentage of persons with a WTP of zero (16.5% for the private program and 29.8% for the public one) was close to that observed in other studies[23,24,29-31,38,43]. To elicit WTP responses, we used payment cards, because of the well-known problems of starting-point bias inherent in the bidding game and of "yea-saying" leading to upwardly-biased estimates in the take-it-or-leave-it approach[20,21,47]. Since the payment card has been associated in some cases with framing bias, we tested for this effect by randomly assigning our patients to one of 3 payments cards. Our results indicate that our WTP estimates from this sample are not contaminated by framing bias.

The presentation of health state information in WTP and utility surveys is often quite short and not standardized, in RA or in other contexts. However, it appears that patient knowledge is often fragmented[48-51]. Thus, in our study we established a process to provide a complete, comprehensible presentation of relevant information about their disease. Research has shown the superior psychometric properties of health status descriptions supplementing textual information with other visual and audio aids[52]. Thus, we added (or omitted) 3 pictures of RA hands to the brochure sent to half of the respondents. A significant relationship was observed between the presence of pictures and the WTP for the private program; this association was not significant for the public one. From our data, it is difficult to speculate if one version of the brochure yields better estimates than the

other; however, as shown by others[57], it emphasizes the need for future research about presentation of health problems in such WTP studies.

As in the previous studies on WTP in patient populations, we cannot be sure how representative our sample is of the population of Quebeckers with RA. In general, RA patients followed in a university hospital outpatient clinic are more likely to have severe disease. However, our study included a substantial number of patients whose regular care was received in the private clinics of the participating physicians. In addition, only a small subset of the patients received anti-tumor necrosis factor (TNF) antibodies. Thus, it is likely that our sample was not restricted to patients with severe RA only. Indeed, the population demographics of our sample showed that they were very similar to those of many randomized controlled trials conducted in established RA.

Previous studies have demonstrated the construct validity of WTP surveys by estimating the association between the WTP responses and characteristics theoretically related to WTP: the respondents' ability to pay (income, other measures of permanent income) and the expected health benefit (disease-related and general health status, utility)[19,45,58]. We also observed a significant relation between WTP for the private program and care in public clinics and the presence of private health or drug insurance. Care in a public clinic and private health or drug insurance are probably, in large part, measures of ability to pay, and are in fact correlated with household income in the sample. However, to the extent that the presence of health insurance represents a choice (of a job with insurance or to purchase health insurance), the effects of the insurance variables represent preferences of health versus other goods for these persons. Other measures of the expected benefit — general health status, number of chronic conditions, HAQ score — did not show a significant effect. Although general health status and the number of chronic conditions may have an ambiguous effect on WTP, it is somewhat surprising that the HAQ did not show an effect. We show in Table 4 that the HAQ score was negatively related to income, but in Table 3 the WTP responses did not decline with worsening HAQ score. This lack of association might be explained by the interpretation of the information given to the patients. Although we told the patients that the treatment was a cure that was 100% effective, it is possible that persons with severe disease did not believe that the cure could reverse their damage. One patient in an open-ended response concerning her WTP of zero explained that her condition was too advanced for her to benefit. Conversely, a patient with mild RA might expect a very big benefit from a cure, if, in the absence of a cure, she/he had expected a substantial decline over time. Also, although we described a variety of possible disease trajectories in the brochure, nonetheless its effect might have been to introduce a certain degree of uniformity in patients' projection of the course of the disease, regardless of the level of severity at the time of the survey.

In previous studies, age has also been associated with WTP, either positively or negatively[26,29,30,33,40-43,46,59-61]. In the present case, elderly people, i.e., older than 65 years, had a lower WTP than younger respondents. This might be related to a lower likelihood of experiencing benefit from the proposed program because their children are old enough to be economically independent. It is also possible that the age effect represents a lower ability to pay, although for the private program, age continued to have a significant negative effect on WTP, even when other measures of ability to pay were controlled for.

One of the innovations of our study lies in the 2-program scenario. The private scenario was designed to capture the direct-benefit and "option" valuations associated with an RA cure, while the public scenario was intended to capture direct-benefit, option, and altruistic valuations. The difference in WTP between the 2 programs was intended to yield the altruistic valuations of the respondents. However, the WTP for the public program turned out to be lower than that of the private program, even though almost two-thirds of the respondents stated their preference for a public healthcare system. For the public program, the respondents' opinion on the performance of the public healthcare system was a significant predictor of the WTP for the public program, indicating the presence of protest responses. In addition, although the referendum-type question with associated taxes has been proposed to correct for free-riding in WTP surveys for public goods[17], it is possible that this approach does not fully address this problem. Thus, unfortunately, the comparison of the WTP for the private and public programs does not tell us anything about the magnitude of altruistic valuations of our sample of patients with RA. Another explanation for the lower public WTP was an ordering effect, since the public program scenario was presented after the private one; this must be addressed in future studies.

It is interesting to compare results from this study to results from previous WTP studies focusing on samples of arthritis patients. In a study conducted in patients with chronic arthritis, either osteoarthritis or RA, in the 1980s[23,24], respondents were asked to provide the percentage of their income that they might be willing to pay to be completely relieved of their symptoms; the patients' responses implied WTP varying from $1820 CAD ($1400 USD) to more than $6500 CAD ($5000 USD) (1984 currency). Providing a WTP as a percentage of income is unusual in WTP studies — almost all WTP studies ask for a dollar amount, and this difference in the framing of the WTP elicitation may explain the higher level of WTP. Also, the studies from Thompson, *et al* excluded from the sample a substantial number of patients expressing a zero WTP. Another report focused on the marginal WTP for anti-TNF-α therapy[25]; the mean annual WTP was estimated between

$1330 CAD ($1100 USD) and $2660 CAD ($2200 USD) (2000 currency). These estimates are substantially higher than those elicited in our study. Again, the method of elicitation may explain the differences with our study, since their study used different elicitation methods[62].

The present work emphasizes the interest of involving patients in complex questions about health and organization of the health care system[63-67]. In general, patients found the survey easy to respond to and expressed a preference for being consulted about health care decision-making. The responses were realistic and were associated with several variables expected to be related to WTP.

Our results show that it is possible to explore with patients the monetary aspects of their cure, even in a context where patients do not have to pay directly for their care, such as in the publicly funded Quebec healthcare system, a result consistent with patients participating in the study in Denmark[62], where healthcare is also publicly funded. Given that currently affected individuals are likely to benefit the most from a cure, the WTP responses of patients may make up an important part of society's WTP (along with the WTP of unaffected individuals), and bring additional information about the perception of the disease by the patients, which may be of relevance in the exchanges between a patient and her/his physician.

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APPENDIX

Instructions to respondents

You have read the brochure about rheumatoid arthritis. We would like to remind you that:

- Rheumatoid arthritis is the most disabling joint disease and it is able to cause extensive damage to joints.
- Rheumatoid arthritis affects approximately 1 out of every 100 people, that is 50,000 to 70,000 Quebeckers, mainly women.
- The functional impact of the disease is due to pain, joint swelling, stiffening and deformation.

There is no way to predict disease occurrence in a given person, or its severity when the disease is diagnosed.

- Currently available treatments can limit the disease consequences but cannot cure it.

The goal of our study is to determine the public's willingness to pay for a hypothetical cure of rheumatoid arthritis, to estimate how important such a cure can be. For this purpose, we would like you to imagine that this treatment, taken regularly, can alleviate all the symptoms described in the brochure for persons who currently are living with rheumatoid arthritis. All answers are valuable for this purpose: there are no right or wrong answers. Before answering, please keep in mind that:

- Such a cure is purely hypothetical. As described in the brochure, although there are many treatments for RA, to our knowledge, no complete cure for RA is available or in development.
- In addition to the amount you will accept to pay on an annual basis for the RA cure, you will have to pay for your other usual expenditures out of your income.

Willingness to pay for a private program

- Imagine that a cure for RA becomes available. This hypothetical cure is 100% effective for all affected individuals.
- For the purposes of this questionnaire, we would also like you to imagine that you live in a country like the USA where people do pay for private health insurance.
- Suppose that you are offered a one-time chance to purchase lifetime coverage for the rheumatoid arthritis cure; that is, if you do not elect to buy coverage now, it will not be possible to do so later. For this insurance, you have to pay an annual premium in addition (a supplement) to any currently private insurance that you may have.

What is the maximum annual premium charge at which you would accept this one-time insurance offer?

Willingness to pay for a public program

- Imagine that a cure for RA becomes available. This hypothetical cure is 100% effective for all affected individuals.
- Now consider the situation in Quebec, where medical care is provided to everyone without charge in a tax-financed public system.
- Recall that some treatments are not covered by the Régie de l'assurance maladie de Quebec [Quebec public health insurance system].
- The government has decided to hold a vote to know whether the public is willing to pay the additional taxes required to finance this RA cure as part of the health care system. If the initiative does not pass, the government will not go ahead with the program and the cure will not be offered.
- The ballot question will be: "Should the government raise your annual taxes by ___ dollars to finance a RA cure provided through the public health care system?"

What is the maximum amount of additional annual taxes for which you would vote yes to this ballot question?

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