Cost-effectiveness of Self-management Methods for the Treatment of Chronic Pain in an Aging Adult Population A Systematic Review of the Literature

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Objective: To determine the cost-effectiveness of self-management techniques for older populations (65 and over) with chronic pain and in the absence of such evidence to investigate this question in an aging adult population (average age 60 and over).

Methods: Systematic review of randomized controlled trials (RCTs) with cost-effectiveness data and at least 6 months' follow-up, up to December 2010.

Results: No RCT studies reported cost-effectiveness of self-management exclusively in the over 65 age group. Ten RCTs reported participants with an average age of 60 years or over and met all other inclusion criteria. All of these studies measured cost-effectiveness as cost per improvement in primary outcome, 7 of them using the Western Ontario and McMaster Universities Osteoarthritis Index score, of which 6 reported the pain dimension. Six studies reported cost per quality-adjusted life year (QALY)-gained information, with a further 1 reporting EQ-5D. In 7 studies, relative to usual care, self-management was effective, and in the remaining 3 studies, there was no significant difference. Among those reporting cost per QALY-gained results, self-management did not lead to statistically significant QALY gains relative to usual care (with only one exception). Eight studies suggested that the cost of developing and delivering self-management interventions may be partly offset by savings from reduced subsequent health care resource use.

Conclusions: Self-management is effective among an aging adult population (mean age over 60) with chronic pain and may be costeffective when outcomes are measured using the Western Ontario

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and McMaster Universities Osteoarthritis Index pain score. Costeffectiveness is less certain when measured using the QALY metric. Uncertainty over conclusions regarding cost-effectiveness exists partly due to lack of information regarding societal willingness to pay for pain improvement. There is a need for large multicentred high-quality RCTs to confirm the findings of this review exclusively among older aged populations, such as those who have already reached the statutory retirement age.

Key Words: chronic pain, older adult, aging adult, economic evaluation, cost-effectiveness, self-management

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Pain is a common problem among many groups in society. There is clear evidence that the prevalence and impact of pain conditions increases with age.^{1–3} In older people, the occurrence of pain (especially disabling pain and/or pain at multiple sites) is a particularly important problem as it threatens independent functioning and functional self-efficacy.⁴ Reduced activity,⁵ reduced social networks, and increased morbidities⁶ are all possible consequences of chronic pain that impact on daily living. The effects of chronic pain may include perceived helplessness, depression, isolation, family breakdown, and disability,⁷ and the impact on the quality of life of older people experiencing chronic pain is therefore significant. Such impact has the potential for high costs in terms of the input of carers and the health care system^{8,9} and social care.¹⁰ Although robust evidence on the economic impact of chronic pain is lacking, there is wide consensus that it is significant with 1 study suggesting that the economic impact of musculoskeletal disorders is greater than that of cancer.11 Current pain-management strategies may be expensive and offer very little choice for a patient apart from regular general practitioner (GP), physiotherapist or consultant meetings, often leading to long-term pharmacological analgesia and its associated side effects.12 Chronic pain conditions account for 4.6 million GP appointments each year in the UK, at a cost of £69 million¹³ (approximately \$140 million USD, 2010). Back pain alone, the most common form of chronic pain, is estimated to cost the UK economy £12.3 billion (\$25.9 billion USD, 2010) annually in direct and indirect costs.14

Self-management is a potentially cheaper form of pain management, reducing physician contacts, which may be at least as effective as other interventions, with a greater emphasis on empowering patients to take control of their condition with the aim of improving quality-of-life outcomes. However, research evidence supporting these arguments is lacking, especially in terms of the cost-effectiveness of such approaches. The aim of this systematic literature review therefore was to assess the cost-effectiveness of selfmanagement strategies for older people with chronic pain, and should such evidence be lacking, to answer this question in an aging adult population; specifically investigating whether, relative to other forms of care or management, enabling aging people to self-manage their pain condition would improve quality of life and/or reduce costs to health service providers.

METHODS

Search

We conducted a broad literature search for health economic evaluations of a range of self-management therapies. Specific search terms such as "self-management," "selfefficacy," as well as those focussing on particular therapies, which may constitute self-management were used to identify relevant studies. Our results were combined with a further broad highly sensitive, nonspecific search, the aim of which was to screen a wide range of titles and identify any additional studies, which may not have been identified by terms such as "self-management," but may still evaluate studies, which we deem as involving a period of self-management. We initially conducted an OVID search of Medline and Embase. Follow-up searches of NHSEED, PedRo, Physiotherapy, Scopus, CINAHL, and the CRD database of economic evaluations (University of York database) were also conducted. All databases were searched electronically from their date of inception up until December 2010, with search strategies adapted to fit the search engine being used. Abstracts were scanned and potentially relevant papers retrieved. Reference lists of relevant papers were hand searched with a view to identifying additional relevant studies.

No time limit was set, and databases were searched for English language studies only. Study selection was quality assessed by regular meetings of the EoPIC project committee. Any discrepancies and/or uncertainties were agreed by discussion and mutual consensus between the authors.

Inclusion/Exclusion Criteria for Studies

Only randomized controlled trials reporting cost utility, cost-effectiveness, cost minimization, or cost-benefit analyses comparing self-management interventions with usual care or another active form of care were included in this review. The original aim of the review was to include studies focussing specifically on older people over the age of 65. However, as this inclusion criterion for the search failed to return any papers, we widened our search strategy to include studies focusing on an aging adult population, where the mean participant age was at least 60 years of age (so that the majority of participants would be within the United Nations agreed cutoff for the definition of "older age"¹⁵). This strategy was adopted to maintain focus on those populations approaching older age. Where a minimum age was reported in the studies, Table 1 shows that the majority of these studies include participant populations aged 45 years and over.

Only studies focussing on self-management interventions for chronic pain were included. There are many different definitions of self-management available in the literature, and self-management very often means different things to different people, sometimes even meaning different things to the same people at different points in time, depending on the state of their condition, their ability to cope, and a range of other factors.²⁷ It is thus extremely difficult to define what selfmanagement really is, and each identified study thus has its own definition, all arguably valid. For the purposes of this review, self-management was defined as "a single approach or combination of approaches that can be initially taught by any health professional or learned by an individual to enable them to minimise the impact their chronic pain can have on everyday life." We developed this definition after a review of the literature and in discussion with a number of pain experts from a variety of clinical fields, GPs and patient opinions. It was decided that self-purchased analgesia and any invasive procedures would not be included under this definition.

To be included in the review, the study in question needed to have an adequate follow-up period after the intervention had been taught in which to evaluate the self-management component. Therefore, a 6-month follow-up period at home (after conclusion of the initial taught phase) was determined to be sufficient to capture the self-management component of the intervention. There is no universally accepted definition of chronic pain, but the International Association for the Study of Pain defines it as "pain which has persisted beyond normal tissue healing time."²⁸ In the absence of a rigorous measure, a period of 3 months is taken as normal tissue healing time.²⁹

Given the heterogeneity between the study groups and interventions prescribed, it was not possible to conduct any meta analyses. Therefore, where possible, a narrative description of the broad categories of intervention is given. Efforts were made to separate results by different age strata where possible.

Health Economic Review

Value for money in the provision of health services is increasingly important in the analysis of clinical trials and many now incorporate health economic analyses as a vital component. Key aspects for the formal conduct of a health economic analysis are presented here, namely: the framework of the analysis undertaken, measurement of costs, measurement of clinical outcome and quality of life, determining cost and outcome differences between trial arms, calculating a measure of cost-effectiveness and addressing uncertainty in the analysis. Each of these is discussed in turn under the relevant subheadings.

Health Economic Analysis Frameworks

In short, health economic studies can be divided into cost-benefit analyses (costs and outcomes measured in monetary terms), cost-effectiveness analyses (eg, a measure of cost per \times % pain reduction) and cost-utility analyses (CUA)—reported as cost per quality-adjusted life year (QALY). CUA is the gold standard analysis for the purposes of health economic evaluation as recommended by the UK National Institute for Health and Clinical Excellence (NICE).

Measurement of Costs

Costs considered as part of this review include the cost of intervention delivery, as well as costs to the health service provider over follow-up. For example, costs of primary and secondary care consultations. One may expect that use of such health care resources may reduce as a result of a selfmanagement intervention, where the patient relies less on primary care services. For the purposes of this review, costs are primarily reported as published in the studies. Estimates from the studies have also been inflated to a common price

TABLE 1. Summary of P	apers Assessing the Cost-effectiveness	s of Chronic Pain Self-mana	agement Technigu	les	
Modality (Reference)	Interventions	Participants (Sample Size, Age, Condition)	Analysis Type	Country	Economic Analysis Results†‡
Exercise interventions (ur Water-based therapy ¹⁶	usupplemented) 18 mo RCT of water-based exercise lasting 1 h including warm-up, strengthening, etc. given over 12 mo, compared with usual care	N = 312 adults Minimum age: 60 Mean age: 70 Condition: low limb osteoarthritis	CUA :	UK	Total costs (water exercise vs. usual care): + £134* (\$265) WOMAC pain 12 mo (water exercise vs. usual care): -0.39* WOMAC pain 18 mo (water exercise vs. usual care): -0.39 Total QALY (water exercise vs. usual care): 0.013* ICER reported as £5008 (\$9904) per QALY or £219 (\$433) per unit of WOMAC pain reduction 75% chance of cost-effectiveness at £30,000 per QALY; 95% probability of cost-effectiveness at a willingness to pay of £580 (\$1147) per unit WOMAC pain improvement
Aerobic and resistance exercise ¹⁷	 15 mo R.CT of: A) Aerobic exercise training (60 min 3 × wk over 3 mo, (60 min 3 × wk over 3 mo, followed by 15 mo home exercise program (supplemented by 4 home visits and telephone calls, frequency reduced over time B) Resistance training program: same time periods, resistance increased as classes progressed A and B) Compared with education provision, monthly 1.5 h education session (1-3 mo), followed by regular nurse contact reduced over time 	N = 439 Minimum age: NR Mean age: 69 Condition: knee osteoarthritis	CEA	USA	Total costs Aerobic vs. education: - \$20 (\$30) Resistance vs. education: - \$19 (\$29) Self-reported disability score Aerobic vs. education: -0.18* Resistance vs. education: -0.16* ICER is dominant (aerobics and resistance are both less expensive and more effective than education) Cost differences are not significant Magnitude of differences between aerobic and resistance is small
Lifestyle interventions ^{18,19}	 2 y RCT of 3 interventions: A) Dietary (home based); B) quadriceps (initially class based, followed up at home); C) A + B Each compared with leaflet control 	N = 389 Minimum age: 45 Mean age: 61 Condition: chronic knee pain and a body mass index > 28	CUA	UK	Total costs (diet vs. leaflet): $+ £766.64$ (\$1331) WOMAC pain (diet vs. leaflet): -0.08 QALY (diet vs. leaflet): -0.048 DALY (diet vs. leaflet): -0.048 ICER: dominated (diet is more expensive and less effective) Total costs (exercise vs. leaflet): $+ £245.73$ (\$427) WOMAC pain (exercise vs. leaflet): -1.34^* QALY (exercise vs. leaflet): -1.34^* DALY (exercise vs. leaflet): -1.34^* QALY (exercise vs. leaflet): -1.34^* QALY (exercise vs. leaflet): -1.34^* QALY (exercise vs. leaflet): -1.34^* QALY (exercise vs. leaflet): -0.65 MOMAC pain (diet + exercise vs. leaflet): -0.65 QALY (diet + exercise vs. leaflet):

Cost-effectiveness	of Self-management	for	Chronic	Pain
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ercise interventions (su	pplemented)	M = 750 cdialec	V E V	2111	10030/#3000 Josephine on an advantage for the T
y nome exercise program ²⁰	zy NC1: exercise turing py/monuny telephone contact/both, compared with no intervention	N = 7.97 aduuts Minimum age: 45 Mean age: 62 Condition: long-term knee pain	CEA	2	WOMAC pain score (exercise vs. no exercise): - + ± ± ± - (3.2.9) WOMAC pain score (exercise vs. no exercise): - 0.74* 80% probability of cost-effectiveness at willingness to pay of £8000 (518,784) per clinically significant improvement in WOMAC pain score; 50% probability at a willingness to pay of £2570 (56034)
lass-based program to supplement home exercises ²¹	12 mo RCT follow-up of home exercise program + 2×weekly physiotherapist visits over 8 wk, compared with home exercise only	N = 214 Minimum age: 41 Mean age: 65 Condition: knee osteoarthritis	CUA	UK	Total costs (supplemented vs. not supplemented): $-\pounds 5$ (- \$11) WOMAC pain (supplemented vs. not supplemented): -1.32^* QALY (supplemented vs. not supplemented): $+0.023$ ICER dominant but not statistically significant 70% to 95% probability of class-based program being cost-effective
ehavioral graded activity ²²	65 wk RCT of combined concepts of operant conditioning and exercise—goal to help integrate the benefits into patient's daily lives; compared with usual care (12 wk physiotherapy) Tx of 12 wk maximum 18 sessions and 5 booster sessions	N = 200 adults Mean age: 65 Minimum age: NR Condition: osteoarthritis of the hip and/or knee	CUA	Nether- lands	Total costs (behavioral graded therapy vs. usual care): -€773 (-\$1260) QALY (behavioral graded therapy vs. usual care): -0.02 WOMAC pain score (behavioral graded therapy vs. usual care): + 0.53 Intervention is on an average less expensive and less effective; ICER: €51,385 (\$83,772 (\$2,712,912)] 90% probability that behavioral graded therapy is cost saving
<i>tidisciplinary interven</i> ntegrated rehabilitation program ²³	<i>tions (ESCAPE interventions)</i> 12 mo RCT of ESCAPE knee pain (regular exercise and changing behaviors to enable self management—involving group discussion, information, advice, etc.) compared with outpatient physiotherapy	N = 64 adults Minimum age: 51 Mean age: 67 Condition: chronic knee pain	CUA— QALYs not formally calculated	UK	Total costs (ESCAPE vs. physiotherapy): $-\pounds 263$ (-\$520) WOMAC pain score (ESCAPE vs. physiotherapy): -0.27 WOMAC function score (ESCAPE vs. physiotherapy): -0.06 EQ-5D (ESCAPE vs. physiotherapy): $+ 0.24$ ESCAPE knee pain program is on an average less expensive and more effective than physiotherapy (dominant). QALYs were however, not calculated
ehabilitation program- integrating exercise SM and active coping strategies ²⁴	6mo RCT of usual primary care + rehabilitation program administered to individuals/ groups of 8 people; intervention consists of 12 supervised sessions 2×wk for 6 wk, aim of teaching good exercises to be progressed at home, compared with usual primary care	N = 418 Minimum age: 50 Mean age: 67 Condition: arthritic knee pain	CUA	UK	Total costs (rehabilitation vs. usual care): £169 (\$324) WOMAC function (rehabilitation vs. usual care, (% change in responders): 4.3* Total QALY (rehabilitation vs. usual care): -0.09 Intervention is dominated; although differences are not significant. Probability of cost-effectiveness of any rehabilitation over UC was 90% at a WTP of £1900 (\$3649) for a 1% increase in the proportion of participants gaining at least 15% improvement in functioning (WOMAC); no differences were evident between individual and group rehabilitation from a health and social care perspective
<i>rr approaches</i> rthritis self- management ²⁵	12 mo RCT incorporating 6 sessions of an arthritis self- management program as well as an educational booklet,	N = 812 participants Minimum age: 50 Mean age: 69 Condition:	CUA	UK	Total costs (self-management vs. booklet): $-\pounds 26$ (- \$51) SF-36 physical (self-management vs. booklet): $+ 0.33$ SF-36 mental (self-management vs. booklet): $+ 1.35$ QALY (self-management vs. booklet): -0.01
					(continued)

TABLE 1. (continued)					
Modality (Reference)	Interventions	Participants (Sample Size, Age, Condition)	Analysis Type	Country	Economic Analysis Results†‡
	compared with educational booklet control	osteoarthritis of hips and/or knees			20% probability of cost-effectiveness at £30,000/QALY Great uncertainty depending on quality-of-life measure used, ranging from 2% (QALY, NHS perspective) to 90% (SF-36 mental health dimension. societal perspective)
Telephone contact ²⁶	1 y RCT of telephone only/clinic	N = 393	CEA	NSA	Total costs (telephone vs. no telephone): + \$183 (\$285 USD,
	$\frac{1}{1000}$ only/telephone \pm chinic contact compared with usual care control	Minimum age: INK Mean age: 63			ZUIU) AIMS (telephone vs. no telephone): -0.48*
		Condition:			ICER: \$381 (\$593) per unit improvement in AIMS pain score
		osteoarthritis			Potentially cost-effective compared with no telephone intervention
*A statistically significat †In the case of pain scor †Where possible, costs h ‡Where possible, costs h AIMS indicates Arthritis enabling self-management at SF-36, 36-question short-for	it difference in a reported value at a 95% lev res, a negative difference indicates less pain i ave been inflated to a common base year (20 i finpact Measurement Scale; CEA, cost-effect and coping with arthritic knee pain through e m quality-of-life survey; Tx, treatment grough	vel of statistical significance. In the intervention (self-manage 010) and adjusted for purchasis tiveness analysis; CEAC, cost-e- sercise; ICER, incremental co cv AS, Visual Analogue Scale	ment) group. ng power parity to a ffectiveness acceptabi st-effectiveness ratio; e: WOMAC, Western	common curre lity curve; C1, c NR, not repor Ontario and M	ncy—US dollars (\$). onfidence interval; CUA, cost-utility analysis; Cx, control group; ESCAPE, ted; QALY, quality-adjusted life year; RCT, randomized controlled trial; tediaster Universities Ostocarthritis index (Canada).

year (2010) and converted to US dollars at a rate of $\pounds 1 = \$1.604$ (average exchange rate, 2010) to account for purchasing power parity.

QALYs

One aim of health economic analysis is to focus on whether and to what extent interventions improve quality of life and/or length of life, as determined by the difference between QALYs with and without an intervention, namely determining the size of any QALY gains. QALYs merge together a patient's length of life, adjusted for the quality of those life years. NICE recommends that quality-of-life values be estimated from the Euroqol or EQ-5D,³⁰ a generic preference-based measure of quality of life. This measure is a patient-administered questionnaire asking respondents to rate their general health and well being at a given point in time. Respondents rate their health over 5 dimensions (mobility, self care, usual activities (eg, house work), pain/ discomfort, and anxiety/depression) using a 3-point scale for each dimension (no problems, moderate/some problems, great difficulty/problems). For example, in relation to the pain and discomfort dimension, the 3 options presented to respondents are: (1) "I have no pain or discomfort"; (2) "I have moderate pain or discomfort"; and (3) "I have extreme pain or discomfort." After completion of the questionnaire, a total of 243 health states are possible (3 levels to the power of 5 dimensions). Each health state can be translated into a quality-of-life utility value using UK population tariffs.³¹ These weights are multiplied by the number of life years to give QALY. A QALY value = 1(best possible scenario) represents 1 year of life in full health, whereas a QALY value = 0 represents a patient who has died.

Estimation of Incremental Costs and Effects

Where available for this review, we present mean cost and outcome differences between intervention and control together with their associated 95% confidence interval (CIs). When the CI includes 0, there is no statistically significant difference between the intervention and control groups. For example, if we found a self-management intervention to be overall cost saving by £200, and a CI surrounding this estimate of -£50 to £400, we would conclude that the evidence was not statistically significant. The same approach is used for the presentation of outcome measures (QALY or effectiveness such as the Western Ontario and McMaster Universities Osteoarthritis Index (WOMAC score).

Incremental Cost-effectiveness Ratios (ICERs)

Both cost and outcome differences are used to calculate the ICER,³² which is given as [(cost of treatment–cost of control)/(effectiveness of treatment–effectiveness of control)]. For CUA, this formula for the ICER is [(cost treatment–cost control)/(QALY treatment–QALY control)]. NICE considers an intervention as cost-effective when the ICER is <£20,000/QALY gained, and will also consider an intervention with an ICER between £20,000 and £30,000 per QALY gained. An intervention with an ICER >£30,000 per QALY gained will need to have some extraordinary beneficial health gain to be accepted (eg, in some rare diseases or in the treatment of cancers). Should a trial report an intervention as reducing costs, and improving quality of life, the ICER value is less meaningful and we report the intervention as being dominant over the control group.³³

Handling Uncertainty in Health Economic Studies

Various analytic techniques are further used to address uncertainty surrounding the reported ICER, including bootstrapping (a statistical technique to account for the non-normality of small sample size data), deterministic sensitivity analysis (varying assumptions such as the values of cost and outcome measures), and the presentation of cost-effectiveness acceptability curves (CEACs).^{34,35} CEACs are particularly useful in presenting the probability of an intervention being cost-effective at various threshold values of willingness to pay for a QALY gain, or in the case of cost-effectiveness analysis, willingness to pay for a given improvement in health outcome. They are often used to show the probability of cost-effectiveness when the ICER fails to meet the traditional level of statistical significance, which in the context of this paper is taken as 95% confidence.

RESULTS

Literature Identified

Initial search strategies failed to return any costeffectiveness studies of chronic pain self-management exclusively in older adults (those aged over 65). The revised search targeted toward an aging adult population that (mean age 60 and over) retrieved a total of 522 potentially relevant titles from Ovid Medline and Embase. The literature search was broad and nonspecific to capture the full range of techniques, which may fit our definition of selfmanagement. Therefore, many were scanned through by title before being rejected. A total of 65 abstracts were further retrieved, of which 14 full-text papers were obtained. Of these 14 papers, 10 were included in the review. Four papers did not meet the inclusion criterion of mean age over 60 or were not deemed to fit with our definition of selfmanagement upon reading the full text. A list of excluded studies from the initial search results is available from the authors on request. Further searches of specialist databases and of identified papers' reference lists did not identify any additional papers. Table 1 details the main characteristics of the patient groups, the interventions received; participant age, follow-up, country of study, and main health economic results.

Types of Economic Evaluation Identified

All included studies measured incremental costs and incremental effects of self-management compared with other therapies and/or usual care. All 10 studies measured incremental costs per incremental effectiveness using a primary trial outcome, the most common of which was the WOMAC score, which was measured in 7 included studies, 6 of which reported the pain dimension. Other measures of primary outcome included the Arthritis Impact Measurement Scale (AIMS), WOMAC function score, self-reported disability scores, and the 36-question short-form qualityof-life survey (SF-36) quality-of-life instrument. Of the 10 included studies, a total of 6 included a cost-utility analysis, measuring incremental costs per incremental QALYs gained between intervention and usual care. Of the 4 remaining studies that did not measure QALYs, 1 reported EQ-5D and only 1 study reported cost per improvement in WOMAC pain score.

Modality of Intervention

Nine of the 10 studies focussed on exercise as the main treatment in the trial, with only 1^{26} focussing on a telephone advice service for patients. All exercise-based studies were taught in classes and followed up over a period of home-based exercises (constituting self-management). Exercises included aerobic, muscle strengthening and water-based exercise. Exercise interventions fit broadly into 2 groups, (1) unsupplemented or (2) supplemented and reinforced over follow-up. Supplemented exercise interventions could take the form of class supplementation, cognitive support, or multi-disciplinary program-integrating coping strategies, education and psychological support, for example. Study results are grouped by modality and discussed in order of presentation in Table 1.

Exercise (Unsupplemented)

Three of the 10 studies involved some form of exercise training taught by a health professional and evaluated after a follow-up period of patient self-management at their own home, but this period of self-management was not reinforced by any other follow-up "booster" sessions or regular supportive contact with/supervision of the participants beyond that offered to the control groups. This includes studies in which some telephone contact was maintained between the health professional and all participants regardless of the randomized group. The interventions evaluated consisted of aerobic exercises, resistance training, a combination of aerobic and resistance training, and water exercise, with 1 study evaluating the impact of combining dietary and exercise interventions together. Two of the 3 studies measured cost per WOMAC pain score reduction, whereas 2 measured cost per QALY gained. The magnitude of pain score reduction on which cost-effectiveness was measured varied across all studies in the review, with some measuring costs against a single-point reduction and others compared with a predefined percentage improvement.

Water-based exercise therapy¹⁶ for lower limb osteoarthritis significantly reduced WOMAC pain scores immediately after delivery of the intervention. Pain reductions were however not sustained at the 6-month follow-up appointment. The intervention was more expensive and significantly increased QALYs using the EQ-5D over the 12month intervention period; however, QALYs were not measured over the additional 6-month follow-up. The reported ICER varied between £3800 (\$7516 USD) and £5900 (\$11,668 USD) per QALY gained and a probability of costeffectiveness of between 60% and 80% (for a willingness to pay of £30,000 per QALY gain), increasing to 95% at a willingness to pay of £580 (\$1147 USD) per 1-unit reduction in WOMAC pain score. The conclusions were robust to sensitivity analyses undertaken.

Aerobic and resistance-based exercises¹⁷ were found to significantly improve self-reported disability and pain frequency scores compared with a simple leaflet provision, with no significant difference between aerobic and resistance exercises. Incremental savings per incremental effect tended to be greater for the resistance group; however, a lack of statistical significance rendered strong conclusions difficult.

An additional study^{18,19} found that while an exercise intervention (delivered at home) reduced pain scores in overweight and obese adults with chronic knee pain, neither exercise nor dietary control (prescribed separately) was con-

sidered cost-effective. In combination, the diet and exercise intervention resulted in an ICER of £10,469 (\$18,168 USD) per QALY gained but only had a probability of 23% of cost-effectiveness at the £20,000 threshold willingness to pay for a QALY gain. The authors concluded that a combination of exercise and diet control is the most cost-effective in improving overall quality of life as measured using the QALY; however, there is a high level of uncertainty surrounding the results, with no statistically significant differences reported for the QALY outcome.

Exercise (Supplemented)

Telephone Support: Home-based exercises²⁰ taught by a research nurse at the patient's own home and supplemented with monthly telephone calls offering advice and support showed a significant increase in the mean costs for a significant reduction in knee pain measured using the WOMAC³⁶ pain score and an 80% probability of costeffectiveness at a willingness to pay of £8000 (\$18,784 USD) for a 50% improvement in knee pain.

Educational: McCarthy et al,²¹ took an alternative approach, using a class-based program to reinforce a homebased exercise intervention compared with home exercise alone. The supplemented group improved WOMAC pain score and locomotor function over the 12-month follow-up. The supplemented group generated slightly greater QALYs, was on average slightly less expensive overall and was thus dominant over no supplementation with a 70% probability of cost-effectiveness (at a £20,000/QALY threshold). The authors concluded that the intervention was likely costeffective but cautioned that there was approximately a 30% uncertainty in this conclusion.

Psychological: The impact of behavioral graded activity,²² combining operant conditioning, and exercise over 12 weeks with 5 booster sessions to integrate the benefits of exercise into the patients' everyday life did not reduce pain outcome, was marginally cost saving and generated very small QALY losses compared with outpatient physiotherapy. The intervention was thus on an average less expensive and less effective with an ICER of €51,385 (\$83,782 USD), but this estimate was highly uncertain with very wide CIs. The authors did not present a probability of cost-effectiveness at any threshold values, although visual inspection of the scatter-plot graphs suggests that there is a 90% probability the intervention is cost saving and approximately 60% probability of cost-effectiveness at a threshold value of £20,000 per QALY. The results remain robust to sensitivity analyses and there seems to be a high probability that the intervention was at least cost saving if not costeffective.

Multidisciplinary Interventions

Three of the reviewed trials focused on multidisciplinary interventions, combining interventions such as exercise training and reinforcement, developing coping strategies, boosting education of self-management processes, and providing strong support for people in persevering with their exercise. Two of these evaluated the enabling selfmanagement and coping with arthritic knee pain through exercise (ESCAPE) knee pain program.

ESCAPE: When compared with outpatient physiotherapy program,²³ there were no differences in WOMAC pain and physical functioning scores with both intervention

and usual care groups sustaining physical and psychosocial benefits. However, a participant-administered questionnaire showed that those in the exercise group had a statistically significantly better impression of the effect of the ESCAPE knee program on their overall health. The program was overall cost saving, (due to reduced secondary care contacts). There were no differences in EQ-5D scores, and QALYs were not calculated. The authors concluded that the ESCAPE program was cost-effective on the basis of cost savings; however, the statistical significance of cost differences or CIs are not reported.

Hurley et al²⁴ also evaluated the ESCAPE intervention (individual or group level implementation) compared with usual primary care. The intervention significantly increased the proportion of participants showing a 15%, or greater improvement in WOMAC function subscore was more expensive and was associated with slightly lower QALY gain compared with usual primary care. The ESCAPE intervention was never likely to exceed a 38% probability of cost-effectiveness when the outcome of choice was QALYs. However, there was a probability of 90% and 50% of cost-effectiveness at willingness to pay threshold values of £1900 (\$3649) and £800 (\$1536) for a 1% change in the proportion of participants improving baseline WOMAC functioning score by at least 15%. The authors concluded that the intervention was cost-effective in improving function but not in increasing QALYs. The group rehabilitation intervention (groups of 8 people) cost £189 (\$362 USD) less to deliver than the individual rehabilitation without compromising clinical effectiveness. Cost savings were not sustained over follow-up, with no evidence of a difference in overall costs from a health and social care perspective.

Both evaluations of the ESCAPE knee program failed to show cost-effectiveness based on QALY outcomes. Jessep found no difference in clinical outcomes, but cost savings compared with outpatient physiotherapy, whereas Hurley found a high probability of cost-effectiveness compared with usual care based on the percentage of participants improving WOMAC function scores.

Other Approaches

The economic costs and QALYs associated with an arthritis self-management program among adults over 50 compared with a basic leaflet education program,²⁵ showed that from a health and social care perspective, probabilities of cost-effectiveness on the basis of QALY were very low; however, results were slightly better for alternative outcomes, namely physical and mental health scores for the SF-36. As differences in outcomes were highly uncertain and associated with wide CIs, the authors concluded that there was insufficient evidence to prove cost-effectiveness for this intervention.

A telephone-based follow-up intervention compared with usual care²⁶ found that the intervention significantly improved AIMS outcome measure for nonsignificant cost increases. It seems that a telephone intervention may be a cost-effective way to encourage self-management with an ICER of \$381 (\$593 USD) per unit improvement in AIMS score. It is unclear, however, how much society would be willing to pay for a 1-unit improvement in AIMS score and thus it is difficult to draw strong conclusions from the analysis with regard to cost-effectiveness.

DISCUSSION

We did not retrieve any studies on cost-effectiveness of self-management techniques in an exclusively older population, such as those aged 65 years or over. Thus, costeffectiveness remains unknown in this group. To retrieve as much relevant evidence as possible, we adopted a pragmatic criterion to include aging adults with a mean participant age of 60 years (UN agreed working definition of older age) or over to preserve a focus on an aging population including "early old age," where needs and approaches are likely to be different from those relevant for younger adults. Although no studies included only older adults (over 65), some did exclude younger adults, having a minimum age of 45 years. The conditions causing pain in the participants who were included (arthritis, knee pain, etc.) are reflective of an aging adult population. We were unable to retrieve any age-stratified results in any of the published studies reviewed or associated publications. Meta-analyses were considered; however, due to the heterogeneity of study participants, interventions and controls, this would have been neither robust nor informative. The evidence presented offers at least some information to guide future trial design, using available methods such as value of information.³

There is evidence that exercise-based interventions may be cost-effective as a self-management strategy for dealing with chronic pain in aging adults, relative to usual care. Further, there is a potential that reinforcing this through an ongoing supervision of participants over follow-up is also cost-effective.

It is notable that exercise emerged as the self-management method studied most often. One question that arises is whether this finding, along with the findings relating to effectiveness, would hold among exclusively older populations, where levels of function and presence of comorbidity might reduce the feasibility (or at least alter the content) of the exercise program. Guidelines, produced by the British Geriatric Society³⁸ and American Geriatric Society,³⁹ recommend exercise for older people with chronic pain. These guidelines are mainly based on studies that include participants over the age of 50 years. Further research is however required to determine whether these findings will translate to older participant groups.

Many exercise-based self-management interventions show cost savings over more intensive control treatments or usual care, suggesting that as participants keep up their exercise, they are less likely to require visits to their GP or hospital, thus leading to savings in terms of health care resource use. Costing evidence was of mixed quality in the reported papers, with studies previous to the year 2000 reporting minimal costing information. All 10 included studies adequately detailed the methods of costing the intervention including staff time, operational costs, and the costs of reimbursement for participation if applicable. However, only 2 studies^{16,24} conducted sensitivity analysis around the methods used to estimate the intervention costs and explored the impact on an overall total cost outcome. Cost data for primary and secondary care contacts were reported in 9 studies and were well presented and costed using standard methodologies. There was however, a lack of reporting of data on prescribed medications in 7 of the studies. For example, not reporting on prescribed medications may underestimate the cost saving associated with a particular intervention as more effective interventions may lead to reduced pain medications. Although there were good-quality-costing data relating to costs falling to the health services, there were limited

data on costs incurred by patients and their families. For example, only 4 reported on direct patient costs including the purchase of over-the-counter pain management products. Only 3 studies detailed any costs to carers and families. Exploration of these cost elements would significantly enhance the usefulness of many of the included studies.

The main clinical outcome measure common to most of the trials was the WOMAC (Western Ontario and McMaster assessment of chronic pain), focussing on pain and physical functioning dimensions.¹⁷ Most papers suggest that self-management intervention improves WOMAC pain score relative to usual care, often reflected in a statistically significant improvement. It is difficult, however, to generalize a cost-effectiveness conclusion for 2 reasons: first, there is no consensus among the papers presented regarding the size of clinically significant improvements as measured by WOMAC, with reported clinically significant improvements mentioned in papers of 1 unit, 15%, 30%, and 50%. Second, compared with the QALY metric, there is no equivalent willingness to pay for a unit improvement in WOMAC. Therefore, use of the WOMAC makes it difficult to determine ceiling thresholds at which we can consider an intervention cost-effective. For this reason, the QALY measure is preferred for the assessment of costeffectiveness analysis. While translation models have been developed to map the WOMAC to utility-based measures of quality of life to enable the calculation of QALYs, such methods have been found to give quite large errors when compared with actual EQ-5D responses.⁴⁰ It is therefore, preferable to conduct utility measures within trial when intending to conduct economic evaluation. Probabilities of cost-effectiveness based on WOMAC scores were quite high; however, wide variation in what was considered a clinically significant improvement in pain meant that these probabilities were unreliable. In addition, some studies did not present CEACs or scatter plots, making it difficult to derive these probabilities.

Although the majority of studies reported QALYs (6 studies) estimated from the EQ-5D, (an additional study reported EQ-5D only) only 1 study detected a statistically significant improvement in QALYs, greatly limiting the ability to draw strong cost-effectiveness conclusions on this measure. This 1 study only measured QALYs over the 12-month intervention phase of the trial and not over the additional 6-month home-based follow-up. Probabilities of cost-effectiveness using the QALY outcome measure were generally quite low. The lack of strong statistically significant cost or QALY differences could be for a number of reasons. First, each study compared the intervention with different controls, some of which may be construed to be more active treatment than others. This lack of clarity on what "usual care" actually is could have contributed to the wide variability in the reported incremental costs and QALYs across trials. Second, there may be issues surrounding the applicability of QALY measures to chronic pain. QALYs are based on a generic preference-based measure of quality of life (usually the EQ-5D) and may not be sensitive enough to show the impact on quality of life of changes associated with improved levels of pain. There is much uncertainty as to the appropriateness of the EuroQol for the assessment of quality of life in pain-related chronic diseases. Hurst et al,41 found that the EuroQol offers good reliability and validity for the assessment of quality of life in rheumatoid arthritis patients. However, Wolfe and Hawley⁴² found that the EuroQol's scoring properties and distributional aspects indicate substantial problems for its use in rheumatic disease patients. Finally, as QALYs were not the primary trial outcome, these trials were not powered around detection of such differences and this may further reduce the likelihood of detecting statistically significant differences. Where QALY differences were not significant, it would be helpful to see CEACs and scatter plots illustrating the uncertainty. These were only reported in a handful of studies.

CONCLUSIONS

Because of the highlighted lack of evidence, it was not possible to draw any conclusions on the cost-effectiveness of self-management for an exclusively older subgroup of the population. However, there is evidence that self-management techniques are effective methods of alleviating pain among aging people with chronic pain, with many significantly reducing WOMAC pain scores relative to other forms of care. There is also evidence in a number of studies to suggest that self-management interventions for chronic pain in aging adults are cost saving, with savings accrued through reduced use of health care resources as patients are more in control of their own pain and require fewer visits to their doctor and other health care services. Although there is insufficient statistical evidence to declare that these selfmanagement interventions are definitely cost-effective based on QALY outcomes, many techniques report a > 50% chance of cost-effectiveness. Self-management interventions (especially those reinforced and supported over the follow-up period) are more likely to be cost-effective when health gain is measured using condition-specific outcome measures such as the WOMAC scale. However, a lack of evidence on societal willingness to pay for an improvement in WOMAC pain means further research is required to make robust judgments for cost-effectiveness. Finally, due to lack of data among an exclusively older population (ie, those exclusively over the age of 65), there is a need for further high-quality randomizedcontrolled trials evaluating the effectiveness and cost-effectiveness of a variety of potential self-management strategies among groups of older people.

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