Health Services Costs and Their Determinants in Women with Fibromyalgia

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ABSTRACT. ***Objective.*** Patients with fibromyalgia (FM) use health services extensively. Knowledge about costs of FM is limited because of non-inclusiveness in assessing direct costs, because attempts to assess indirect costs are largely absent, and because determinants of costs have yet to be identified. We investigated the 6-month costs (direct and indirect) in women with primary FM, and we identified determinants of direct costs.

Methods. Subjects (n = 180 women) completed a health resource questionnaire as well as measures of pain, psychological distress, comorbidity, and disability. Unit costs for resources were obtained from government, hospital, laboratory, and professional association sources. Regression modeling for 6-month direct cost included age, disability, comorbidity, pain intensity, psychological distress, education, and work status.

Results. The average 6-month direct cost was $CDN 2298 (SD 2303). The largest components were medications ($CDN 758; SD 654), complementary and alternative medicine (CAM; $CDN 398; SD 776), and diagnostic tests ($CDN 356; SD 580). Our most conservative estimate of average 6-month indirect cost was $CDN 5035 (SD 7439). Comorbidity and FM disability were statistically significant contributors to direct costs in the multivariate analysis. Costs increased by approximately 20% with each additional comorbid condition.

Conclusion. Women with FM are high consumers of both conventional and CAM services. Our estimates of costs exceed those from most other studies; this may be due to our inclusion of a broader set of health services, medications, and indirect costs. Although in univariate analyses the number of comorbidities and indices of the effect of FM, psychological distress, and pain intensity were associated with higher direct cost, in a multiple regression analysis, only the measure of FM disability and the number of comorbidities were significant direct-cost determinants. FM also imposes important indirect costs, which were nearly 70% of the economic burden. (J Rheumatol 2004;31:1391–8)

Key Indexing Terms:
FIBROMYALGIA DETERMINANTS HEALTH SERVICES COSTS

Fibromyalgia (FM), a disorder characterized by widespread pain and fatigue, has an estimated prevalence of 3.4% for women and 0.5% for men[1], and is a major cause of morbidity[2]. No clear diagnostic markers exist and no single treatment is consistently successful[3,4]. Clinical trials have suggested some benefit of numerous therapies, including exercise, electromyographic biofeedback, hypnotherapy, and electroacupuncture[1]. Medications, (antidepressants, anxiolytics, hypnotics, analgesics, and others) are commonly taken[5], but benefit only 30–50% of patients[6]. Longitudinal studies indicate FM symptoms remain stable over years, with a few studies showing minor improvements over time[7-9].

Patients with FM use health services extensively, incurring high costs[10-12]. However, the current literature on the cost of FM is deficient, because of the divergent and relatively non-inclusive means of assessing direct costs, and because attempts to assess indirect costs are largely absent. In assessing economic burden, both direct costs (resulting from health care use) and indirect costs (resulting from output loss due to cessation or reduction of both labor-market and non-labor-market productivity) must be incorporated[13]. Given that patients with FM are mostly female, a large portion of output loss will accrue from losses to non-labor-market activities such as housekeeping and child-care[14]. A final flaw in the existing literature is that although complementary and alternative medicine (CAM) use in FM is ubiquitous[15,16], the cost of these interventions has rarely been taken into account.

Our objectives were to describe comprehensively

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Supported by The Arthritis Society.

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Submitted March 6, 2003; revision accepted December 24, 2003.

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(including complementary care as well as conventional health services use) both direct and indirect costs of FM over a 6-month period, using a sample made up of both clinical and community subjects. An additional objective was to identify the determinants of direct costs in FM.

MATERIALS AND METHODS

Subjects and procedures. Two approaches were used in the recruitment of subjects. First, 10 rheumatologists working in hospitals and private practice settings were asked to identify subjects with FM who were scheduled for an office visit, and to invite them to participate in the study. Second, advertisements were run in English and French newspapers seeking women with widespread body pain and fatigue. This latter method included a structured telephone screening interview developed by White, *et al*[2] that identified subjects most likely to have FM. Those who screened positive were examined by a rheumatologist to confirm the diagnosis according to American College of Rheumatology criteria. The research protocol was approved by the McGill University Faculty of Medicine Institutional Review Board and by hospitals not affiliated with McGill University.

Eligibility criteria included age 18 years or older, a diagnosis of primary FM, and fluency in English or French. Written informed consent was obtained. Subjects completed questionnaires within 72 hours after the initial visit with the physician (baseline); demographic, clinical, and psychosocial data were obtained then. In a second set of questionnaires mailed out 2 weeks later, data on family income and health care utilization during the preceding 6 months were obtained.

Costs. The Cost Assessment Questionnaire (CAQ), a version of the Stanford Health Assessment Questionnaire (HAQ), modified for the Canadian health system context and for FM, was used to collect data on health service use. It has been validated[17-20] for various rheumatic diseases. It inquires about the use of all health services during the preceding 6 months without asking the respondent to make attributions to any one disease or condition. Subjects reported on outpatient use of physicians, laboratory tests, imaging procedures, medications, emergency room (ER) visits, outpatient surgery, inpatient stays, all medications (prescription and nonprescription), and complementary and alternative medicine (CAM) services. The latter category included health services of the professional (rather than popular or folk) sector[21]: psychologists, chiropractors, ergonomists, physiotherapists, occupational therapists, podiatrists, acupuncturists, massage therapists, naturopaths, osteopaths, homeopaths, and dieticians.

Annual direct cost estimates were calculated by multiplying health service utilization levels by the appropriate unit prices. Data are expressed in 2001 Canadian dollars. At the purchasing power parity exchange rate, 1.2 Canadian dollars equal one US dollar[22]. In cases where 2001 costs data were not available, the most recent available data were converted according to the health component of the consumer price index. To develop unit costs, we used methods previously employed by ourselves and others[17,19,20,23]. The prices for physician services, and the technical and professional component of outpatient laboratory tests, imaging procedures, and assistive devices were assigned according to provincial reimbursement for these services when provided in private offices. When provincial reimbursement data for private office laboratory tests or imaging procedures were not available, we based the costs on charge data from private laboratories. Data on the cost of outpatient visits to nonphysician health professionals (nurses) were obtained from the public outpatient health clinic closest to the Montreal General Hospital (CLSC Metro) and the Ordre des infirmières du Québec. To estimate the cost of other CAM services, we obtained recommended or usual prices from the following relevant professional associations: Collège des naturopathes du Québec, Ordre des naturothérapeutes du Québec, Association des acupuncteurs du Québec, Ordre des podiatres du Québec, Association des massothérapeutes professionels du Québec, Société des ostéopathes du Québec, Association des chiropracticiens du Québec, Ordre des psychologues du Québec, Ordre professionnel des physiothérapeutes du Québec, Association des ergothérapeutes en pratique privée du Québec, Ordre professionnel des travailleurs sociaux, and the Ordre professionnel des diététistes du Québec.

Costing of medical equipment was done using prices suggested by the provincial insurance agency, the Quebec association of medical technologies, device manufacturers, and other sources. Acute care hospital costs were estimated according to the Canadian Institute for Health Information[24], which assigns costs based on the Case Mix Group of the admission. Costs of inpatient physician services were based on provincial reimbursement schedules and were counted in the inpatient category. Ambulance service costs were based on data provided by Urgence-Santé of Montreal. The nonphysician ER costs were based on the last available data from Statistics Canada on the nonphysician diagnostic and therapeutic costs of an ER visit as well as data on the percentage of diagnostic and therapeutic costs as a percentage of total nonphysician ER visit costs in an urban hospital[25,26]. Physician ER visit costs were based on provincial reimbursement schedules and were counted as part of the ER costs rather than under the physician visit category.

We calculated the cost of prescription medications, using the data we had recorded of the subjects' medication consumption according to 11 drug classes. We derived a weighted average of the cost of that drug class using data from the Intercontinental Medical Statistics Compuscript Audit; the weights that we used represented the frequency at which specific drugs within that class are prescribed in Quebec. These data are based on a sample of retail prices, so they are inclusive of the wholesale price as well as any pharmacy markups and dispensing fees.

Indirect costs were calculated for productive (market work or nonmarket work) time lost by our subjects, as identified in the CAQ. The opportunity cost method was used to estimate the value of lost time in market work[27]. Though non-paid household work has economic value, a "wage" to value this time is not observed. To assign values to unpaid work losses, we used the replacement cost method[28,29], which provides a conservative estimate of these costs and is based on the compensation of persons working for pay in a variety of household activities. We have previously described the implications of applying 5 commonly used methods of calculating the indirect costs of illnesses affecting primarily women[14].

Putative Predictors of Direct Costs

Disability was measured by the Fibromyalgia Impact Questionnaire (FIQ)[30], a validated self-report measure of physical functioning, work status, psychological distress, pain, stiffness, fatigue, and well-being within the past week. FIQ total scores range from 0 to 100, with higher scores indicating greater disability.

Present pain intensity (PPI) was assessed using the McGill Pain Questionnaire[31], in which subjects described their present pain according to the scale 0 (no pain) to 5 (excruciating). For the analyses, this variable was dichotomized to represent the presence or absence of high pain intensity (corresponding to a score $\geq$ 4).

Comorbidity. Subjects indicated the presence of other medical conditions; in total, 44 distinct conditions were reported. We constructed a categorical variable, ranging from 0 (no other condition) to 5 ($\geq$ 5 conditions); 14 subjects indicated 5 or more conditions. Analyses assumed linear effects across the 6 values.

Psychological distress. The Symptom Checklist-90-R, a validated measure of psychological symptoms during the past week[32], was completed by the subjects. Symptom clusters include somatization, obsessive-compulsion, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, and psychoticism. Items are combined in a Global Severity Index. Clinically important psychological distress corresponds to T scores $\geq$ 63[32], and this cutoff was used to dichotomize the variable in analyses.

Statistical analyses. For a descriptive analysis of subjects, we calculated means (or proportions, where appropriate) and standard deviations of relevant demographic and socioeconomic determinants (age, years of educa-

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tion, family income, and work status) as well as health-related variables (comorbidity, disability, pain intensity, and clinically important distress).

We then described the costs of specific resources such as physician visits, hospital stays, indirect costs, etc. Next, we estimated an aggregated model where total direct costs were modeled as a function of subject characteristics. The unadjusted coefficients and the adjusted coefficients of these variables were based on models using the natural logarithm of the dependent variable, since a logarithmic transformation of the total direct cost data more accurately reflected a normal distribution. Independent variables for our analyses included age, years of education, and work status (whether the subject worked during the study period), as well as the health-related variables listed above.

The planned sample size (160 to 200 patients) ensured adequate statistical power at ≥ 80% to detect the adjusted effect of a putative determinant that accounts for at least 8% of the total variance in health care utilization scores, at the corrected 0.01 level of significance. Statistical analyses were performed with SPSS for Windows 9.0 software and Stata™ 6.0 software.

RESULTS

Subjects. Among the community subsample who screened positive in the telephone interview, about one-third failed to appear for their medical examination to confirm diagnosis of FM. Among those examined (n = 104), 95% (n = 99) received a diagnosis of FM. Among the tertiary subsample (n = 106), 85.8% (n = 91) agreed to participate after being informed about the study. From this initial sample of 190 participants, 4.2% (n = 8) dropped out at baseline. At the 2-week followup period, 1% (n = 2) failed to return their questionnaire package, leaving a working sample of 180.

Rheumatologists recruited 91 (45.5%) of the subjects, constituting the tertiary care subsample. We examined the possibility that this subsample might differ from their 97 counterparts recruited from the community (54.5%) on outcomes and predictors. Independent t tests and chi-square tests revealed no statistically significant differences between the 2 subsamples in terms of outcomes and predictor variables; data were thus collapsed across the subsamples for all subsequent analyses.

Many of the subjects were French-speaking (57.3%), Caucasian (88.2%), and married (56.7%). Table 1 summarizes standard statistics for outcome and predictor variables. The median age was 51 years (range 21–78). Forty-two percent of subjects worked in the previous 6 months, and 25% were receiving disability assistance or retired because of FM, implying a desired labor force participation ratio of 67%. The mean and median for other comorbid conditions was 2. The mean and median FIQ scores for the sample (57.74 and 58.46, respectively) indicated significant FM-related disability. When asked to rate their present pain intensity, 14 patients (7.9%) reported no or mild pain, 59 (33.1%) described their pain as discomforting, 54 (30.3%) as distressing, and 51 (28.7%) as horrible or excruciating.

Health services utilization. The health services utilization data (Table 2) revealed that subjects reported, on average, 7 visits to physicians and 7 visits to CAM service providers during the preceding 6 months. The vast majority of subjects reported visits to both physicians and CAM providers in the preceding 6 months.

Table 1. Health, demographic, and socioeconomic variables (n = 180).

Variable	Mean ± SD (range)
Demographic	
Age, yrs	50.78 ± 10.22 (20.60, 78.7)
Education, yrs	12.97 ± 3.33 (3, 18)
Family income < $CDN 30,000*, %	42
Family income > $CDN 50,000*, %	33
Worked during study period, %	42
Worked before FM diagnosis, %	78
On disability because of FM, %	16
Retired because of FM, %	9
Health status	
No. of comorbid conditions	2.10 ± 1.37 (0, 5)
Disability (FIQ)**	57.74 ± 16.50 (0.50, 92.51)
High pain intensity†, %	29
Psychological distress††, %	72

* Data available on 168 subjects. Median family income corresponded to the income bracket $CDN 30,000–40,000. ** Fibromyalgia Impact Questionnaire (179 responses). † High pain corresponds to 4 or 5 on the McGill Pain Questionnaire (179 responses). †† Global Severity Index ≥ 63, Symptom Checklist-90-R (% of subjects classified as cases).

Table 2. Health services utilization in the 6-month study period (n = 180).

Variable	Mean ± SD (range)
All physician visits	7.12 ± 4.73 (0, 30)
General practitioner	3.29 ± 2.96 (1, 18)
Specialist	3.83 ± 3.53 (0, 30)
Complementary and alternative medicine visits	7.09 ± 13.48 (0, 84)
Imaging and laboratory procedures	4.56 ± 4.49 (0, 28)
One or more hospitalizations, acute or longterm	0.08 ± 0.27 (0, 1)
One or more outpatient surgeries or procedures	0.01 ± 0.30 (0, 1)
No. of medications taken	6.98 ± 3.64 (0, 16)
Drug doses per day in study	6.63 ± 5.32 (0, 26)

The average total 6-month direct costs (Table 3) of health services was $CDN 2298 (SD 2303). The largest components of costs were for medications ($CDN 758, SD 654), CAM services ($CDN 398, SD 776), imaging and laboratory ($CDN 356, SD 580), physician visits ($CDN 348, SD 264), and inpatient stays ($CDN 300, SD 1539).

Our conservative estimate of indirect costs (Table 3) was $CDN 5035 (SD 7439) for the 6-month period. Market work accounted for 78% of this ($3936 ± 6630). Of note, only 42% of our subjects were working women; 22% of the sample said their work status was retired (9%) or disabled (16%) due to FM. Over half the working women in our sample missed some work because of FM (average 77 hours, over 6 months, or about 4 weeks annually). Adding to this loss of work time due to women exiting the work force due to FM, the 6-month work losses for the total sample were 251 hours, or 12.5 weeks annually. An additional 80

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Table 3. Direct, indirect, and total costs reported over 6 months ($CDN).

Variable	Mean ± SD (range)
Direct costs	2298 ± 2303 (0, 16,566)
Physician visits	348 ± 264 (0, 2078)
Complementary and alternative medicine visits	398 ± 776 (0, 4600)
Imaging and laboratory procedures	356 ± 580 (0, 5694)
Inpatient stays, acute or longterm	300 ± 1539 (0, 16,566)
Outpatient procedures	62 ± 229 (0, 1939)
Medications	758 ± 654 (0, 3892)
Other health services*	77 ± 150 (0, 826)
Indirect costs	5034 ± 7439 (0, 37,519)
Market work**	3936 ± 6630 (0, 30,635)
Nonmarket (household) work***	1098 ± 1911 (0, 13,567)
Total costs (direct and indirect)	7333 ± 8053 (31, 40,320)

* Other health services include emergency room visits, ambulance services, and equipment. ** Market work costs are based on the average wages of adult Canadian women. *** Nonmarket costs are based on the replacement cost method.

hours of household work were lost over the 6 months because of FM limitations.

The results of our examination of the determinants of direct costs are presented in Table 4. The first column gives the unadjusted estimates of the relationship between the candidate variables and the natural logarithm of direct costs. Since the dependent variable is expressed as a natural logarithm, the estimated coefficient can be interpreted as an approximation of the percentage change in direct cost with a unit change in the independent variable. For example, the unadjusted estimate of the effect of the FIQ score indicates that direct costs increase roughly 1.7% with each additional FIQ point. The unadjusted coefficients show an economically and statistically significant relationship between direct costs and the number of comorbid conditions, the FIQ score, the presence of high pain intensity, and psychological distress, and the effects are in the expected direction (poorer health is associated with higher expenses). The estimated unadjusted coefficients of ≥ 16 years of education and work status variables were economically significant, but the confidence intervals are large and cover zero; the magnitudes of their univariate associations with direct expenses remain to be determined in a larger study. In the multiple regression analysis (Table 4, second column), of all the variables included, only comorbidity ($p < 0.001$) and FIQ score ($p = 0.024$) were statistically significant contributors to direct costs. Since the dependent variable is the natural logarithm of direct costs, the estimate of the comorbidity coefficient indicates that costs increase by roughly 20% with each additional comorbid condition. Whether this increase in costs is due to the treatment costs associated uniquely with the comorbid conditions or whether the FM severity increases with these comorbid conditions cannot be separated out unless FM severity is perfectly controlled for. Therefore, the coefficient on comorbidity must be interpreted as a composite effect of increasing costs of the comorbid conditions themselves and the possible increasing severity of FM accompanying these comorbidities. Although in the univariate analyses work status, pain intensity, and psychological distress were significantly associated with the natural logarithm of direct costs, they were not statistically significant in the multivariate regression. Our power calculations for the multivariate regression were valid for variables that explained at least 8% of the variance of the logarithm of costs in univariate analyses. Comorbidity and FIQ score exceeded this limit; however, work status, pain intensity, and psychological distress explained only 5%, 4%, and 3%, respectively, of the overall variance in the logarithm of costs. Therefore, a larger study is required to examine the effects of these variables with adequate power. Given their importance in explaining direct costs, we describe in Table 5 the most common comorbidities reported by subjects in our sample.

Table 4. Ordinary least-squares regressions of the logarithm of direct costs (n = 178).

Variable	Unadjusted Estimates Coefficient (95% CI)	Multiple Regression Estimates Coefficient (95% CI)
Age	–0.0014 (–0.014, 0.011)	–0.008 (–0.020, 0.042)
16 or more years of education	–0.13 (–0.43, 0.16)	0.050 (–0.23, 0.33)
Worked during study period	–0.32 (–0.58, –0.068)*	–0.17 (–0.42, 0.083)
No. of comorbid conditions	0.24 (0.15, 0.32)*	0.20 (0.11, 0.29)*
FIQ score	0.017 (0.0098, 0.023)*	0.011 (0.0014, 0.20)*
High pain intensity**	0.44 (0.16, 0.71)*	0.13 (–0.17, 0.42)
Psychological distress***	0.39 (0.11, 0.67)*	0.04 (–0.27, 0.35)
Constant	—	6.8 (6.0, 7.5)
R^2	—	0.23
Adjusted R^2	—	0.20

* Indicates statistical significance at the 0.05 level. ** High pain corresponds to 4 or 5 on the Present Pain Index of the McGill Pain Questionnaire. *** Global Severity Index ≥ 63, Symptom Checklist-90-R. FIQ: Fibromyalgia Impact Questionnaire.

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Table 5. Comorbidity in women with FM at baseline.

Comorbidity Prevalence in Sample	N	%
No comorbid condition	16	8.7
One condition	53	29.1
Two conditions	53	29.1
Three or more	60	33.1
Types of comorbidity*		
Medical	120	66.7
Osteoarthritis	72	40.0
Osteoporosis	4	2.2
Hypertension/vascular	18	10.0
Thyroid	9	5.0
Diabetes	6	3.3
Hypercholesterolemia	4	2.2
Anemia/hematological	6	3.3
Psychiatric	10	5.6
Depression	9	5.0
Anxiety	1	1.0
Functional	70	38.9
Irritable bowel	65	36.1
Chronic fatigue syndrome	4	2.2
Irritable bladder	1	1.0

* Some subjects had more than one comorbid condition. Other medical conditions included 4 with cardiac valve disease, 7 allergies, 4 gastrointestinal reflux disease, and 2 with cirrhosis. One each reported a sinus problem, a vision disability, a hearing disability, diverticulitis, a pituitary problem, skin cancer, psoriasis, Paget's disease, carnityle transferase deficiency, hypophosphatemia, hypoglycemia, bladder incontinence, and obesity.

DISCUSSION

Our findings substantiate previous work[11] describing that individuals with primary FM are heavy consumers of physician and CAM services. In our sample, women reported, on average, 7 visits to physicians during the preceding 6 months, 14 visits per year. This number was split almost equally between general practitioners and specialists. Fourteen visits contrasts strikingly with the mean number of annual physician visits in Quebec[34], which is documented at 3 visits per citizen (SD 0.8). Our results are somewhat higher than reports from other studies of health resource use in FM, particularly those based in the United States. Wolfe, *et al*[11] estimated from a large FM sample that the average was 10 physician visits per year; Alexander, *et al*[35] estimated the figure to be 11 outpatient visits per year (but this rate included both physicians and other providers). One may speculate that patients with access to universal health care use, such as the women in our study, use more physician services than those being treated within other types of health care systems. White's Canadian FM sample[36] (which included men and women) reported 12 physician visits per year; this approaches our estimate. Altogether, the finding of high frequencies of physician visits across different FM samples is consistent.

Our subjects reported, on average, 7 visits to CAM providers in the preceding 6 months. The vast majority of women who used these types of services visited both physicians and other health care providers. It is difficult to directly compare our results to those in the literature because of the heterogeneity of the definition of CAM services across studies. However, similar to our findings, subjects in Wolfe's study[11], when assessed for the use of "non-traditional" medical services (massage, acupressure, acupuncture, herbal therapy, and homeopathy), averaged 6 visits per 6-month period. Bombardier and Buchwald reported[37] that in their small sample (n = 28), FM patients had a mean of 26 visits per year to a wide variety of health-care providers, both physician and complementary (e.g., chiropractor, osteopath, naturopath, psychologist). Their figure is similar to our estimate for combined physician and complementary services (28 visits per year).

It is instructive to compare our estimated average annual direct costs ($CDN 4596, $US 3830 at purchasing power parity) to estimates from other FM samples shown in Table 6. The only other published Canadian estimates of the annual direct costs of FM[36] were substantially lower than our data. However, the previous Canadian study incorporated only the costs of physicians and other health professionals, laboratory tests, and imaging procedures. In our sample, the estimated annual cost of physician visits, laboratory tests, and imaging procedures was $CDN 1408. This figure is remarkably similar to the previous Canadian study[36] ($1082 in 1994 Canadian dollars, or $1243 in 2001 Canadian dollars), but represents only 31% of total direct FM costs, as estimated here.

In a study of the direct costs of FM in the United States, Wolfe's estimate of $US 2274 per year[11] ($2545 in 2001 US dollars) is somewhat lower than ours, possibly due to our inclusion of a broader range of services in costing health care. However, Oliver, *et al*[12], in a recently published study of 590 FM patients being treated in a health maintenance organization, calculated an average direct cost at $US 4570 per year, which is similar to our figure. Comparison between our work and Oliver, *et al* is especially pertinent as the average levels of disability were similar (FIQ Total 57.74 and 61.03, respectively). Some part of this difference may result from higher utilization of services by American patients or from higher unit costs of resources used by patients with FM. However, since the American study[12] describes the costs of the various categories of services and not the utilization of services, it is not possible to say how much of the difference is due to each of these factors.

As Table 3 indicates, the largest component of direct costs in our subjects was medications (an average of $CDN 758 over the study period, or $1518 per year). Wolfe, *et al*[11] reported medications as the second largest contributor to direct costs (an average of US$ 716 per year, or $981 in 2001 Canadian dollars). In both our sample and Wolfe's sample, medications represented one-third of the average total direct cost. The number of medications taken by indi-

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Table 6. Research pertaining to cost of healthcare services in FM.

Study	N	Services	Cost	Comments
Present study	180	Physicians; inpatient care; outpatient procedures; emergency; ambulance; equipment; CAM services; prescription/nonprescription drugs; imaging; laboratory tests	$CDN 4596/yr	2001 dollars, CAM = psychologists, chiropractors, physiotherapists, occupational therapists, podiatrists, acupuncturists, massage therapists, naturopaths
White[36]	100	Physicians; other health professionals covered by government insurance; imaging; laboratory tests	$CDN 1028/yr	1993 dollars ($CDN 1243 in 2001 dollars) Community-based cohort 1994 dollars
Wolfe[11]	538	Physicians; inpatient care; outpatient procedures; CAM services; prescription drugs; imaging, laboratory tests	$US 2274/yr ($CDN 2729*)	CAM = physiotherapy, occupational therapy, chiropractor, "nontraditional" Clinic cohort 1997 dollars
Oliver[12]	590	Physicians; inpatient care; emergency; mental health; nurse, technician and rehabilitation services; prescription drugs; radiology; laboratory tests	$US 4570/yr ($CDN 5484*)	CAM care not included HMO setting

* Conversion at the purchasing power parity exchange rate. CAM: Complementary and alternative medicine, HMO: health maintenance organization.

viduals with FM is remarkable: the average number of medications taken over the 6-month period was almost 7. Many of the women were taking various different courses of medications of a given type; for example, over half reported taking antidepressants at some point during the 6 months, and of that number, 42% had taken 2 different antidepressants, and a further 9% had taken 3 different antidepressants over the interval. (Use and adherence to medications in this sample have been reported[38].) The large number of medications taken by persons with FM likely reflects several phenomena, including (1) no successful therapy for FM is known, (2) FM can be very disabling, and (3) it is common for FM patients to suffer from comorbid conditions. Failure of first-line drug therapies appears to give way to a practice of trying first one then another medication, which seems to contribute significantly to the high cost of medications in FM.

This phenomenon of ineffective prescription medications may lead to seeking complementary and alternative care, further increasing costs. Interestingly, as a category, the amount spent on CAM visits exceeded spending on all other categories except for medications. Typically, this cost is incurred by the patient, as these services are not covered by government health insurance in Quebec, and these patients often do not have private insurance.

In Wolfe's sample, hospitalizations generated the highest direct cost category, at $US 882 per year ($1215 in 2001 Canadian dollars), and represented almost 40% of the total direct cost. In contrast, inpatient stays contributed to only 13% of the mean total direct cost in our sample. Part of this difference is because the set of resources included in our study was broader. Also, since our study is more recent, it may result partly from the trend in North America toward shorter hospital stays. Moreover, it may indicate important structural differences in the health care systems in the 2 countries.

Our estimate of indirect costs of FM over 6 months was $CDN 5035 (SD 7439), or $CDN 10,070 annually. Most of these costs were attributable to the 25% of the study sample that were receiving disability assistance or retired from market work because of FM. The remaining costs were due to the loss of workdays for working women and household work losses. The estimate of the total cost, direct and indirect costs, of FM in our sample was $CDN 14,666. Clearly, it is imperative to include indirect costs in assessing the economic impact of FM, as we found that these constituted nearly 70% of the economic burden of the disease. Taking the group of musculoskeletal diseases as a whole, the Canadian Burden of Illness Study Group[39,40] estimates a similar proportion of the burden of disease due to indirect costs.

Average health care costs in FM may be influenced by intensive users[41]. In previous studies, the intensity of health service use in FM has been related to various sociodemographic, clinical, and psychosocial variables, including physical and sexual abuse[42,43], psychological distress[44], and psychiatric history[41]. In our study, the number of comorbid conditions and the FIQ score were statistically significant determinants of total direct costs, consistent with other studies[11,41]. Walen, *et al*[41] indicated that low-cost FM patients had less disease severity, better functioning, and lower depression scores than the high-cost users, where low-cost and high-cost patients were defined as having below-median and above-median costs, respectively. Wolfe, *et al*[11] also reported that disability (measured with the HAQ) and FM severity were predictors of total direct costs and utilization. Although the sign of the coefficients of other health status variables (high pain intensity and psychological distress) in our multivariate regression was as predicted by theory and previous research, the confidence intervals were wide enough to include zero effect as well as economically and clinically important effects. Physicians treating patients may wish to focus more on comorbid conditions and FM disability, offering interventions that may improve health and thereby reduce healthcare costs.

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While we had a relatively large sample size and employed validated, standardized measures of disability, psychosocial factors, and resource utilization, we acknowledge study limitations. First, the period of time over which costs were determined was relatively short, such that we did not study the evolution of costs over time. Second, although much of the health utilization data from the CAQ have been validated for patients with rheumatic illness, the data on CAM services have not been validated. Given the uneven insurance coverage and/or medical record coverage for these services, it is not clear how such a validation could be carried out. Third, although our study is the most comprehensive in the scope of costs of illness in FM patients to date, we did not determine some costs that may be associated with the cost of illness in this population, measured from a societal perspective. The costs omitted include caregiving provided by family or friends and the time cost and transportation costs associated with seeking medical care. Also, although we estimated the costs of CAM services, since standardized dosing and cost lists do not exist for alternative medicines, we did not include these costs in our analysis. Fourth, despite our relatively large sample size, the confidence intervals around our point estimates of many of the determinants of direct costs include economically significant effects as well as zero. Moreover, we cannot assume that the sample is representative of all individuals with FM; men were excluded, as were individuals who chose not to participate. Nonetheless, those who were included appear to be "typical" FM patients in that they reported high levels of psychological distress, comorbidity, disability, and medication use[45].

The strengths of our study include the comprehensive description of both direct (including complementary care as well as regular health-service use) and indirect costs of FM. In addition, our sample was made up of both clinical and community subjects, enhancing external validity. Finally, to elucidate the determinants of direct costs in women with FM we included not only demographic and clinical variables but also psychological factors.

We have shown that the substantial impairment imposed by FM is associated with high direct and indirect costs. As an illness resulting in substantial pain and disability but without a well understood pathophysiology, the failure of current medical therapies to provide relief to patients is an important force behind the economic burden of FM. Research to promote better understanding of the biopsychosocial causes of FM symptomatology and health care use could lead to more cost-effective treatment[46,47]. In addition, careful evaluations of the types of complementary and alternative treatments for FM should be undertaken, to help health service providers direct their patients to potentially beneficial therapies, while avoiding costly and ineffective ones. Finally, the large economic burden associated with FM highlights the need for better programs for the treatment and rehabilitation of those who suffer from this syndrome[48].

REFERENCES

1. Goldenberg DL. Fibromyalgia syndrome a decade later: what have we learned? Arch Intern Med 1999;159:777-85.
2. White KP, Harth M, Speechley M, Ostbye T. Testing an instrument to screen for fibromyalgia syndrome in general population studies: The London Fibromyalgia Epidemiology Study Screening Questionnaire. J Rheumatol 1999;26:880-4.
3. Yunus MB. Fibromyalgia syndrome: Is there any effective therapy? Consultant 1996; June:1279-85.
4. Simms RW. Fibromyalgia syndrome: current concepts in pathophysiology, clinical features, and management. Arthritis Care Res 1996;9:315-28.
5. Wolfe F, Zhoe S, Lane N. Preference for nonsteroidal antiinflammatory drugs over acetaminophen by rheumatic disease patients: a survey of 1,799 patients with osteoarthritis, rheumatoid arthritis, and fibromyalgia. Arthritis Rheum 2000;43:378-85.
6. Lautenschlager J. Present state of medication therapy in fibromyalgia syndrome. Scand J Rheumatol 2000;29 Suppl:32-6.
7. Da Costa D, Fitzcharles M-A. Predictors of improved health status in fibromyalgia: A two-year prospective study. The 20th Annual Meeting for the Society of Behavioral Medicine, San Diego, CA, March 21[Supplement], S041. 1999.
8. Granges G, Zilko P, Littlejohn GO. Fibromyalgia syndrome: assessment of the severity of the condition 2 years after diagnosis. J Rheumatol 1994;21:523-9.
9. Wolfe F, Anderson JP, Harkness D, et al. Health status and disease severity in fibromyalgia: results of a six-center longitudinal study. Arthritis Rheum 1997;40:1571-9.
10. Dobkin PL, De Civita M, Bernatsky S, Kang H, Baron M. Does psychological vulnerability determine health care utilization in fibromyalgia? Rheumatology 2003;42:1-8.
11. Wolfe F, Anderson JP, Harkness D, et al. A prospective, longitudinal, multicenter study of service utilization and costs in fibromyalgia. Arthritis Rheum 1997;40:1560-70.
12. Oliver K, Cronan TA, Walen HR, Tomita M. Effects of social support and education on health care costs for patients with fibromyalgia. J Rheumatol 2001;28:2711-9.
13. Fitzcharles M-A, Esdaile JM, Clarke AE. Employment status and household disability in Canadian women with fibromyalgia syndrome [abstract]. J Rheumatol 1996;23:1462.
14. Clarke AE, Penrod J, St. Pierre Y, et al. Underestimating the value of women: assessing the indirect costs of women with systemic lupus erythematosus. Tri-Nation Study Group. J Rheumatol 2000;27:2597-604.
15. Pioro-Boisset M, Esdaile JM, Fitzcharles M-A. Alternative medicine use in fibromyalgia syndrome. Arthritis Care Res 1996;9:13-7.
16. Nicassio PM, Schuman C, Kim J, Cordova A, Weisman MH. Psychosocial factors associated with complementary treatment use in fibromyalgia. J Rheumatol 1997;24:2008-13.
17. Clarke AE, Esdaile JM, Bloch DA, Lacaille D, Danoff DS, Fries JF. A Canadian study of the total medical costs for patients with systemic lupus erythematosus and the predictors of costs. Arthritis Rheum 1993;36:1548-59.
18. Clarke AE, Petri MA, Manzi S, et al. An international perspective on the well-being and health care costs for patients with systemic lupus erythematosus. J Rheumatol 1999;26:1500-11.
19. Clarke AE, Zowall H, Levinton C, et al. Direct and indirect medical costs incurred by Canadian patients with rheumatoid arthritis: a 12 year study. J Rheumatol 1997;24:1051-60.
20. Lubeck DP, Spitz PW, Fries JF, Wolfe F, Mitchell DM, Roth SH. A

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multicenter study of annual health services utilization and costs in rheumatoid arthritis. Arthritis Rheum 1986;29:488-93.
21. Silenzio VMB. What is the role of complementary and alternative medicine in public health. Am J Publ Health 2002;92:1562-4.
22. Organisation for Economic Cooperation and Development. Purchasing Power Parities (PPPs) for OECD Countries 1970-2002. Paris: Organization for Economic Cooperation and Development. [cited January 21, 2004]. Available from http://www.oecd.org/dataoecd/61/56/1876133.xls
23. Gironimi G, Clarke AE, Hamilton VH, Danoff DS, Bloch DA, Fries JF. Why health care costs more in the US: Comparing health care expenditures between systemic lupus erythematosus patients in Stanford and Montreal. Arthritis Rheum 1996;39:979-87.
24. Canadian Institute for Health Information. DAD resource indicators. Ottawa: Canadian Institute for Health Information; 1997.
25. Statistics Canada. Hospital indicators, 1993-1994. [83-246-XMB]. Ottawa: Statistics Canada; 1996.
26. Baraff LJ, Cameron JM, Sekhon R. Direct costs of emergency medical care: a diagnosis-based case-mix classification system. Ann Emerg Med 1991;20:1-7.
27. Gold MR. Cost-effectiveness in health and medicine. Oxford: Oxford University Press; 1996.
28. Jackson C. The value of household work in Canada. Statistics Canada Catalogue No. 11-010. Canadian Economic Observer June 1992. Ottawa: Statistics Canada; 1986:3.1-3.19.
29. Statistics Canada. Households' unpaid work: Measurement and valuation. Catalogue No. 13-603E, No. 3. Ottawa: Statistics Canada; 1995.
30. Burckhardt CS, Clark SR, Bennett RM. The Fibromyalgia Impact Questionnaire: Development and validation. J Rheumatol 1991;18:728-33.
31. Melzack R. The McGill Pain Questionnaire: major properties and scoring methods. Pain 1975;1:277-99.
32. Derogatis LR. Symptom Checklist-90-R. Towson, MD: Clinical Psychometric Research; 1994.
33. Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed. Hillsdale, NJ: Erlbaum Associates; 1988.
34. Demers M. Frequent users of ambulatory health care in Quebec: The case of doctor-shoppers. CMAJ 1995;153:37-42.
35. Alexander RW, Bradley LA, Alarcon GS, et al. Sexual and physical abuse in women with fibromyalgia: Association with outpatient health care utilization and pain medication usage. Arthritis Care Res 1998;11:102-15.
36. White KP, Speechley M, Harth M, Ostbye T. The London Fibromyalgia Epidemiology study: Direct health care costs of fibromyalgia syndrome in London, Canada. J Rheumatol 1999;26:885-9.
37. Bombardier CH, Buchwald D. Chronic fatigue, chronic fatigue syndrome, and fibromyalgia. Med Care 1996;34:924-30.
38. Sewitch MJ, Dobkin PL, Baron M. Determinants of nonadherence to medication in fibromyalgia. Submitted to Rheumatology 2003.
39. Health Canada. Economic burden of illness in Canada. [H21-136/1998E]. 1998. Ottawa: Minister of Public Works and Government Services Canada; 2002.
40. Lajas C, Abasolo L, Bellajdel B, et al. Costs and predictors of costs in rheumatoid arthritis: A prevalence-based study. Arthritis Care Res 2003;49:64-70.
41. Walen HR, Cronan TA, Bigatti SM. Factors associated with health care costs in women with fibromyalgia. Am J Manag Care 2001;7:SP39-47.
42. Walen HR, Oliver K, Groessl E, Cronan TA, Rodriguez VM. Traumatic events, health outcomes, and health care use in patients with fibromyalgia. J Musculoskel Pain 2001;9:19-38.
43. Aaron LA, Bradley LA, Alarcon GS, et al. Perceived physical and emotional trauma as precipitating events in fibromyalgia. Associations with health care seeking and disability status but not pain severity. Arthritis Rheum 1997;40:453-60.
44. Kersh BC, Bradley LA, Alarcon GS, et al. Psychosocial and health status variables independently predict health care seeking in fibromyalgia. Arthritis Rheum 2001;45:362-71.
45. Boissevain MD, McCain GA. Toward an integrated understanding of fibromyalgia syndrome. II. Psychological and phenomenological aspects. Pain 1991;45:239-48.
46. Friedman R, Sobel D, Myers P, Caudill M, Benson H. Behavioral medicine, clinical health psychology, and cost offset. Health Psychol 1995;14:509-18.
47. Katon W, Sullivan M, Waker E. Medical symptoms without identified pathology: relationship to psychiatric disorders, childhood and adult trauma, and personality traits. Ann Intern Med 2001;1:917-25.
48. Okifuji A, Turk DC. Fibromyalgia: Search for mechanisms and effective treatments. In: Gatchel RJ, Turk DC, editors. Psychosocial factors in pain. New York: Guilford Press; 1999:227.

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