Cost Assessment Instruments in Rheumatology: Evaluation of Applied Instrument Characteristics

JORG RUOF, SONIA MERKESDAL, JAN L. HUELSEMANN, OLIVER SCHOEFFSKI, ANDREAS MAETZEL, WILFRED MAU, and HENNING ZEIDLER

ABSTRACT. We compared the major characteristics of internationally applied cost assessment instruments (CAI) in rheumatic conditions. Fifteen utilization questionnaires were identified and assessed using a structured approach. The forms differed considerably with respect to applied characteristics: length (3–113 items), recall period (between 1 week and 1 year), format (2 interview, 13 self-administered), response categories, cost units (physical vs monetary), and cost domains covered. While all included a gross assessment of outpatient and inpatient costs, the level of disaggregation differed. Only a few CAI included an assessment of other direct disease related costs (e.g., home remodeling or home health care services) and out-of-pocket expenditure. Productivity costs were included in all but 2 CAI. Efforts to further standardize the applied CAI should (1) be based on sound psychometric data, (2) define a required core set of cost domains covered, (3) discriminate between generic and relevant disease related cost components, and (4) examine the feasibility of developing international standards for cost data. (J Rheumatol 2001;28:662–5)

Key Indexing Terms:
COST OF ILLNESS ECONOMIC EVALUATION QUESTIONNAIRES

BACKGROUND

While clinical outcome measures of rheumatic diseases have undergone considerable efforts of standardization, measures of resource utilization and costs are often applied on an ad hoc basis. It is no surprise then that cost evaluations were often found to be incomplete and very heterogeneous with respect to covered cost domains and the costing methods used[1]. So far, no standardized cost assessment instrument (CAI) has been published nor any form of agreement on a core set of minimally required cost items. Additionally, there is a lack of studies exploring psychometric properties (truth, discrimination, feasibility[2]) of patient derived cost data. Consequently, comparability of study results may be very limited.

We examined and compared the major characteristics of internationally applied cost questionnaires in rheumatic conditions.

METHODS

Three approaches were applied: (1) identification of authors/research groups who published patient derived cost data in rheumatic conditions; (2) establishing direct contact with the authors that included a request for the CAI they used; and (3) structured analysis of instruments received.

The identification of authors who published patient derived cost data was performed by review of all articles that were cited in 5 recently published literature reviews[1,3-6] on economic evaluation in rheumatic conditions. The review also included the articles cited in our synthesis of cost domains in rheumatic conditions[7]. All articles were analyzed and authors/research groups who (1) reported original cost data, (2) provided a detailed cost analysis, (3) explicitly discussed the source of cost data in the methods section, and (4) primarily relied on CAI derived cost data were identified.

All authors received a set of 4 questions: (1) Was the CAI developed originally for this study; (2) Do you have any published information on the development process; (3) Do you have data documenting validity and reliability of the CAI; and (4) May we receive a copy of the CAI. Received questionnaires were examined according to a structured protocol covering characteristics such as disease focus, length of the forms, units of cost assessment (monetary versus physical), recall period, availability of published psychometric data (truth, discrimination, feasibility), original language, and predominant response category. Additionally we used the matrix of cost domains to document the number and detail of cost components covered by the individual questionnaires.

Many of the questionnaires were provided for confidential use only. We therefore decided not to disclose the authors by questionnaire, but rather to mention all authors

From the Division of Rheumatology, Hannover Medical School, Forschungsstelle Gesundheitsökonomie, Universität Hannover, Hannover, Germany; and the Arthritis and Autoimmunity Research Centre, University Health Network, Toronto, Ontario, Canada.

J. Ruof, MD, MPH; S. Merkesdal, MD; J.L. Huelsemann, MD; W. Mau, MD, PhD; H. Zeidler, MD, Professor of Internal Medicine and Rheumatology, Division of Rheumatology, Hannover Medical School; O. Schoeffski, MPH, PhD, Associate Professor of Health Economics, University of Hannover; A. Maetzel, MD, MSc, Arthritis and Autoimmunity Research Centre, University Health Network.

Address reprint requests to Dr. J. Ruof, Abteilung für Rheumatologie, Medizinische Hochschule Hannover, Carl Neuberg Str. 5, 30625 Hannover, Germany.

who provided a questionnaire in the Acknowledgment. Moreover, only CAI of authors who agreed with this approach were included in the results section of this article.

RESULTS

Through review of the literature 26 articles that met the criteria outlined in the Methods section were identified. Eighteen authors covering all the identified articles were contacted. Seven authors did not respond [6 university groups/authors and one from a contract research organization; the university authors were located in rheumatology divisions (3 authors), orthopedic divisions (1 author), department of family and preventive medicine (1 author), and department of physical therapy (1 author)]. One of the nonresponding authors has published detailed costing studies. However, several attempts to reach him by either phone or mail did not succeed. CAI were obtained from 11 authors/research groups (see Acknowledgment). Four authors provided more than one CAI.

A total of 15 CAI were subject to review. Table 1 summarizes the major characteristics of the 15 questionnaires. Published data related to psychometric properties of the forms were available for only 2 CAI[8,9]. However, 2 other groups are currently working on such data. Table 2 displays the cost domains covered by the individual CAI. While all of them included a gross assessment of outpatient and inpatient costs, the level of disaggregation differed considerably. Additionally, only a small number of CAI included a comprehensive assessment of other direct disease related costs, i.e., costs incurred for items such as home remodeling or home health care services and out-of-pocket expenditure. Productivity costs were included in all but 2 CAI. However, opportunity costs and lost wages were covered only by a minority of the reviewed instruments.

DISCUSSION

Standardization of cost assessment in rheumatic conditions is a major concern of the OMERACT task force on economic evaluation[10]. An important obstacle in this context is the usage of a great variety of objective (e.g., hospital data or the Medicare data base) and subjective (patient derived) cost data sources, which limits the comparability of study results[1]. While objective data sources are tied to the highly specific features of each health care system and to the char-

Table 1. Major characteristics of the received cost/utilization questionnaires (Due to confidentiality of some questionnaires all questionnaires are only cited with anonymous numbers).

	Questionnaire Number														
	1	2	3	4	5	6	7	8	9	10	11	12	#13	#14	15
Disease focus	RA	RA	Variable	RA	SLE	Variable	LBP	RA	RA	RA	RA & OA	RA	AS	AS	Variable
Total items*	32	113	65	69	15	N/A	73	11	23	24	30	30	12	5	3
Self-administered format	+	–	+	+	+	+	–	+	+	+	+	+	+	+	+
Direct costs units	m&p	m&p	p	m&p	p	m&p	p	m&p	p	m&p	m&p	m&p	m&p	m&p	p
Productivity costs units	p	p	p	p	m&p	m&p	n/a	n/a	p	m&p	m&p	p	m	p	n/a
Predominant recall period	Variable	1 year	1 year	Variable	6 month	6 month	Variable	Variable	Variable	Variable	1 year	Variable	1 year	1 week	6 mo
Psychometric data available	–	–	–	–	(+)	–	+	–	–	–	–	–	(+)	(+)	(+)
Population/language	Australian	US	German	German	US/ UK/ Canadian	US	Canadian	Canadian	US/ Canadian	Canadian	US	Dutch	Dutch	Dutch	US
Predominant response category	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ Likert scales	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Item lists/ free space/ Yes-no items	Yes-no items/ Free space	Item lists/ free space/ Yes-no items	Yes-no/ Free space	Free space	Free space

N/A: not applicable.
#Both instruments are designed to be used together: 13 is a more comprehensive baseline questionnaire and 14 is a followup diary.
*the numeration provided by the authors was used; questionnaires 2-4 covered more than pure socioeconomic questions.
m: monetary units; p: physical units; RA: rheumatoid arthritis; SLE: systemic lupus erythematosus; AS: ankylosing spondylitis; LBP: low back pain; OA: osteoarthritis.

Table 2. Cost domains included in the various questionnaires. Listed cost domains are derived from Reference 3.

	Questionnaire Number														
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
1 Health care costs (direct)															
Outpatient costs	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Visits to physicians	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Outpatient surgery	(+)	+	(+)	(+)	+	(+)	(+)	–	(+)	–	–	(+)	–	–	–
Emergency room visits	–	–	–	–	+	–	+	+	(+)	–	–	–	–	–	+
Non physician service utilization	+	+	+	+	+	+	+	–	+	–	–	+	+	–	–
Medication	+	+	+	+	+	+	+	–	–	+	–	–	+	+	–
Diagnostic/therapeutic procedures and tests	+	+	–	–	+	+	+	+	+	+	–	–	+	–	–
Devices and aids	+	–	–	+	+	–	+	–	+	+	+	+	+	–	–
Inpatient costs	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Acute hospital facilities (without surgery)	+	+	(+)	(+)	+	+	+	+	(+)	+	–	+	–	–	–
Acute hospital facilities (surgery)	(+)	+	(+)	(+)	+	+	(+)	–	(+)	–	–	+	–	–	–
Non acute hospital facilities	(+)	+	+	+	+	+	(+)	+	–	+	–	+	+	–	–
2 Other disease related costs (direct)															
Transportation	+	–	–	(+)	+	–	–	+	–	+	+	–	+	–	–
Home health care services	+	–	–	–	+	–	–	–	–	–	+	–	+	+	–
Home remodeling	+	–	–	–	–	–	–	–	–	+	+	+	–	–	–
Medical equipment	+	–	–	–	–	–	–	+	–	+	–	+	–	–	–
Non medical practitioner, alternative therapy	+	+	+	+	+	–	+	–	+	–	–	+	+	–	–
3 Productivity costs															
Loss of productivity in employed patients	+	+	+	+	+	+	–	+	+	+	+	+	+	+	–
Opportunity costs	–	+	+	+	+	–	–	–	+	+	–	+	–	–	–
Lost wages	–	–	(+)	–	+	–	–	–	–	–	–	+	–	–	–
Out of pocket*															
Out-of-pocket expenses	(+)	+	–	+	–	–	–	+	–	+	+	+	–	–	–
Data related to health insurance	+	+	–	–	–	+	–	–	–	–	+	+	–	–	–

+/–: cost domain covered/not covered.
(+): cost domain covered but not explicitly mentioned.
*May overlap with costs listed under 1-3.

acteristics of the data source provider, patient derived measures may provide an opportunity to further standardize cost assessment. However, our review revealed that currently a variety of cost/utilization questionnaires is used internationally, and most of them differ considerably with respect to major questionnaire characteristics.

While reviewing and analyzing the various CAI we identified 4 areas that may be useful to consider when discussing further standardization of cost assessment in rheumatic conditions:

(1) So far, there is only very limited information on psychometric properties (i.e., truth, discrimination, and feasibility — as suggested by the OMERACT filter[2]) of presently used CAI. After comparing patient diaries and provider records one author concluded that the developed CAI is a sufficiently valid source of health care utilization data in low back pain[8]. Clarke, *et al*[9] reported an overall agreement of > 90% of patient self-reports with (1) the Montreal General Hospital database that records all hospitalizations within the institution, and (2) the Quebec Ministry of Health data on all physician billings and reimbursements for prescription medications. However, unpublished data of 2 other questionnaires (Table 1, CAI No. 13 and 15) indicate differences between patient utilization reports and provider records. Further research is needed to identify those areas where patients' reports provide more valid and accurate utilization information and those where provider data are needed.

(2) A core set of cost domains that should be included in each clinical trial and/or observational study should be

identified. As a first step the domains that were covered by the majority of the reviewed CAI may be used. However, further research is required to identify the appropriate level of disaggregation for each cost domain and the effect of the selected response category on the utilization data provided. This point may be illustrated by comparing how the collection of medication data is performed by the various CAI. While CAI No. 6 (Table 1) included a detailed list of all available antirheumatic medication and dosing patterns on which patients were asked to mark their drug regimes, other CAI included only free text areas to actively list medication and dosage. It remains to be examined which of these approaches provides best access to the real drug consumption patterns. Similar problems occur in most of the other cost domains shown in Table 2.

(3) While many of the examined CAI focused on rheumatoid arthritis, some were designed to assess costs in other rheumatic conditions such as ankylosing spondylitis, low back pain, osteoarthritis, and systemic lupus erythematosus. Some CAI were also designed to cover more than one disease. Current approaches to estimating cost, such as the reference case analysis, favor a disease related cost assessment[11]. However, some of the items used in the various reviewed CAI were virtually identical (e.g., almost all CAI applied a list of physicians to capture data on outpatient physician visits). This supports the development of a core set of generic cost items that could be supplemented with disease related questions when examining costs in an individual rheumatic condition.

(4) Finally, international comparability of CAI derived cost data should be examined. This may be of particular relevance when considering the perspective of the cost analysis performed. For instance, our review revealed a detailed assessment of insurance coverage in some of the forms, while others completely excluded this aspect. It may be assumed that out-of-pocket expenditures differ considerably from country to country. In particular, international multicenter clinical trials should cover such aspects.

In conclusion, the reviewed CAI differed considerably with respect to the examined characteristics. Efforts to further standardize the applied CAI should (1) be based on sound psychometric data, (2) define a required core set of cost domains covered, (3) discriminate between generic and relevant disease related cost components, and (4) examine the feasibility of developing international standards for cost data.

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