

Specialized Rheumatology Nurse Substitutes for Rheumatologists in the Diagnostic Process of Fibromyalgia: A Cost-Consequence Analysis and a Randomized Controlled Trial

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ABSTRACT. Objective. To perform a cost-consequence analysis of the substitution of specialized rheumatology nurses (SRN) for rheumatologists (RMT) in the diagnostic process of fibromyalgia (FM), using both a healthcare and societal perspective and a 9-month period.

Methods. Alongside a randomized controlled trial, we measured costs and consequences of a nurse-led diagnostic consult (SRN group, n = 97) versus a rheumatologist-led diagnostic consult [usual care (UC) group, n = 96]. Patients were followed for 9 months. Every second month a questionnaire on medical consumption and social participation was filled out. Satisfaction was measured 1 week after the first consultation. During followup, health status was measured by health-related quality of life (EQ-5D), functional status (Fibromyalgia Impact Questionnaire), fatigue (Checklist Individual Strength), and self-efficacy (Generalized Self-Efficacy Scale).

Results. Patients in the SRN group were significantly more satisfied. Improvements in health status were similar in both groups after 9 months of followup. Total costs for healthcare consumption and patient and family costs were significantly lower in the SRN group (€1298 vs €1644; difference €346; 95% CI -€746 to -€2). Total societal costs were €3853 per patient for the SRN group and €5293 for the UC group after 9 months of followup (difference €1440; 95% CI -€3721 to €577).

Conclusion. From both a healthcare and societal perspective, the nurse-led diagnostic process can be recommended. Patients in the SRN group were significantly more satisfied, improvements in health status were similar in both groups, and total societal costs were lower for the SRN group compared to the RMT group after 9 months' followup. Registered with Current Controlled Trials, no. ISRCTN77212411. (J Rheumatol First Release April 1 2011; doi:10.3899/jrheum.100753)

Key Indexing Terms:

FIBROMYALGIA
OUTCOME AND PROCESS ASSESSMENT

COSTS AND COST ANALYSIS
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Fibromyalgia (FM) is a chronic pain disorder characterized by generalized musculoskeletal pain and concomitant symptoms such as fatigue, sleep disturbances, cognitive dysfunction, and depression. The prevalence has been estimated at 2% to 4% in the general population, forming 10% to 20% of rheumatologic consultations and 5% to 8% of primary care consultations^{1,2}.

Patients with FM rate their quality of life extremely low compared with other groups of patients^{3,4,5,6}. FM diminishes social and occupational functioning⁷. The economic burden of FM is considerable because of reduced productivity or ability to work and a high use of healthcare resources^{8,9,10,11,12,13,14,15}.

Patients may repeatedly present to the general practition-

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er (GP) with various symptoms before a diagnosis of FM is made. This delay may result in repetitive drug prescriptions, multiple diagnostic tests, and referrals to medical specialists and other healthcare providers¹⁶.

Little is known about the effect of labeling a patient with a diagnosis of FM. There are 2 schools of thought¹⁶. A diagnosis of FM may lead to increased illness behavior, dependence on healthcare providers, and increased health service costs^{17,18}. A diagnosis of FM, however, may also reduce the number of referrals, use of healthcare providers, and costs^{16,19}.

The importance of a prompt diagnosis in patients who are at risk of developing persistent pain and pain-related behavior is increasingly recognized²⁰. Early diagnosis and intervention may reassure the patient and reduce or prevent disability, which in turn will reduce societal and medical costs. Cost savings allowed by a diagnosis are estimated at €126 (2007 values) to €200 (2003 values) per patient per year^{16,21}.

Limitations in healthcare capacity and a high prevalence of FM may jeopardize an early approach. In Maastricht, in the southern part of The Netherlands, this problem was addressed by designing a nurse-led diagnostic process, in which trained, specialized rheumatology nurses (SRN) assist in the diagnostic process and simultaneously provide nursing care such as information, education, and support to this group of patients.

Over the last decade, the role of specialized nurses has evolved in undertaking such extended activities as patient assessments, formulating and carrying out plans of disease management, and making referrals to other health professionals^{22,23}.

In our previous study on the feasibility of substituting SRN for rheumatologists (RMT) in the diagnostic process of FM, we have shown that this approach is safe (no misclassifications), avoids waiting time, provides better patient satisfaction, and is cheaper from a health service perspective²⁴. Insight into the full societal consequence of this nurse-led diagnostic process is important for health professionals, healthcare management, health policy decision makers, and third-party payers. Our study investigates the effect of a timely diagnosis for FM by an SRN on health outcomes, social participation, and costs from both a healthcare and societal perspective.

MATERIALS AND METHODS

Patients. Between December 2003 and November 2005, we performed a 9-month pragmatic, prospective, randomized controlled trial with patients who were referred by their GP to the rheumatology outpatient clinic of the Maastricht University Hospital with a referral letter describing symptoms of FM. Inclusion criteria were suspicion of FM, first referral to the rheumatology outpatient clinic, age between 18 and 65 years, and the ability to read and write the Dutch language. Exclusion criteria were severe comorbidity and involvement in a legal procedure because of a disability pension. The protocol was approved by the local ethics committee of the University Hospital and University of Maastricht and all participants gave written informed consent. Current Controlled Trials no. ISRCTN77212411.

Intervention. The procedure of this RCT has been described in detail²⁴. Patients were randomly assigned to either the nurse-led diagnostic consultation (SRN group) or to the regular physician-led diagnostic consultation [usual care (UC) group]. The SRN patients were seen within 3 weeks by experienced SRN (n = 2) who were trained in the diagnosis of FM. During the consultation, the SRN used a checklist in detecting symptoms of FM as well as conditions that should be excluded. Also, a routine blood test was done. In a standardized 5-min supervision session immediately following the SRN consultation, an RMT who was involved in the study was informed by the SRN about the medical history. Further, the RMT performed a brief physical examination, and confirmed or rejected the diagnosis made by the SRN.

Patients in the UC group were seen in a regular clinical visit by an RMT after a waiting period of about 3 months. This visit included extensive history-taking, physical examination, and additional tests if considered necessary by the RMT. In both groups, FM was diagnosed according to the American College of Rheumatology criteria²⁵.

Study design. A prospective cost-consequence analysis²⁶ was performed to be able to present an array of outcome measures alongside cost for the 2 procedures of diagnostic process. Such an analysis is opportune if it is not feasible or practical to value all costs and benefits in monetary terms²⁷. In this situation, available monetary values can be augmented by other measures of cost and benefit such as waiting time and patient satisfaction²⁷. Presenting the results of our economic evaluation in a disaggregated format allows readers and decision makers to select the information most relevant to their perspective, while the overview of all aspects reflects the societal perspective.

Health assessments. Outcome measures recorded were patient satisfaction, health-related quality of life (HRQOL), functional status, fatigue, self-efficacy, medical consumption, and social participation. Patient satisfaction was measured 1 week after the first consultation. HRQOL and participation were assessed by 2 monthly questionnaires. Functional status, fatigue, and self-efficacy were assessed at baseline (before randomization) and after 3 weeks, and at 3, 6, and 9 months of followup.

Patient satisfaction was measured with questions derived from the QUOTE (Quality of Care Through the Patient's Eyes)-Rheumatic Patients²⁸, where Q-values ≥ 1 reflect care aspects that could be improved. Further details have been described²⁴.

HRQOL was measured by the EuroQol-5D (EQ-5D), a self-administered, generic instrument that incorporates descriptions and valuations of health states²⁹. The EQ-5D was developed and validated in a number of European countries, including The Netherlands^{30,31,32}, and has been used several times with patients with FM^{4,6}. We used the British (for reasons of comparison to foreign studies) and the Dutch utility tariff, with results in possible utilities ranging from -0.59 and -0.33, respectively (worst imaginable health state), to 1 (best imaginable health state, equal to full health)^{31,32,33,34}. The EQ-5D includes a visual analog scale (VAS) on which patients rate their current health state with endpoints of 100 (best imaginable health state) to 0 (worst imaginable health state). So the EQ-5D utility is a reflection of how society values the patient's health state, and the EQ-VAS is a reflection of how patients value their own health state.

Social participation was assessed by questions on productivity as well as unpaid activities. A self-developed questionnaire measured paid labor (e.g., hours of paid employment, hours of sick leave). Time spent on unpaid tasks, chores, leisure, and social activities in the past 2 months was measured by an adapted activity questionnaire based on the Utrechtse Activiteiten Lijst^{35,36}, a Dutch adaptation of the Craig Handicap Assessment Rating Technique³⁷. The unpaid participation was divided into unpaid tasks and chores (hours per week spent on study, housekeeping, odd jobs around the house, and voluntary work) and leisure and social activities (hours per week spent on sports, club life, social activities, and other leisure activities).

Functional status was measured with the Fibromyalgia Impact Questionnaire (FIQ), a self-administered 10-item instrument that measures

physical functioning, number of days felt well, number of days unable to work because of FM symptoms, work difficulty, pain, fatigue, morning tiredness, stiffness, anxiety, and depression³⁸. All items of the FIQ were standardized on a scale ranging from 0 to 10, with 10 indicating greater impairment. The total FIQ score was calculated by adding up the 10 items (range 0–100)³⁹.

Fatigue was assessed by the Checklist Individual Strength (CIS-20), a 20-item self-report questionnaire⁴⁰. Each item was scored on a 7-point Likert scale, and a CIS total score is calculated by adding up the score of the 20 items (range 20–140). Higher scores indicate more problems.

The Generalized Self-Efficacy Scale is a 10-item scale designed to assess optimistic self-beliefs to cope with a variety of difficult demands in life⁴¹. Responses are made on a 4-point scale and are summed to yield the final score, ranging from 10 to 40, with 10 indicating lower self-efficacy.

Cost assessment. Costs during the 9-month followup period were assessed

from the societal perspective, including healthcare consumption, patient and family costs, and productivity costs. Two monthly cost diaries [t1 (before randomization) – t6] completed by the patients were used to estimate healthcare costs (e.g., consultations, medication, home care) and non-healthcare costs (e.g., home help, informal care, medical aids, health activities, and productivity costs). Table 1 shows the cost items defined and the price value used. Prices were generally obtained from Dutch standard prices that were defined to reflect societal costs and to standardize economic evaluations⁴². The calculation of costs of the diagnostic process has been described in detail²⁴. We used the tariffs of the Dutch National Health Tariffs Authority of 2006. Overhead costs were not taken into consideration.

All costs were presented in 2007 prices and inflated where appropriate, using the general Dutch consumer price index rate (Centraal Bureau voor de Statistiek, Den Haag, Netherlands; website: www.cbs.nl). The costs of the diagnostic process were published in 2006 prices²⁴ and were also

Table 1. Costs per unit by categories, and sources of cost estimates.

Cost Categories	Source of Estimate	Cost per Unit, € (2007)
Hospital		
Diagnostic process of fibromyalgia	Dutch National Health Tariffs authority ²⁴	Various
Outpatient	Oostenbrink ⁴²	110.83/visit ^a
Specialized nurse	Oostenbrink ⁴²	84.38/visit ^a
General practitioner		
Practice	Oostenbrink ⁴²	21.98/contact ^b
Home	Oostenbrink ⁴²	42.75/contact
Telephone	Oostenbrink ⁴²	10.69/contact
Out-of-hours services (practice)	Dutch National Health Tariffs Authority	65.02/contact ^a
Out-of-hours services (telephone)	Dutch National Health Tariffs Authority	25.00/contact
Healthcare professionals		
Physical therapist	Oostenbrink ⁴²	24.68/contact ^b
Mensendieck and Cesar therapy	Oostenbrink ⁴²	24.95/contact ^b
Occupational therapist	Oostenbrink ⁴²	24.95/contact ^b
Psychologist	Oostenbrink ⁴²	76.31/contact ^b
Social worker	Dutch National Health Tariffs Authority	55.61/contact ^b
Activity therapy	Dutch National Health Tariffs Authority	55.61/contact ^b
Dietician	Oostenbrink ⁴²	27.06/contact ^b
Alternative medicine	Patient-reported costs	Various ^b
Medication		
Prescribed drugs	Pharmacotherapeutic compass 2007 ⁶³	Various/DDD
OTC drugs	Patient-reported costs	Various
Day care	Oostenbrink ⁴²	128.03/day
Professional domestic care	Oostenbrink ⁴²	22.96/h
Informal care		
Various	Oostenbrink ⁴²	8.78/h
Paid housekeeper	Oostenbrink ⁴²	8.78/h
Meal service	Estimated market price	7.00/day
Expenses for health activities	Patient-reported costs	Various
Expenses for medical aids	Patient-reported costs or estimated market price ^c	Various
Productivity costs	Oostenbrink ⁴²	Men
	Standard hourly wage by age for mean and women	15–24 yrs: €20.49
		25–34 yrs: €32.74
		35–44 yrs: €40.86
		45–54 yrs: €45.37
		55–65 yrs: €47.82
		Women
		15–24 yrs: €20.07
		25–34 yrs: €29.88
		35–44 yrs: €33.60
		45–54 yrs: €34.21
		55–65 yrs: €36.41

^a Including €5.02 travel expenses (mean distance to hospital 7 km): $14 \times 0.17/\text{km} + €2.65$ (parking)⁴²; ^b Including €0.61 travel expenses (mean distance to GP/PT 1.8 km): $3.6 \times 0.17/\text{km}$; ^c by various websites, e.g., www.thuiszorgwinkel, www.medireva.nl. OTC: over the counter; DDD: daily defined dose.

indexed. For 8 patients in the RMT group, costs for the diagnostic process could not be calculated because they canceled their appointment. Therefore, the mean costs of the diagnostic process of the remaining 88 patients were used.

Productivity costs were calculated by using the human capital limited approach (HClim). This approach estimates the value of all potentially lost production, in contrast with the friction cost method, in which productivity costs are only counted as long as it takes to replace someone^{43,44}. The difference between the HClim and the HC extended approach (HCext) is that in HClim, disease-related work disability at baseline is not taken into account, while in HCext, work disability at baseline is included in the estimation of productivity costs⁴⁵. We chose HClim because FM leads to disease-related work disability for a substantial number of patients. In the cost diary, patients reported their official working hours per week and the number of days and hours of absenteeism. Because FM is highly prevalent in middle-aged women, paid work was valued at age-dependent and sex-dependent standard hourly costs, ranging from €17 to €41 per hour (including 80% production elasticity)⁴². Costs associated with paid labor were calculated for each patient as the difference between the official working hours reported and the number of hours actually worked, valued at the patient's value per hour.

Sample size. The power calculation was based upon the acceptability of a 3-month waiting time for a first visit led by a rheumatologist, as described²⁴.

Statistical analysis. All analyses were conducted on the basis of intention-to-treat. Descriptive statistics were used for demographic and clinical variables and included percentages, means, and standard deviations.

Because of the rather large number of missing values in the cost and social participation data at t3 to t5 (31%–42%), we used a nonparametric multiple imputation method, which replaces each missing value with a set of “m” plausible values to generate 5 replacement values ($m = 5$) for each of the missing cells in these datasets, using multiple linear regression models. Means presented for cost and social participation are an average of the means from the 5 datasets created.

Average total costs were calculated for patients in each group. Given that cost data are often positively skewed, a nonparametric bootstrap method was used to obtain uncertainty intervals for the mean differences in costs⁴⁶.

Because of the random assignment, differences in health outcome at baseline were considered to occur by chance⁴⁷. For consistency, health outcome data at 9 months of followup were treated in the same way as the cost data: a multiple imputation method was used to generate 5 replacement values for each of the missing cells and the nonparametric bootstrap was applied to calculate uncertainty intervals.

Statistical analysis was performed using SPSS 17.0 (SPSS, Chicago, IL, USA). Bootstrapping was performed using Excel.

RESULTS

Patient characteristics. A total of 193 patients were randomized (97 to the SRN group and 96 to the UC group). Comparison of patient characteristics showed no meaningful differences between the SRN and the UC group. The majority of the patients were women. The mean (SD) age was 44.1 (11.1) and 44.7 (11.9) years in the SRN and UC groups, respectively. The mean (SD) duration of complaints at presentation was 6.1 (7.2) years in the SRN group and 5.7 (6.8) years in the UC group. Most patients (86% in the SRN and 79% in the UC group) indicated additional health problems.

Resource use 2 months prior to randomization is described in Table 2. The UC group reported more GP con-

tacts (3.3 vs 2.5), but fewer physical therapy consultations (1.8 vs 2.9). A majority of the patients used prescribed medications (58.8% in the SRN and 52.1% in the UC group). Mean costs for prescribed medications were €34.1 in the SRN group and €25.5 in the UC group. Use of over-the-counter medications was higher in the SRN group (47.4% vs 39.6%). Costs for (paid and unpaid) help and medical aids were higher in the UC group (help: €62 vs €34; medical aids: €25.0 vs €3.6). In the SRN group, more money was spent on health activities (€20.7 vs €10.4).

More patients in the SRN group had a job (47.4% vs 36.8%), especially a full-time job (22.7% vs 11.7%) and fewer of those patients received a disability insurance benefit (22.7% vs 34.7%). The percentage of patients with sick pay was 21.6% in the SRN group and 25.0% in the UC group. The SRN group had more contractual hours (15.1 vs 11.7 per week), worked a higher number of hours (12.2 vs 8.2 per week), and reported a lower number of sick leave hours during the 2 months prior to randomization (25.4 vs 35.2).

Health outcomes. Table 3 shows the results on waiting time and patient satisfaction of the 2 approaches, as published²⁴. The mean waiting time after randomization was 2.8 weeks in the SRN group and 12.1 weeks in the UC group ($p \leq 0.0001$). In the UC group, 8 patients canceled their appointments because of a too-long waiting time. Patients in the SRN group were significantly more satisfied than patients in the UC group with regard to nearly all items.

The changes in health status during the 9 months of followup are presented in Table 4. The UC group scored lower than the SRN group at baseline and at 9 months of followup on nearly all health outcomes. The improvements are fairly similar across the 2 groups.

Healthcare and productivity costs. The costs of the diagnostic process have been published²⁴ and are presented in Table 3. Mean total costs of the diagnostic process were lower in the SRN group than in the RMT group (€219 per patient vs €282 per patient; 95% uncertainty interval €–103 to €–20).

The use of healthcare resources during the 9 months of followup is summarized in Table 2. The resource use is generally higher in the UC group. The UC group reported significantly more contacts with GP, medical specialists, and other paramedical professionals and significantly more hours of paid housekeeping help.

Table 5 shows a comparison of paid and unpaid activities between the SRN group and the UC group. After 9 months of followup, a slight decrease in contractual hours and a small increase in actually worked hours is seen in both groups. Also, a substantial decrease in sick leave is observed, especially in the UC group. The decrease of absenteeism in the UC group occurred only in the last month of followup, resulting in higher costs for absenteeism in this group (€–1109; 95% uncertainty interval €–3581 to €–1094).

Table 2. Average resource use per patient 2 months before baseline and during 9 months' followup (mean) and difference in resource use during 9 months' followup between UC group and SRN group. Boldface type indicates significant difference.

Cost Components	UC Group, n = 96		SRN Group, n = 97		Difference (95% UI) Resource Used During 9 Mo Followup
	2 Mo Before Baseline	During 9 Mo Followup	2 Mo Before Baseline	During 9 Mo Followup	
Healthcare consumption					
Total GP contacts	3.3	6.1	2.5	4.6	-1.5 (-2.6 to -0.2)
Outpatient specialist care contacts	0.6	0.8	0.3	0.5	-0.3 (-0.6 to 0.0)
Physical therapy contacts	1.8	9.8	2.9	8.0	-1.8 (-4.6 to 1.4)
Psychological therapy contacts	0.4	1.8	0.4	1.7	-0.1 (-1.1 to 1.0)
Other therapy contacts	0.5	3.4	0.3	1.9	-1.5 (-2.9 to -0.3)
Multidisciplinary day care (no. contacts)	0.0	0.9	0.0	0.6	-0.3 (-1.4 to 0.6)
Home help (h/wk)	0.1	0.7	0.1	0.8	0.1 (-0.5 to 0.7)
Patient and family costs					
Paid housekeeping help (h/wk)	0.3	2.7	0.2	1.5	-1.2 (-2.5 to -0.1)
Unpaid help from family/friends (h/wk)	0.2	4.2	0.0	2.5	-1.7 (-4.1 to 0.4)
Meal provision (no. meals)	0.0	0.5	0.3	1.7	1.2 (-0.1 to 3.3)

UC: usual care; SRN: specialized rheumatology nurses; UI: uncertainty interval based on bootstrap replications; GP: general practitioner.

Table 3. Cost consequence of diagnostic procedure (means): comparison between UC group and SRN group. Boldface type indicates significant difference.

Cost Components	Usual Care, n = 96*		SRN Group, n = 97		Difference (95% UI**)
	Units	Costs (€)	Units	Costs (€)	
Waiting time, wks	12.1		2.8		-9.3 (-10.0 to -8.4)
No. "no shows"	8		0		-8
No. FM diagnoses (%)	66 (75)	n = 88	89 (92)		23 (17)
	70 (73)	n = 96			19 (19)
		n = 88			
Total consultations (RMT and/or SRN)	3.1		2.5		-0.6 (-1.1 to 0.0)
Total length of consultation (min)	124		144		21 (-2 to 42)
Costs of consultations with RMT and/or SRN		210		162	-48 (-75 to -22)
Costs of blood tests		23		44	21 (12 to 30)
Costs of function tests		48		14	-34 (-54 to -13)
Total costs		281		219	-62 (-103 to -20)
Patient satisfaction***					
	n = 85		n = 94		
Take seriously	0.17		0.00		-0.17 (-0.47 to 0.00)
Know problems very well	1.86		1.58		-0.28 (-1.29 to 0.71)
Take enough time for me	0.74		0.00		-0.74 (-1.26 to -0.33)
Take care that I can tell story	0.95		0.11		-0.84 (-1.52 to -0.29)
Tell findings at the end of consultation	0.85		0.25		-0.60 (-1.30 to 0.01)
Pay attention to (psycho)social aspects of illness	2.42		0.48		-1.94 (-2.71 to -1.19)
Give clear information about disorder	1.47		0.24		-1.23 (-1.99 to -0.49)
Give useful advice	4.96		1.59		-3.37 (-4.53 to -2.23)
Acceptable waiting time	1.54		0.20		-1.34 (-1.74 to -0.95)

* 8 patients canceled the diagnostic consultation at the outpatient clinic. ** UI: uncertainty interval based on bootstrap replications. *** Quality effect by QUOTE: Quality of Care Through the Patient's Eyes measurement instrument. Higher values indicate less satisfaction; values ≥ 1 reflect care aspects that could be improved. UC: usual care; SRN: specialized rheumatology nurse; RMT: rheumatologist.

Table 4. Health outcome measurements after 9 months' followup per patient. Boldface type indicates significant difference.

Outcome Measure	UC Group, n = 96			SRN Group, n = 97		
	Baseline	9 Mo Followup	95% UI*	Baseline	9 Mo Followup	95% UI*
EQ-5D UK tariff	0.35	0.43	0.08 (−0.01 to 0.16)	0.43	0.49	0.06 (−0.03 to 0.14)
EQ-5D Dutch tariff	0.43	0.47	0.04 (−0.05 to 0.13)	0.51	0.54	0.03 (−0.05 to 0.11)
EQ-VAS	47.7	47.4	−0.3 (−5.0 to 4.4)	49.2	49.8	0.6 (−4.6 to 5.6)
FIQ total	59.9	58.4	−1.5 (−5.9 to 3.0)	57.0	56.1	−0.9 (−5.4 to 3.4)
CIS-20 total	99.0	93.9	−5.1 (−11.1 to 1.4)	90.4	90.7	0.3 (−6.5 to 7.1)
CIS-20 fatigue severity	47.3	42.5	−4.8 (−7.0 to −2.4)	44.4	41.9	−2.5 (−5.4 to 0.2)
Self-efficacy	26.5	26.2	−0.3 (−1.6 to 1.0)	27.4	26.7	−0.7 (−2.1 to 0.8)

* UI: uncertainty interval based on bootstrap replications. UC: usual care; SRN: specialized rheumatology nurses; EQ-VAS: EuroQol visual analog scale; FIQ: Fibromyalgia Impact Questionnaire; CIS-20: Checklist Individual Strength.

Table 5. Amount of paid and unpaid participation per patient (mean h/week): comparison between UC group and SRN group.

Outcome Measure	UC Group, n = 96			SRN Group, n = 97		
	Baseline	9 Mo Followup	95% UI*	Baseline	9 Mo Followup	95% UI*
Official working hours	11.9	10.4	−1.5 (−5.4 to 2.6)	15.2	14.3	−0.9 (−5.1 to 3.3)
Hours actually worked	8.3	10.4	2.1 (−1.3 to 5.8)	12.2	13.3	1.1 (−3.0 to 5.2)
Absent hours (mean, last 2 mo)	36.6	9.4	−27.2 (−44.6 to −11.6)	26.1	13.7	12.4 (−26.8 to 1.0)
Absent hours (HClim; sum 9 mo followup)		99.5			76.1	−23.4 (−86.6 to 32.2)
Cost of absenteeism from work (HClim; €)		3674			2565	−1109 (−3581 to 1094)
Unpaid participation						
Education	1.6	2.3	0.7 (−1.1 to 2.4)	1.0	1.8	0.8 (−0.4 to 2.1)
Housekeeping	23.4	22.2	−1.2 (−7.4 to 4.5)	23.0	20.5	−2.5 (−8.1 to 2.9)
Odd jobs around the house	2.6	3.2	0.6 (−0.7 to 1.9)	3.1	4.0	0.9 (−0.5 to 2.3)
Volunteer work	1.0	1.6	0.6 (−0.4 to 1.6)	0.5	1.0	0.5 (0.0 to 1.0)
Sport	1.5	2.2	0.7 (−0.1 to 1.3)	1.7	2.4	0.7 (0.0 to 1.5)
Club life	0.4	0.9	0.5 (0.1 to 0.9)	0.7	0.9	0.2 (−0.3 to 0.6)
Leisure time	7.0	7.0	0.0 (−2.3 to 2.1)	7.3	5.7	−1.6 (−3.2 to 0.2)
Social activities	2.9	3.6	0.7 (−0.1 to 1.7)	3.5	4.0	0.5 (−0.4 to 1.4)
Other activities	0.2	1.0	0.8 (0.0 to 2.1)	0.2	0.4	0.2 (−0.1 to 0.6)
Unpaid tasks and chores	28.7	29.2	0.5 (−5.5 to 6.8)	27.7	27.3	−0.4 (−5.8 to 5.0)
Leisure and social activities	11.9	13.8	1.9 (−0.9 to 4.6)	13.2	13.1	−0.1 (−2.4 to 2.2)

* UI: uncertainty interval based on bootstrap replications. UC: usual care; SRN: specialized rheumatology nurses; HClim: human capital limited approach.

Concerning unpaid activities, the UC group spent, in contrast to the SRN group, slightly more hours on unpaid tasks and chores and leisure and social activities.

Table 6 presents the mean healthcare and societal costs per patient during 9 months of followup. Cost differences were nearly all in favor of the SRN group. Costs of the diagnostic process and GP contacts were significantly lower in the SRN group compared to the UC group. Expenses for health activities, conversely, were higher in the SRN group compared to the UC group. Total costs excluding absenteeism were significantly lower in the SRN group (€1298 vs €1644; 95% uncertainty interval €−746 to €−2). Mean total costs from a societal perspective were €3853 per patient for the SRN group and €5293 for the UC group after 9 months of followup. Productivity costs accounted for two-thirds of the total societal costs and had a big effect on the incremen-

tal costs. The mean cost difference was €1440 per patient (95% uncertainty interval €−3721 to €577) in favor of the SRN group.

DISCUSSION

We investigated the health and economic consequences of substituting SRN for RMT in the diagnostic process of FM. Patients in the SRN group were significantly more satisfied, but differences in health status between the 2 groups during 9 months of followup were small and insignificant. Mean total costs from a societal perspective (including absenteeism) were €1440 per patient lower in the SRN group than in the UC group after 9 months of followup (95% uncertainty interval €−3721 to €577).

We could not prove that a timely diagnosis had a positive effect on health outcomes and social participation in terms

Table 6. Mean costs of healthcare consumption during 9 months' followup per patient (€). Boldface indicates significant difference.

Cost Components	UC Group, n = 96	SRN Group, n = 97	Difference (95% UI)*
Healthcare consumption			
Total costs diagnostic process	287	223	-64 (-103 to -26)
Total GP contacts	123	83	-40 (-76 to -1)
Outpatient specialist care contacts (excluding contacts in Table 2)	103	63	-40 (-79 to 0)
Therapist contacts	436	355	-81 (-224 to 70)
Prescribed medications	148	115	-33 (-80 to 7)
OTC medications	30	24	-6 (-20 to 8)
Multidisciplinary day care	98	62	-36 (-169 to 82)
Patient and family costs			
Home help (paid/unpaid)	295	216	-79 (-276 to 109)
Meal provision	15	16	1 (-12 to 20)
Expenses for health activities	82	118	36 (-6 to 86)
Expenses for medical aids	19	19	0 (-20 to 23)
Total costs (excluding absenteeism from work)			
Absenteeism from work (HClim)	3674	2565	-1109 (-3581 to 1094)
Total societal costs	5293**	3853**	-1440 (-3721 to 577)

* UI: uncertainty interval based on bootstrap replications. ** Total costs deviate from the sum of cost items because of bootstrap random variation. UC: usual care; SRN: specialized rheumatology nurses; GP: general practitioner; OTC: over the counter; HClim: human capital limited approach.

of hours spent. However, medical and productivity costs were significantly lower in the SRN group. Since the time-span of our analysis was only 9 months, a longer followup may be necessary to confirm these effects.

Our results suggest that healthcare use in terms of contacts with GP, medical specialists, and physical and psychological therapists and in medication costs was similar in both groups before and after diagnosis. The literature is not in agreement on the effects on healthcare use of making a diagnosis. Annemans, *et al* suggest that making a diagnosis leads to a decrease of resource use and costs, which is confirmed by other studies^{16,48,49}. However, Hughes, *et al* found that following diagnosis, visits for most symptoms and healthcare use markers declined, but within 2–3 years most visits rose to levels the same as or higher than those at diagnosis²¹. On the other hand, Maugars, *et al* described a decrease in referrals and tests after diagnosis, and an increase in drug use and GP visits for the first 2 years after diagnosis⁴⁸. White, *et al* illustrated that healthcare costs rose immediately after diagnosis⁵⁰. In our study, we saw an increase after diagnosis in contacts with other therapists, paid housekeeping help, informal care, multidisciplinary daycare, and expenses for health activities.

Notwithstanding a considerable decrease in sick leave hours in both intervention groups, productivity costs based on absenteeism accounted for two-thirds of the total societal costs in our study. This is much higher than described in pre-

vious studies, in which about 20%–33% of the total societal costs of FM are related to productivity costs^{10,14,51,52,53}. This could be due to the way that productivity costs were calculated. Huscher, *et al* showed that indirect costs differ by a factor of 3, based on whether the human capital approach or the friction costs approach is used⁵⁴.

The percentage of indirect costs (74% and 77% of the total societal costs for the SRN group and the RMT group, respectively) is in line with other studies¹⁰. In Boonen, *et al*⁵¹, also based on patient diaries, the annual direct medical costs were €1311 (2002 values), which is in line with our results converted to total annual costs (€1233 and €1633 in the SRN and the RMT groups, respectively). Also, the total annual costs per patient are comparable: €7813 (2002 values) in Boonen, *et al*⁵¹, €5137 (converted to annual costs) in the SRN group, and €7057 in the RMT group (converted to annual costs). Although Annemans, *et al*¹⁶ showed that the total costs for FM, reported by Boonen, *et al*⁵¹ and 12 other studies, are quite similar, a direct comparison of Dutch data with data from other countries should be done cautiously⁵⁵.

Our study has several difficulties. First, the choice for a cost-consequence analysis has disadvantages. Ideally, all outcomes should be integrated into 1 overall index of benefit. This is important when comparing different interventions or comparing the results of a study with other studies or other diseases in all facets. A cost-minimization analysis was not relevant in our study, as it assumes outcomes to be equivalent, which was not the case. In a cost-effectiveness analysis, consequences of programs are measured in the most appropriate natural effects or physical units, while in a cost-utility analysis, the consequences of interventions are adjusted by health state preference or utility weights. In our situation, with utility as one of the consequences and outcomes, we found no difference between improvement in quality of life in the intervention groups. Besides, these analyses, from a societal perspective, include all costs, irrespective of who bears them. A cost-benefit analysis measures costs in monetary terms as well as consequences of an intervention, which was not feasible for most of our consequences and therefore not a useful option.

In our study it is difficult to integrate the outcomes into 1 overall measure of benefit since they concern both consequences at the patient level (e.g., satisfaction, health status) and at the healthcare system level (e.g., waiting time, number of no-shows). A cost-consequence analysis overcomes this problem by presenting information on both costs and outcomes in a disaggregated form, allowing decision makers to make the necessary value judgments (implicitly weighing the relative importance of the outcomes) and tradeoffs that are relevant from their particular perspective. However, in our study costs from both healthcare and societal perspectives as well as consequences seem to point in the same direction, so we can formulate one overall conclusion and related recommendation.

Second, notwithstanding randomization, we found baseline differences for the EQ-5D index score, CIS-20, the percentage of employed patients, and hours of absenteeism. Concerning the between-group differences in work situation at baseline, costs of absenteeism at baseline were calculated and the difference was not found to be statistically significant. Adjustment for cost differences at baseline is not necessary in this situation, as the cost differences at baseline for other than productivity costs are very small and go in both directions. Besides, the recommendation of Van Asselt, *et al* for dealing with cost differences at baseline concerns baseline differences in total costs and not baseline differences in components of costs⁵⁶. However, the finding that productivity costs were €1109 lower in the SRN group should be interpreted cautiously. Using the most conservative adjustment, this amount should be reduced, with the difference in productivity costs at baseline (€445).

Third, some data were missing from our study. We have carefully checked whether these missing values were random. Patients with lower scores in HRQOL at baseline were generally more likely to be missing from the data at some point. Since patients with FM seem to have lower scores than patients who do not have FM, and the SRN group contained more patients with FM, the HRQOL scores of the SRN group could be an underestimation. Therefore, the incremental difference between the RMT group and the SRN group could also be a conservative difference. Although we also found more missing values in patients who did not have FM and therefore in the RMT group, we assume the effect on the scores is limited because of the small number of missing patients who did not have FM on the total of the group.

Fourth, an unknown number of patients in the RMT group underwent medical consultations in another hospital because of waiting time. The visits to RMT and SRN are, as far as reported, included in the costs of outpatient specialist care contacts, but we have no data on diagnostic tests performed and therefore those costs could not be included. This lack of information could have caused an underestimation of the medical consumption in the RMT group. This situation is closely connected with the next point. Nearly all cost data are based on cost diaries, which may not necessarily reflect actual patterns of use because of problems with patient recall. However, patient-reported healthcare consumption data are considered to be relatively reliable regarding formal care⁵⁷ and if biased, we do not expect systematic reporting differences between the groups in our study.

In addition, the estimated productivity costs did not include costs associated with reduced productivity on the job (presenteeism) or with replacement costs^{58,59}. In a future study, it is worth taking into account these costs because one of the consequences of FM is work loss, and those patients who do not lose their jobs still obviously experience difficulties in their working life as a direct result of FM^{60,61}.

Next, in our study we could not assess a possible difference in placebo effect of the nurse-led diagnostic process versus usual care. Within a cost-consequence analysis, however, the aim is to assess a difference in effectiveness, including a possible placebo effect instead of clinical efficacy as in clinical trials.

An important disadvantage of the nurse-led diagnostic process was the fact that more than 8% of the patients in the SRN group refused to participate because they indicated that they would only accept a rheumatologist consultation. However, given our analytic approach based upon intention-to-treat, this issue is incorporated in our findings and conclusion. This is a study based upon Dutch data, and the generalizability of our conclusion, and especially the issue of compliance to consultation by a nurse specialist, should be carefully considered, for instance by explicit assessment of transferability issues⁶².

Finally, given the incremental approach of our analysis, we did not consider overhead costs of hospital facilities; those were considered to be sunk costs, retrospective costs that have already been incurred and cannot be recovered. When an FM clinic is to be initiated, it is important to be aware that such costs might be relevant. One should note as well that our results were obtained in a trial setting and possibly can differ from real-life data, as is commonly the case in a protocolized trial. Therefore, we advise monitoring the outcomes after the implementation of such a nurse-led diagnostic process.

The nurse-led diagnostic process can be recommended from a healthcare and societal perspective. Patients in the SRN group were significantly more satisfied, and no differential changes in health status were observed between the 2 groups during 9 months of followup. Total healthcare consumption costs and patient and family costs were significantly lower in the SRN group. Also, costs from a societal perspective (including absenteeism) were lower in the SRN group compared to the RMT group.

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