

Managing chronic oedema in community settings

KEY WORDS

- ▶ Chronic oedema
- ▶ Community setting
- ▶ Lymphoedema
- ▶ Literature review
- ▶ Self-management

Aim: The aim of this rapid-evidence assessment of the literature is to assess the existing evidence relating to the management of cancer and non-cancer related chronic oedema/Lymphoedema in the community setting. **Methods:** Using the PICO (Patient/Population/Problem Intervention Comparison Outcomes) method online journal databases MEDLINE, CINAHL and PUBMED, were searched between June 2016 and August 2016. Additional studies were identified through searching bibliographies of related publications and using the Google Scholar internet search function. The following search terms were used in line with the different PICO domains: chronic oedema; Lymphoedema; exercise; compression hosiery; compression bandages; bandages; hosiery; stockings; self-care; self-management; Health-related quality of life; managing chronic oedema; cost of; burden of; economic evaluation; QALY. **Results:** Fifty nine potentially relevant abstracts were found from the search. Twenty one papers were chosen for inclusion in the final review – eight US studies, four UK studies, one joint UK and Canadian study, four Australian studies, two German studies and one study each from Japan and Italy. The papers were thematically categorised (based on health-related quality of life and cost burden outcomes of the study) by Self-managing chronic oedema, Exercise, Compression devices, Dressings/Stockings, Quality of life and Cost studies/Economic burden. **Conclusion:** There is sufficient evidence in the literature that suggests promoting self-management of lymphoedema, the quality of life of lymphoedema patients and the correct use of bandages and stockings is vitally important to clinical practice. Moreover, these issues have a major impact in the health outcomes of patients in the community. This review is a valuable addition to the evidence base for both the care and management of lymphoedema, and an opportunity to add valuable research into the economic burden of the disease.

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The term chronic oedema acts as an umbrella description of abnormal swelling of tissues which lasts for more than 3 months, regardless of whether the aetiology is lymphatic or venous in origin. There are a number of causes of chronic oedema, including dependency oedema, that is to say oedema associated with inactivity and immobility, venous oedema (swelling mainly caused by the vascular insufficiency) and lymphoedema, a chronic swelling due to lymphatic failure. There are two main types of lymphoedema, primary and secondary. Primary lymphoedema is associated with abnormal development of the lymphatics for

either congenital or genetic reasons. Secondary lymphoedema is connected with the lymphatics being damaged by surgery, radiotherapy, infection (such as cellulitis), burns or trauma. In many cases lymphoedema and chronic oedema are intrinsically linked, as chronic oedema is caused by the failure of the lymphatic system (Mortimer and Rockson, 2014). Lipoedema is a condition that alters the fat cell distribution; causing an abnormal build up of fat in the lower limbs. Chronic lipoedema can progress into lipo-lymphoedema.

Chronic oedema can affect people of all ages. However, it is most prevalent in older people. It can

occur in all limbs and, more rarely, the head, neck, trunk or genital area. An incurable, progressive and debilitating condition that often requires lifelong management chronic oedema has a profound effect on people's quality of life and their ability to engage in normal daily activities. It also has a significant impact on NHS resources as patients can have a number of hospital admissions for cellulitis. Cellulitis occurs more in patients with chronic oedema as the fluid component in the limbs contains proteins and other molecules, which is an ideal medium for bacteria. In Wales, it is estimated that the prevalence rate for the lymphoedema alone is in excess of 5.49 per 1,000 population, with over 10,500 people with the condition.

There is poor awareness and understanding of chronic oedema amongst healthcare professionals. Consequently, chronic oedema is frequently unrecognised, resulting in diagnostic delay and even inappropriate treatment. However, awareness of the need for innovative approaches to education for healthcare professionals on the various genres of chronic oedema, their treatments and available resources is improving internationally.

Lymphoedema Network Wales has been proactive in developing innovative methods to support the management of chronic oedema within the community setting. One innovation is through a community-based education model that involves the use of video prescription films as well as a practice educator to support community health professionals and patients in the management and care of the patient's chronic oedema. Whilst this requires investment in terms of resources to deliver the model of care, it could result in more effective management of chronic oedema and improved efficiency of correct prescribing of dressings and more effective products from the All Wales Lymphoedema Compression Garment Formulary. This literature

review was the precursor to obtaining funding from the Welsh Government's Efficiency Through Technology Fund for the implementation of this new education model with community nurses.

AIM

The aim of this rapid evidence assessment of the literature was to assess the existing evidence relating to the management of cancer and non-cancer related chronic oedema/lymphoedema in the community setting.

METHODS

We used the PICO process to set up the framework of the literature strategy since this technique is especially useful in answering clinical and/or health care related questions (*Table 1*). Online journal databases MEDLINE, CINAHL and PUBMED, were searched between June 2016 and August 2016. Additional studies were identified through searching bibliographies of related publications and using the Google Scholar internet search function. The inclusion criteria used can be seen in *Table 2*. The exclusion criteria used were:

- ▶▶ Non English language publications
- ▶▶ Abstracts presented at conferences
- ▶▶ Editorials
- ▶▶ Studies not available in full text.

To keep the research relevant to up-to-date practice, publications prior to 2000 were excluded by the searches. The following search terms were used in line with the different PICO domains: chronic oedema; lymphoedema; exercise; compression hosiery; compression bandages; bandages; hosiery; stockings; self-care; self-management; health-related quality of life; managing chronic oedema; cost of; burden of; economic evaluation; QALY.

Figure 1 summarises the search terms (key words) used during the literature searches. The literature

Table 1. PICO strategy

Patient or Population or Problem	Intervention	Comparison	Outcomes
Patients with chronic oedema in community settings	A community-based education model to support community health professionals and patients in the management and care of the patient's chronic oedema	No education model/usual care	1. More effective management of chronic oedema; economic and patient benefits 2. Health-related quality of life; cost of; burden of; economic evaluation, QALY

Figure 1. Summary of search terms used during the literature searches according to different domains of the PICO

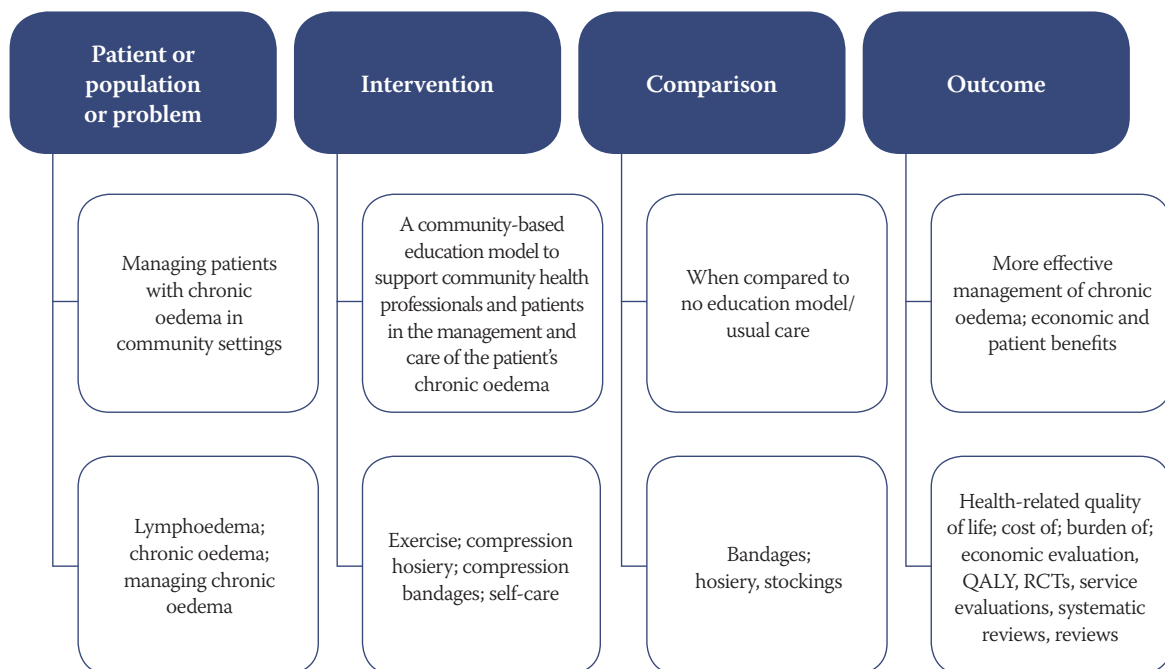


Table 2. Inclusion criteria applied in literature search

Inclusion criteria		Implications
Study population	Adult patients (>18 years)	Studies involving only a population aged less than 18 years were rejected during filtering stages
	Patients with chronic oedema in community settings	Studies on patients with other diseases were rejected during filtering stages
Study design	RCTs, economic evaluations, service evaluations, systematic reviews, reviews	All other study types were either excluded by the searches or rejected during filtering stages, e.g. editorials, letters and comments
Outcomes	Clinical	Studies reporting clinical outcomes were included during filtering stages
	Economic	Studies reporting economic/cost outcomes, economic evaluations, cost studies, cost effectiveness studies, ICERs etc, were included
	Patient reported outcomes	Studies reporting health related quality of life; effective management of chronic oedema; patient benefits; QALY outcomes were included
Other	English language	All other language publications were excluded and rejected during the filtering process
	Since 2000	To keep the research relevant to up to date practice, publications prior to 2000 were excluded by the searches

search results were screened by checking the article's title and abstract against the inclusion criteria (Table 2). Studies that did not match the eligibility criteria were excluded and duplicates were removed. The full articles of non-excluded studies were then obtained and reviewed again for inclusion. Publications that did not fit the PICO at this stage were excluded. The reference lists of included papers were hand-searched to identify any publications that might have been missed during the database searches.

RESULTS

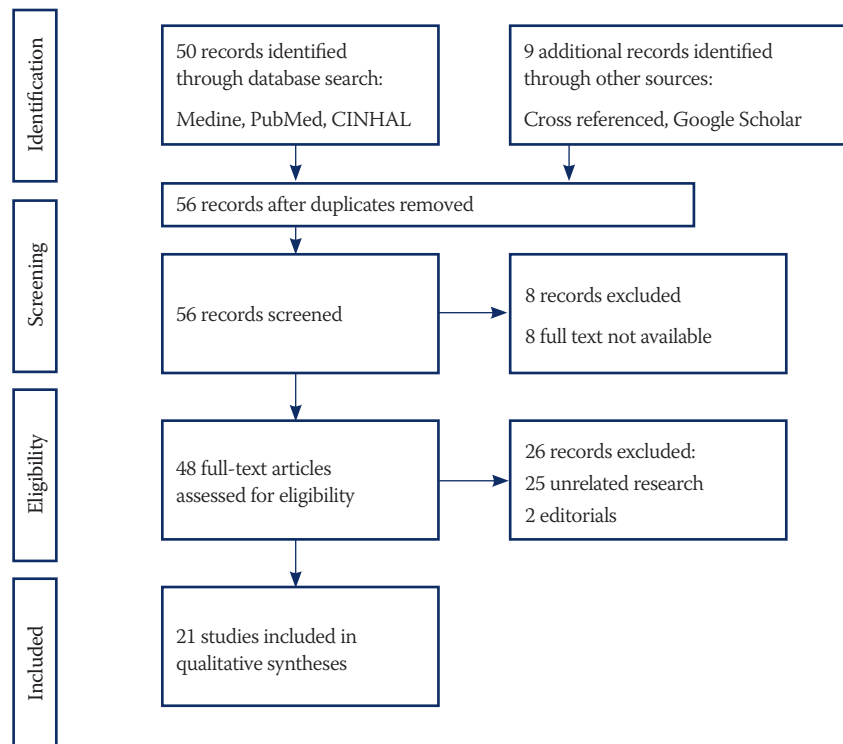
Using Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA), a flow diagram was generated displaying the results of the search (Figure 2). Fifty-nine potentially relevant abstracts were found from a search of the journal databases. Eight papers were excluded as their full text was not available either electronically or via our inter library loan document service. A further 18 papers were excluded as they did not specifically study the management of chronic oedema or lymphoedema in a community setting. Two papers were editorials, three papers were duplicates and the other six papers were deemed to

be unrelated research. Thus 21 papers were chosen for inclusion in the final review — eight US studies, four UK studies, one UK and Canadian study, four Australian studies, two German studies and one study each from Japan and Italy (Table 3). The papers found have been thematically categorised (based on health-related quality of life and cost burden outcomes of the study) into the following sections: Self-managing chronic oedema; exercise; compression devices; dressings/stockings; quality of life and cost studies/economic burden.

SELF-MANAGING CHRONIC OEDEMA

Ahmed et al (2011) used the self-reported data collected by the Iowa Women's Health Study. The data consisted of information for diagnosed lymphoedema in 2004 and data for cancer diagnosis, treatment, behavioural and health characteristics between 1986 and 2003. The main aim of the study was to use this longitudinal data to look at the risk factors for lymphoedema and related arm symptoms in breast cancer survivors that the authors felt had not been examined. The study examined the data from 1,287 women from the cohort and they found that obesity, poorer general

Figure 2. Summary of literature search



health and markers of more advanced cancer were risk factors for lymphoedema and related arm symptoms in breast cancer survivors.

The aim of Jeffs and Purushotham's (2016) study was to identify factors influencing patient perception of success and benefit with self-management of breast cancer-related lymphoedema (BCRL). They also wanted to explore how patients decided whether their swollen limb had improved or deteriorated. The authors identified seven enablers and blocks to self-management: routine, recognising benefit of self-management and consequences of non-treatment, owning treatment, knowledge and understanding, problem-solving, time required for treatment and aesthetics of compression garments.

The study found that lymphoedema practitioners and oncology nurses have a valuable role in providing knowledge and support to patients transitioning to independent self-care. This was down to the women who participated in this study demonstrating varying degrees of acceptance and adjustment to life with lymphoedema. This in turn appeared to directly impact their ability to self-manage lymphoedema. The authors concluded that a better understanding of factors facilitating patients to become experts in their condition and its management may improve long-term outcomes and reduce cost pressures on lymphoedema services.

A more recent systematic review (Douglass et al, 2016) looked at the current evidence for effective lymphoedema self-care strategies that might be applicable to management of all types of secondary lymphoedema. The authors found that there was evidence to support the adoption of remedial exercises in the management of filariasis-related lymphoedema and for a greater emphasis on self-treatment practices for people with cancer-related lymphoedema. Furthermore, empowerment of people with lymphoedema to care for themselves with access to supportive professional assistance has the capacity to optimise self-management practices and improve outcomes from limited health resources. There was no emphasis on the cost burden of lymphoedema, just the physical burden and quality of life of the patient.

EXERCISE

A second 2016 study by Jeff's et al examined BCRL

outcomes and perceived benefit for attendees at a 'reducing your risk of lymphoedema' class between 2000 and 2005. All women found the class beneficial, reporting increased confidence to return to normal life and a wide range of activities/exercise. The authors conclude that prevalence of BCRL should be determined by both clinical judgement and objective measurement to avoid underestimation. Further, the benefit of group education with a lymphoedema expert and of exercise instruction should be further explored, and the potential for exercise to reduce BCRL prevalence should be examined.

COMPRESSION DEVICES

Mulek et al's 2013 study aimed to examine the effectiveness of an advanced pneumatic compression device (APCD) in reducing limb volume, and to evaluate clinician and patient-reported outcomes. Patient-reported outcomes showed a significant increase in ability to control lymphoedema through APCD treatment, with an increase in function and a reduction in the interference of pain. The researchers found that APCD use was associated with consistent reductions in limb volume and had favourable patient-reported outcomes. The results demonstrated that reduction in limb volume and pain enhanced the ability to complete activities of daily living. Combined with functional improvement, patient satisfaction can be achieved.

In 2016, Williams looked at the current evidence surrounding adjustable compression devices. Through reviewing the current literature, the author concluded that research evidence for the use of adjustable compression wrap devices in people with lymphoedema, chronic oedema, venous ulceration and lipoedema is very limited, with the evidence mostly from descriptive papers, case studies or small research studies. These studies were undertaken over a short period of time and the author did not concede that these reflect the long-term nature of the treatment. The review also highlighted the lack of robust economic evaluations that have been undertaken. However, the review conceded that the claims regarding cost savings by some of the studies is 'compelling' and warrants further research.

The 2012 study by Morgan et al explored the challenges of treating patients with complex/severe forms of chronic oedema/lymphoedema with compression therapy. The study identified that in

Canada, services tend to be more rural and remote than in the UK, autonomous specialist practice is less developed and practitioners were generally less confident and felt more vulnerable than their UK colleagues. The study concluded that there is a need for integrated, multidisciplinary services in both countries, with improved education and training, as well as the development of cost-effective compression bandaging systems that can make a major contribution to meeting the challenges of contemporary lymphoedema practice.

The study by Brayton et al (2014) aimed to examine lymphoedema prevalence among cancer survivors and to characterise changes in clinical outcomes and costs associated with a defined therapeutic intervention (use of pneumatic compression devices [PCD]) in a representative, privately insured population. The impact of PCD use was evaluated by comparing rates of a pre-specified set of health outcomes and costs for the 12 months before and after, respectively, PCD receipt. Lymphoedema prevalence among cancer survivors increased from 0.95% in 2007 to 1.24% in 2013. PCD use was associated with decreases in rates of hospitalizations (45% to 32%, $p<0.0001$), outpatient hospital visits (95% to 90%, $p<0.0001$), cellulitis diagnoses (28% to 22%, $p=0.003$) and physical therapy use (50% to 41%, $p<0.0001$). The average baseline health care costs were high (\$53,422) but decreased in the year after PCD acquisition (-\$11,833, $p<0.0001$). The study concluded that lymphoedema is associated with high health care costs; appropriate treatment (in this instance, use of PCD) is associated with significant decreases in adverse clinical outcomes and costs.

COMPRESSION GARMENTS/TAPES

Mosti et al's (2013) randomised controlled pilot study aimed to investigate if a kit consisting of a liner and outer stocking, each exerting 20 mmHg of pressure, would be equally effective in achieving and maintaining volume reduction compared with short-stretch bandaging (2 weeks) followed by a class II (23–33mmHg) stocking (2 weeks) for patients with leg oedema. The authors found that the initial improvement in leg volume (1 week) was independent of the pressure applied and the reduction was maintained by superimposing a second stocking. They concluded that this liner

and outer stocking kit offers a simple alternative for managing leg oedema and could allow for a considerable cost saving.

A meta-analysis of the effectiveness and safety of kinesiology taping in the management of cancer-related lymphoedema was undertaken by Gatt et al (2016). Kinesiology tape is a thin, stretchy, elastic cotton strip with an acrylic adhesive. Therapeutic kinesiology tape that can benefit a wide variety of musculoskeletal and sports injuries, plus inflammatory conditions. This study aimed to determine the effectiveness and safety of kinesiotaping (KT) in the management of CRL compared to compression bandaging or hosiery. A systematic search of the literature was conducted until July 2015. The primary outcomes were reduction in body part volume or circumference and adverse effects of the interventions. The secondary outcomes were subjective experience of the treatment, severity of lymphoedema-related symptoms and patients' quality of life. The authors found that where lymphoedema-related symptoms were reported, KT was found to be superior to compression. However, patients receiving bandaging reported a higher quality of life. KT was not found to be more comfortable than bandaging. The authors recommend KT should only be used with great caution where bandaging cannot be used.

QUALITY OF LIFE

Blome et al (2013) developed and then validated a specific version of the Patient Benefit Index (PBI) that measures patient-relevant benefit in lymphoedema and lipoedema treatment (PBI-L). The authors found that the PBI-L was an internally consistent, valid, and responsive instrument for the assessment of patient-relevant benefits of oedema treatment.

Luchi and colleagues aimed to examine the associations between lymphoedema treatments and outcomes using a cross-sectional observational study of lymphoedema outpatient clinics in Japan (Luchi et al, 2015). The study outcomes that were evaluated were: circumference measurements, Stemmer sign, cellulitis, and skin hardness. Additionally, subjective outcomes were evaluated: satisfaction with treatment, subjective symptoms, EuroQol Five-dimensions and a quality of life measure for limb lymphoedema (LYMQOL). Multiple regression analysis was performed to examine the associations

between lymphoedema treatments and their outcomes. The study concluded that the progression of swelling can be evaluated using the Stemmer sign with regard to compression therapy. The degree of satisfaction can be evaluated as patients' satisfaction with lymphoedema regarding compression garments and simple lymphatic drainage, and improvements in quality of life can be evaluated using the LYMQOL with regard to complete decongestive therapy.

The systematic literature review by Leung et al (2015) looked at the available evidence for the treatment of secondary lower limb lymphoedema in patients with malignancies. The authors found that few studies have evaluated the clinical effectiveness and potential side effects of treatments for lower limb lymphoedema. Symptoms and quality of life assessments were inconsistently reported and all included studies report lower limb volume reduction after treatment, which includes complex decongestion therapy, graded compression stockings and lymphovenous microsurgical shunts. The study concludes that adequately powered randomised controlled trials of these interventions are recommended and effort should be made to establish standardised outcomes, to minimise bias and to improve reporting quality in future trials of treatment for lower limb lymphoedema.

In 2016, Boyages et al (2016) examined the impact of BCRL on women's work and career and claimed to address a significant knowledge gap regarding the additional impact of lymphoedema on breast cancer survivors. Both breast cancer and lymphoedema had a significant negative influence on women's work and career. Respondents reported changes in employment resulting from stress and/or physical impairment, which affected attendance and work performance. The perceived negative impact of breast cancer on respondents' work and career was noticeably greater in Group 2 (63 %) than Group 1 (51 %) ($p=0.03$). The study identifies an additional detrimental effect of lymphoedema on women's work and career over and above the initial impact of breast cancer and provides empirical evidence for future prospective studies and policy improvement.

The 2013, Bell et al looked at the incidence and prevalence of self-reported lymphoedema in breast cancer survivors between 2 and 4 years following diagnosis, the factors associated with the development of lymphoedema and

the impact of lymphoedema on psychological wellbeing. The researchers found that the presence of lymphoedema was associated with lower psychological general wellbeing. The study concludes that lymphoedema after breast cancer treatment frequently has a dynamic pattern and may emerge as an issue for women several years after their initial treatment. It is also associated with a lower level of general wellbeing.

Ezzo et al's Cochrane review aimed to assess the efficacy and safety of manual lymphatic drainage (MLD) in treating BCRL (Ezzo et al, 2015). The authors conclude that MLD is safe and may offer additional benefit to compression bandaging for swelling reduction. Compared to individuals with moderate-to-severe BCRL, those with mild-to-moderate BCRL may be the ones who benefit from adding MLD to an intensive course of treatment with compression bandaging. This finding, however, needs to be confirmed by randomized data. In trials where MLD and sleeve were compared with a non MLD treatment and sleeve, volumetric outcomes were inconsistent within the same trial. Research is needed to identify the most clinically meaningful volumetric measurement, to incorporate newer technologies in lymphoedema assessment, and to assess other clinically relevant outcomes such as fibrotic tissue formation. Findings were contradictory for function (range of motion), and inconclusive for quality of life. For symptoms such as pain and heaviness, 60% to 80% of participants reported feeling better regardless of which treatment they received. One-year follow-up suggests that once swelling had been reduced, participants were likely to keep their swelling down if they continued to use a custom-made sleeve.

Kahn et al (2012) examined factors impacting long-term functional outcomes and psychological sequelae in survivors of breast cancer. The research found that factors associated with poorer current level of functioning and wellbeing included: younger participants, recent diagnoses, aggressive tumour types, receiving chemotherapy, shoulder limitation due to pain, and lymphoedema. Breast cancer survivors require long-term management of psychological sequelae impacting activity and participation.

One study looked at living with lymphoedema in older women with over 50% of breast cancer cases

(Clough-Gorr et al, 2010). The results showed that women with symptoms of persistent lymphoedema consistently reported worse general mental health and physical function. Symptoms of persistent lymphoedema were common in this population of older breast cancer survivors and had a noticeable effect on both physical function and general mental health. Further, their findings provided evidence of the impact of symptoms of persistent lymphoedema on the quality of survivorship of older women. Clinical and research efforts focused on risk factors for symptoms of persistent lymphoedema in older breast cancer survivors may lead to preventative and therapeutic measures that help maintain their health and wellbeing over increasing periods of survivorship.

In 2010, Fu et al investigated how providing information about BCRL affects the cognitive and symptomatic outcome of breast cancer survivors. The researchers found that breast cancer survivors who received information about BCRL had significantly reduced symptoms and increased knowledge about BCRL. In clinical practice, breast cancer survivors should be engaged in supportive dialogues so they can be educated about ways to reduce their risk of developing BCRL (Fu et al, 2010).

COST STUDIES/ECONOMIC BURDEN

The 2013 systematic review by Stout et al sought to examine the policy and economic impact of caring for patients with lymphoedema. As part of a large scale literature review aiming to systematically evaluate the level of evidence of contemporary peer-reviewed lymphoedema literature (2004 to 2011), publications on care delivery models, health policy, and economic impact were retrieved, summarized, and evaluated by a team of investigators and clinical experts (Stout et al, 2013). The authors concluded that the review substantiates lymphoedema education models and clinical models implemented at the community, health care provider and individual level that improve delivery of care. The review also exposed the lack of economic analysis related to lymphoedema. Medical and community-based disease management interventions, taking on a public approach, are effective delivery models for lymphoedema care and demonstrate great

potential to improve cancer survivorship care. The researchers also concede that more research is needed to identify costs associated with the treatment of lymphoedema and to model the cost outlays and potential cost savings associated with management of chronic lymphoedema.

Basta et al (2016) aimed to estimate complicated lymphoedema incidence after breast cancer surgery and calculate associated hospital resource utilization. The study found a 2.3% incidence of complicated lymphoedema after breast cancer surgery. Increased health care utilization for these patients resulted in hospital charge accrual of more than \$180 million in two years; or approximately \$140,000 per patient. Complicated lymphoedema develops in a quantifiable number of patients. The health care burden of lymphoedema mandates further investigation into targeted, anticipatory management strategies for BCRL.

DISCUSSION

This rapid assessment of the literature highlights an encouraging amount of studies and research looking at the management and treatment of lymphoedema and chronic oedema in the community. These studies range from self-management, compression devices, bandages/stockings and quality of life. However, what this review does lack is evidence to highlight the research undertaken to estimate the cost/economic burden of the management and treatment of chronic oedema in the community. There is a troubling paucity of studies addressing the costs associated with treating lymphoedema in both primary and secondary care, social and community care and patient family costs. There are only two studies; Stout et al (2013) and Basta et al (2016) that attempt to research the monetary cost of lymphoedema associated to breast cancer, and neither of these are UK based studies. Therefore, the results from these studies are helpful, but are not necessarily generalizable to the current Welsh setting.

SUMMARY OF EVIDENCE

Six of the studies looked at the clinical effectiveness of either compression bandages (Mulek et al [2013]; Williams [2016]; Morgan et al [2011] and Brayton et al [2014]) or stocking/bandages (Mosti et al [2013]

and Gatt et al [2016]). This is particularly important with regards to the authors' current research study as the correct and timely use of bandages and compression garments in the community is key to the intervention's effectiveness and outcomes.

In addition, the ten studies relating to quality of life — Bell et al (2013), Blome et al (2013), Iuchi et al (2015), Leung et al (2016), Boyages et al (2016), Jeff's et al (2016), Ezzo et al (2015), Kahn et al (2012), Clough-Gorr et al (2010) and Fu et al (2010) — indicated that the health-related quality of life is an important factor in the study of people living with chronic oedema and this also reflects the burden of the disease on family and friends. Further, another encouraging factor is that seven of the studies have been published in the last four years (half of them in 2016). This also indicates that the lymphoedema element of chronic oedema is very much at the forefront of researchers' agendas.

However, what the review also highlights is the paucity of research undertaken looking at the benefits of diet and promotion of exercise in patients with chronic oedema. Exercise is seen as one of the most important factors of the 'Four cornerstones of care', i.e. skin care, movement, simple lymphatic drainage and garments), therefore further research and promotion needs to be undertaken in this area.

CONCLUSION

There is sufficient evidence in the literature that suggests promoting self-management of lymphoedema, the quality of life of lymphoedema patients and the correct use of bandages and stockings is vitally important to clinical practice. Moreover, these have a major impact in the health outcomes of patients in the community. This review is a valuable addition to the evidence base for both the care and management of lymphoedema, and an opportunity to add valuable research into the economic burden of the disease.

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Table 3. Summary of studies

Title	Date	Author	Country	Study Aim	Study Design	Population	Intervention	Comparator	Outcomes	Theme Comments
Complicated breast cancer-related lymphedema: evaluating health care resource utilization and associated costs of management. <i>Am J Surg</i> 211(1): 133–41	2016	Basta MN, Fox JP, Kanchwala SK, Wu LC, Serletti JM, Kovach SJ, Fosnot J, Fischer JP.	USA	Estimate the incidence of complicated lymphedema incidence after breast cancer surgery and calculate associated hospital resource utilization	Burden of care study using the Healthcare Cost and Utilisation Project (HCUP)	Women > 18 years	No intervention	No lymphoedema present	Increased health care utilization for these patients resulted in hospital charge accrual of more than \$180 million in two years; or approximately \$140,000 per patient. Complicated lymphoedema develops in a quantifiable number of patients.	Cost Studies
Worse and worse off: the impact of lymphedema on work and career after breast cancer. <i>SpringerPlus</i> . 5: 657	2016	Boyages J, Kalfa S, Xu Y, Koelmeyer L, Mackie H, Viveros H, Taksa L, Gollan P.	Australia	Impact of BCRL on women's work and career	Longitudinal qualitative study	Women >18 years	No intervention	No comparator	The study identifies an additional detrimental effect of lymphedema on women's work and career over and above the initial impact of breast cancer and provides empirical evidence for future prospective studies and policy improvement.	Quality of life
Self-Care for Management of Secondary Lymphedema: A Systematic Review. <i>PLoS Negl Trop Dis</i> 10(6): e0004740	2016	Douglass J, Graves P, Gordon S.	Australia	Current evidence for effective lymphedema self-care strategies that might be applicable to management of all types of secondary lymphedema	Systematic Review	All participant with filariasis-related lymphoedema or cancer-related lymphoedema	Self-care management	Usual care	There is evidence to support the adoption of remedial exercises in the management of filariasis-related lymphoedema and self-treatment practices for people with cancer-related lymphoedema. No emphasis on the cost burden, just the physical burden and quality of life of the patient.	Self-Management
A meta-analysis of the effectiveness and safety of kinesiology taping in the management of cancer-related lymphoedema. <i>Eur J Cancer Care (Engl)</i> doi: 10.1111/ecc.12510	2016	Gatt M, Willis S, Leuschner S.	Germany	To determine the effectiveness and safety of kinesiotaping (KT) in the management of CRL compared to compression bandaging or hosiery	Meta-analysis	Lymphoedema Pop.	Kinesiotaping (KT) in the management of CRL	Compression bandaging or hosiery	KT was not found to be more comfortable than bandaging. The authors recommend KT should only be used with great caution where bandaging cannot be used.	Stockings/ Bandages
The prevalence of lymphoedema in women who attended an information and exercise class to reduce the risk of breast cancer-related upper limb lymphoedema <i>SpringerPlus</i> . 5: 21	2016	Jeffs E, Purushotham A.	UK	Breast-cancer related lymphoedema (BCRL) outcomes and perceived benefit for attendees at a 'reducing your risk of lymphoedema' class	Cross-sectional study	Women who attended a 'reducing your risk of lymphoedema' class	Information and exercise class to reduce the risk of upper limb BCRL.	Usual care	All women found the class beneficial, reporting increased confidence to return to normal life and a wide range of activities/exercise.	Exercise

Exploring patient perception of success and benefit in self-management of breast cancer-related arm lymphoedema. <i>Eur J Oncol Nurs</i> 20(2016): 173–83	2016	Jeffs E, Ream E, Shewbridge A, Cowan-Dickie S, Crawshaw D, Huit M, Wiseman T.	UK	Identify factors influencing patient perception of success and benefit with self-management of BCRL	Qualitative study using focus groups	Men and Women > 18 years with BCRL	Self-management of BCRL	Usual care	The authors conclude that a better understanding of factors facilitating patients to become experts in their condition may improve longer term outcomes and reduce cost pressures on lymphoedema services.	Quality of life
A review of the evidence for adjustable compression wrap devices. <i>J Wound Care</i> 25(5): 242–7	2016	Williams A	UK	Review of the evidence for adjustable compression wrap devices	Review	Lymphoedema Pop.	Adjustable compression wrap devices	No comparator	The review also highlights the lack of robust economic evaluations that have been undertaken. However, the review concedes that the claims regarding cost savings by some of the studies is 'compelling' and warrants further research.	Compression bandages
Manual lymphatic drainage for lymphedema following breast cancer treatment. <i>Cochrane Database Syst Rev</i> 21(5): CD003475	2015	Ezzo J, Manheimer E, McNeely ML, Howell DM, Weiss R, Johansson KI, Bao T, Bily L, Tuppo CM, Williams AF, Karadibak D	USA	Cochrane review aimed to assess the efficacy and safety of manual lymphatic drainage (MLD) in treating BCRL	Cochrane review	Lymphoedema Pop.	Manual Lymphatic Drainage	Usual care	One-year follow-up suggests that once swelling had been reduced, participants were likely to keep their swelling down if they continued to use a custom-made sleeve	Quality of life
Associations between the treatments and outcomes of patients with upper and lower lymphoedema in Japan: a cross-sectional observational study. <i>Int J Nurs Stud</i> 52(5): 913–9	2015	Iuchi T, Dai M, Sanada H, Okuwa M, Nakatani T, Sugama J.	Japan	Associations between lymphoedema treatments and outcomes such as limb circumference measurements, Stemmer sign, cellulitis, and skin hardness	Cross-sectional observational study	Lymphoedema Pop.	No intervention	No comparator	The subjective outcomes were not associated with every lymphoedema treatment in this study, and the effectiveness of lymphoedema treatment can be evaluated using several different outcomes.	Quality of life
The management of secondary lower limb lymphoedema in cancer patients: a systematic review. <i>Palliat Med</i> 29(2): 112–9	2015	Leung EY, Tirilapur SA, Meads C.	UK	Available evidence for the treatment of secondary lower limb lymphoedema in patients with malignancies	Systematic review	Patients with secondary lower limb lymphoedema	Management of secondary lower limb lymphoedema	No comparator	Adequately powered randomised controlled trials of these interventions are recommended and effort should be made to establish standardised outcomes, to minimise bias and to improve reporting quality in future trials of treatment for lower limb lymphoedema.	Quality of life
Evaluation of patient-relevant outcomes of lymphedema and lipedema treatment: development and validation of a new benefit tool. <i>Eur J Vasc Endovasc Surg</i> 47(1): 100–7	2014	Blome C, Augustin M, Heyer K, Knöfel J, Cornelsen H, Purwins S, Herberger K.	Germany	Development and validation of a specific version of the Patient Benefit Index (PBI) for the assessment of benefit in lymphoedema and lipedema treatment (PBI-L)	Development and validation of tool	All lymphoedema and lipedema patients	No intervention	No comparator	The authors found that the PBI-L was an internally consistent, valid, and responsive instrument for the assessment of patient-relevant benefit of oedema treatment.	Quality of life

Table 3. Summary of studies (continued)

Lymphedema prevalence and treatment benefits in cancer: impact of a therapeutic intervention on health outcomes and costs. <i>PLoS One</i> . 9(12): e114597	2014	Brayton KM, Hirsch AT, O'Brien PJ, Cheville A, Karaca-Mandic P, Rockson SG.	USA	Examine lymphoedema prevalence among cancer survivors and to characterise changes in clinical outcomes and costs associated with a defined therapeutic intervention (use of pneumatic compression devices [PCD])	Retrospective analysis of large private insurance claims database	Lymphoedema Pop.	Pneumatic Compression Devices [PCD]	Usual care	The study concluded that lymphoedema is associated with high health care costs; treatment (in this instance, use of PCD) is associated with significant decreases in adverse clinical outcomes and costs.	Compression bandages
Lymphedema: experience of a cohort of women with breast cancer followed for 4 years after diagnosis in Victoria, Australia. <i>Support Care Cancer</i> . 21(7): 2017–24	2013	Bell RJ, Robinson PJ, Barallon R, Fradkin P, Schwarz M, Davis SR.	Australia	Incidence and prevalence of self-reported lymphedema in breast cancer survivors between 2 and 4 years following diagnosis	Longitudinal study	Women diagnosed with invasive breast cancer	No intervention	No comparator	The study concludes that lymphoedema after breast cancer treatment frequently has a dynamic pattern and may emerge as an issue for women several years after their initial treatment. It is also associated with a lower level of general wellbeing.	Quality of life
Bandages or Double Stockings for the Initial Therapy of Venous Oedema? A Randomized, Controlled Pilot Study. <i>European Journal of Vascular and Endovascular Surgery</i> 46(1): 142–8	2013	Mosti G, Partsch H.	Italy	A kit consisting of a liner and outer stocking and the clinical effect of different pressures	Randomized controlled pilot study	Patients with chronic venous oedema	Liner and outer stocking	Short stretched stocking	The authors found that the initial improvement in leg volume (1 week) was independent of the pressure applied and the reduction was maintained by superimposing a second stocking. They concluded that this offers a simple alternative for managing leg oedema and could allow for a considerable cost saving.	Stockings/ Bandages
Pneumatic compression device treatment of lower extremity lymphedema elicits improved limb volume and patient-reported outcomes. <i>Eur J Vasc Endovasc Surg</i> 46(4): 480–7	2013	Muluk SC, Hirsch AT, Taffe EC.	USA	Examine the effectiveness of an APCD in reducing limb volume	Device registry study	196 lower extremity lymphoedema patients	APCD	Usual care	The results demonstrate that reduction in LV and pain and enhance the ability to complete activities of daily living. Combined with functional improvement, patient satisfaction can be achieved.	Compression bandages
A systematic review of care delivery models and economic analyses in lymphedema: health policy impact (2004–2011). <i>Lymphology</i> 46(1): 27–41	2013	Stout NL, Weiss R, Feldman JL, Stewart BR, Armer JM, Cormier JN, Shih YC.	USA	Examine the policy and economic impact of caring for patients with lymphoedema	Systematic Review	Lymphoedema Pop.	No intervention	Usual care	The review exposes the lack of economic analysis related to lymphoedema. The researchers also concede that more research is needed to identify costs associated with the treatment of lymphoedema and to model the cost outlays and potential cost savings associated with management of chronic lymphoedema.	Cost Studies

Factors associated with long-term functional outcomes and psychological sequelae in women after breast cancer. <i>Breast</i> 21(3): 314–20	2012	Khan F, Amatya B, Pallant JF, Rajapaksa I.	Australia	Examine factors impacting long-term functional outcomes and psychological sequelae in survivors of breast cancer	Prospective rehabilitation outcome study	RMH Database pop.	No intervention	No comparator	Breast cancer survivors require long-term management of psychological sequelae impacting activity and participation.	Quality of life
The challenges of managing complex lymphoedema/ chronic oedema in the UK and Canada. <i>Int Wound J</i> 9(1): 54–69	2012	Morgan PA, Murray S, Moffatt CJ, Honnor A.	UK/ Canada	The challenges of treating patients with complex/ severe forms of chronic oedema/ lymphoedema with compression	Qualitative study using focus groups	Lymphoedema Pop.	No intervention	No comparator	The study concludes that there is a need for integrated, multi-disciplinary services in both countries, with improved education and training, as well as the development of cost-effective compression bandaging systems that can make a major contribution to meeting the challenges of contemporary lymphoedema practice.	Self-management
Risk factors for lymphedema in breast cancer survivors, the Iowa Women's Health Study December. <i>Breast Cancer Res Treat</i> 130(3): 981–91	2011	Ahmed RL, Schmitz KH, Prizment AE, Folsom AR.	USA	Look at the risk factors for lymphedema and related arm symptoms in breast cancer survivors	Population-based prospective cohort study	Women aged 55–69 years	No intervention	No comparator	Obesity, poorer general health and markers of more advanced cancer were risk factors for lymphedema and related arm symptoms in breast cancer survivors.	Self-management
Older Breast Cancer Survivors: Factors Associated with Self-reported Symptoms of Persistent Lymphedema Over 7-years of Follow-up. <i>Breast J</i> 16(2): 147–55	2010	Clough-Gorr KM, Ganz PA, Silliman RA.	USA	Longitudinal study	Women > 65 years diagnosed with primary stage I-IIIa	Lymphoedema Pop.	No intervention	No comparator	Clinical and research efforts focused on risk factors for symptoms of persistent lymphedema in older breast cancer survivors may lead to preventative and therapeutic measures that help maintain their health and well-being over increasing periods of survivorship.	Quality of life
The effect of roviding information about lymphedema on the cognitive and symptom outcomes of breast cancer survivors. <i>Ann Surg Oncol</i> 17(7): 1847–53	2010	Fu MR, Chen CM, Haber J, Guth AA, Axelrod D.	USA	Providing information about breast-cancer related lymphoedema (BCRL) affects the cognitive and symptomatic outcome of breast cancer survivors	Cross-sectional study	Lymphoedema Pop.	Information about lymphoedema on the Cognitive and Symptom Outcomes of Breast Cancer	No information	In clinical practice, breast cancer survivors should be engaged in supportive dialogues so they can be educated about ways to reduce their risk of developing BCRL.	Quality of life