The Impact of Economic Evaluation on Lymphoedema Services Research in Wales

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Dedication

Firstly, I would like to dedicate this thesis to my Mam, Dad, Brother, and Sister. They have always been incredibly supportive of every endeavour (academic or not) that I have embarked on throughout my 50 years. Of course, there have been eyes rolled, and heads shaken at some decisions in my life, but the support has always been there.

Secondly, I would like to dedicate this thesis to my daughters, Nancy, and Jessica. Thank you for always putting up with Daddy when he was grumpy or stressed due to his work and research commitments. It meant the world to me. I promise to be less stressed from now on!

Finally, I would like to dedicate this thesis to my partner Maria. Her support of my research and academic pursuits (through my Master's degree and now this PhD), has been utterly unwavering and must have been pretty exhausting at times. Again, thank you for putting up with my grumpiness, and having my laptop on my lap pretty much every night answering emails and working on papers, etc. Your support has been amazing, and 100% I could not have done this without you.

Declarations and Statements

1. I, Ioan Humphreys, hereby declares that the work presented in this thesis has not previously been accepted in substance for any degree and is not being concurrently submitted in candidature for any degree.

2. I, Ioan Humphreys, hereby declares that the thesis is the result of my own investigations, except where otherwise stated and that other sources are acknowledged by footnotes giving explicit references and that a bibliography is appended.

3. I, Ioan Humphreys, understand that the electronic version will be deposited by the repository administrator in Cronfa, the Swansea University institutional repository. The bibliographic metadata and abstract will be made immediately available. The full-text version of the thesis will be published online.

Signed: __[redacted]

Date: 17th January 2024

Acknowledgements

This thesis represents the culmination of many years of work within the field of health economics, economic evaluations, lymphoedema and cellulitis. Firstly, I would like to thank Dr Melanie Thomas, Karen Morgan, Dr Cheryl Pike, and Dr Rhian Noble-Jones of Lymphoedema Wales whom without their support and belief in me as a researcher and collaborator, this body of work and subsequent thesis would not have happened. Thank you all. I would also like to thank Professor Ceri J Phillips for being a fabulous mentor to me between 2006 and 2020. My career as a health services researcher and health economist would not have happened without you. Finally, I would like to thank my supervisor (and dedicated maggot collaborator) Professor Yamni Nigam who also believed in me as a researcher and health economist. You have guided me throughout this process with resolute support and belief in my abilities, and for this I will be forever grateful.

Abstract

Lymphoedema is a chronic and progressive disease of the lymphatic system characterized by inflammation, increased adipose deposition, and tissue fibrosis. There are two types of lymphoedema, primary and secondary lymphoedema, which have different causes. This thesis comprises of eight papers, a research body that examines the impact of economic evaluations on lymphoedema research and practice in Wales via the Lymphoedema Wales clinical service.

These research papers present a body of evidence of the demonstrably positive impact that economic evaluation research is having on people living with lymphoedema in Wales. This evidence has enabled WG to permanently allocate the funds needed for both the LVA and OGEP programme. This has enabled LW to continue to implement their interventions and innovative practice. This has also given LW the time and resources to bring their experiences and evidence of best practice to other health practitioners.

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List of key abbreviations

A&E	Accident & Emergency
ABMUHB	Abertawe Bro Morgannwg University Health Board
ABUHB	Aneurin Bevan University Health Board
AWLCGF	All Wales Lymphoedema Compression Garment Formulary
BCUHB	Betsi Cadwalader University Health Board
BCRL	Breast Cancer-Related Lymphoedema
BLS	British Lymphology Society
BNF	British National Formulary
BOE	Bank of England
C&VUHB	Cardiff & Vale University Health Board
CC	Complications or Comorbidities
CCG	Clinical Commissioning Groups
CHHS	College of Human Health Sciences
CI	Confidence Intervals
CNO	Chief Nursing Officer
COA	Clinical Outcome Assessment
CP	Community Pharmacists
CTUHB	Cwm Taf University Health Board
DLT	Decongestive Lymphoedema Treatment
DN	District Nurse
DNA	Did Not Attend
GEM	Graduate Entry Medicine
GP	General Practitioner
HB	Health Board
HCSW	Health Care Support Worker
HDUHB	Hywel Dda University Health Board
HEOR	Health Economics and Outcomes Research
HRQoL	Health Related Quality of Life

ICS	Impact Case Study
IH	Ioan Humphreys
IP	In Patient
LIMPRINT	IMpact and PRevalence INTernational Lymphedema Framework
LW	Lymphoedema Wales
LNW	Lymphoedema Network Wales
LU	Leg Ulcer
LVA	lymphaticovenous Anastomosis
MLLB	Multi-Layer Lymphoedema Bandaging
MP	Member of Parliament
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health and Care Research
OGEP	On the Ground Education Program
OP	Outpatient
PROM	Patient reported Outcome Measure
PSS	Paid carers/Social Care
PSSRU	Personal Social Services Research Unit
PTHB	Powys Teaching Health Board
QALY	Quality Adjusted Life Year
QoL	Quality of Life
RCT	Randomised Controlled Trial
REF	Research Excellence Framework
RUQ	Resource Utilisation Questionnaire
SAIL	Secure Anonymised Information Linkage
SBUHB	Swansea Bay University Health Board
SD	Standard Deviation
SF	Short Form
SPQ	Specialist Practitioner Qualification

SWEMWBS	Short Warwick-Edinburgh Mental Wellbeing Scale
TVN	Tissue Viability Nurse
UHB	University Health Board
UK	United Kingdom
UOA2	Unit of Assessment 2
UTA	Unable To Attend
VAS	Visual Analogue Scale
VLU	Venous Leg Ulcer
WAG	Welsh Assembly Government
WEMWBS	Warwick-Edinburgh Mental Wellbeing Scale
WG	Welsh Government
WHSSC	Welsh Health Specialised Services Committee
WLP	Wet Leg Pathway
WNB	Was Not Brought

Chapter 1

Summary of Published Work and author contributions

The first chapter of this thesis is a formal presentation of the papers included in this PhD by published works. In the following sections I will give the title of the paper, the date it was published, a very brief overview of the paper and the order of my authorship on the paper. Additionally, I will also present a detailed author contribution declaration that outlines the percentage contribution that each author presented in each of the papers.

Section 1:1 Paper 1 Managing chronic oedema in community settings

First Author

An original paper initiating a rapid-evidence assessment of the literature to assess the existing evidence relating to the management of cancer and non-cancer related chronic oedema/Lymphoedema in the community setting.

Published: September 2017

Humphreys, I; Morgan, K; Thomas, M (2017) Managing chronic oedema in community settings. Wounds UK, Vol 13, 22-35

Section 1:2 Paper 2 Evaluation of the economic impact of a national lymphoedema service in Wales

First Author

An original paper investigating the economic impact of a national lymphoedema service on the NHS Wales budget. Work was undertaken to determine the care pathway within Lymphoedema Network Wales and develop a hypothetical ‘world without’ the service as a comparator. This evaluation provided the first estimates that suggest that the service is likely to be cost saving when people with lymphoedema are managed within Lymphoedema Network Wales rather than in a ‘world without’ the service.

Published: 10th November 2017

Ioan Humphreys, Melanie J Thomas (2017) Evaluation of the economic impact of a national lymphoedema service in Wales British Journal of Nursing Vol. 26, No. 20 Clinical Focus doi.org/10.12968/bjon.2017.26.20.1093

Section 1:3 Paper 3 Pilot evaluation of the management of chronic oedema in community settings project

First Author

An original paper which aimed to estimate the economic impact of the On the Ground Education Programme (OGEP) within one local University Health Board (UHB) in Wales. The study was a pilot Evaluation of the OGEP using retrospectively and prospectively collected patient recalled data. We collected health care service use data prior to receiving the OGEP (baseline) and at 3 months follow-up from 97 patients during the period June 2016 and January 2017. Our research shows health care resource use and costs decreased, while health-related quality of life scores increased.

Published: 30th November 2017

Ioan Humphreys, Melanie J Thomas, Karen M Morgan (2017) Pilot evaluation of the management of chronic oedema in community settings project British Journal of Community Nursing Vol. 22, No. 12 Long-term Conditions doi.org/10.12968/bjcn.2017.22.12.578

Section 1:4 Paper 4 The benefits of raising awareness of lymphoedema among care home staff

Third Author

An original paper looking at the patients with lymphoedema referred to a lymphoedema service from care homes in one health board area in Wales. A pilot project was initiated to develop education and raise awareness of lymphoedema among care home staff. Forty-four care homes agreed to participate in the project with 1216 education packs being issued to care home staff. Only 13% (35/262) of residents with lymphoedema were known to the local lymphoedema service. Of the 31 residents reporting cellulitis, 81% had lymphoedema; of the 11 residents identified with a wound, 100% had lymphoedema and of the 40 residents reporting falls, 70% had lymphoedema. This educational project has identified the value of raising awareness of lymphoedema within care homes.

Published: 27th February 2020

Melanie Thomas, Karen Morgan, Ioan Humphreys, Karl Hocking, Diane Jehu (2020) The benefits of raising awareness of lymphoedema among care home staff British Journal of Nursing Vol. 29, No. 4 Focus doi.org/10.12968/bjon.2020.29.4.190

Section 1:5 Paper 5 Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation

Third Author

An original paper on the service evaluation conducted to estimate the potential impact of changing the way compression garments are accessed from a prescription to a procurement process. There was a total of 5392 completed patient data forms were included, which when compared to the processes indicated that using a prescription route is overwhelmingly more costly than procuring. Overall costs suggest the potential for substantial savings to NHS Wales (£71.10 per patient) were statistically significant ($P<0.001$). Potential for improved patient outcomes was observed as garments were provided directly on appointment instead of significant delays promoting Value-Based healthcare.

Published: 10th December 2021

Melanie Jayne Thomas, Karen Morgan, Ioan Humphreys, Rhian Newton (2021) Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation Journal of Prescribing Practice Vol. 3, No. 12 Clinical Focus doi.org/10.12968/jprp.2021.3.12.490

Section 1:6 Paper 6 A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs

Second Author

An original paper which aimed to estimate the financial impact of non-attendance on a nation-wide hospital lymphoedema service. The characteristics of 870 patients from 2019/2020 were evaluated suggesting that those with a wound alongside complex lymphoedema were less likely to DNA appointments. Two-thirds of patients were managing two or more comorbidities—obesity, cardiac conditions and diabetes being the most common. Modernising appointment processes and identifying patient value may help minimise DNA costs in the future.

Published: 21st November 2022

Melanie Jane Thomas, Ioan Humphreys, Rhian Wyn Noble-Jones (2022) A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs International Wound Journal doi.org/10.1111/iwj.13999

Section 1:7 Paper 7 Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study

First Author

An original paper which aimed to estimate costs associated with managing patients with cellulitis from the UK National Health Service (NHS) perspective. The analysis was undertaken through the SAIL (Secure Anonymised Information Linkage) Databank. The estimated direct annual costs to the Welsh NHS (£28 554 338) are considerable. Initiatives to support patients and healthcare professionals in identifying early signs/risks of cellulitis, improve the accuracy of initial diagnosis, prevent cellulitis recurrence, and improve evidence-based treatment pathways would result in major financial savings, to both the Welsh and UK NHS. In light of these findings, Wales has developed the innovative National Lymphoedema cellulitis Improvement Programme to address these burdens, providing a proactive model of cellulitis care.

Published: 17th January 2023

Ioan Humphreys, Ashley Akbari, Rowena Griffiths, Dave Graham-Woollard, Karen Morgan, Rhian Noble-Jones, Marie Gabe-Walters, Melanie Thomas Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study International Wound Journal doi.org/10.1111/iwj.14088

Section 1:8 Paper 8 The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months

Third Author

An original paper which aimed to explore the impact and outcomes after LVA over 24-months follow up. Data on 150 patients with upper and lower limb lymphoedema were prospectively captured before and after LVA surgery. All patients were assessed and operated on by the same team involving two surgeons and three lymphoedema specialists. Data captured included health related quality of life tool (EQ5D5L), circumferential measurements using tape measure and Perometer, usage of compression garments, occurrence of cellulitis and a range of patient reported outcome measures. LVA resulted in significant improvements in patient reported outcome measures, reduced need for compression garments and substantial reduction in cellulitis episodes. Limb circumference via tape measure or Perometer did not alter, yet patient's quality of life significantly improved.

M Thomas, C Pike, I Humphreys, T Bragg and A Ghattaura, The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months, Journal of Plastic, Reconstructive &Aesthetic Surgery, (2023)
doi:https://doi.org/10.1016/j.bjps.2023.06.068

Section 1:2 Author Contribution Declaration

The following people and institutions, listed in the table below, contributed to the publication of work undertaken as part of this thesis. Therefore, we the undersigned agree with the below stated "proportion of work undertaken" for each of the above published peer-reviewed manuscripts contributing to this thesis.

Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 1: Managing chronic oedema in community settings					
1	Ioan Humphreys	Swansea University	IH undertook 80% of the rapid literature search in the above paper	80	[redacted]
2	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 40% of the writing of the above paper	10	[redacted]
3	Karen Morgan	Swansea Bay UHB - Lymphoedema Wales Clinical Network	KM undertook 20% of the writing of the above paper	10	[redacted]
Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 2: Evaluation of the economic impact of a national lymphoedema service in Wales					
1	Ioan Humphreys	Swansea University	IH undertook all the economic evaluation costing work, all the statistics and around 60%	60	[redacted]

			of the writing of the above paper.		
2	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 40% of the writing of the above paper	40	[redacted]
Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 3: Pilot evaluation of the management of chronic oedema in community settings project					
1	Ioan Humphreys	Swansea University	IH undertook all the economic evaluation costing work, all the statistics and around 50% of the writing of the above paper.	50	[redacted]
2	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 30% of the writing of the above paper	30	[redacted]
3	Karen Morgan	Swansea Bay UHB - Lymphoedema Wales Clinical Network	KM undertook 20% of the writing of the above paper	20	[redacted]
Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 4: The benefits of raising awareness of lymphoedema among care home staff					

1	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 30% of the writing of the above paper	30	[redacted]
2	Karen Morgan	Swansea Bay UHB - Lymphoedema Wales Clinical Network	KM undertook 30% of the writing of the above paper	30	[redacted]
3	Ioan Humphreys	Swansea University	IH undertook all the costing work, all the statistics and around 30% of the writing of the above paper.	30	[redacted]
4	Karl Hocking	Swansea Bay UHB - Lymphoedema Wales Clinical Network	KH undertook 5% of the writing of the above paper	5	[redacted]
5	Diane Jehu	Cwm Taf UHB - Lymphoedema Wales Clinical Network	DJ undertook 5% of the writing of the above paper	5	[redacted]

Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 5: Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation					
1	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 35% of the writing of the above paper	35	[redacted]

2	Karen Morgan	Swansea Bay UHB - Lymphoedema Wales Clinical Network	KM undertook 20% of the writing of the above paper	20	[redacted]
3	Ioan Humphreys	Swansea University	IH undertook all the costing work, all the statistics and around 35% of the writing of the above paper.	35	[redacted]
4	Rhian Newton	Swansea Bay UHB - Head of Prescribing and Medicine Management	RN undertook 10% of the writing of the above paper	10	[redacted]
Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 6: A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs					
1	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 40% of the writing of the above paper	40	[redacted]
2	Ioan Humphreys	Swansea University	IH undertook all the costing work, all the statistics and around 25% of the writing of the above paper.	25	[redacted]

3	Rhian Noble-Jones	Swansea Bay UHB - Lymphoedema Wales Clinical Network	RN-J undertook 35% of the writing of the above paper	35	[redacted]
Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 7: Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study					
1	Ioan Humphreys	Swansea University	IH undertook all the costing work, all the statistics and around 45% of the writing of the above paper.	45	[redacted]
2	Ashley Akbari	Swansea University	AA undertook 10% of the writing of the above paper	10	[redacted]
3	Rowena Griffiths	Swansea University	RW undertook 100% of the SAIL Databank analysis and around 10% of the writing of the paper	10	[redacted]
4	Dave Graham-Woollard	Swansea Bay UHB - Lymphoedema Wales Clinical Network	DG-W undertook 5% of the writing of the above paper	5	[redacted]
5	Karen Morgan	Swansea Bay UHB - Lymphoedema Wales	KM undertook 5% of the writing of the	5	[redacted]

		Clinical Network	above paper		
6	Rhian Noble-Jones	Swansea Bay UHB - Lymphoedema Wales Clinical Network	RN-J undertook 10% of the writing of the above paper	10	[redacted]
7	Marie Gabe-Walters	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MG-W undertook 5% of the writing of the above paper	5	[redacted]
8	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 10% of the writing of the above paper	10	[redacted]

Author Number:	**Author Name:**	**Author Institute:**	**Author Contribution:**	**Proportion of Contribution (%):**	**Author Signature:**
Publication 8: The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months					
1	Melanie Thomas	Swansea Bay UHB - Lymphoedema Wales Clinical Network	MT undertook 30% of the writing of the above paper	30	[redacted]
2	Cheryl Pike	Swansea Bay UHB - Lymphoedema Wales Clinical Network	CP undertook 30% of the writing of the above paper	30	[redacted]
3	Ioan Humphreys	Swansea University	IH undertook all the costing work, all the statistics and around 30%	30	[redacted]

			of the writing of the above paper.		
4	Thomas Bragg	Swansea Bay UHB - Plastic and Reconstructive Department	TB undertook 5% of the writing of the above paper	5	[redacted]
5	Amar Ghattaura	Swansea Bay UHB - Plastic and Reconstructive Department	AG undertook 5% of the writing of the above paper	5	[redacted]

Chapter 2

Background and context

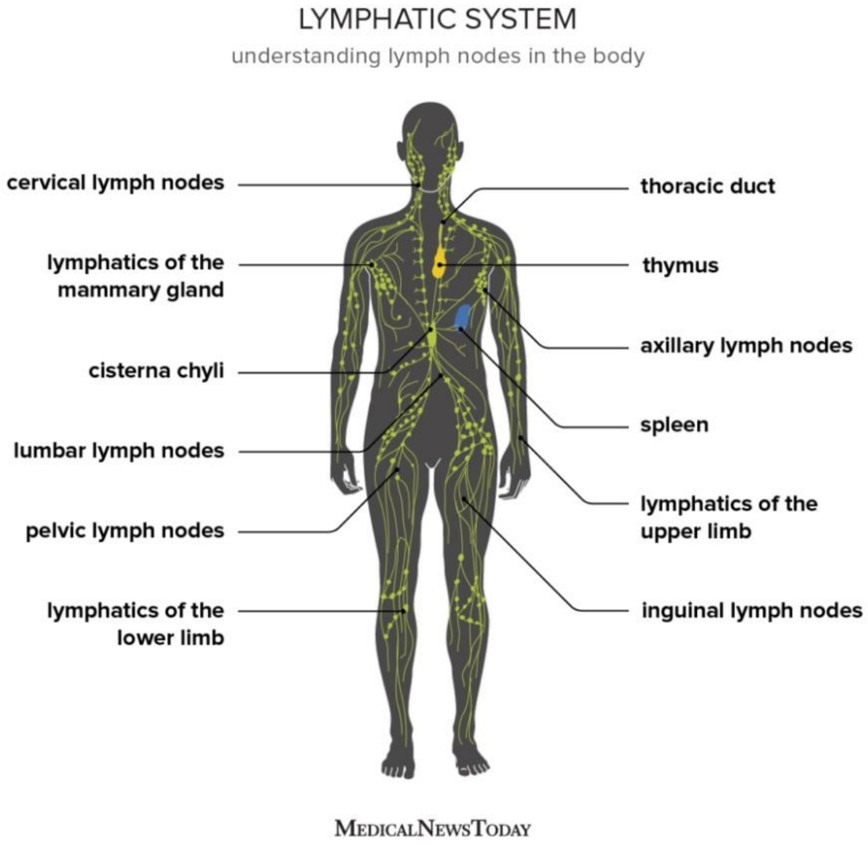
The next chapter of this thesis will give a background to the lymphatic system, lymphoedema, the causes of lymphoedema and the current assessment and diagnosis of the condition. I will also discuss the discipline of health economics and economic evaluations, incorporating health related quality of life and patient related outcomes. Finally, the chapter will consider the challenges of healthcare in Wales and will discuss Welsh Government initiatives and principles such as Prudent Healthcare and Value-Based Healthcare.

Section 2:1 The Lymphatic System

The lymphatic system is an important and often underappreciated component of the circulatory, immune, and metabolic system (Ozdowski and Gupta 2023). It is composed of lymphatic fluid, lymphatic vessels, lymphatic cells, and organs. Lymphatic cells include macrophages, dendritic cells and lymphocytes, and Lymphatic organs include the spleen and thymus. The lymphatic system carries out three primary functions:

- Tissue drainage and maintenance of fluid balance in the body. The lymphatic vessels reabsorb excess interstitial fluid from tissues to return it to the intravascular space, which prevents fluid build-up in peripheral tissues.
- Facilitation of the absorption of dietary fats from the gastrointestinal tract to the bloodstream for metabolism or storage
- The enhancement and facilitation of the immune system. The lymphatics allow for the immune system to function properly carrying antigens to lymph nodes, and immune cells, such as macrophages, to sites of infection to begin the immune process (Ozdowski and Gupta 2023)

Figure 1 The Lymphatic System (www.medicalnewstoday.com)



Section 2:2 Lymphoedema

Lymphoedema is a chronic and progressive disease of the lymphatic system characterized by inflammation, increased adipose deposition, and tissue fibrosis. (Bowman C 2024) There are two types of lymphoedema, primary and secondary lymphoedema, which have different causes. Primary lymphoedema is caused by alterations (mutations) in genes responsible for the development of the lymphatic system. (NHS UK 2025) The faulty genes cause the parts of the lymphatic system responsible for draining fluid to not develop properly or not work as they should. Secondary lymphoedema develops in people who previously had a normal lymphatic system that then becomes damaged. (NHS UK 2025) It can have a number of different causes, however, the latter can be acquired through damage to the lymphatic system often due to cancer, skin disorders, venous disease, obesity, or trauma.

Risk factors and/or causes linked to secondary lymphoedema diagnosis are shown in Table 1 below (Chaput et al 2020).

Table 1. Secondary lymphoedema Risk Factors (Chaput et al 2020)

Lymphoedema risk factors	
Lymphoedema site	
Upper limb or trunk	**Lower limb**
Axillary lymph node dissection	Lymphatic vessel compression because of tumour location (pelvic or abdominal tumours)
Radiotherapy to the breast	Inguinal or pelvic lymph node dissection
Radiotherapy to lymph nodes (axillary, internal mammary, or subclavicular)	Pelvic radiotherapy
Chemotherapy (taxanes)	Deep vein thrombosis and post-thrombotic syndrome
Skin changes after axillary radiotherapy (fibrosis, radiation dermatitis)	Comorbid conditions (cardiac or renal disease)
Complications from wounds or drains	Advanced cancer
Axillary web syndrome	Overweight or obesity
Lymphocele	Chronic venous insufficiency
Advanced cancer	Varicose vein treatments (stripping)
Overweight or obesity	Cellulitis and inflammatory skin conditions
Cellulitis and inflammatory skin conditions	Orthopedic surgery
High blood pressure	Prolonged limb immobilization
Procedures such as pacemaker insertion and arteriovenous shunt for dialysis	Genetic predisposition
Filariasis (nematode worm infection)	Filariasis (nematode worm infection)

Inflammation can also lead to the development of lymphoedema. (NHS UK 2024, Bowman C 2024) Medical conditions that cause tissue to become red and swollen can permanently damage the lymphatic system and enable lymphoedema to develop. Two of these medical conditions are Psoriasis and atopic eczema (NHS UK 2024) both of which cause inflammation of the skin. These skin condition related lymphoedema causes the patient to experience pain, recurrent cellulitis, wounds, immobility, and heaviness (Burian et al 2021). The impact of lymphoedema also affects quality of life (QoL) extending into all aspects of normality (Thomas et al 2020, Moffatt et al 2007, Neuner et al 2014 & Gabe-Walters & Thomas 2021). Lymphoedema also influences health outcomes in relation to wound care, ulceration, and other co-morbidities and is considerable burden to the UK National Health Service (NHS) (Phillips et al 2016 & Phillips et al 2020). Conservative lymphoedema treatment including compression bandaging, skin care, exercises, weight management and massage, can improve the effects and symptoms of the condition. However, this requires lifelong patient compliance in wearing the compression garments to maintain the improvements made and does need continual self-management (Thomas et al 2020, Mihara et al 2014, Rosian & Stanak 2019 & Cornelissen et al 2018).

Lymphoedema is estimated to affect more than 450,000 people in the UK (Lymphoedema Wales Clinical Network 2023), with a recent estimated prevalence in Wales of 23,000 people with the condition (Lymphoedema Wales Clinical Network 2023).

Section 2:3 Assessment, diagnosis, and management

Section 2:3:1 Assessing and diagnosing lymphoedema

A lymphoedema specialist will carry out a full assessment of the patient's oedema. This includes measuring the amount of swelling, checking the patient's skin, and taking a medical history. The lymphoedema specialist will usually stage the patient's lymphoedema. Staging is a way of describing lymphoedema severity. This helps the specialist create a treatment plan that is right for the patient (Cancer Research UK 2023).

According to the International Society of Lymphology (https://isl.arizona.edu/) Lymphoedema stages fall between 0 and 3:

Stage 0: This is when someone is at risk of developing lymphoedema but does not have signs of swelling. For example, a person who has had cancer treatment involving lymph nodes. This could be surgery, radiotherapy, or both. This stage may last months or years and some patients will not progress to develop lymphoedema.

Stage 1 (mild lymphoedema): The swelling starts to appear, and the skin can look puffy. In this stage, the swelling might be relieved, for example when a limb is raised. The skin feels soft and may leave an impression when pressed. This is known as pitting oedema.

Stage 2 (moderate lymphoedema): The swelling increases and does not go away when the limb is raised. The skin may become dry and feel thick. The skin becomes tighter as the subcutaneous fat under the skin increases. Pitting oedema may be seen but this is more difficult to assess due to the thickening (fibrosis) of the skin.

Stage 3 (severe lymphoedema): The skin can become very dry and thick and may look darker in places. Skin changes can appear and wart like growths (papilloma) can form.

With swelling in an arm or a leg (limb), the specialist can compare the swollen limb with the unaffected one. The specialist can assess how much swelling the patient has and they can also measure the swelling of the affected limbs in different ways. The most common way is with a tape measure. Starting with a point on the hand or foot, the specialist can make marks at regular intervals up the affected arms or legs. They can measure around the limb (the circumference) at those intervals (circumferential limb measurements).

They will work out whether the swelling is:

- Mild
- Moderate
- Severe

Section 2:4 Current management of lymphoedema

Lymphoedema Network Wales have produced the following 5 important issues relating to the management of lymphoedema. (Appendix 1).

This current medical treatment is supported by many Lymphoedema and cellulitis and societies, both nationally and internationally. The British Lymphology Society (BLS) (www.thebls.com) was set up in 2017 and hosts several events throughout the year. Their annual conference is attended by both members and Lymphoedema patients, as well as industry exhibitors and sponsors. They also promote Lymphoedema Awareness week every March, and they also recognise and mark World Lymphoedema Day (6th March) with online promotion and dissemination of current Lymphoedema. For example, in February 2023, the BLS issued major revisions to the guidelines on the management of cellulitis in Lymphoedema: (https://www.thebls.com/documents-library/guidelines-on-the-management-of-cellulitis-in-lymphoedema) and in October 2024 they launched new guidance documents on; BLS Manual Lymphatic Drainage Position Document (https://www.thebls.com/documents-library/british-lymphology-society-manual-lymphatic-drainage-position-document) and Practical Issues in Applying Compression Garments and Wraps for Management of Lymphoedema (https://www.thebls.com/documents-library/practical-issues-in-applying-compression-garments-and-wraps-for-management-of-lymphoedema) and finally, Managing Lymphoedema in the Presence of Heart Failure - an educational resource (www.thebls.com). Additionally, to the medical guidance and network that the BLS has created, they also offer research grants to further Lymphoedema research throughout the UK.

Another influential and important charity/society that raises awareness of Lymphoedema and its co-morbidities is Legs Matter (www.legsmatter.org). This organisation is a coalition of health professionals, partners and charities who come together to offer advice on feel, legs, swelling, wounds and everything linked to Lymphoedema, venous insufficiency and podiatry. Legs Matter hold UK wide ‘Roadshows’ to disseminate the up-to-date guidance and research to health professionals and patient groups. Similarly to the BLS, Legs Matter also have an annual awareness week where patients are encouraged to download resources, share stories, and are also encouraged to talk to their respective Member of Parliament (MP) about the quality of care they are currently receiving.

In March 2019, the Commissioning Guidance for Lymphoedema Services for Adults in the United Kingdom was published by The National Lymphoedema Partnership. This document, which was supported by Macmillan Lymphoedema Association, Breast Cancer Care, Welsh Wound Innovation Centre, BLS and the Lymphoedema Support Network. The document's aim was to "provide guidance for commissioners to improve commissioning of Lymphoedema services across the UK regardless of cause". They would do this by explaining what lymphoedema/chronic oedema is, including risk factors and the impact it has on the individual; explore why providing care matters, including likely prevalence and the economic case; evidence the current provision across the UK; describe what 'good lymphoedema/chronic oedema care' looks like; and outline the short and long-term recommendations of the National Lymphoedema Partnership. This guidance references Paper 2 of this thesis and also refers to the future publication of the Paper 3 of this thesis.

In 2020, Brown and Sneddon published 'Lymphoedema service provision across the UK: a national survey'. This paper aimed to understand how Lymphoedema services are delivered and funded across the UK. The authors also aimed to quantify the services' levels of resource. Members of the BLS were asked to complete a survey consisting of 19 questions about their services. The study concluded that yet more information is needed to understand the scope of the issues faced by Lymphoedema services. However, two outcomes of the survey were the production of a Tariff Guide (www.thebls.com) and a Cost Calculator (www.thebls.com) With the limitations of this survey acknowledged, the authors intend on the BLS repeating this survey before the revision date of the National Lymphoedema Tariff Guide.

In terms of current Lymphoedema research, LIMPRINT (www.lympho.org/) which stands for IMpact and PRevalence INTernational Lympoedema Framework, is the international leader and disseminator of current Lymphoedema research.

Initially set up as the UK Lymphoedema Framework in improving the management of the condition in the UK, the work led by Christine Moffatt CBE and her team attracted the interest of many leading international experts who led Moffat to set up the International Lymphoedema Framework (ILF) as a charity and which is currently formed of 19 international networks. In terms of LIMPRINT, this is an international study aimed at capturing the size and impact of chronic oedema in different countries and health services across the world.

The focus of LIMPRINT is to provide evidence to support the development and reimbursement of lymphoedema services. Carried out in close collaboration with participating national lymphoedema frameworks, LIMPRINT has demonstrated the possibility of global research within a charitable framework and the power of strong academic partnerships to ensure delivery of robust evidence from each country.

Between 2014 and 2017 9 countries with 40 sites have contributed to an international data set of over 13,000 patients. The capturing of new data is now focused on low resource countries. In 2019, Moffat et al published their paper The Concept of Chronic Edema—A Neglected Public Health Issue and an international Response: The LIMPRINT Study which laid out the key principals of what the LIMPRINT study aimed to achieve via a systematic review and to what its exact goals were. The follow up 2019 study by Moffatt et al then laid out the development and validation of the LIMPRINT methodology and its creation of 9 stages. 2025 sees the 12th International Framework Conference which will be held in Niagara Falls, Canada. Wounds International (www.woundsinternational.com) also have an international visibility with annual conferences and webinars concentrating on different aspects of wounds, as well as also having an emphasis on Lymphoedema research through their own Journal of Lymphoedema which is currently on its 19th volume.

Other notable journals that focus on Lymphoedema research include Journal of Lymphology (https://journals.librarypublishing.arizona.edu/lymph/) which is currently on its 57th volume. The European Journal of Lymphology (https://www.eurolymphology.org/about/the-european-journal-of-lymphology) is currently on its 34th volume. There are also the British Journal of Nursing (https://www.britishjournalofnursing.com/) and the British Journal of Community Nursing (https://www.britishjournalofcommunitynursing.com/) both of which I published in and are featured in this thesis. Lastly, Wounds International (https://woundsinternational.com/) Wounds UK (https://wounds-uk.com/) , The Journal of Wound Care (https://www.journalofwoundcare.com/) and the International Wound Journal (https://onlinelibrary.wiley.com/journal/1742481x) are all wounds focused journal, but which also feature heavily Lymphoedema and Cellulitis research as a sequelae of wounds and infection.

One of the first real attempts to quantify the cost burden of Lymphoedema was undertaken by Moffat CJ et al (2003) where the authors estimated that patients hospitalised by Lymphoedema with a mean length of stay of 12 days, was around £2,300 per patient. Atkins L (2016) aimed to quantify the cost burden of cellulitis. The author estimated that an estimated 400,000 bed days annually are spent on the admission and treatment of patients with cellulitis. This equated to between £172 - £254 million pounds per year.

A recent letter (Pinar Karaca-Mandic et al 2023) discussed the benefits of patient-centred innovations for individuals with BCRL (Breast Cancer-Related Lymphoedema). The authors stated that these innovations can improve adherence to self-care and reduce complications and costly healthcare utilization. A systematic review by De Vrieze et al (2020) looked to quantify the economic burden (both on patients and society) of BCRL. The authors concluded that the review found a lack of recent BCRL related economic analyses in Europe, Asia, Africa and South America. Additionally, the authors recommended that further scrutiny of the economic impact of DLT (Decongestive Lymphoedema Treatment) for BCRL in clinical settings is needed worldwide. A Belgium randomised controlled study by the same authors (De Vrieze et al (2021)) stated that their study was one of the first health economic analyses of BCRL treatment in Europe. The researchers estimated per patient direct healthcare costs of €2,248.93, with 80% (€1,803.35) of costs accounting for statutory health insurances, and 20% (€445.58) identified as patients' out-of-pocket expenses.

Section 2:5 Health Economics and Economic Evaluation

Health economics is concerned with the problem of allocating health care resources under conditions of scarcity and uncertainty. With increasing demands on NHS resources and limited funding, the economic impact of new treatments has never been more important or influential (NIHR 2023). Economic evaluation seeks to inform the difficult range of options and unavoidable decisions in healthcare management (Drummond et al 2015). Economic evaluation has two features. It deals with the inputs and outputs that form part of the evaluation. These can be referred to as the 'costs' and 'consequences' of the proposed alternative courses of action. A health economist conducting an economic evaluation is usually interested in the resources needed to deliver a new treatment and any comparative treatments i.e., cost of control intervention versus cost of new intervention. Any resulting possible savings (or increase in costs) seen in terms of need for future health and social care and/or benefits to patients, their families or society as a whole, are reported in a disaggregated way to allow for full transparency for decision makers. The results of economic evaluations are used by health care decision makers such as government bodies, (National Institute for Health and Care Excellence (NICE)), to determine whether to recommend the adoption of new treatments into routine practice (National Institute for Health and Care Research (NIHR) 2023).

For the development of NICE guidelines, the preferred measure of health outcome is the Quality Adjusted Life Year (QALY) calculated using the EQ-5D-5L (EUROQOL 2023). The EQ-5D-5L is a brief, five-dimension measure of Health-Related Quality of Life (HRQoL). The EQ-5D-5L has five response levels for each dimension and the aim of the EQ-5D-5L is to improve the sensitivity of the EQ-5D to smaller changes in health status (NIHR 2023). A health economist can advise on alternative measures of HRQL or health outcome that may be more relevant to the disease area (NIHR 2023).

Economic evaluation can be used to inform decisions about the economic impact and relative value for money of digital health products. It can tell whether differences in costs between the product and competing alternatives can be justified in terms of health and non-health benefits (Gov.UK 2023). The study perspective is the point of view that will be taken for the economic evaluation. This is important because it determines which costs and effects are relevant to analysis and need to be captured. For example, the reduction of psychiatric hospital beds might seem cost-effective from the perspective of the NHS, but less so from the perspective of society, including patients or carers. A societal perspective helps detect cost shifting between different sectors, for example formal and informal care, or health and economy sectors (Gov.UK 2023).

Public health professionals can use economic evaluation to identify, measure, value, and compare the costs and consequences of different public health interventions. Allocating resources and implementing these interventions—whether policies or programs—requires an understanding of the relationships between resources used and health outcomes achieved by the program or intervention (Centre for Disease Control and Prevention 2023).

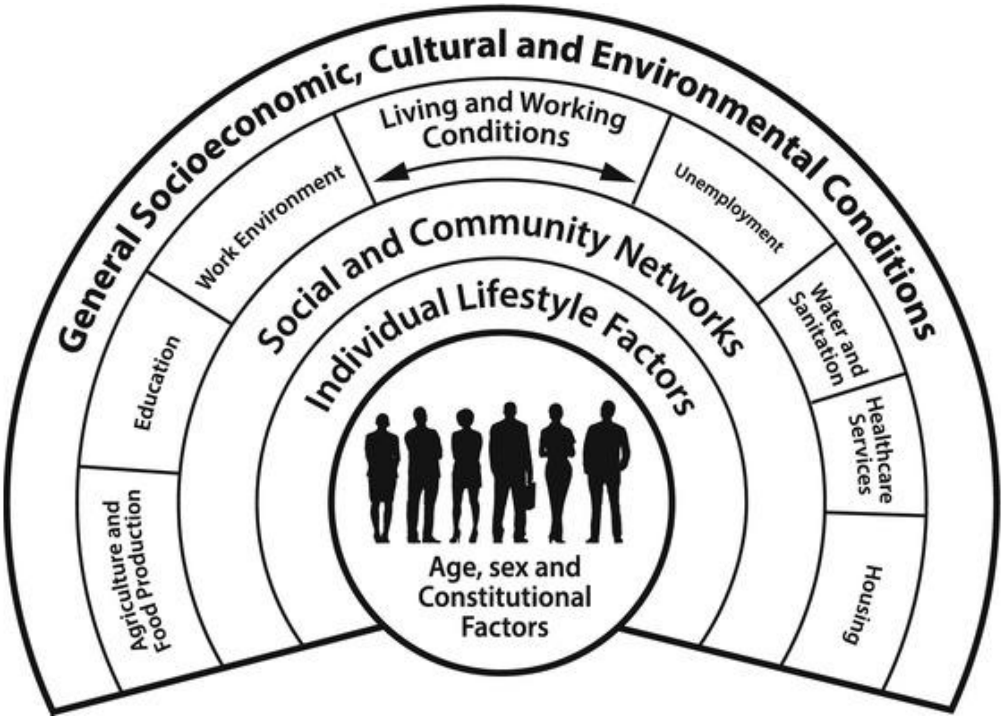
Section 2:6 Quality of Life, Health Related Quality of Life (HRQoL)

Quality of life (QoL) is a broad and multidimensional concept that includes and combines subjective evaluations of both positive, and negative aspects of a person’s life (Centers for Disease Control and

Prevention (CDC) (2023). What makes QoL challenging to measure is that although the term 'quality of life' has a meaning for nearly every person, every academic principle, individuals, and groups can define 'quality of life' differently (CDC 2023). Although health is one of the most important domains of overall 'quality of life', there are other domains/influences that that will have an impact (positively or negatively) on a persons' perceived 'quality of life'. These domains can include, housing, schools, transport infrastructure, accommodation, etc.

In 1991 two researchers Dahlgren & Whitehead developed the infographic shown in Figure 1. The infographic depicted all the elements and domains that the researchers felt had an effect on a person's health and wellbeing. The authors believed that it was all these factors that had a real impact and not just the perceived effect of a person's health and their access to health care services. From figure 1, it can be seen Health care services actually form quite a small part of the elements that the authors define as the determinant of health.

Figure 1. The main determinants of health (Dahlgren & Whitehead 1991)



Additionally, aspects such as culture, the arts, values, religion, and spirituality can also add an extra layer of complexity to the 'quality of life' measurement (CDC 2023).

The concept of health-related quality of life (HRQoL) and its determinants have evolved and grown since the 1980s to show that the overall 'quality of life' of a person, can affect their health – either physically, mentally, or both (CDC 2023). Broadly, a person's HRQoL includes physical and mental health as perceived by that person at that moment in time. This will include, but not limited to:

- Energy
- Anxiety
- Mood
- Tiredness
- Pain
- Mobility
- Financial security

- Job satisfaction
- Family life

All these issues will have a direct link to not only an individual's functional status, but their current socioeconomic status, social support status and overall health risks (CDC 2023). Time off work for health reasons is a significant economic burden at both a healthcare system and societal level throughout the world. For example, in Nagata et al's 2018 cost burden study, the researchers estimated the absenteeism, presenteeism, and medical/pharmaceutical expenses in four Japanese pharmaceutical companies. Absenteeism was estimated at $520 per person per year, with presenteeism estimated at $3055 per person, per year. Additionally, two of the highest total cost burdens from chronic illness were related to mental health conditions and musculoskeletal disorders (Ngata et al 2018).

In terms of health economics and economic evaluations, tracking a person's HRQoL over time is invaluable as it can help identify poorly perceived health in certain subgroups of people in the population and aid health care services to target those people in an efficient and possibly more cost-effective way, rather than continuing without specialist care.

The impact of health-related problems on people's QoL, and assessment of the HRQoL, has been a complimentary aspect to health care discipline and integral to most economic evaluations for the last 30 years. Many health care interventions and services do not directly affect life expectancy but can have a major impact on the ability of the patients to function and undertake normal duties. The HRQoL embraces a range of dimensions relating to a person's physical and mental capacities and can provide an indication of the utility that a person derives from receipt of a service.

Section 2:7 Patient Reported Outcome Measure (PROM)

Patient-reported outcome measures (PROMs) are used by healthcare specialists, clinicians, and researchers to assess the quality of healthcare experiences, specifically focusing on patients. These measures help healthcare providers, policy makers, commissioners, and other stakeholders to make informed changes to their services, based on the statistical results of the PROMs. Evaluating the effectiveness and experience of care using patient-reported outcomes is fast becoming standard practice in healthcare. It has been widely used in randomised controlled trials and service evaluations assessing the effect of new interventions, medications, medical procedures, and system changes. Utilising Patient Reported Outcome Measure (PROM) instruments like the aforementioned EQ-5D-5L (EUROQOL 2023), the Short Form 36 (SF-36) (SF-36 2023) or the Short Form 12 (SF-12) (SF-12 2023) (which both produce the SF-6D score) can help researchers and clinicians map out and track a persons perceived HRQoL longitudinally.

Survival analysis is concerned with studying the time between entry to a study and a subsequent event (BMJ 2023). Originally the analysis was concerned with time from treatment until death, hence the name, but survival analysis is applicable to many areas as well as mortality (BMJ 2023). Utilising the Kaplan-Meier estimate provides the simplest way of computing the survival over time in spite of all the difficulties associated with subjects or situations. For each time interval, survival probability is calculated as the number of subjects surviving divided by the number of patients at risk. (Goel et al 2010)

Any changes in this utility score can be combined with the survival analysis outcome to generate the Quality Adjusted Life Year gained (QALY), which embraces both quality and quantity of life and provides a common currency for measuring health gains resulting from a health care intervention. The above-mentioned instruments are generally regarded as generic PROM instruments that can be used to track a person’s HRQoL, but they can also be validated for certain diseases or health conditions.

As well as these ‘generic’ PROM instruments, certain disease and conditions specific PROMs have been developed and validated by researchers across the globe. These instruments can vary in length and scope and can address several dimensions such as physical health, mental health, self-care, emotional health and social health.

A good example of a disease specific PROM is the Multiple Sclerosis Quality of Life-54 (MSQOL-54) This is a multidimensional HRQoL measure that combines both generic and MS –specific items into a single instrument (Vickery et al, 1995). In developing the instrument, the researchers actually combined the SF-36 as the generic component, and then added 18 items to tap into MS-specific issues such as fatigue, cognitive function, etc. (Vickery et al, 1995).

There is no overall single score for the MSQOL-54. Instead, two summary scores - physical and mental health – can be derived from a weighted combination of scales scores (Vickery et al, 1995).

Being able to calculate these patients perceived scores at differing time points can allow researchers to quantify and also track the trajectory of disease over time during a randomised trial or some other clinical research study.

When administering PROMs to patients and/or research participants, the researchers or clinicians must be mindful that they are not overburdening the respondent with numerous PROMs and other health related questionnaires. This can lead to participant response fatigue, and possible study drop out. However, administering more than one PROM can be advantageous as seen in the study by Humphreys et al (2013) when several questionnaires were administered to the cohort of participants. These included the General Health Questionnaire 12 (GHQ12) (Goldberg 1972), Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith 1983), EQ-5D (EUROQOL 2023) and Beck's Depression Inventory II (BDI-II) (Beck et al 1996), which is a measure of the severity of depression. Once the measures were all collected and analysed, there was a significant effect of the intervention on mood (as collected by the Beck Depression Inventory) but not on quality of life. This was because the intervention was targeted at mood, and the EQ-5D assesses other aspects of quality of life that would not be affected by the intervention. Therefore, the authors were able to report more positive results relating to the more specific mood related PROM, rather than the results generated by the more generic EQ-5D PROM (Humphreys et al 2013).

The original EQ-5D three-level (3L) version was introduced in 1990. The standard paper-based, self-complete version consists of a title page, the descriptive system (on page 2), and the EQ VAS (on page 3). [1] The EQ-5D was rooted in the desire to “create a way of measuring health status to inform resource allocation decisions by enabling the application of cost-effective analysis to health care” [2]. The measure grew in popularity in 2004 after the National Institute for Health and Care Excellence (NICE) in the UK identified it as a preferred instrument for assessing the economic value of new medical technologies [3].

One of the most common criticisms of the EQ-5D-3L/5L was its potential lack of specificity and sensitivity to certain conditions and diseases. It was billed as a generic HRQoL prom tool, even though it was validated for certain diseases and conditions as its usage got more extensive. A 2016 study investigating it’s validity and responsiveness for Chronic obstructive pulmonary disease (COPD) [12] found there were moderate-to-strong correlations between utility index and EQ-VAS with disease-specific questionnaires and that they found it to be a valid and responsive measure for COPD. Additionally, a more recent 2022 study [13] found that fatigue and cognitive problems of Q-Fever or COVID-19 patients was partially captured by the EQ-5D-5L dimensions. They also concluded that adding a fatigue dimension to the EQ-5D-5L would increase the sensitivity and specificity of the measure, in particular, it’s relationship with the VAS. A 2023 study [14] found similar, but more mixed results when it came to patients with long-term post COVID-19 symptoms. The study again found a positive relationship with the VAS, but very little agreement with the EQ-5D-5L index.

Gnanasakthy and DeMuro’s 2024 editorial discusses the EQ-5D-5L’s limitations as a Clinical Outcome Assessment (COA) Tool when using it for certain health issues. In particular they reference the HER2CLIMB breast cancer study [4] where the Taucatinib Safety Profile [5] said that the “HRQoL was preserved”. Gnanasakthy and DeMuro’s 2024 editorial however, discuss that the conclusion ignored most common side effects of tucatinib (e.g., diarrhoea, nausea, vomiting, hand-foot syndrome) [5] and that these are not specifically covered by EQ-5D.

Janssen MF et al (2022) aimed to compare the responsiveness of EQ-5D-3L (3L) with EQ-5D-5L (5L) descriptive systems and value sets in two independent samples (rehabilitation and stroke patients). The researchers concluded that descriptive results were the main driver of the differences between EQ-5D-3L and EQ-5D–5L responsiveness. The responsiveness of EQ-5D-3L was influenced by the 'confined to bed' label and the overestimation bias of EQ-5D-3L, which affected all responsiveness results. The authors surmised that this limitation on behalf of that item within the dimension of 'Mobility' may on the calculation of the quality-adjusted life-year (QALY) estimations. And that may lead to an over or underestimations of QALYs gained, depending on the health condition and its severity. They concluded that QALY calculations based on EQ-5D-5L data will result in more accurate estimates.

Snowden DA et al (2024) looked at the 'consumer acceptability of routine use of the EQ-5D-5L' within a clinical care healthcare setting. The authors concluded that the EQ-5D-5L is "acceptable for use in care but does not capture all aspects of health relevant to clients".

Keetharuth AD et al (2022) looked to assess the psychometric performance of EQ-5D-5L in dementia vis a systematic review. The authors conclude that the "EQ-5D-5L seems to capture the health of people with dementia on the basis of known-group validity and convergent validity. But yet the "evidence is inconclusive regarding the responsiveness of EQ-5D-5L" as the disease progresses. Tellingly, within this cohort of patients, "the ability to self-complete EQ-5D-5L is diminished", which is a big concern for the EQ-5D-5L's ability to represent all patients.

It's also important to consider the relationship between the EQ-5D-5L and other 'generic' PROMs that have been developed. It's important to see how well the instruments 'map' with each other and also how the 'sensitivity' and 'specificity' changes between the instruments' scoring mechanisms. Mulhern BJ et al (2023) study 'Understanding the measurement relationship between EQ 5D 5L, PROMIS 29 and

PROPr' looked at and assess the measurement relationship between the EQ-5D-5L descriptive system and value sets, the PROMIS-29 and PROPr (PROMIS value set). The authors witnessed some consistencies in what the EQ-5D-5L, PROMIS-29 and PROPr measure and produce. The authors also found that the differences between value set characteristics can be linked to differences in what is measured and the valuation approaches used. They saw the implications of this for the use of assessing health outcomes, and they concluded that 'the results can inform decisions about which instrument should be used in which context'.

Verstraete J and Scott D (2022) investigated the comparison of the EQ-5D-Y-5L, EQ-5D-Y-3L, with the PedsQL in children and adolescents. The researchers found that both the EQ-5D-Y-3L instrument and EQ-5D-Y-5L showed comparable psychometric validity to the PedsQL overall. Additionally, when the researchers looked at the choice between the EQ-5D-Y-5L and the EQ-5D-Y-3L, they found no systematic difference in the validity between these instruments or between the EQ-5D-Y-5L and the EQ-5D-Y-3L and the PedsQL

Finally, Churchill K et al (2021) looked at implementing the EQ-5D-5L as a routine outcome measure in Community Outpatient and Specialized Rehabilitation Services and their Rehabilitation Model of Care (R-MoC). This is very similar to what LW were wanting to implement within their OGEP service as their HRQoL tool, and what would eventually become the LYMPROM tool. Even though the researchers encountered particular barriers to the implementation of the EQ-5D-5L as part of the R-MoC, they found that implementing as part of routine data collection in community rehabilitation, has increased the quality improvement culture with the clinical teams collecting and analysing data. It has informed both the characteristics and the health-related quality of life of patients accessing community rehabilitation and will help direct where future efforts need to be focused.

As can be seen, the EQ-5D-5L maps well to some disease/condition specific outcome measures and is generally seen as a useful generic PROM for measuring the health-related quality of life at certain points in time. However, there are some issues with sensitivity and specificity that makes it unsuitable for some conditions. Finally, some PROMs are designed and developed to specifically measure the mental health and wellbeing of the population. One of the most widely utilised PROM in this field is the Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS) (Tennant et al 2007). The WEMWBS was developed to enable researchers to monitor the mental wellbeing of the general population and to be used in the evaluation of projects, programmes and policies that aim to improve mental wellbeing (Tennant et al 2007). The WEMBWS is a 14-item scale of positively worded statements covering feelings and functioning aspects of mental wellbeing. The 14 statements have five response categories:

Table 3. Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS)

STATEMENTS	**None of the time**	**Rarely**	**Some of the time**	**Often**	**All of the time**
I've been feeling optimistic about the future	1	2	3	4	5
I've been feeling useful	1	2	3	4	5
I've been feeling relaxed	1	2	3	4	5
I've been feeling interested in other people	1	2	3	4	5
I've had energy to spare	1	2	3	4	5
I've been dealing with problems well	1	2	3	4	5
I've been thinking clearly	1	2	3	4	5
I've been feeling good about myself	1	2	3	4	5
I've been feeling close to other people	1	2	3	4	5
I've been feeling confident	1	2	3	4	5
I've been able to make up my own mind about things	1	2	3	4	5

I've been feeling loved	1	2	3	4	5
I've been interested in new things	1	2	3	4	5
I've been feeling cheerful	1	2	3	4	5

The WEMWBS has also been developed into a shorter version (SWEMWBS) which has reduced the 12 statements to 7 (Shah et al 2021). This version of the WEMWBS uses 7 of the statements about thoughts and feelings, but which relate more to functioning, than feelings. This therefore gives a slightly different perspective on mental wellbeing in the population being asked (Shah et al 2021).

Section 2:8 Prudent Healthcare

The challenge to improve health and drive forward an excellent and fair health and social care system for Wales within an era of increasing demand, increasing inequality and severe financial restraint, is considerable. This dilemma continues to exercise Welsh Government, health boards, trusts and local government (Bevan Commission 2014). However, this is not unique to Wales, these same issues face other healthcare systems within the UK and internationally. Immediate collective action is required to ensure that the best outcomes are obtained for individuals, most fairly within the resources available. This needs a comprehensive understanding of what is already being achieved, where the gaps are and how health services can be improved. A fundamental culture change is also required to embed a prudent approach to health at scale and pace, not only across the health and social care systems, but also with members of the public and the media (Bevan Commission 2014).

In response to this fundamental need for change at a health services level, the Bevan Commission outlined its approach and thinking through its concept of Prudent Healthcare. In its initial discussion paper entitled 'Simply Prudent Healthcare – achieving better care and value for money in Wales'. In this paper, Prudent Healthcare was defined as ‘healthcare which is conceived, managed, and delivered in a cautious and wise way, characterised by forethought, vigilance and careful budgeting which achieves tangible benefits and quality outcomes for patients’. A prudent approach to health aims to support health and well-being, providing, when needed, care that fits the needs and circumstances of the individual and actively avoids ineffective or duplicative care that is not to the patients’ benefit or fair. It is about being wise and accountable for how public money is spent. By placing greater value on patient outcomes rather than the volume of activity and procedures delivered, prudent healthcare aims to rebalance the NHS around the patient or population it serves. An NHS based upon prudent healthcare principles aims to ensure that patients receive the most appropriate care or treatments to achieve mutually agreed goals, reflecting the contribution individuals and communities can make to their own health and wellbeing. This requires healthcare and other professionals working with government to use resources effectively and efficiently, and where wasteful allocation of scarce resources is generally regarded as being unacceptable and an injustice. There is also a requirement for the public to be mindful of the way in which they use NHS resources. Prudent healthcare requires co-production at an individual and population level where patients and the NHS each contribute to improve health and wellbeing (Bevan Commission 2014).

Section 2:9 Value-Based Healthcare

In Wales, value-based care is underpinned by Prudent Healthcare, first launched as a philosophy and a policy in January 2014. The key principles of coproduction, equity, intervening gently and effectively (and only as much as is needed) and reducing unwarranted variation (including under and over-treatment), are all vital to achieving value for patients and citizens across a whole system of health and social care. Prudent Healthcare has provided a strong foundation for healthcare improvement in Wales, and it is believed that a Value-Based healthcare approach will help to make the aims of Prudent Healthcare a reality (Value-Based Healthcare for Wales 2021)

In their paper published in 2021, ‘A Healthier Wales’, the Welsh Government outlined their plan for health and social care. The paper set out the need for health services to change and become fit to meet the challenges that an ageing population presents and how individuals can all help deliver better health outcomes for themselves and others. One key component in this plan was to improve value for patients by giving a greater focus to the outcomes that matter to individuals and considering their own role in contributing to the costs of achieving those outcomes.

The value for the patient relates to the whole cycle of care not just a single intervention. Frequently in healthcare, the outcomes that matter to patients, such as consistency, quality, timeliness, therefore Value-Based healthcare encourages the focus on patient-reported outcome measures (PROMs) as well as clinical outcomes.

A National Action Plan for Value-Based healthcare in Wales was launched in the autumn of 2019, setting out a three-year programme to embed the Value-Based healthcare approach as part of making Prudent Healthcare philosophy a reality (Value-Based Healthcare for Wales 2019).

Health and care organisations in Wales are committed to developing their Value-Based healthcare approaches. This includes better collection and reporting of outcome data for a range of medical conditions and looking at unwarranted variation in services and outcomes to reveal the under and over-use of different aspects of health care (Value-Based Health Care for Wales 2019).

Coming up in the next section of my thesis are PDF copies of each of the papers, the critical review, the discussion and the conclusion of the thesis.

Chapter 3

Publications

The next chapter of this thesis is a PDF copy of each page of all eight papers presented here in this PhD by published works.

Section 3:1 Paper 1 Managing chronic oedema in community settings I HUMPHREYS, MJ THOMAS, KM MORGAN - Wounds UK, 2017 Vol. 13 Issue 3, p22-33. 13p.

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

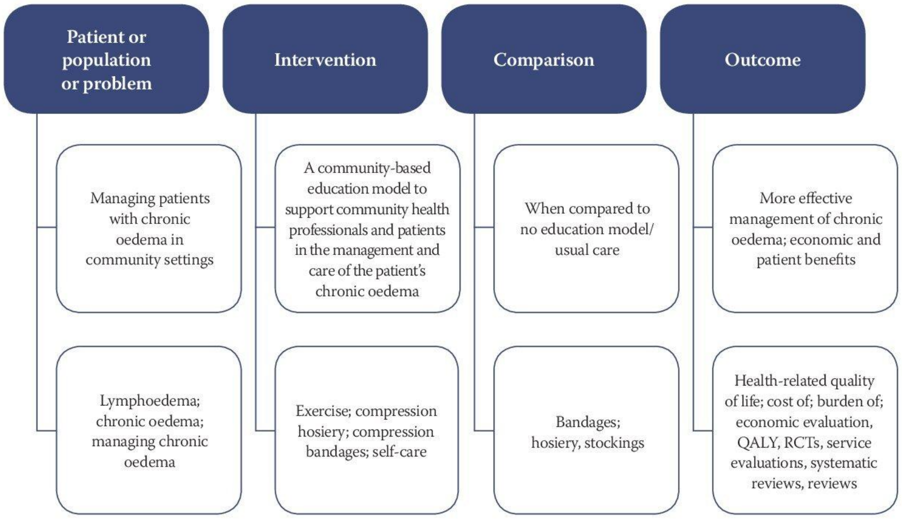


Table 2. Inclusion criteria applied in literature search

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.

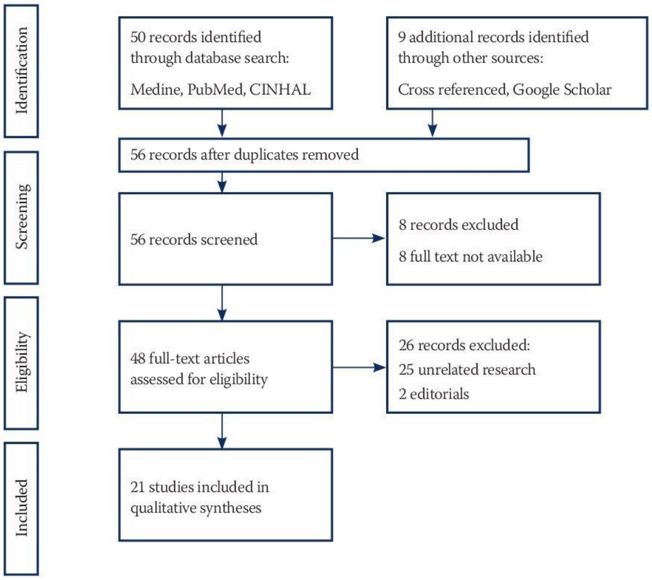
RESULTS

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 (23–33 mmHg) 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. *Am J Surg* 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. *SpringerPlus*. 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. *PLoS Negl Trop Dis* 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. *Eur J Cancer Care (Engl)* 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 *SpringerPlus*. 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. *Eur J Oncol Nurs* 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. *J Wound Care* 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. *Cochrane Database Syst Rev.* 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. *Int J Nurs Stud* 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. *Palliat Med* 29(2): 112–9	2015	Leung EY, Tirlapur 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. *Eur J Vasc Endovasc Surg* 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 lipoedema treatment (PBI-L)	Development and validation of tool	All lymphoedema and lipoedema 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. *PLoS One.* 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. *Support Care Cancer.* 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. *European Journal of Vascular and Endovascular Surgery* 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. *Eur J Vasc Endovasc Surg* 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 extremety 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). *Lymphology* 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. *Breast* 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. *Int Wound J* 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. *Breast Cancer Res Treat* 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. *Breast J* 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. *Ann Surg Oncol* 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

Section 3:2 Paper 2 Evaluation of the economic impact of a national lymphoedema service in Wales I Humphreys, MJ Thomas British Journal of Nursing 26 (20), 1093-1100

Evaluation of the economic impact of a national lymphoedema service in Wales

Ioan Humphreys and Melanie J Thomas

Lymphoedema is a chronic condition that results in swelling in the body's tissues, caused by a failure of the lymphatic system. It can affect any part of the body, but usually develops in the arms or legs (NHS Choices, 2017). It is an incurable condition and requires lifelong management (Moffatt et al, 2003). Lymphoedema can affect people of all ages and can occur in limbs, the head and neck, trunk or genital area. It may occur after trauma or surgery, sometimes not becoming apparent for some time, and patients may remain at risk of developing lymphoedema for life (Morgan et al, 2005).

ABSTRACT

Lymphoedema Network Wales has focused on maximising the impact of its service through the effective use of available resources to ensure high-quality and consistent care for people with lymphoedema across Wales. The aim of this evaluation was to estimate the economic impact of a national lymphoedema service on the NHS Wales budget. Work was undertaken to determine the care pathway within Lymphoedema Network Wales and develop a hypothetical 'world without' the service as a comparator. The four groups of patients that made up the pathways were group 0: 'at risk', group 1–2: 'uncomplicated lymphoedema', group 3: 'complicated/complex' and group 4:

[redacted]

The aim of this evaluation was to estimate the economic (cost) impact of Lymphoedema Network Wales on the NHS Wales budget. The objectives were to:

- Quantify the likely cost burden associated with lymphoedema in Wales
- Assess the wider costs to patients and families resulting from lymphoedema
- Estimate the financial benefits associated with the services provided in Wales.

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Table 1. Selected differences in resource utilisation

Resource pre-lymphoedema service involvement	Cost	Resource: post-lymphoedema service involvement	Cost	Cost difference
Baseline GP (surgery)	£15136	6 months GP (surgery)	£6192	-£8944
Baseline GP (home visits)	£8470	6 months GP (home visits)	£1980	-£6490
Baseline district nurse (home visits)	£71540	6 months district nurse (home visits)	£20160	-£51380

[redacted]

that was seen was asked if they wanted to participate, until a maximum of seven participants from each service had been recruited. Abertawe Bro Morgannwg University Health Board recruited eight participants, giving a total of 50.

All data were fully anonymised prior to analysis and ethical approval was granted for analysis of the data from both Abertawe Bro Morgannwg University Health Board and Swansea University.

Development of model structure

The design and structure of the lymphoedema service costing model was based on several inputs, each informed by the patient-related information obtained from interviews with lymphoedema service clinical leads from the university health boards (UHBs) and the RUQs administered to patients. In addition, based on information from the clinical leads, estimated

in the baseline target group. Time and resource constraints meant that this does not include medication and/or appliance costs (such as garments or dressings). However, further studies by the authors have illustrated significant cost savings in dressings with lymphoedema service collaboration (Thomas and Morgan, 2017).

Results

Resource utilisation

The mean resource utilisation for selected categories is shown in *Table 1*. The overall cost savings and resource utilisation between the 6-month pre-lymphoedema service entry and the 6-month post-utilisation for the 50 patients studied is highlighted (*Table 1*) .

There were significant savings to be seen, in particular GP surgery visits (£8944), GP home visits (£6490), practice nurse

Table 2. Lymphoedema service pathway results (all-Wales)

Group 0: 'at risk' total per person cost	**£903**
Population	1015 (11%)
Total estimated resource utilisation cost	£916 053
Intervention cost	£356 012
Overall cost	£1272065
Estimated no pathway cost	£9862409
Cost savings	**£8590344**
Group 1–2: 'uncomplicated oedema' total per person cost	**£1218**

[redacted]

also included community- and hospital-based physiotherapy and occupational therapy, and community pharmacist and ambulance and paramedic costs.

Costing model

The baseline pre-lymphoedema service entry cost per person was £4859. However, this was for 6 months so, to extrapolate to an annual cost, this was multiplied by two to get £9718.

Lymphoedema service pathway costs were combined and compared with the individual running costs of each of the lymphoedema groups, showing cost savings (see *Table 2*). The population of the active lymphoedema caseload in 2015 was 9226 patients in Wales. Estimated annual savings of £8590344 were seen for the group 0 patients and savings of £34148010 for the group 1–2 patients. An estimated £21674569 savings compared with the 'world without' the service estimates were seen for group 3 and an estimated £270644 savings for group 4 patients.

Summary of findings

The potential costs associated with lymphoedema and its

savings are seen for visits to practice nurses, district nurses and physiotherapists and when these are formally analysed in a costing model, these appear to be the main drivers of cost savings. However, caution must be exercised, given the moderate to high uncertainty in these savings being realised, given the threshold analysis undertaken, especially for group 4 (palliative) patients. This small study suggests an immediate need to prospectively evaluate lymphoedema services over a longer period of time;

KEY POINTS

- Untreated lymphoedema is a burden on the NHS
- The commissioning of lymphoedema services across the UK not only improves quality of life of people with lymphoedema but makes health economic savings for the NHS
- This research article provides the first steps in suggesting the potential economic improvements in developing lymphoedema services
- Lymphoedema services are an exemplar of prudent health care

and consider the different models of care delivery across Wales to develop an understanding of what elements drive efficiencies. This evaluation has focused on the healthcare resources used and

[redacted]

£30 000 cost per QALY threshold) (NICE, 2013), were not possible and need to be considered for future evaluations.

The main cost drivers compared with the 'world without' the service are the GP surgery visits, practice nurse and district nurse visits. This will no doubt vary from centre to centre based on local practice. Lymphoedema Network Wales, which has standardised all the care provided in all the Welsh lymphoedema services, uses a system of a one-stop clinic to initiate assessment and treatment immediately in collaboration with other community nursing staff. This may be different to other lymphoedema services in the UK and the authors suggest the differences between the services need to be explored further. All services follow the same protocols and processes, but some may see only cancer patients or do not provide all treatment options.

Conclusion

The evaluation of cancer-related lymphoedema treatment programmes (Shih et al, 2009; Stout et al, 2012; 2013) and the possible consequential post-treatment economic burden provides an excellent scenario for illustrating the complexities involved in attempting to integrate the evidence relating to

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CPD reflective questions

- What types of data do you collect in your department and how could they be used to prove the effectiveness of your service?
- Considering that the NHS has to make efficiency savings, how can you demonstrate that your service provides 'prudent healthcare'?
- How can you showcase the service improvements for your patients as well as the cost improvements to help managers understand the difference your service can make?

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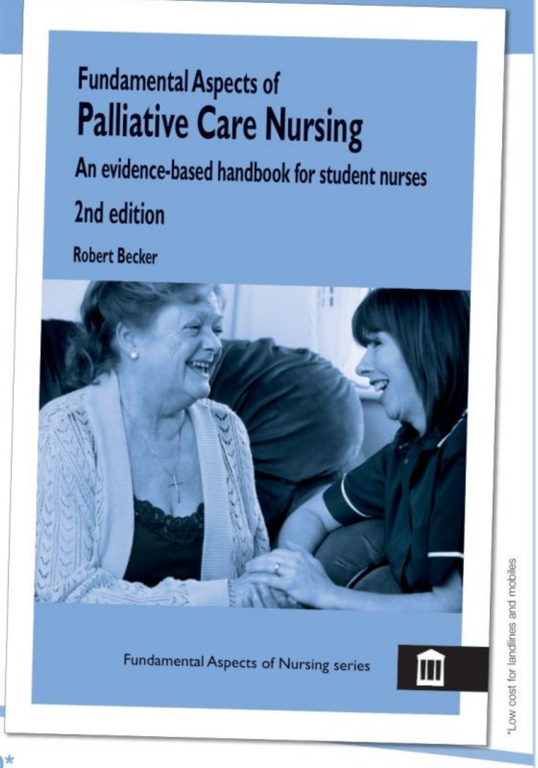

Section 3:3 Paper 3 Pilot evaluation of the management of chronic oedema in community settings project I Humphreys, MJ Thomas, KM Morgan British journal of community nursing 22 (12), 578-585

Pilot evaluation of the management of chronic oedema in community settings project

Ioan Humphreys MSc, Melanie J Thomas FCSP, Karen M Morgan RGN

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Chronic oedema is a debilitating, enduring condition connected with several chronic conditions, primary and secondary lymphoedema, obesity and immobility, with prevalence greatest within the older population (Todd, 2013). It is characterised by atypical swelling lasting for more than 3 months. Those affected may experience skin changes, recurrent cellulitis, superficial ulceration, exudate, lymphoedema and enduring pain, reduced mobility and discomfort (Todd, 2013). Evidence suggests that the impact of chronic oedema/leg ulcers on an individual's health, wellbeing, sense of self and quality of life may be profound and extends to all those who are important to the individual.

As a chronic debilitating condition, chronic oedema can have significant impact on health outcomes and result in a significant burden to the NHS. Ineffective prevention and management alongside inappropriate prescribing of dressing and garments have been identified as significant issues and with appropriate management based on clinically and

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alongside improving outcomes and experiences for people living with chronic oedema.

Aim and objectives

The aim of this economic analysis, based on the pilot evaluation of the OGEP, was to estimate the economic impact of the OGEP within one local University Health Board (UHB) within Wales. Specific objectives were to:

at 3 months follow-up.

Conclusions: Our research show health care resource use and costs decreased, while health-related quality of life scores increased.

KEY WORDS

◆ lymphoedema ic oedema ◆ nursing ◆ Wales
◆ district nurse ◆ cancer ◆ economic ◆ impact

- Assess the health care resource use and related costs associated with the delivery of the OGEP compared to 'no OGEP' i.e. the status quo
- Estimate changes in the profile of health services delivered to patients as a result of the OGEP model
- Provide a description of preference-based patient health

(e.g. primary care, secondary care, medication and dressing costs) and valued in pounds sterling using a price year of 2016. The costs were determined from national published sources of unit costs including All Wales Lymphoedema Compression Garment and Wound Care 2017 (All Wales Lymphoedema Compression Garment and Wound Care

[redacted]

Nurse (Band 7) 0.8 FTE	£52	12 hours per week	£17 472	PSSRU (2016) Band 7 - Page 137	Banding as advised by study team
Overall cost of OGEP intervention			**£35 812**		
Cost of OGEP intervention per participant			**£358.12**	**Based on 100 participants**	

Table 2. Summary of direct NHS/Personal Social Services (PSS) health care costs

		Baseline			3 month follow-up			
Time point	**n**	**Sum**	**Mean (SD)**	**Time point**	**n**	**Sum**	**Mean (SD)**	**Difference in means (95% confidence interval)**
Total costs baseline (including PSS costs)	97	£56 729	£5812 (£5870)	Total costs 3 months (including intervention costs)	97	£445 098	£4589 (£5465)	-£1222.90 (-£344.50, -£2101.50)
Total baseline health care costs (excluding PSS costs)	97	£282 539	£2912.77	Total health care costs 3 months (including intervention costs)	97	£173 385	£1787	-£1125.77 (-£1987, -£264)

[redacted]

after. As would be expected, there were numerical differences seen when comparing the mean and median costs, but overall the pattern of observed differences was similar.

domains of the EQ-5D 5L, 43 participants (44%) increased their mobility score; 39 participants (40%) increased their self-care score; 29 participants (30%) increased their usual

Table 3. Patient health-related quality of life outcomes at baseline and 3 months

	n	Mean	SD	95% confidence interval of the difference	*p*-value
EQ-5D 5L utility baseline	97	0.401	0.254	-0.136 (-0.098, -0.174)	<0.0001
EQ-5D 5L utility 3 months	97	0.537	0.231		
	n	**Mean**	**SD**	**95% confidence interval of the difference**	***p*-value**
EQ-5D 5L VAS baseline	97	47.07	15.17	14.69 (10.75, 18.63)	<0.0001
EQ-5D 5L VAS 3 months	97	61.76	18.41		

KEY POINTS

- Chronic oedema is a debilitating, enduring condition connected with several chronic conditions.
- As a chronic debilitating condition, chronic oedema can have significant impact on health outcomes and result in a significant burden to the NHS.

[redacted]

- Reflect on how many patients you have referred to lymphoedema services previously. Do you think this will increase after reading this article?
- If a patient is not improving with their current management, would you reassess? Consider if you would refer or collaborate with other services.
- Do you think in a proactive or reactive manner? When you see patients, check their legs for any oedema and advise all patients on skin care, movement and exercise and refer to lymphoedema services early.
- To Doppler or not to Doppler a patient with chronic oedema? How accurate would an ankle brachial pressure index or toe brachial pressure index be?
- How can the Doppler tool be used as part of an holistic assessment? Remember, doing nothing can lead to an increased risk of cellulitis and complex wounds.

cost reduction when direct health care and PSS costs are considered, with suggestion that resources (and associated costs) are shifted from key areas such as district nursing and dressings. While the small numbers in secondary care resource and costs are limited, there may be areas where observed trends in cost reductions warrant further explora-

OGEP could optimise service capacity is important for the OGEP team when discussing next steps.

Evidence of understanding where the potential cost drivers could be and whether health gains can be measured using instruments such as the EQ-5D 5L are fundamental components of pilot/feasibility work. A further, subsidiary research question from the analysis which may help future evaluations is formal examination of the validity, reliability and sensitivity of the EQ-5D 5L within this patient population. While this was outside the scope of this current pilot evaluation period, further analysis of the data collected could yield important insights into the psychometric and clinical utility of using this preference-based generic health measure in patients with chronic oedema.

[redacted]

Conclusions

The economic analysis has provided a first in-depth examination on the economic and patient impact that the OGEP could make in providing an innovative solution to delivering best standard of care practice. While our findings suggest an observed trend for reductions in cost to NHS Wales when disaggregated resource use and costs are examined, it also shows the possibility for a possible shift of health care resources across key areas of primary care and secondary

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Section 3:4 Paper 4 The benefits of raising awareness of lymphoedema among care home staff M Thomas, K Morgan, I Humphreys, K Hocking, D Jehu British Journal of Nursing 29 (4), 190-198

The benefits of raising awareness of lymphoedema among care home staff

Melanie Thomas, Karen Morgan, Ioan Humphreys, Karl Hocking and Diane Jehu

ABSTRACT

Background: Patients with lymphoedema referred to a lymphoedema service from care homes in one health board area in Wales were often complex cases, with repeated cellulitis, a history of falls and other complications. A pilot project was initiated to develop education and raise awareness of lymphoedema among care home staff. Aims: To enable care staff to identify residents with lymphoedema, promote prompt referrals, raise the importance of skin care and exercise in the management of lymphoedema and estimate the likely costs from complications associated with lymphoedema. Methods: An education tool was developed and 47 care homes were asked

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congenital abnormalities or through the removal of lymph nodes because of malignancy. Obesity, cellulitis, cardiovascular conditions, immobility and ageing are among the factors involved in developing lymphoedema. In an epidemiological study in the UK by Moffatt et al (2017), the crude prevalence of lymphoedema was 3.93 per 1000 of the population rising to 28.75 per 1000 in those aged 85 or over.

The impact of lymphoedema can affect individuals physically, psychologically, emotionally and socially (Thomas et al, 2020). Reports of increasing falls, superficial wounds, difficulty in accessing footwear, anxiety and pain are commonly encountered.

attend and refer. Educating and raising awareness of lymphoedema is very important in maintaining the health and wellbeing of residents of care homes. It could also decrease the risk of further costly complications such as cellulitis, superficial wounds and the incidence of falls requiring possible hospital admissions.

Working with the Welsh Government/1000 Lives Campaign, an education pilot was established for LNW to deliver existing teaching tools developed by the On the Ground Education Programme (Humphreys et al, 2017) directly within the care home setting in one health board area in Wales with a population size of 290 000. The funding in January 2019 supported the employment of a lymphoedema specialist (a role shared between two specialists) working 1 day a week for 6 months. This specialist would be able to disseminate the lymphoedema education tool, consisting of an e-learning package, lymphoedema videos, and lymphoedema literature, including the LNW Wet Leg Pathway (Morgan and Thomas, 2018), and provide interactive group

[redacted]

anonymous data on gender, age, presence of lymphoedema, wound dressings in situ, mobility and footwear. The scoping review would allow greater discussions (away from the residents) and the building of confidence in recognising lymphoedema among non-registered-nurse care staff. Care staff initiated conversations with residents and asked them how their legs were feeling and whether they had any swelling. Carers were asked not to include any residents who they felt would be unsuitable for inclusion, for example if they were unwell, sleeping or were unable to answer questions because of cognitive impairment.

If a carer identified a resident with lymphoedema, a referral form was immediately completed jointly with the lymphoedema specialist instead of referring to a GP. Data were also collected on whether the resident had experienced cellulitis or falls and if a district nurse was attending the home. A second educational visit would be planned for all care homes, including an additional dedicated group education session to consolidate learning.

Data collection measures

All data were collected on-site in the care homes via paper

published sources of NHS unit costs (NHS Improvement, 2018) and the Personal and Social Services Research Unit (PSSRU) (Curtis and Burns, 2018). The analysis was undertaken in MS Excel and SPSS Version 22 for Windows.

Results

Forty-four out of 47 care homes within one health board area in Wales responded to the initial emails/letters and were

Table 1. Intervention costs

Cost items	Unit cost per hour	Total cost	Unit cost source/ description	Comments/ assumptions made
1 nurse (band 7)	£53 x 2hours = £106	£2544	PSSRU band 7	Based on 1day a week over 6months (24 days)

[redacted]

Care staff and the lymphoedema specialist jointly reviewed 960 residents and completed the anonymous data form. Not all residents were included due to cognitive impairments, illness or unwillingness to answer a carer's questions. As shown in *Table 3*, 73% of residents were from a residential home and 27% from a nursing home. Nearly three-quarters were female, and 45% were between the ages of 80 and 89 years.

Lymphoedema identification

Of the 960 care home residents, 262 (27%) were identified as having lymphoedema in the scoping review. Of those, 261 (99.6%) had lymphoedema in their lower limbs, and additionally, 91% of those 261 had bilateral oedema. If the care staff identified lymphoedema, the accompanying lymphoedema specialist coded the severity of swelling based on the LNW Outcome Severity Scale (Thomas and Morgan, 2017). Of the 262 residents, 162 (62%) (17% of 960) were considered to have 'mild' lymphoedema; 67 (25%) (7% of 960) 'moderate'; 16 (7%) (2% of 960) 'severe'; and 17 (6%) (2% of 960) 'complex'. Due to age and immobility factors,

twice; 3 (8%) had fallen 3 times; 2 (5%) had fallen 6 times and lastly, 1 person (3%) had fallen 12 times. Of those 40 residents identified as falling, 28 (70%) had lymphoedema, and only one was attending the lymphoedema service.

Moreover, 31 residents were identified as being unable to wear shoes due to lymphoedema. Of the 960 residents, 134 (14%) were identified as wearing inappropriate shoes (too big/small, flip flops, etc) and had lymphoedema thus would be at an increased risk of a fall. The cost implications of these falls could be estimated at over £170 000 (NHS Improvement, 2018).

Referrals to a lymphoedema service

Based on the education received on lymphoedema, all carers now had increased awareness of the benefits of skin care, exercise and the importance of positioning/elevation. During the scoping review, when a resident was identified as having lymphoedema, a referral was immediately completed. Of the 960 residents, 35 (4%) were already under a lymphoedema service; 702 (73%) did not require a referral, 223 (23%) were referred by the care home/lymphoedema staff. Although 262 residents were identified as having lymphoedema, two

[redacted]

As shown in *Table 6*, reviewing the data further suggests that costs do not start to accrue until the 70–79 age bracket, with the greatest cost usage seen in residents aged 80–89 years (£115 502). Residents identified with lymphoedema show almost twice the level of resource usage compared to a patient with no lymphoedema (£124 404 vs £74 256).

Statistical analysis

A series of Pearson correlation coefficient analyses were undertaken to identify any relationships between the variables collected. The analysis indicated strong correlations between lymphoedema and a risk of falls (*Table 7*).

Table 3. Resident characteristics

Characteristic	Number	Percentage
Type of home		
Nursing home	263	27%
Residential home	697	73%
Total	**960**	**100%**
Gender		

Severe	16	2/16 (13%)
Complex	17	6/17 (35%)
Total		35/262 (13%)

Discussion

This education pilot indicated that there are several benefits to raising awareness and increasing knowledge of lymphoedema for care staff in nursing and residential homes. First, the education itself is inexpensive, and over 6 months can be calculated as around £2 per person educated and £2.65 per resident

Table 5. Wound care visits by district nurses and tissue viability nurses

Wounds requiring district nurses/ tissue viabiity nurse attendance	Number (*n*=11)	Percentage
Yes	6	55%
No	5	45%
Number of wounds requiring visits	**Number of DN/ TVN visits**	**Percentage of patients with wounds requiring visits (*n*=6)**

within care homes in this health board in Wales. Secondly, this education project highlighted the extensive costs attributed to lymphoedema, in particular, the high number of district nurse visits involved (£18 368), cellulitis hospitalisations (£7756), and the cost of falls (£172 536). Additionally, the education intervention instils confidence in care staff, allowing them to undertake referrals rather than first referring to a GP. From this project, 223 lymphoedema referrals should have been made to the local service, identifying an unmet clinical need. Care staff

[redacted]

Lymphoedema under lymphoedema service	Pearson correlation	0.920*
	Sig (2-tailed)	*P*<0.01
	n	960
Lymphoedema severity	Pearson correlation	0.668*
	Sig (2-tailed)	*P*<0.01
	n	960

* Correlation is significant at the 0.01 level (2-tailed)

collect the scoping data. However, if a resident had obvious lymphoedema a referral was suggested where a full assessment could take place

- In some of the homes where families were present, they were keen that their family member who had dementia were involved and answered questions to support the data scoping
- Patients who were sleeping, unwell or receiving palliative care were not included in the scoping review, and this was fully explained to care staff at the start of the education pilot as it would not be ethical. However, if the carer did think that

KEY POINTS

- More than a quarter of residents in a care home setting in one health board area in Wales were identified as having lymphoedema and only 13% of those were known to the local lymphoedema service, portraying an unmet need
- Many of the patient identified with lymphoedema had experienced falls, cellulitis and wounds

[redacted]

as part of the lymphoedema education pilot provides valuable information for further studies.

Conclusion

This educational project has identified the value of raising awareness of lymphoedema within care home settings in a health board in Wales. By improving carers' confidence in identifying lymphoedema they were able to refer residents to a local lymphoedema service directly, negating the need for GP visits. Furthermore, enabling care home staff to understand the importance of skin care and exercises to enhance the role of the calf muscle pump, will decrease the risk of further lymphoedema in other residents.

The analysis has provided the first in-depth examination of the potential economic benefits of education in a residential and

Thomas MJ, Morgan K. The development of Lymphoedema Network Wales to improve care. Br J Nurs. 2017;26(13):740-750. https://doi.org/10.12968/bjon.2017.26.13.740

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CPD reflective questions

- Considering the themes in this article, how many of the patients on your caseloads may have an unmet lymphoedema need?
- What can you do to enhance the safety of patients with lymphoedema in your own clinical setting?
- Consider whether your patients have repeated cellulitis, wounds or are at risk of falling—could they have 'a little bit of limb swelling' that needs treatment to help the other problems?
- If you feel that many of your patients do have suspected lymphoedema, explore what training is available for you locally

Section 3:5 Paper 5 Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation Melanie Jayne Thomas, Karen Morgan, Ioan Humphreys, Rhian Newton 2021 https://doi.org/10.12968/jprp.2021.3.12.490

Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation

Melanie Jayne Thomas, Karen Morgan, Ioan Humphreys, Rhian Newton

Abstract

Compression garments are the mainstay of lymphoedema treatment, yet the process of obtaining by prescription is lengthy, governed by funding, involves numerous professionals with little patient value and often involes copious inaccuracies, delays and errors. This service evaluation estimates the potential impact of changing the way compression garments are accessed from a prescription to a procurement process. The main objective was to estimate the impact and costs of altering the process within the NHS Wales setting. A service evaluation was formally supported by Medicine Management and Lymphoedema Wales Strategy Board to compare the prescription process to a new procuring route. A form was devised so that data was could be collected over 12 months by lymphoedema therapists on each compression garment issued to patients attending two lymphoedema services in NHS Wales. The data collected included information on the garment ordered, costs (prescribed compared to procured), timing between garment ordered and received plus a comparison of the processes. All non-patient identifiable data was entered into a database and comparison analysis was completed to identify any correlations between the variables collected. The project was ratified by research and development as service evaluation. There was a total of 5392 completed patient data forms were included, which when compared to the processes indicated that using a prescription route is overwhelmingly more costly than procuring. Overall costs suggest the potential for substantial savings to NHS Wales (£71.10 per patient) were statistically significant (*P*<0.001). Potential for improved patient outcomes was observed as garments were provided directly on appointment instead of significant delays promoting value-based healthcare.

Key words: Lymphoedema compression garments | prescribing versus procurement | efficiencies | service evaluation | value-based healthcare

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Lymphoedema is a debilitating, enduring condition connected with several diseases including cancer. It is characterised by atypical swelling lasting for more than 3 months and can occur in any part of the body. Those affected may experience skin changes, pain, heaviness, recurrent cellulitis, reduced mobility and psychological distress (Morgan et al, 2005; Greene and Meskell, 2016; Thomas et al, 2020). Evidence suggests that the impact of Lymphoedema on an individual's health, wellbeing, sense of self and quality of life may be profound (Thomas et al, 2020; Greene and Meskell, 2016). As a chronic condition, lymphoedema can have a significant impact on health outcomes and results in a substantial burden to the NHS (Atkin, 2016; Moffatt et al, 2016; Guest et al, 2016; Thomas et al, 2017). Lymphoedema requires ongoing management including skincare, exercise and the daily use of compression garments (Lymphoedema Framework, 2006). Since 2007, compression garments have been accessed through Part 1XA of the drug tariff in the UK, which covers appliances (NHS Business Services Authority, 2021a) . Ineffective management of the prescription process alongside inappropriate prescribing of garments have been identified by patients and healthcare professionals as important issues.

Throughout Wales and in the UK, many patients' access compression garments via a prescription that is completed by the general practitioners (GPs) based on a request from a lymphoedema professional (Woods, 2018). This prescription process has many unnecessary steps, which does not focus on quality of care, safety, and is not a patient-centred experience. These superfluous steps include writing prescriptions or referral to GPs for prescriptions, collecting and taking prescriptions to community pharmacists (CPs), and returning to collect the prescribed compression garment and lastly, for 80% of patients, they have to return to the lymphoedema service for a fitting appointment. The processes on average takes 4 weeks, but some are delayed for over six, resulting in treatment delays. Further, when the garment is wrongly dispensed, the process begins again (*Figure 1*).

As the incidence and prevalence of lymphoedema increases because of improved awareness so does

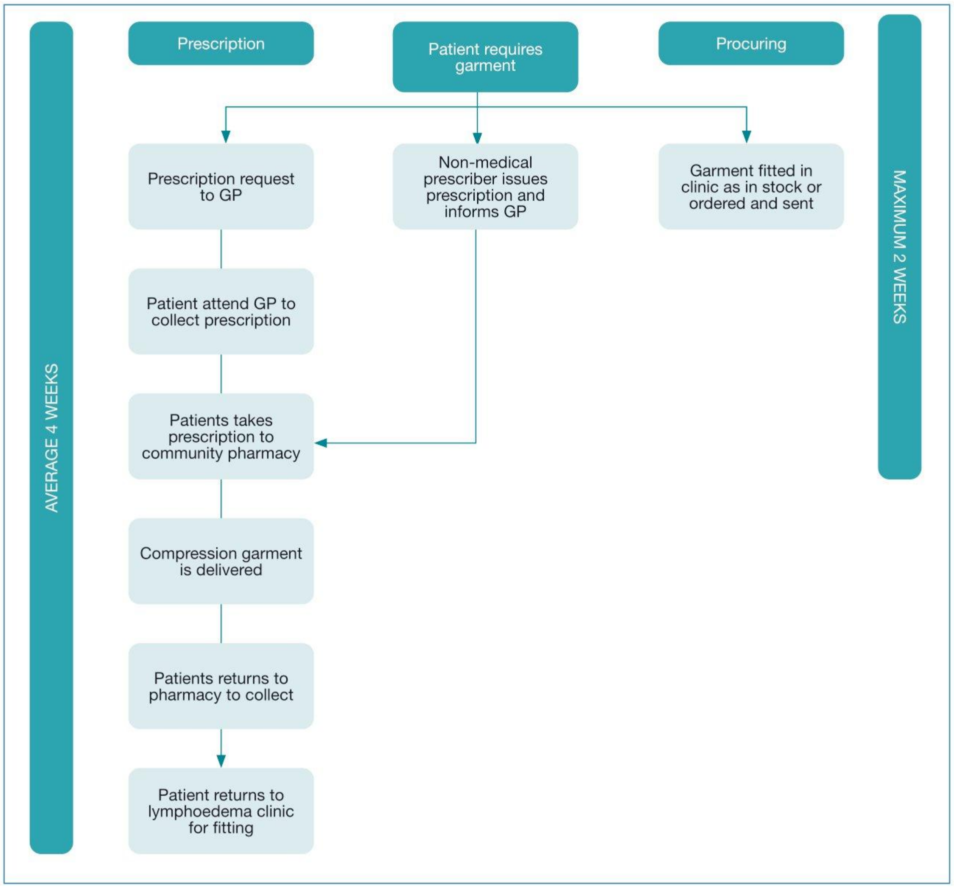

Figure 1. Prescription versus procuring patient pathway

the annual demand for lymphoedema compression garments accessed from the NHS drug tariff. During the last 6 months compression garment prescriptions fulfilled in the UK amount to nearly £20 million from the NHS drug tariff with a potential spend of over £40 million per annum and is still growing exponentially.

Unfortunately, waste, harm and variation in garments being prescribed via NHS prescription forms have been identified nationally (Board and Anderson, 2018; Woods, 2018). Incorrect dispensing of garments occurs frequently and has even caused harm in patients being issued with the wrong sizes and style of garments exacerbating their oedema (Woods, 2018). The delays and errors in garments being dispensed have negatively affected patient outcomes by preventing prompt treatment and impacts on compliance. Moreover, the lymphoedema services receive numerous phone calls from patients, GPs, prescription clerks and CPs regarding the garment prescription requests because of confusion over the wide variation (tens of thousands of compression garment lines on the drug tariff due to numerous sizes, colours and designs) (NHS Business Services Authority, 2021b).

In addition to prescriptions, compression garments are also procured via secondary care and these costs are also mounting. In 2014, NHS Wales Shared Services and Lymphoedema Network Wales (LNW) completed an all wales lymphoedema compression garment contract (Thomas and Morgan, 2017)ensuring best garment, best price and best outcomes. The contract was developed following collaboration with lymphoedema clinicians, stakeholders, surgical material testing laboratory and NHS Wales shared services. This contract guarantees that patients receive the best product (as it has been tested) for the best price from a procurement perspective. Subsequently, the All Wales Lymphoedema Compression Garment Formulary was created in 2015 for primary and secondary care and renewed again in

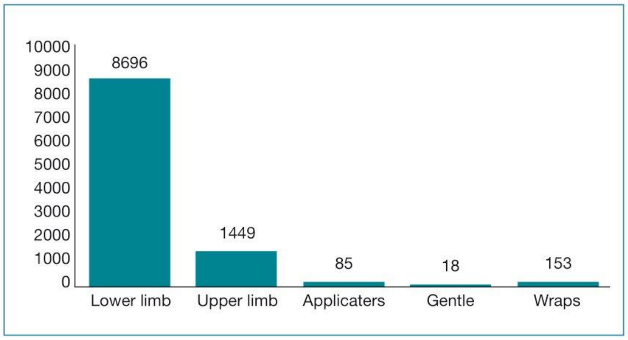

Figure 2. Types and numbers of compression prescribed

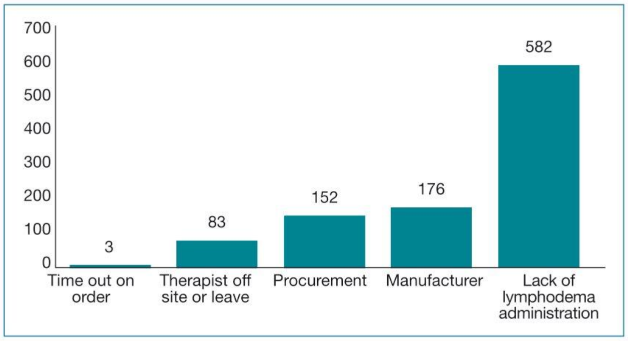

Figure 3. Delays in receiving garments

2018. The formulary was rolled out nationally and provides evidence-based compression information for all professionals. The current expenditure in NHS Wales for procuring compression garments is in excess of £1.4 million per annum.

Following value-based healthcare principles (Gray, 2017) it transpired that many lymphoedema compression garments cost much less to procure in secondary care than the drug tariff reimbursement charge. However, it is important to note that the funding route of the NHS is different in each of the four nations in the UK. As NHS Wales is a devolved nation, all NHS funding involves primary and secondary care. In England, the Clinical Commissioning Groups (CCGs) are responsible for all funding thus the change in process could be targeted at a local level.

To clarify the pricing differences, it is essential to understand that there are two types of compression garments. Flat knit garments are manufactured flat and then sewn together with a seam. They are used for patients who have skin folds, skin conditions and complicated oedema. A circular knit garment is made on a spherical knitted machine and has no seams. It is mainly used for mild/moderate oedema (Lyhmphodema Framework, 2006). Compression garments can also be categorised into ready to wear or made to measure and the costs of these garments are all different. For example, lymphoedema circular compression tights are around £48 on the drug tariff and £26 when procured directly. A thigh circular knit compression garment is around £54 on drugs tariff and is £25 cheaper when procured. Flat knit garments are not available ready to wear on the drugs tariff thus the only way to access would be via a made to measure, which introduces significant additional costs of around £30 per garment including VAT (NHS Business Services Authority, *no date* . Thus, if compression garments were procured directly instead of being accessed via the drugs tariff, opportunities for cost savings could be available.

Subsequently, an alternative process was considered adhering to value-based healthcare principles ensuring that resources used are sustainable and achieve better outcomes and experiences for patients. Accordingly, LNW, GPs, CPs and medicine management staff agreed to pilot an alternative process so that lymphoedema services could procure lymphoedema garments direct from the pharmacy budget, removing the need for GPs and CPs, thereby reducing the need for prescriptions. This article will present the data on the changed process and discuss the permanent service redesign that could be replicated.

Aims and objectives

The main aim of this evaluation was to estimate the potential impact of changing the way compression garments are accessed from a prescription to a procurement process. The main objectives were to:

- Estimate the costs of the service redesign
- Investigate any delays in patients receiving compression garments.

Methods

Ready to wear compression garment stock was purchased at a cost of £15000 in each of the two piloted services. The stock was based on the high usage of different types of garments across these services and included a variety of commonly used sizes and styles. If a made to measure garment was required, then this was ordered following the same procuring process. A data form was devised by the team so that information was collected by lymphoedema therapists for each compression garment issued to patients attending the twoservices in Wales over 12 months. The data collection included details on the garment ordered: the costs if prescribed (based on drug tariff) compared to procured (based on NHS contract), the date the garment was ordered and received and a comparison of the processes.

There were slight differences within the two lymphoedema services undergoing the service evaluation, as service A had two non-medical prescribers who wrote the prescriptions for patients and service B requests that GPs issue prescriptions. Team meetings were held with all staff so all were aware of the new process and service evaluation. All non-patient identifiable data from the form was emailed to a project manager and entered into a Microsoft Excel database. All data were checked and costings verified by two of the authors. Any missing data was presumed to be zero resource usage. Analysis was undertaken in Microsoft Excel and SPSS Version 22 for Windows to identify any correlations and descriptive counts between the variables collected. This study design was ratified as service evaluation by the local Joint Service Research Committee and no formal NHS ethics permissions were required to conduct the evaluation. Swansea University College of Human and Health Sciences (CHHS) ethics committee also deemed the study a service evaluation for a senior researcher (IH) to analyse the anonymised datasets.

Results

For this study, 5392 forms were completed on patients receiving compression garments from the two lymphoedema services over 12 months (3432 patients (64%) from service A and 1960 patients (36%) from service B). Across both services, 957 (18%) were new patients, compared to 4435 (82%) who were follow up patients. Of the 5392 patient data forms, this equated to a total of 10 401 compression garments (an average of two garments per patient). A large proportion of the garments ordered were for lower limb lymphoedema (84%) followed by upper limb (14%) and miscellaneous items, as highlighted in *Figure 2*. Of the garments required, 4915 (47%) were flat knitted and 5128 (49%) were circular knit. Positively, 95% (9881) of the garments requested (ready to wear and made to measure) were part of the NHS Wales compression garment contract compared to 520 (5%) being non-contracted items.

Table 1. Cross tabulation between new /follow up patients by fitted/ ordered

Garment fitted at appointment or ordered?				
New patient or follow up	**Fitted ready to wear**	**Ordered made to measure**	**Ordered ready to wear**	**Total**
New patient	683 (71%)	30 (3%)	244 (26%)	957
Follow up	2131 (48%)	318 (7%)	1986 (45%)	4435

Table 2. Cross tabulation between collected, fitted or posted and ready to wear

Was the garment fitted at the appointment or ordered compared with ordered m2m or ready to wear?		
How was the garment delivered?	**Ordered made to measure**	**Ordered ready to wear**
Collected	112 (32%)	1171 (52%)
Fitted	115 (33%)	346 (16%)
Posted	121 (35%)	713 (32%)
Total	348	2230

Over half of the garments issued (52%) were fitted on the day of the appointment as they were in stock; 7% were ordered as the patient required made to measure garments and 41% were ordered although they were ready to wear the product but were not available in the service. Of the patients (2578) requiring garments that were ordered, 1283 (50%) were collected; 834 (32%) were posted by the lymphoedema service direct and 461 (18%) required an additional appointment at the lymphoedema service as the garment had to be fitted. In contrast, no patients were fitted on the same day when accessing garments via the prescription route.

Reviewing the new and follow-up patients, more new patients (71%) were fitted with garments on their appointment than follow-up patients (48%) as highlighted in *Table 1*. Data showed that even though 2230 patients could have been fitted with a garment at the time of their appointment they were not in stock and 16% (346) had to have an additional appointment for a fitting (*Table 2*). Compared to the prescription route, 80% required a return to the service for fitting.

Table 3. Time lags between ordered garments (made to measure and ready to wear)

		Time lag between appointment and item requested (days)	**Time lag between appointment and item ordered (days)**	**Time lag between appointment to date dispatched (days)**	**Time lag between appointment to date received (days)**
Collected	Mean	0.4	3	11	14
Fitted	Number	348	348	348	348
Posted	Sum	160	1080	3812	4729
Total	Mean	1	3	6	10
	Number	2230	2230	2230	2230
	Sum	2222	6571	13 478	22 475

Table 4. Costs to Service A for Prescribing process

Cost items	Unit cost source/ description	Cost per task	Total cost based on 3432 patients	Task
Healthcare practitioner (Band 7)	PSSRU (Curtis and Buns, 2020) Band 7 (15 minutes)	£14.50	£49 764	Writing script
Community pharmacist (Band 6)	PSSRU (Curtis and Buns, 2020) Band 6 (30 minutes)	£22.50	£77 220	Ordering, processing order, telephone queries and invoicing checking order/ receipting with patients
			£126 984	£37 per patient

Table 5. Costs to Service B for prescribing process

Cost items	Unit cost source/ description	Cost per task	Total cost based on 1960 patients	Task
Lymph specialist (Band 6)	PSSRU (Curtis and Burns, 2020) Band 6 (30 minutes)	£22.50	£44 100	Completing Prescription Request form /sending to GP with letter for a prescription
General practitioner	PSSRU (Curtis and Burns, 2020) General Practitioner (10 minutes)	£7.30	£14 308	Signing scripts - (actual cost per GP prescription)
Prescription clerk (Band 2)	https://www.nhsemployers.org/pay-pensions-and-reward/nhs-terms-and-conditions-of-service---agenda-for-change/pay-scales/hourly (30 minutes)	£4.95	£9 692	Writing, searching for items, telephone queries
Community Pharmacist (Band 6)	PSSRU (Curtis and Burns, 2020) Band 6 (30 minutes)	£22.50	£44 100	Ordering, processing order, telephone queries and invoicing checking order/ receipting with patients
			£112 200	£57 per patient

Data were also collected on the procuring time delays (more than 11 working days as within the contract is a 10 day turnaround) if garments had to be ordered which amounted to 996 out of 2578 (39%). As reported in *Figure 3*, the main reasons for the delays were a lack of administrators in the lymphoedema clinic followed by manufacturing delays (making/distributing) and procurement (awaiting a price if noncontract/ manpower). *Table 3* details the analysis of the time lag between different garments ordered. Made to measure garments had the highest time lag with an average of 14 days, compared to 10 days for ordered ready to wear. Further, it transpires that non-contracted items take on average 12 days to arrive compared to 7 days for contracted items.

Comparing the costs of prescribing versus procuring garments

The staffing costs associated with prescribing garments were estimated for lymphoedema service A and B. As previously described, service B uses GPs for issuing prescriptions and service A has non-medical prescribers, which therefore bypasses the need for GP involvement. For service A, the staffing element of prescribing a garment was estimated as £37 (*Table 4*) and for service B, this was projected to be £57 as it includes GPs (*Table 5*). Personal Social Services Research Unit (PSSRU) and Agenda for Change NHS costs were used for calculations of staffing costs (Curtis and Burns, 2020; NHS Employers, 2021). In calculating the costs for the procurement process, a new Band 3 Garment Administrator would be employed to order, restock, send and provide patients with garments, which would be at a cost of £1.80 per patient (*Table 6*). The difference in staffing costs indicates that using the prescribing process is overwhelmingly more costly than using procurement (*Table 7*). Total costs across the two lymphoedema services for staffing only (n=5392) show a mean per-patient cost of £106 (SD £74) for prescription process and £77 (SD £54) for the procuring process (including the band 3 administrator).

Analysing the costs of the compression garment highlights that procuring is far more economical than prescribing (*Table 7*). Procuring garments suggests an average per patient cost of £40.40 compared to £55.60 for prescribing. When the costs include the garments and staffing, a mean per-patient cost of £149.90 (SD £75) for the prescription process and £78.80 (SD £54) for the procuring process. The difference in overall costs is then estimated at -£71.1 (SD £40.8) and this difference is also seen as statistically significant (*P*-value <0.001).

A one-way sensitivity analysis (±30%) was undertaken (*Table 8*) to assess the extent of altering the main cost parameters on the cost of the impact of the service evaluation. This indicated that the results remained consistent and in favour of altering the process from prescription led to a procuring service redesign.

Table 6: Proposed costs if the service redesign was implemented

Cost Items	Unit cost source/ description	Cost per task	Total cost	Comments
Band 3	https://www.nhsemployers.org/pay-pensions-and-reward/nhs-terms-and-conditions-of-service---agenda-for-change/pay-scales/hourly	£1.81	£9759	Every patient for the ordering process - 10 minutes (based on 5,392 Patients)
		Cost per oracle	£9759	£1.80

Limitations

A limitation of this study is the narrow perspective adopted on direct NHS costs through PSSRU and Agenda for Change (Curtis and Burns, 2020). As a result of the available resources, it was not possible to include the indirect costs relating to the prescription route for patients for example travelling, time off work. The authors were also unable to estimate the costs incurred with the procuring process. Although pragmatically a reduced impact seems logical as the need for patients to travel to collect prescriptions from their GP, take to pharmacy and collect is eliminated. Postal prescription service is not used within lymphoedema services in Wales, however, this approach along with direct ordering with their local pharmacy could have sped up the process. If the journey on average was 10.8 miles for each patient (Iredale et al, 2013) (5392), there is a reduction for substantial out of pocket expenses. Only the costs associated to the ordering of garments were reviewed and did not include the costs associated with additional fitting appointments or postal charges for the prescription or procuring route. Another limitation may be that not all data forms were completed by therapists, although 5392 is a positive number of returns. Lastly, this first attempt at costing the prescription route for compression garments does not include a patient's qualitative perspective. This requires a further study to establish if the change in the process does indeed impact the patient experiences of obtaining compression garments.

Discussion

This analysis of 5392 patients across two lymphoedema services indicates there are differences between the timing and quality of care the patients receive comparing the prescription to procuring route. Comparable to other research (Board and Anderson, 2018; Woods, 2018) waiting unacceptable lengths of time for compression garments affect care and concordance to treatment.

Furthermore, the risk of error with the additional steps in prescribing will likely further impact concordance and patient outcomes. The amount of time waiting for a garment via the prescription route is on average 4 weeks (Board and Anderson, 2018; Woods, 2018). Other solutions such as the postal prescription

Table 7: Summary of costs comparing the two processes

Item *n*= 5392	Total cost of garments across both services	Mean cost per patient	Standard deviation	95% xonfidence interval of the difference	*P*-value
Procuring cost of garments	£344 943	£33.70	£45.10	£64 (£62.8, £65.2)	<0.001
Procuring cost including VAT	£413 932	£40.40	£54.10	£76.8 (£75.3, £78.2)	<0.001
Prescribing cost of garments	£553 259	£54.00	£71.80	£102.60 (£100.7, £104.5)	<0.001
Prescribing cost including dispensing charge	£569 540	£55.60	£74.00	£105.6 (£103.7, £107.6)	<0.001
Overall costs procuring route	£424 716	£41.50	£54.10	£78.80 (£77.3, £80.2)	<0.001
Overall costs prescribing route	£808 244	£78.90	£74.90	£149.90 (£147.9, £151.9	<0.001
Overall difference in costs	-£383 528	-£37.40	£40.8	-£71.10 (-£72.2, -£70)	<0.001

could be used but it still could induce errors as the garments are not fitted at the right place and time.

Positively, the procurement route allowed 52% of patients to be fitted on the actual day of their appointment and for those patients waiting, the average time was 10 days for ready to wear and 14 days for made to measure garments. Reductions in the time waiting for garments would not only have positive health implications for the patient enabling prompt management of their lymphoedema and possibly improved outcomes and experiences, but it would also indicate a cost-saving due to the avoidance of any health-related consequences due to time lags such as superficial wounds and cellulitis.

However, this evaluation highlighted that even though 2814 patients were fitted on the day a further 2230 could have been fitted if the ready to wear stock had been available. Nevertheless, stock analysis requires dedicated administration staff, which would be required if this service design was accepted as business as usual. The analysis also showed that delays in receiving compression garments longer than 11 days was also identified as 'admin' problems. Therefore, a band 3 administrator would support even more efficiencies if this was a permanent service redesign.

Research also suggests that the prescription route for garments is not efficient and has reported numerous risks of waste, harm and variation (Woods, 2018; Board and Anderson, 2018). Although each compression garment does have a unique code for ordering these are not readily available on the GP and CPs systems, thus errors can occur in what was prescribed and what was dispensed. Throughout the procuring process all patients received the correct garment. There is a benefit in being able to try garments on patients and easily alter the size needed based on clinical expertise.

This evaluation is the first to investigate the costs of the prescribed and procured process for compression garments. It was established there is a statistically significant difference in costs when using prescription to procuring. When all costs are considered, there is an individual patient cost saving of £71.10 (SD £40.80) when procuring is compared to prescription. This difference in cost is also seen as statistically significant (*P*-value <0.001). The differences between the processes indicate the potential for substantial savings to be made if the prescription method was switched to procuring the garments bypassing GPs, clerks and CPs. This would ensure patients are seeing the right people in the right place and not involving superfluous layers in a process and supports value-based healthcare (Gray, 2017). Sustainability of GP and CPs time is vital and the procuring process ensures that they are not being asked to perform tasks with little value.

The prescription process will also minimise patients having to travel unnecessarily for additional fitting appointments. This will help reduce unnecessary spending for patients and limit the reported stress of parking at acute hospital sites. Looking to the environment, there may be capacity to positively influence climate control (use of fossil fuels and emissions). If an average of 10.8 miles per patient was calculated (Iredale et al, 2013), then changing to the procurement process would decrease annual travel (58 000 miles based on 5392 contacts using a prescription based service compared to 5000 miles based on the 461 fitting appointments in this procurement evaluation). This is likely a conservative estimate given that access to garments will likely improve as the procurement initiative embeds.

Table 8. Results of one-way sensitivity analysis

Parameter	Base-case (mean)	Lower range (mean)	Upper range (mean)	Result
Overall costs prescribing	£149.90	£104.93	£194.87	In favour of altering process and a service redesign
Overall costs procuring	£78.80	£55.16	£102.44	
Overall difference in costs	-£71.10	-£49.77	-£92.43	

Key Points

- Compression garments are vitally important in the management of lymphoedema
- Accessing compression garments via prescriptions can be long-winded and provides little value for patients in superfluous tasks
- Improving the process of procuring instead of prescribing garments may offer many benefits including efficiencies and more effective care.

CPD reflective questions

- Think about the current process for patients to gain compression garments- is it effective? Have you encountered waste or harm?
- Have you experienced difficulties or confusion in obtaining the correct compression garments for patients as there are thousands of different options?
- What education have you received in prescribing compression garments? Is it from independent (non-biased) sources or based on manufacturers guidance?
- Are there other areas that may benefit from direct purchase instead of prescribing? For example stoma products? Incontinence? Nutrition supplements?

Although CPs may have developed knowledge in compression for venous ulceration, the number of garments available for lymphoedema is vast; tens of thousands of lines because of an abundance of sizes, manufacturers, designs, colours and fabrics. This lack of clarity in ordering with unknown manufacturers' causes costly mistakes especially for locally owned pharmacists and not all offer pharmacy discounts. The financial element may not be a burden when the garment costs pounds but when compression can be hundreds it does impact. So much so, that prior to this service evaluation, some CPs were reluctant to dispense prescriptions, causing unnecessary anxiety with patients.

In making this service evaluation a permanent service redesign, a dedicated administration is required. Even when a new band 3 administrator was costed into the analysis, there still remained efficiencies compared to the prescription route. Subsequently, six out of the seven health board services in Wales now have supported the service redesign and compression is procured instead of prescribed. This service evaluation has led to improved patient outcomes, avoided waste, harm and variation.

Conclusion

The analysis has provided a first in-depth examination on the potential health and economic benefits of changing the process of accessing lymphoedema compression garments from prescribing to procuring. The findings suggest substantial differences in costs when comparing prescribing to procuring processes. There is also a substantial time lag in the prescribing process with the adding of healthcare workers with no patient value and is purely based on finance budgets involving primary and secondary care.

The analysis indicates the potential for substantial savings to the NHS (£71.10 per patient), and benefits to patients in terms of timely and quality of care. For the two lymphoedema services alone over one year, the potential cost avoidance would be £38 3371. Further analysis and evaluation is needed, but it seems very likely that using the procurement process would be highly efficient to the NHS. It would also be cost-saving to the patient due to out of pocket expenditure savings incurred by travel between GP practices and CPs. This would also enhance patient care, patient safety and effectively reduce wastage and streamlining this process is in line with delivering value-based healthcare. **JPrP**

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Section 3:6 Paper 6 A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs MJ Thomas, I Humphreys, RW Noble-Jones - International Wound Journal, 2022

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ORIGINAL ARTICLE

A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs

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Abstract

This unique evaluation aimed to estimate, the financial impact of non-attendance on a nation-wide hospital lymphoedema service. Along with gaining some understanding of patient characteristics of those who Did Not Attend (DNA) and were subsequently discharged. The evaluation design interrogated existing performance data from 2012 to 2022. This information was used to estimate the costs incurred based on national published sources and pay scales. Staffing costs of over £1.1 m in one decade related to the financial impact of over 23 000 unattended lymphoedema appointments. The characteristics of 870 patients from 2019/2020 were also evaluated suggesting that those with a wound alongside complex lymphoedema were less likely to DNA appointments. Two-thirds of patients were managing two or more comorbidities—obesity, cardiac conditions and diabetes being the most common. It seems likely that some DNAs are avoidable by adapting appointment administrative processes and greater understanding of patients' perception of value. However, the reasons for DNA are likely to be varied and nuanced so potentially a small proportion are unavoidable. Modernising appointment processes and identifying patient value may help minimise DNA costs in the future.

KEYWORDS

comorbidity, Did Not Attend (DNA), financial workforce impact, lymphoedema, wounds

Key Messages

- the workforce financial costs of over 23 000 Did Not Attend (DNA) events, in a decade of lymphoedema hospital appointments was examined at a national level
- closer examination of an anonymised data set of 870 patients, who DNA scheduled appointments from 1 year, gave an indication of the characteristics of this population with lymphoedema
- a financial cost relating to staffing of over £1.1 m was attributed to patient non-attendance of hospital lymphoedema appointments in one decade
- patients with a wound alongside complex lymphoedema were less likely to fail to attend appointments

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- two-thirds of patients were managing two or more comorbidities—obesity, cardiac conditions and diabetes being the most common in people who DNA their lymphoedema appointment

1 | INTRODUCTION

A spotlight on the workforce costs of missed appointments and demographics of people who 'Did Not Attend' (DNA) lymphoedema hospital appointments has not previously been undertaken at national level. People with lymphoedema have a progressive condition, causing swelling (oedema), skin changes, poor wound healing and pain in the affected limb(s), face, trunk or genitalia. Comorbidity with lymphoedema has not been quantified in national DNA data. This paper is the first of a series to gain greater understanding of the DNA phenomenon in Lymphoedema Services nationally, leading to a future economic impact assessment including patient reported outcomes and wider health system costs.

Unattended appointments produce a considerable impact on the resources and daily schedules of hospitals, GP clinics, NHS services and treatment centres. Indicated in records as DNA, DNAs decrease the productivity and efficiency of health services and increase the costs of delivery. The financial burden of missed appointments in the NHS is significant. The NHS Benchmarking Network, which includes 129 NHS Organisations across England and Wales, reported an 8% DNA rate costing £1 billion in their 2019 Outpatients project.[1] Also in 2019, missed GP appointments in England alone were estimated to be in excess of £216 million.[2] In Wales, missed appointments between 2015 and 2019 were estimated to have cost £2.5 million, with regional variation of DNA rates ranging from 7.7% to 10%.[3]

Health care research has sought to uncover human characteristic or demographic factors for 'no show' behaviours, as well as process issues that may affect the DNA rates. A US systematic review in 2018 concluded that DNAs were most common in adults of a younger age group, lower socioeconomic status, increased travel distance to the clinic and a prior history of no shows; whilst a long lead time to the appointment was also significant.[4] Appointment process methods such as reminder letters and electronic methods of communication have been tested. A meta-analysis of the use of SMS text messaging in 2016 concluded it to be an effective way of improving attendance in health care appointments.[5] However, they could not conclude whether the effect was greatest in certain groups of people nor the ideal frequency or time interval of reminders.[5] A more recent study of young people with type 2 diabetes found that an SMS reminder message more than doubled attendance.[6] Similarly, in a different hospital, implementing such reminders to dermatology outpatients reduced the mean DNA from 10.66% to 4.77%.[7] It may be that greater use of technology and understanding the demographics of those who are most likely to DNA would allow consideration of targeted processes to support patient attendance.

The economic burden of lymphoedema services have been examined for particular sub-group populations, for example, breast cancer-related,[8] and by specific provider groups, for example, independent hospices.[9] However, there appear to be no studies of the financial impact, in terms of staff costs, of patients who DNA scheduled outpatient appointments at Lymphoedema Services. Since the inception of a national lymphoedema service in Wales[10] lymphoedema services in each NHS health board provide performance data on numbers of patients attending and those who DNA. The data includes whether those individuals who DNA are new patients (NP) or follow-up (FU) patients. This information is already used for local service development, but has not been reviewed longitudinally nor has the financial impact been considered. These data also provide an opportunity to explore the demographics of people who DNA, that is, were discharged from the services for non-attendance of lymphoedema appointments with lack of response to correspondence offering another appointment.

Management of lymphoedema involves reducing the risk of infection (cellulitis) through good skin care, compression of the oedema and other interventions aimed at improving lymph flow and reducing consolidation into fibrosis and the occurrence of wounds.[10] The impact of daily life can be physical and psychosocial.[11] As with other progressive chronic conditions, non-attendance can lead to delay in diagnosis or lack of appropriate care, which in the long-term can mean increased demand on health care, for example, requiring unplanned admissions with infection, wounds or repeated GP episodes with recurrent cellulitis and increased risk of falls.[12] The impact of a lymphoedema appointment DNA is likely therefore be more than the burden to the service; the costs for the patient may be risk to employment, increased use of personal resources as a result of disability and reduction in quality of life.[13] However, this initial study focussed on staff costs and is therefore likely to be of interest to lymphoedema service providers and similar outpatient health care providers.

2 | AIMS AND OBJECTIVES

The aims of this evaluation study were to:

- estimate the financial workforce costs of DNAs on Lymphoedema Services across an entire nation,
- gain more understanding, from an anonymised data set, of patients who DNA scheduled appointments and were discharged from Wales lymphoedema services during 2019/2020.

The specific objectives were to:

- explore the numbers of DNAs since 2012 for NP and FU patients in Lymphoedema Wales (LW) providing an estimate of costs incurred to lost workforce hours,
- examine an anonymised data set of demographics from individuals who DNA and failed to respond to correspondence so were discharged from LW lymphoedema services in 2019/2020,
- estimate the lost workforce costs associated with DNAs at an individual health board level compared with population and lymphoedema activity,
- consider other factors that could influence DNA appointments in LW.

3 | METHODS

3.1 | Evaluation design and sample

The design of this study was a two-pronged observational data audit.

Firstly, the evaluation design interrogated existing performance data from LW from 2012 to 2022 on patients who attend or DNA appointments captured monthly and stored on the NHS Wales database. This information was used to estimate the lost workforce costs incurred owing to DNAs based on national published sources of unit outlays[13] and the Agenda for Change pay scale for NHS employees (www.healthcareers.nhs.uk). To estimate the costs, the following specifics within LW were applied:

- NP lymphoedema assessments in Wales were allocated 1.5 hours of a registered professional. A Band 6 (average between Band 5-8a) has been pragmatically submitted plus 30 minutes of an administrator (Band 3). The administrator time includes inputting data into the NHS databases, generating a letter of appointment, creating a case file, checking patient status if DNA, sending a DNA letter, updating databases, producing another letter of discharge and filing.
- FU lymphoedema patients were allocated 45 minutes of a Band 6 and 30 minutes of a Band 3 administrator (Table 1).

Secondly, anonymous case note data (870 patients) on individuals who DNA and were discharged from LW (2019/2020) were collected from Lymphoedema Services in the NHS Wales Health Boards. These were entered into an MS Excel database indicating sex, age, type of lymphoedema (cancer/non-cancer), number of previous appointments attended, mobility, employment status, lymphoedema severity outcome including presence of wound, miles travelled to clinic location and any comorbidities noted. No personal identifiable data was gathered.

The perspective taken was that relating to NHS Wales, which could be inferred for other similar NHS environments.

3.2 | Data collection measures

The Lymphoedema Project Manager entered all the data and the authors of this paper (M.T. and R.N.J.) were responsible for data cleaning, checks and running any queries before the data were locked and transferred to the Swansea University researcher (I.H.) for statistical analysis. The data set was anonymous.

3.3 | Ethics and Research Governance

This study design was reviewed by the Joint Study Review Committee at Swansea Bay University Health Board and deemed a service evaluation/data audit in relation to ethical approval requirements. Swansea University College of Human and Health Sciences (CHHS) ethics committee provided permission to analyse the anonymised data.

3.4 | Resource use

Resource use associated with DNAs was summarised into relevant categories and valued in £ sterling using a price year of 2019/2020. The costs were determined from national published sources of unit costs from the Personal and Social Services Research Unit[14] (Table 1) and the Agenda for Change pay scale for NHS employees.[13]

3.5 | Data and statistical analysis

Data and statistical analysis was undertaken in MS Excel and SPSS Version 26 for Windows. Basic descriptive demographic statistics were collected alongside the resource use and cost data.

TABLE 1 Unit costs for new patient (NP) and follow-up (FU) appointments

NP cost items	Unit cost per hour	Unit cost source/description	Total cost	Comments
Lymphoedema specialist (band 6)	£48.00	PSSRU[14] Band 6—Page 119	£72.00	Based on 1 h 30 min needed for NP appointment
Band 3 (mid-scale)	£10.81	NHS[15]	£5.41	30 min administration
		Cost per DNA (NP)	£77.41	
FU cost items	**Unit cost per hour**	**Unit cost source/description**	**Total cost**	**Comments**
Lymphoedema Specialist (Band 6)	£48.00	PSSRU[14] Band 6—Page 119	£36.00	Based on 45 min needed for FU appointment
Band 3 (Mid-scale)	£10.81	NHS[15]	£5.41	30 min administration
		Cost per DNA (FU)	£41.41	

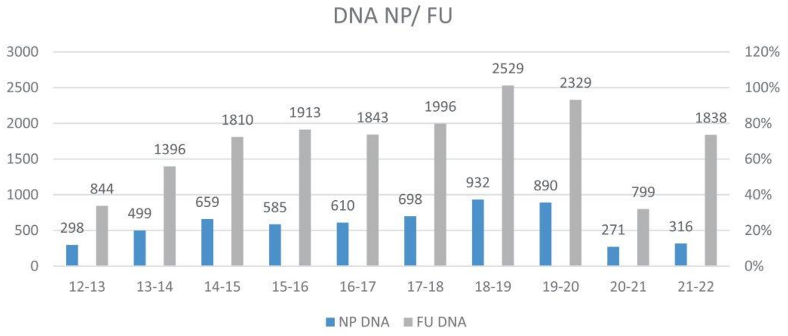

FIGURE 1 Numbers of Did Not Attend in Lymphoedema Clinical Network Wales (LW) 2012 to 2022

4 | RESULTS

4.1 | Number of unattended appointments (DNA)

Since the inception of LW the number of unattended appointments has grown proportionally each year as the caseloads increase (except for 2020 to 2022 which was influenced by the Covid19 pandemic). Since 2012, 5758 NP and 17 297 FU DNA events were recorded. This is broken down for each of the 10 years in Figure 1.

In the first year of the Covid19 pandemic (2020/2021), activity level (patient contacts) reduced by 9% compared with the previous year (39 978 to 36 373), however, the reduction in DNA was obvious (NP 890 to 271, a 69% reduction on the previous year; FU 2329 to 799, a 66% reduction). When seen as a percentage of overall activity for each of the 10 years (Figure 2), the NP DNA had remained consistently at 1% to 2% of all activity. Over the same period, the FU DNA rate had increased from 3% to a peak of 7%. The FU DNA rate in 2020/2021 reduced to 2%, resuming in following year to 5% (Figure 2). During 2020/2021 (first year of Covid19 pandemic) 60% of appointments were offered as virtual appointments (phone/video), compared with only 2% previously. This was a significant difference in the service process. This may account for part of the change in DNA rate; however, the wider domestic context for UK patients was also unique because of the Covid19 pandemic.

4.2 | Staffing costs of unattended appointments

Using the resource use described in the Section 3, the DNA data suggest that over the 10 years (2012-2022) in lymphoedema, staffing cost of over £1.1 million pounds for these unattended appointments (Table 2).

The populations in the NHS Wales Health Boards range from 133 030 to 703 360 people. Some are low population rural areas (eg, Powys Health Board, PHB), others high population urban areas (eg, Cardiff and Vale University Health Board) or mixed (Health Board) or mixed (eg, Betsi Cadwaladr University Health Board). This is demonstrated using population density in Table 3. Each of the health boards also differ in their deprivation. As highlighted one of the health board has four areas in the top 10 of the Welsh Index Multiple Deprivation and two

FIGURE 2 Percentage of new patients and follow-up Did Not Attend in Lymphoedema Wales

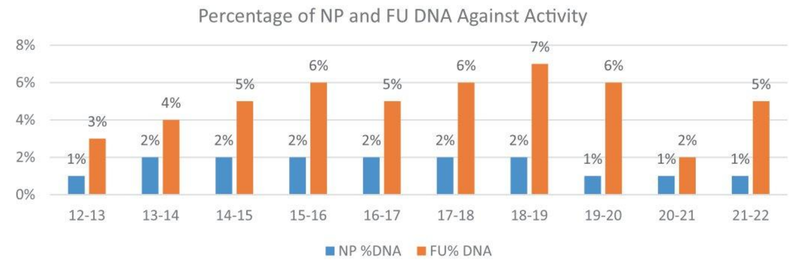

TABLE 2 Numbers of NP and FU DNA in LW, and related staffing costs 2012 to 2022

Year	Patient activity of LW	Numbers of NP DNA	Cost @ £77.41	Numbers of FU DNA	Cost @ £41.41	Total costs owing to DNA
2012-2013	25 286	298	£23 068.18	844	£34 950.04	£58 018.22
2013-2014	32 943	499	£38 627.59	1396	£57 808.36	£96 435.95
2014-2015	37 569	659	£51 013.19	1810	£74 952.10	£125 965.29
2015-2016	32 665	585	£45 284.85	1913	£79 217.33	£124 502.18
2016-2017	35 164	610	£47 220.10	1843	£76 318.63	£123 538.73
2017-2018	33 395	698	£54 032.18	1996	£82 654.36	£136 686.54
2018-2019	38 066	932	£72 146.12	2529	£104 725.89	£176 872.01
2019-2020	39 978	890	£68 894.90	2329	£96 443.89	£165 338.79
2020-2021	36 372	271	£20 978.11	799	£33 086.59	£54 064.70
2021-2022	37 202	316	£24 461.56	1838	£76 111.58	£100 573.14
Total	348 640	5758	£445 727	17 297	£716 269	£1 161 996

Abbreviations: DNA, Did Not Attend; FU, follow-up; LW, Lymphoedema Wales; NP, new patients.

other health boards have three areas each. Using the year 2019/2020 as an example, the total lymphoedema activity (including DNAs) by health board is presented (Table 3). The DNA as a percentage of activity (events) differs from 3% to 13% across the different Health Boards giving a mean across Wales of 8% of appointments being unattended (Table 3). The health board with the highest DNA percentage also have the highest deprivation. The 3219 DNA events represent 870 patients who did not respond to reminder correspondence and were subsequently discharged from the service.

4.3 | Descriptive data analysis of 2019/2020 DNA discharged patients

In 2019/2020, a total of 870 people were discharged from LW as they DNA an agreed appointment, failed to respond to a formal notification letter of DNA and were therefore removed from the active caseload. Non-identifiable information on these 870 individuals were analysed. The mean age was 62 (range 25-98) where 73% (632) were female, and 27% (238) male.

Just under half of the discharged patients identified themselves as married (43%), 29% were retired and 16% were unemployed (Table 4). Half of the patients reported their mobility as 'independent' with only 3% recorded as house bound, requiring a hoist or using a wheelchair. However, these three pieces of information were missing for 289, 267 and 193 patients respectively (Table 4). One reason for missing data on NP is a lack of information on referral communication, another, that on attendance patients can decline to give their marital or employment status.

Of the 870 discharged patients, 30% (258) had failed to attend a first appointment (NP) while 70% (612) DNA a subsequent appointment (FU). Using the unit costs from Table 1 this equates to over £45 000 lost in staffing costs (Table 5). Of the 612 FU patients, data show that they had attended, on average, five times each prior to the DNA event (range 1-59). Amongst these, 31% attended only once and 37% had attended more than four time previously.

Cancer and non-cancer related lymphoedema status was included within the data set. Nearly a quarter of the discharged patients (209, 24%) had a cancer-related

TABLE 3 Health Board population, density, deprivation, Lymphoedema activity data and Did Not Attend (DNA) data 2019 to 2020

Health Board	Population of health board and density per square kilometre	Welsh Index multiple deprivation WIMD top 10 areas	Total activity (including DNAs)	Total DNA events and % of activity	Total of patients discharged owing to DNA
Aneurin Bevan University Health Board (ABUHB)	598 194 (597)	Includes 3 of the top 10 deprived areas in Wales	6539	439 (7%)	107
Betsi Cadwaladr University Health Board (BCUHB)	703 361 (164)	Includes 3 of the top 10 deprived areas in Wales	7956	251 (3%)	72
Cardiff &Vale University Health Board (C and VUHB)	505 497 (1514)	None	4795	478 (10%)	146
Cwm Taff Morgannwg University Health Board (CTMUHB)	449 836 (567)	Includes 4 of the top 10 deprived areas in Wales	4021	510 (13%)	113
Hywel Dda University Health Board (HDUHB)	389 710 (66)	None	4985	331 (7%)	135
Powys Health Board (PHB)	133 030 (26)	None	1776	92 (5%)	17
Swansea Bay University Health Board (SBUHB)	390 949 (490)	None	9806	1018 (10%)	280
All Wales total	3 169 586 (153)		39 978	3219 (8%)	870

TABLE 4 Demographic data of 870 discharged patients 2019/2020

Marital status	N	Employment status	N	Mobility status	N
Divorced	21	Employed	208	Hoist	4
Married	378	Housewife	5	House bound	3
Single	154	Retired	249	Independent	434
Widow	28	Unemployed	141	Wheelchair	20
Not recorded	289	Not recorded	267	Not recorded	193
				With Aids	216
Total	870	Total	870	Total	870

TABLE 5 Costs attributed by new patient (NP)/follow-up (FU) 2019/2020

NP or FU	N	SD	Cost
New patient	258	0.0	£19 972
Follow-up	612	0.0	£25 343
Total	870	16.5	£45 315

TABLE 6 Costs of Did Not Attends (DNAs) by cancer/non-cancer related lymphoedema status

Cancer/non-cancer status	N	SD	Cost
Cancer related lymphoedema	209	17.0	£11 175
Non-cancer lymphoedema	661	16.3	£34 140
Total	870	16.5	£45 315

lymphoedema, of these, over half were breast cancer-related (56%), 9% were of the head and neck region, and both skin cancer-related and gynaecological-related accounted for 5%, while the sum of various other cancers amounted to 5%. Twenty percent had missing data on cancer type. The majority (661, 76%) of DNA discharged patients in 2019/2020 had non-cancer related lymphoedema. Table 6 shows the relative costing attributed to cancer to non-cancer related lymphoedema types for 1 year.

Each time a patient attends a lymphoedema clinic the severity of their lymphoedema is recorded. Of those with data available (Table 7), 24% had Complex lymphoedema, defined as total volume difference between limbs of ≥50%, and/or limb shape distortion (proximal to distal ratio ≥0:3/0:4) and/or skin changes including fibrosis,

TABLE 7 Classification of Lymphoedema severity 2019/2020

Lymphoedema classification	Did Not Attends (DNAs) 2019/2020 N	%	Overall attendances 2019/2020	%
At risk of Lymphoedema	71	8	3277	9
Mild Lymphoedema	166	19	7477	21
Moderate Lymphoedema	152	17	8403	23
Severe Lymphoedema	29	3	3647	10
Complex Lymphoedema	205	24	9853	28
Complex with a wound	16	2	3338	9
Not recorded	231	27	0	0
Total	870	100	35 995	100

TABLE 8 Mean miles from patient home to clinic by Health Board 2019/20

Health board	N	SD	Sum	Mean miles to clinic
Aneurin Bevan University Health Board (ABUHB)	107	7.62	1186	11
Betsi Cadwaladr University Health Board (BCUHB)	72	6.82	536	7.4
Cardiff and Vale University Health Board (C&VUHB)	146	3.74	1001	6.9
Cwm Taff Morgannwg University Health Board (CTMUHB)	113	4.34	645	5.7
Hywel Dda University Health Board (HDUHB)	132	7.12	1096	8.3
Powys Health Board (PHB)	17	5.58	64	3.8
Swansea Bay University Health Board (SBUHB)	280	7.73	2813	10
Total	867	6.84	7340	8.5

papillomatosis, hyperkeratosis, skin folds, acute cellulitis or red leg syndrome. Mild lymphoedema was identified in 19%, described as 5% to 10% volume difference between limbs and/or limb shape is normal (proximal to distal ratio 0:0/0:1). Between these, the lymphoedema was categorised 'moderate' in 17% of patients meaning overall volume difference 11% to 20% between limbs and/or some limb shape distortion (proximal to distal ratio 0:1/0:2). Patients that were deemed 'At Risk of lymphoedema', who were referred for advice and support only, accounted for 8% of the DNA data. Only 16 patients who were DNA discharged were categorised as having Complex Lymphoedema with a Wound (2%). The category was 'not recorded' for 27% (231), NP who failed to attend account for the majority of these, as they were yet to be categorised. In Table 7 the DNA discharges are compared with the overall attendances for the same year, these latter figures give a snapshot of the spread of classifications across the caseload for that year. From this, we can see that those least likely to DNA seem to be the patients with severe lymphoedema and those classified 'Complex with a Wound'. However, the high proportion of DNA with classification 'not recorded' means direct comparison is not possible.

4.4 | Distance from home area to clinic

We examined the distance from home area to clinic. The postcode area (not specific to individual house) of each patient was used to approximate the distance to the nearest Lymphoedema clinic. The mean miles to clinic for DNA discharged patients was 8.5 miles (SD 6.84). There was no statistically significant difference in mean distance travelled by age group.

In order to establish if there were regional differences, the mean distance to clinic for each of the Health Board Lymphoedema Services was calculated. Only three data sets were missing, providing 867. This showed that for DNA discharged patients the mean distance varied between 3.8 miles in Powys Health Board (PHB), a predominantly rural region, and a mean of 11 miles in Aneurin Bevan University Health Board (ABUHB) a mixed urban-rural health board (Table 8). Whilst individuals may find the distance to a clinic problematic, there was no statistical relationship between distance to clinic and rate of DNA.

Of the DNA discharged patients over Wales in 2019/2020, those who attended at least once (FU), lived a mean distance of 7.8 miles from the clinic, whereas those who did not even attend their first appointment (NP) lived a mean 10.1 miles from their appointed clinic (Table 9). It may therefore be that distance to clinic is an inhibiting factor for some people so that these patients are not seen within the service.

4.5 | Comorbidity

The data regarding each patient included whether they had co-existing health conditions (comorbidity) (Table 10). Obesity (BMI over 30) was common, recorded in just under half of the DNA population, followed by Cardiac issues (30%) and Diabetes (21%). The number of comorbidities were counted for each person, showing 11% had no comorbidities, 26% had one, meaning almost two-thirds of patients were managing two or more comorbidities (20% had two, 15% had three and 28% had four or more).

TABLE 9 Mean miles from patient home area to clinic by new patient/follow-up 2019/20

New/follow-up	N	SD	Sum	Mean miles to clinic
New patient	256	7.17	4741	10.1
Follow-up	611	6.58	2599	7.8
Total	867	6.84	7340	8.5

TABLE 10 Co-existing morbidities for Did Not Attend (DNA) discharged patients

Health condition	No	Yes	Missing data
Obesity	239	422 (49%)	206
Cardiac	405	259 (30%)	203
Diabetes	486	178 (21%)	203
Mental Health	573	96 (11%)	198
Neurological	589	72 (8%)	206
Renal	603	21 (2%)	243
Dementia	655	13 (1%)	199

Lastly, of the 870 patients who were discharged only 50 (6%) were re-referred to the Lymphoedema Service within the year. Twenty-seven were FU patients and 23 were NP. Analysis of this subset showed no correlations between any of the variables.

4.6 | Sensitivity analysis

A one-way sensitivity analysis was undertaken (Tables 11 and 12) to assess the extent to which reducing and increasing the unit cost estimates of the cost per DNA (FU) and the cost per DNA (NP) by 30% would have on the results seen in Table 2. Table 11 shows the new estimated costs when the cost of NP DNAs is reduced by 30% to £54.19. This equates to total estimated costs of £312 026.02.

When the estimated costs of follow-up patient (FU) DNAs is reduced by 30% to £28.99, this equates to total estimated costs of £501 440.03. The total combined costs of this reduction in unit costs by 30% is £813 466 (Table 11).

Conversely, Table 12 shows the new estimated costs when the cost of NP DNAs is increased by 30% to £100.63. This equates to total estimated costs of £579 427.54. When the estimated costs of follow-up

TABLE 11 Sensitivity analysis of decrease of unit estimates in Table 2

Year	Patient activity of LW	Numbers of NP DNA	Cost @ £54.19	Numbers of FU DNA	Cost @ £28.99	Total costs owing to DNA
2012-2013	25 286	298	£16 148.62	844	£24 467.56	£40 616
2013-2014	32 943	499	£27 040.81	1396	£40 470.04	£67 511
2014-2015	37 569	659	£35 711.21	1810	£52 471.90	£88 183
2015-2016	32 665	585	£31 701.15	1913	£55 457.87	£87 159
2016-2017	35 164	610	£33 055.90	1843	£53 428.57	£86 484
2017-2018	33 395	698	£37 824.62	1996	£57 864.04	£95 689
2018-2019	38 066	932	£50 505.08	2529	£73 315.71	£123 821
2019-2020	39 978	890	£48 229.10	2329	£67 517.71	£115 747
2020-2021	36 372	271	£14 685.49	799	£23 163.01	£37 849
2021-2022	37 202	316	£17 124.04	1838	£53 283.62	£70 408
Total	348 640	5758	£312 026.02	17 297	£501 440.03	£813 466

Abbreviations: DNA, Did Not Attend; FU, follow-up; LW, Lymphoedema Wales; NP, new patients.

TABLE 12 Sensitivity analysis of increase in unit estimates in Table 2

Year	Patient activity of LW	Numbers of NP DNA	Cost @ £100.63	Numbers of FU DNA	Cost @ £53.83	Total costs owing to DNA
2012-2013	25 286	298	£29 987.74	844	£45 432.52	£75 420
2013-2014	32 943	499	£50 214.37	1396	£75 146.68	£125 361
2014-2015	37 569	659	£66 315.17	1810	£97 432.30	£163 747
2015-2016	32 665	585	£58 868.55	1913	£102 976.79	£161 845
2016-2017	35 164	610	£61 384.30	1843	£99 208.69	£160 593
2017-2018	33 395	698	£70 239.74	1996	£107 444.68	£177 684
2018-2019	38 066	932	£93 787.16	2529	£136 136.07	£229 923
2019-2020	39 978	890	£89 560.70	2329	£125 370.07	£214 931
2020-2021	36 372	271	£27 270.73	799	£43 010.17	£70 281
2021-2022	37 202	316	£31 799.08	1838	£98 939.54	£130 739
Total	348 640	5758	£579 427.54	17 297	£931 097.51	£1 510 525

Abbreviations: DNA, Did Not Attend; FU, follow-up; LW, Lymphoedema Wales; NP, new patients.

patient (FU) DNAs is increased by 30% to £53.83, this equates to an estimated £931 097.51.

The total combined costs of this increase in unit costs by 30% is £1 510 525 (Table 12).

5 | DISCUSSION

This analysis has provided a spotlight examination of the financial workforce burden of DNAs on Lymphedema Services; this is the first time this has been investigated at this scale. Approximately £1.1 million of 'lost staff hours' expenditure is accounted for by patients missing valuable appointment slots over the last 10 years of activity. DNAs cause delays in lymphoedema treatment for other patients. Improving the worst region from 13% (Table 3) to the national average of 8% could free up around 188 appointments per year in that health board alone. Furthermore, staff time is underused, creating negative impact on capacity and demand. The staff cost is mitigated to some extent as lymphoedema staff use the time to catch up on other tasks; however, it remains an inefficiency since this work cannot be planned.

In our study, most of the expenditure was on FU patients despite being allocated half the time of a first appointment; accounting for over £716 000 compared with NP at nearly £446 000. Managing non-attendance in people who are known to the service (FU) is likely to need a different approach to those who have never attended (NP). Despite a wide range of number of appointments being attended before DNA (1-59), almost a third had only attended one appointment. It would be useful to investigate whether this signalled dissatisfaction with the service, or indeed, that the patient had adopted the self-management approach promoted in the clinics and felt further attendance was unnecessary. In relation to DNA management, this is an important point, because with chronic conditions, such as lymphoedema, improving the self-management capacity of patients has been shown to be associated with lower health care use and less wasteful utilisation across primary and secondary care.[16] This explanation is most likely for the 9% of patients classified 'at risk' of lymphoedema who were referred for advice and education. For others, such as the two-third of patients who were juggling two or more comorbidities, the lymphoedema may have become a lower priority once the condition and its management had been understood. This is where patient initiated FU (PIFU) appointments may be more beneficial instead of routine 6 month FUs and is a key recommendation for the NHS going forward[17]; although the evidence for cost savings was found to be weak.[18] In the overall caseload, 20% are cancer-related lymphoedema, whereas we found that 24% of the DNA discharged patients had cancer-related lymphoedema, this may again signal competing health care demands on the patients' time. Only 16 patients with a co-existing wound (2% of DNA) missed their appointment; this may highlight that patients recognised the role of lymphoedema management in their wound care making it a priority for them. Mental health as a key factor for DNA, described in primary care-based studies, does not seem to be as significant our data.[19-21] Almost half of the DNA discharged in 2019/2020 were living with obesity, much higher than the 24% prevalence of adult obesity in Wales.[22] Diabetes is an unsurprising comorbidity in an obese population but Wales' prevalence of diabetes as a proportion of its population is 7.4%[23] compared with 21% in our DNA cohort of 2019/2020. Similarly, around 11% (340 000) of the Wales population have heart and circulatory disease[24] whereas this was noted in 30% of our DNA patients. Whilst the link between diabetes and cardiac/circulatory problems are recognised,[24] the link between these comorbidities and lymphoedema is less clear, except that they commonly co-exist. Further work is now required on examining the rates of these comorbidities in our wider caseload and in global research to better understand the physiological connections involved. Following on from this study, a new investigation has commenced to further understand why patients attending the Lymphoedema Service do in fact DNA appointments. Understanding patient's reasons may initiate necessary service improvements supporting a reduction in the DNA rates.

During the first year of the Covid19 pandemic 2020 to 2021, service activity reduced by 10% but proportionally, more people attended their appointments, that is, the proportion of DNA appointments greatly decreased, the DNA rate reduced from 6% to 2%. One reason for this could be that planned implementation of virtual consultations were expedited[25] with proven benefits including travel/waiting time and costs for the patient (time off work and childcare) being eliminated. The DNA percentage increased back up to 5% in 2021 to 2022 despite maintaining the availability of virtual consultations. It may be that contact with any health care professional during that first unsettling year of the pandemic was welcome or that other commitments were fewer during 'lockdowns' on social movement. One local finding was that during 2021 to 2022, some patients said they had assumed that the government reporting system for Covid infection would notify the NHS automatically that they were unable to attend appointments as a result of isolation regulations.

Previous studies have reported many reasons for people not attending appointments including forgetting, transportation, work commitments, oversleeping or illness of children.[21] The complexity of living in social deprivation and with comorbidity is a cause of higher DNA rates.[19,20] This is reflected in our findings with a higher than average DNA rate (13% compared with an 8% national average) in one Health Board (Cwm Taff Morgannwg University Health Board [CTMUHB]). This health board scores highest (ie, worst) for almost all Government indicators for deprivation, including income (18%), and, (in rates per 100), GP recorded chronic condition (15), limiting long-term illness (26.4) and second highest for GP recorded mental health condition (24.7).[26] These issues cannot be resolved by an individual service but focusing increased activity on increasing their capability for self-management may reduce future DNAs.[16]

In addition to making virtual consultation mainstream within the lymphoedema service, the recent implementation of a lymphoedema-specific patients reported outcome measure (LYMPROM)[27] is hoped to further improve the patients' perception of value from the service. The patient rated aspects on the impact of lymphoedema on their life enables the service to prioritise those patients with the highest of level of need for face-to-face appointments.[27] Repeating the LYMPROM as care proceeds allows input to be titrated to the patients' perception of value from the service. These new data are now being evaluated and will be interesting to see in future if this changes the DNA rate based on patient outcomes.

Throughout the different Health Boards in Wales, there was a variety of standard operating procedures for making appointments. Some used appointments letters with a date specified, others invited the patient to telephone to make an appointment, others pro-actively telephoned the patient to agree a date, and FU appointments were made in person while a patient was in clinic or later by telephone. During the analysis of the 870 patients we did not analyse which method had been used. Some of the services had tried text reminders prior to the appointment but with their existing systems these had been disproportionately time consuming. It is hoped that with recent digital health care communication implementations in secondary care, a cheaper automatic reminder system will be possible such as patients have become familiar with from their GP practices. Furthermore, in supporting Value-Based health care the onus of when patients need to be reviewed (PIFU) could be initiated by them. Although we collected FU data we did not specify if these were annual or six monthly appointments. It may be that those patients waiting longer periods of time did not need to attend and PIFU may reduce subsequent DNA appointments.

Surprisingly, only 6% of DNA patients were referred back into the system within the same year (roughly half being FU and half NPs). As lymphoedema is a chronic condition it may have been expected that these patients would have been re-referred within the year as they would require further compression garments. However, this was not seen in the data but may have been affected by the Covid19 pandemic.

5.1 | Limitations

As with any study that incorporates a financial evaluation in its primary outcomes, there will be a level of uncertainty regarding the costing estimates used within the methodology used. Therefore, we feel that using the PSSRU[14] to estimate the 'per hour cost' of a lymphoedema specialist is justifiable as it takes into account all the estimated overheads and indirect costs relating the per hour cost. Lymphoedema therapists/nurses regardless of discipline are salary banded in the same way. Furthermore, the lack of Band 3 (mid-scale) costing work within the PSSRU,[14] led us to use the next best estimate from the NHS agenda.[15] This source provided us with the most accurate rate of hourly pay available at the time of writing that we could have used. We have tried to address any uncertainty with the base case unit costs estimates used by conducting a one-way sensitivity analysis +−30% shown in Tables 11 and 12. The results of this sensitivity analysis show that at the lowest estimate, the costs are still extensive at £813 466 (Table 11).

A wider economic impact would include the opportunity costs to staff and could include patient incurred costs. However, this narrow focus produced information which informed the direction of further evaluation. This is already underway and includes patients reported outcomes and additional health care costs to further inform service development.

Comparison of DNA patient demographics with our overall/attending caseload was not the purpose of this project since the focus was on DNA costs but it would be useful to do as a next stage. Seasonal variations and days of the week when DNA occurred were not investigated, neither was length of time waiting for FU appointments, this may offer potential areas for improvement and system cost effectiveness. For example, would a patient initiated FU reduce DNA rates instead of routine annual appointments and appreciate potential the cost benefits. Although gaining information on why DNA patient's DNA is difficult and is a limitation of this study, it is vital that we understand the barriers and how service improvements may support people attending appointments in the future.

6 | CONCLUSION

DNAs absorb a significant portion of health resources, including those of lymphoedema services. It seems likely that some DNAs are avoidable by adapting appointment-making processes and by working more closely with patients to understand their perception of value. However, when lymphoedema clinic attendees have a mean age over 60 and a higher than average prevalence of comorbidities, there is likely to be a percentage of DNA that is unavoidable. The reasons for DNA are likely to be varied and nuanced but modernising appointment processes, for example, digital and PIFU, and using PROMs to identify patient value may help minimise DNA costs in the future.

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CONFLICT OF INTEREST

The authors had no conflict of interests in this study.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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Section 3:7 Paper 7 Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study I Humphreys, A Akbari, R Griffiths, D Graham-Woollard, K Morgan, R Noble-Jones, M Gabe-Walters, M Thomas - International Wound Journal, 2023

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ORIGINAL ARTICLE

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Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study

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Abstract

This study aimed to estimate costs associated with managing patients with cellulitis from the UK National Health Service (NHS) perspective. The analysis was undertaken through the Secure Anonymised Information Linkage Databank, which brings together population-scale, individual-level anonymised linked data from a wide range of sources, including 80% of primary care general practices within Wales (population coverage ∼3.2 million). The data covered a 20-year period from 1999 to 2019. All patients linked to the relevant codes were tracked through primary care settings, recording the number of general practice visits (number of days with an event recorded) and number of in-patient stays. Resources were valued in monetary terms (£ sterling), with costs determined from national published sources of unit costs. These resources were then extrapolated out to reflect UK NHS costs. This is the first attempt to estimate the financial burden of cellulitis using routine data sources on a national scale. The estimated direct annual costs to the Welsh NHS (£28 554 338) are considerable. In-Patient events and length of stay costs are the main cost drivers, with annual Welsh NHS estimates of £19 664 126 with primary care events costing £8 890 212. Initiatives to support patients and healthcare professionals in identifying early signs/risks of cellulitis, improve the accuracy of initial diagnosis, prevent cellulitis recurrence, and improve evidence-based treatment pathways would result in major financial savings, to both the Welsh and UK NHS. In light of these findings, Wales has developed the innovative National Lymphoedema cellulitis Improvement Programme to address these burdens; providing a proactive model of cellulitis care.

KEYWORDS
cellulitis, economic burden, longitudinal data, lymphoedema, SAIL databank

Key Messages

- the estimated costs associated with managing patients with cellulitis across two decades was examined at a national level.
- the estimated direct annual costs to the Welsh NHS are £28,554,338.
- in-Patient events and length of stay costs are the main cost drivers, with annual Welsh NHS estimates of £19,664,126.
- initiatives to support patients and healthcare professionals in identifying early signs/risks of cellulitis, improve the accuracy of initial diagnosis, prevent cellulitis recurrence, and improve evidence-based treatment pathways would result in major financial savings.
- wales has developed the innovative National Lymphoedema cellulitis Improvement Programme to address these burdens, providing a proactive model of cellulitis care.

1 | INTRODUCTION AND BACKGROUND

Cellulitis is a skin infection involving the dermis and sub cutaneous tissue and is usually caused by a break or wound in the skin, commonly allowing either staphylococcus or streptococcus bacteria to enter. Cellulitis is commonly interchanged with the term erysipelas and can affect any part of the body, however, 70%–80% are encountered in the lower limbs.[1-3] All age groups can be affected by cellulitis but it is more common in those over 60 years of age.[4] The signs and symptoms of a cellulitis include redness, erythema, pain, oedema, bullae, blisters, bruising, petechiae as well as nausea, vomiting, lethargy and rigours. Not all signs and symptoms may be present and mild cases that are detected promptly can be effectively managed in primary care with oral antibiotics. However, more severe cases with systemic toxicity, uncontrolled morbidities or tissue necrosis may require intravenous antibiotics and admittance to hospital. Antibiotics are administered to prevent the infection from spreading and reduce the risk of sepsis, which is potentially fatal. The link between cellulitis occurrence and lymphoedema is well documented with nearly 50% of people with lower limb lymphoedema reporting an episode of cellulitis and half of those experiencing another cellulitis within a year. Recurrent cellulitis is associated with reduced patient quality of life as it caused physical, psychological, and functional impacts. Other risk factors evidenced include wounds, obesity, fungal infections, and venous insufficiency.[5-9]

Antimicrobial resistance is a challenge globally; thus people with cellulitis should be accurately diagnosed and treated promptly and the risk factors adequately supported. Further, if these risk factors were reduced and the incidence of cellulitis was less then this would lead to reduced admissions, antibiotic costs, and financial savings.

There remains a dearth of knowledge concerning the economic costs associated with cellulitis from a whole National Health Service (NHS) system. In 2019–2020 over 37 000 Wales hospital bed days were recorded for cases of cellulitis admissions, but there is little understanding of the burden of cellulitis within primary care services.[4] This is surprising, given that proactive models of care support prompt and appropriate care, with the potential to mitigate the 1.4% of emergency admissions owing to cellulitis in the United Kingdom (UK) in 2018–2019.[10] Understanding the underlying contributory factors and comorbidities that increase the risk of cellulitis, including sex, age or social deprivation is also limited from a national perspective. By gaining a better understanding of these factors including the economic angle, it may be possible to prioritise care and reduce financial encumbrance on the NHS as a whole.

2 | AIMS AND OBJECTIVES

This study aimed to estimate the costs associated with managing patients with cellulitis from the perspective of the Welsh/UK NHS using routine electronic health record data sources available within the Secure Anonymised Information Linkage (SAIL) Databank.

Specific objectives of the study were as follows:

- To understand the financial burden of cellulitis on the Welsh/UK NHS.
- To establish the numbers of cases attending primary care.
- To better understand the incidence of cellulitis in Wales and the implicated costs for UK NHS.

3 | METHODS

The analysis was undertaken through the SAIL Databank,[11-13] sources within a privacy-protecting

Trusted Research Environment. These data include primary care events from 80% of general practices around Wales (population coverage ∼3.2 million people), General Practitioner (GP) records and secondary care in-patient hospital episodes. All data are anonymised within SAIL, but the individual-level linkage is possible through an encrypted anonymised linking field which allows associations between data sources and longitudinal patient pathway analyses. The patient cohort was identified through relevant clinical codes and the resource implications of their management were collected and estimated using published sources.[14-18]

This approach provided an in-depth inventory of the contacts, consultations, and resources utilised in the current management of patients with cellulitis in NHS Wales.

3.1 | Inclusion/exclusion criteria

Patients with a cellulitis diagnosis in their Welsh Longitudinal General Practice (WLGP) records from 1999 to 2019 were included in the primary care cohort. Patients admitted and coded with cellulitis during their In-Patient stay were included in the secondary care cohort.

3.2 | Data analysis

The SAIL Databank was interrogated to catalogue health service resource utilisation by this cohort of patients. The WLGP data was used to identify and quantify all events in order to measure service usage. The Welsh Demographic Service Dataset (WDSD)[19] was utilised to gather basic demographics, information on follow-up time (residency in Wales and GP registrations), and the Welsh Index of Multiple Deprivation (WIMD)[20] version 2014 quintile to measure deprivation.

3.3 | Resource use

All patients linked to the relevant cellulitis codes (Table A1) were tracked through primary care settings, recording their level of general practice visits (number of days with an event recorded), and separately In-Patient admissions were captured including length of stays.

3.4 | Cost data

As highlighted in Table 1, resources were valued in monetary terms (£ sterling), and the costs were determined from nationally published sources of unit costs, including the Personal and Social Services Research Unit (PSSRU) unit costs[16] and costings derived from the NHS Wales Financial Delivery Unit.[17] Where costs were unavailable, local costs were utilised (eg, from local financial records or NHS Wales formulary). The currency year was 2020 and an inflation calculator (Bank of England-BOE) was used to convert previous years' costs to current prices.

TABLE 1 Age distribution of cellulitis in a primary care cohort Wales 1999–2019 and population distribution by the Welsh Index of Multiple Deprivation (WIMD)

Age group	**N**
0–10	28 712
11–20	21 284
21–30	21 628
31–45	46 420
46–60	58 083
61–75	60 983
76–90	34 586
91+	2999
N/A + Missing data	<800
WIMD Quintile 2014	**N**
1 Most deprived	60 690
2	56 535
3	56 801
4	48 190
5 Least deprived	52 487
Total	274 703

3.5 | Perspective

The perspective taken was from NHS Wales and extrapolated to the UK NHS.

3.6 | Statistical analysis

Statistical analysis was undertaken in SPSS Version 25 for Windows. Further, basic descriptive demographic statistics were also collected alongside the resource use and cost data. Survival analysis of treatment duration was conducted using R 3·5.

3.7 | Ethics approval and consent to participate

Approval for the use of anonymised data in this study, provisioned within SAIL Databank was granted by an

TABLE 2 Estimated cost of WLGP events over 20-years

		GP EVENTS	General Practitioner	NURSE
SURGERY VISIT	UNIT COST	-	£39	£42
	%	82%	62%	38%
	NO.	2 858 607	1 772 336	1 086 271
	COST	-	£69 121 115	£45 623 366
		GP EVENTS	**General Practitioner**	**NURSE**
TELEPHONE	UNIT COST	-	£16	£7.80
	%	15%	62%	38%
	NO.	522 916	324 208	198 708
	COST	-	£5 031 706	£1 549 923
		GP EVENTS	**General Practitioner**	**NURSE**
HOME VISIT	UNIT COST	-	£134	£84
	%	3%	62%	38%
	NO.	104 583	64 842	39 742
	COST	-	£8 688 771	£3 338 295
	TOTAL NO.	3 486 106	2 161 386	1 324 720
	TOTAL COST	-	£82 841 592	£50 511 584

independent Information Governance Review Panel (IGRP) under project 1061. The IGRP has a membership comprised of senior representatives from the British Medical Association (BMA), the National Research Ethics Service (NRES), Public Health Wales, and NHS Wales Informatics Service (NWIS).[18] Usage of additional data was granted by data owner. The SAIL Databank is General Data Protection Regulations and the UK Data Protection Act compliant.

The report utilised primary care data between January 1st 1999 and December 31st 2019 from the SAIL Databank, WLGP[19] records, in-patient hospital admissions from the PEDW[14] data, week of birth and sex were obtained from the WDSD[20] and deaths from the Annual District Death Extract (ADDE) based on the Official for National Statistics (ONS) deaths dataset (ADDE). Around 6% of the data, (approximately 18 400 records) were lost to quality assurance and missing Lower Layer Super Output Area code (LSOA) and hence deprivation quintiles. A small number were also lost due to the quality assurance for missing data carried out on the in-patient hospital admission data. In addition, patient cases that had several duplicate in-patient hospital admissions on the same day were removed, only keeping the one admission with the longest duration. Some cases may still have two admissions for the same date but have different ICD-10 cellulitis codes. These records have been kept but are only counted as one admission.

The index date is the first date found in either the WLGP records or the hospital records using PEDW for a diagnosis of cellulitis. Most of the index dates are found in the WLGP records but a small number (just under 4500) have a hospital admission for cellulitis before they are seen by a GP, so for those cases, the first hospital admission date is used.

4 | RESULTS

4.1 | Cohort demographics

Of the cohort of 274 703, 47% (n = 128 464) were male and 53% female (n = 146 238). The overall mean age of the cohort was 63.9 (Median 68 years). By sex, the mean age of females was 66.9 and 60.6 years for males. Table 1 shows the spread of the cohort across the 8 age groups assigned. The biggest share of the cohort was 61–75 (n = 60 983), 46–60 (n = 58 083), and 31–45 (n = 46 420).

Of the cohort of 274 703, 22% (n = 60 690) were in the WIMD Quintile 1 (Most Deprived); 21% (n = 56 535) were in the WIMD Quintile 2; 21% (n = 56 801) were in the WIMD Quintile 3; 17% (n = 49 190) were in the WIMD Quintile 4 and 19% were in the Quintile 5 (Least Deprived) (n = 52 487) as shown in Table 1.

4.2 | Cost of general practice event days

Not all days with an event represent consultations with a GP. Some are prescription renewals, receipt of letters from

TABLE 3 Incidence of cellulitis in GP Events and hospital admissions

Year	GP Patient events	Incidence per 1000	Hospital events	Incidence per 1000
2010	96 449	39	3116	1.3
2011	97 075	39	3245	1.3
2012	100 496	41	3159	1.3
2013	96 610	39	3198	1.3
2014	93 958	38	3507	1.4
2015	90 382	36	3601	1.5
2016	88 129	36	3819	1.5
2017	83 167	34	3704	1.5
2018	80 226	32	3906	1.6
2019	75 493	30	3902	1.6

hospital and other activities. There is no reliable way from the data held in SAIL to identify the number of patient consultations with a GP. Therefore, a pragmatic approach to defining and costing a primary care contact was taken. With a breakdown of GP and nurse-led contacts, based on previous research[21] it was assumed that 82% of consultations were conducted at the surgery, 12% of consultations were over the telephone and 3% were either home visits or conducted at other locations.[21] For ease of analysis and to combat the unknown quantity of 'other locations', the 3% conducted at other locations were added to the telephone consultations to make them 15% of all contacts.

Additionally, the cited study[21] disaggregated the numbers further, by assuming that 62% of the above-mentioned consultations were undertaken by GPs, 34% were undertaken by Practice Nurses and 4% by other clinicians. Again, for ease of analysis and the unknown quantity of 'other clinicians', the 4% of other clinicians were classed as being undertaken by practice nurses (Table 2).

Table 3 shows the incidence of cellulitis during the last 10 years in primary and secondary care in NHS Wales as reported by WLGP records using the population of Wales as 2.48 million as only 80% of GP data are captured.

The unit costs for health care utilisation were obtained from several PSSRU sources[16] and are shown in Table A2.

The number of GP events was observed from the SAIL Databank were 3 486 106 (£133 353 176) over the 20-year period. Using the assumption as laid out above, a breakdown of those events is shown in Table 4. Of the 3 486 106 GP events identified over the 20-years, 2 858 607 were deemed a surgery visit, with 1 772 336 (£69 121 115) deemed a GP visit and 1 086 271 (£45 623 366) deemed a Practice Nurse visit.

Additionally, 522 916 were deemed a Telephone call, with 324 208 (£5 031 706) estimated as GP Telephone call and 198 708 (£1 549 923) considered a Telephone call with the Practice Nurse.

Finally, 104 583 were deemed a Home Visit, with 64 842 (£8 688 771) supposed a GP Home Visit and 39 742 (£3 338 295) considered a Practice Nurse Home Visit.

The total resource use over the 20 years demonstrates that female patients accounted for £240 936 917 compared to £170 887 185 for male patients (Table 4).

When the resource use is reviewed alongside the age group, the biggest share of the expenditure was for the 76–90 age group with £132 217 754 followed closely by the age group 91+ with £124 928 422 across the 20-years (Table 4).

Overall cellulitis expenditure by WIMD quintiles in Table 5 showed that total resource usage expenditure was highest in WIMD Quintile area 1 (Most Deprived) with £97 939 134 (or a mean per-person cost of £1614). For the cost of WLGP Events/Contacts the total resource usage was £32 075 934 (or a mean per-person cost of £529) and for the cost of PEDW Admissions, the total resource usage was £65 863 200 (or a mean per-person cost of £6728).

This is in comparison to the overall cellulitis expenditure WIMD Quintile area 5 (Least Deprived) with £66 056 449 (or a mean per-person cost of £1259). For the cost of WLGP Events/Contacts the total resource usage was £22 482 945 (or a mean per-person cost of £428) and for the cost of PEDW Admissions, the total resource usage was £43 573 504 (or a mean per-person cost of £6632).

Table 6 shows an overall summary of resource usage costs as observed in the SAIL Databank over the 20-year period. With the observed number of patients of 274 703,

TABLE 4 Estimated overall resource use costs by sex and age-group over 20-years

Sex		Total cost	Cost admissions (PEDW)	Cost of GP events (WLGP)
Male	Mean	£1330	£5838	£416
	N	128 464	20 116	128 464
	Sum	£170 887 185	£117 440 544	£53 446 641
	Std. deviation	£5429.36	£12 346.50	£555.92
Female	Mean	£1648	£8203	£546
	N	146 238	19 631	146 238
	Sum	£240 936 917	£161 030 688	£79 906 229
	Std. deviation	£6753.47	£16 501.09	£695.46
Age group		**Total cost**	**Cost PEDW admissions**	**Cost of WLGP events**
0–10	Mean	£214	£779	£145
	N	4667	411	4667
	Sum	£999 077	£320 320	£678 757
	Std. deviation	£356.05	£789.68	£143.37
Nov-20	Mean	£311	£878	£253
	N	12 883	855	12 883
	Sum	£4 006 960	£750 464	£3 256 496
	Std. deviation	£391.19	£922.51	£217.81
21–30	Mean	£385	£1093	£319
	N	18 428	1105	18 428
	Sum	£7 088 508	£1 208 064	£5 880 444
	Std. deviation	£532.99	£1361.42	£300.78
31–45	Mean	£557	£1941	£381
	N	31 636	2873	31 636
	Sum	£17 635 658	£5 576 896	£12 058 762
	Std. deviation	£1494.20	£4145.18	£428.61
46–60	Mean	£955	£3743	£456
	N	45 802	6097	45 802
	Sum	£43 726 339	£22 820 096	£20 906 243
	Std. deviation	£4231.25	£10 717.80	£598.12
61–75	Mean	£1442	£5700	£571
	N	56 304	8605	56 304
	Sum	£81 216 952	£49 044 736	£32 172 216
	Std. deviation	£5900.48	£13 756.85	£757.39
76–90	Mean	£2172	£8733	£629
	N	60 882	10 756	60 882
	Sum	£132 217 754	£93 927 392	£38 290 362
	Std. deviation	£7370.88	£15 343.31	£783.97
91+	Mean	£2833	£11 590	£456
	N	44 093	9044	44 093
	Sum	£124 928 422	£104 819 520	£20 108 902
	Std. deviation	£9643.64	£18 490.59	£552.72

TABLE 4 (Continued)

Age group		Total cost	Cost PEDW admissions	Cost of WLGP events
Total	Mean	£1499	£7006	£485
	N	274 703	39 747	274 703
	Sum	£411 824 408	£278 471 232	£133 353 176
	Std. Deviation	£6171.70	£14 595.27	£637.40

TABLE 5 Estimated overall cost by WIMD Quintile 2014 over 20-years

WIMD Quintile 2014		Total cost	Cost PEDW admissions	Cost of WLGP events
1 Most deprived	Mean	£1614	£6728	£529
	N	60 690	9789	60 690
	Sum	£97 939 134	£65 863 200	£32 075 934
	Std. deviation	£6670.77	£15 172.03	£681.07
2	Mean	£1629	£7126	£508
	N	56 535	8896	56 535
	Sum	£92 098 102	£63 388 832	£28 709 270
	Std. deviation	£6636.23	£15 093.59	£670.38
3	Mean	£1489	£7135	£489
	N	56 801	7957	56 801
	Sum	£84 565 043	£56 772 768	£27 792 275
	Std. deviation	£5996.46	£14 314.02	£635.59
4	Mean	£1477	£7479	£463
	N	48 190	6535	48 190
	Sum	£71 165 680	£48 872 928	£22 292 752
	Std. deviation	£6029.26	£14 552.05	£608.40
5	Mean	£1259	£6632	£428
	N	52 487	6570	52 487
	Sum	£66 056 449	£43 573 504	£22 482 945
	Std. deviation	£5299.72	£13 346.23	£567.69
Total	Mean	£1499	£7006	£485
	N	274 703	39 747	274 703
	Sum	£411 824 408	£278 471 232	£133 353 176
	Std. Deviation	£6171.70	£14 595.27	£637.40

WLGP Events/Contacts were estimated at £133 353 176; PEDW Events were estimated at £294 961 888 and the total cost over the 20 years was estimated to be £428 315 064. Using the observed number of patients, the estimated annual cost of treatment was £21 415 753 per annum.

When extrapolated from the observed SAIL Databank to an all-Wales cohort of 3.2 million, the extrapolated cohort is estimated at 366271 patients. WLGP Events/Contacts were estimated at £177 804 235; PEDW Events were estimated at £393 282 517 and the total cost over the 20 years was estimated to be £571 086 752. Using the observed number of patients, the estimated annual cost of treatment was £28 554 338.

Finally, when extrapolated from the observed SAIL Databank to an estimated all UK cohort of 7 325 413 patients.

WLGP Events/Contacts were estimated at £3 556 084 697; PEDW Events were estimated at £7 865 650 347 and the total cost over 20 years was estimated to be £11 421 735 044. Using the observed number of patients, the estimated annual cost of treatment was £571 086 752 for the UK.

TABLE 6 Estimated overall summary of all costs as observed from the SAIL Databank, all Wales and UK perspective over 20-years

SAIL observed data	
No. of patients	**274 703**
WLGP events/contacts	£133 353 176
PEDW events	£294 961 888
Total over 20 years	£428 315 064
Estimated annual cost of treatment (n = 274 703)	£21 415 753
Wales	
No. of patients	366 271
WLGP events/contacts	£177 804 235
PEDW events	£393 282 517
Total over 20 years	£571 086 752
Estimated annual cost of treatment (n = 366 271)	£28 554 338
UK	
No. of patients	7 325 413
WLGP events/contacts	£3 556 084 697
PEDW events	£7 865 650 347
Total over 20 years	£11 421 735 044
Estimated annual cost of treatment (n = 7 325 413)	£571 086 752

5 | DISCUSSION

Economic perspectives on clinical conditions can be important at local, national and global levels. This is the first attempt to estimate the economic burden of cellulitis using the SAIL Databank for both primary and secondary care costs in the Welsh NHS. The direct costs are considerable (£28 554 338) and would represent 0.35% of the annual budget in Wales. In-Patient events and length of stay costs are the main cost drivers with annual estimates of £19 664 126. This is followed by primary care costs of £8 890 212. At a UK level, the cellulitis burden would amount to around £571 086 752 per annum with an average of £1499 per patient. One American paper[22] estimated cellulitis discharges in 2013 cost $3.74 billion (95% CI, $3.65 billion–$3.83 billion) with a median cost per visit of $5159. These costs are not directly comparable since only hospital data were used. Hospital admissions costs for Wales would be £7006 per patient (plus inflation).

Our study also reviewed the overall cellulitis expenditure by WIMD quintiles which demonstrated that resource usage expenditure was highest in WIMD Quintile area 1 (Most Deprived) with £97 939 134 (or a mean per-person cost of £1614), compared to £66 056 449 (or a mean per-person cost of £1259) for WIMD Quintile area 5 (Least Deprived). WIMD measures relative deprivation by geographical areas and takes into account factors such as income, employment, education, housing, and crime. More understanding is needed, but as cellulitis is associated with deprivation due to poor diet and nutrition, challenges with personal skin care, oedema management,[23-26] clothing, and environment, along with recreational substance misuse, the data presented here provides the impetus for risk reduction strategies.

One American study[27] reviewed cellulitis incidence using regional data from health insurance claims and found the rate to be 24.6 per 1000 people. They suggested that of male sex and increasing age were potential risk factors. We did not stratify the data per year to age and sex, but over the 20 years, we found that older age was a factor incurring most cellulitis costs in the 76–90 age group, with £132 217 754 followed closely by the age group 91+ with £124 928 422. In contrast, we did not establish that male sex was a risk factor with cellulitis incidence as our data suggested female sex was more dominant with 146 238 cases compared to males with 128 464 episodes. This is also contrary to a longitudinal Australian cohort study that also found male sex to be a risk factor for cellulitis.[27] However, our study was over 20-years, whereas others were over 10 and 3 years. Capturing incidence data via databanks uses codes rather than case reviews, so there is a possibility of data or coding errors. In this initial trial through the data, we did not distinguish between where on the body the cellulitis was located. Further data analysis may support or refute previous studies reporting a higher prevalence of lower limb cellulitis, but this could be a further paper.

Ellis Simonsen et al[28] also suggested that 78% of care for cellulitis was provided in an outpatient setting, whereas our data suggested that over the two decades, primary care provided 95% of the treatments. This is important and demonstrates that only those requiring emergency treatment were seen through acute hospital settings reducing the pressures on unscheduled care. However, with increasing pressures on general practice, this trend may alter as the incidence of hospital admissions has risen from 3116 to 3902 over the last 10-years.

Concern over antimicrobial resistance is important. Reducing inappropriate antibiotic use while expanding essential access is a difficult challenge at the international level and especially for low and middle-income countries. For UK-based health care, it remains a concern in secondary and primary care, therefore approaches that reduce the need for antibiotics are important. For example, the increasing evidence base that compression rather than prophylactic antibiotics is most useful in preventing recurrence of cellulitis, especially in the previously difficult to treat/higher-risk patients is relevant. Cellulitis is integrally linked to lymphoedema/chronic oedema, not only as a precursor but one can significantly exacerbate the other in a vicious circle.[8] In a recent international study[1] involving 40 sites and 9 countries of 7477 chronic oedema patients, 16% had encountered cellulitis in the last 12 months with a prevalence of 37%. Other risk factors in Burian et al's study[1] included wounds, morbid obesity, obesity, midline oedema, male sex, and diabetes. Given the demonstrated financial encumbrance of cellulitis, a proactive model of cellulitis care, which includes lymphoedema/chronic oedema management to reduce the risk of cellulitis recurrence would seem logical.

Indeed, if prevention of cellulitis schemes could decrease the incidence by a modest 5%, the savings could be nearly 1.5 million per annum for the Welsh NHS alone (5% of £571 086 752, when extrapolated to the UK population = £28 554 337.6 for the UK). This information may nudge a change of practice, providing significant financial impact for health care providers and improved care for the patients at the fore. Possibly the best way of attacking these challenges is ensuring that there are local, regional, national, and international guidelines, protocols, algorithms and cellulitis care pathways with implementation monitoring.

Two decades ago Byford et al[29] criticised cost-of-illness (COI) studies for over-simplification, overestimation of savings and a lack of consideration of outcomes achieved. In this study, the assumptions have been made clear and acknowledging that not all costs can be saved, and that we propose a modest 5% saving. Methods of estimating the burden of costs of specific episodes of illness or particular conditions have expanded since then as access to larger data banks and more sophisticated software have developed. It is therefore important to acknowledge the strengths and limitations of the data used. SAIL data was previously used by members of the research team in 2016[29] and 2020.[30] Lessons learned in those previous studies (including issues with the use of READ codes, diagnosis, GP event definition, etc.) allowed for greater awareness of limitations in this study.

The purpose of this study was not to divert funding from one condition to another as Byford et al feared but to focus on one particular aspect which was considered 'changeable' by clinicians as a first step in understanding where efficiencies and improved patient experience could occur within a national service.

5.1 | Strengths and limitations

This study used data available within the SAIL Databank, covering nearly 80% of primary care data of the Welsh population, and 100% of secondary care data. Extrapolations made from this are likely to represent a realistic estimate of the problem. Although clinical coding may be an issue, we have tried to improve data quality using all cellulitis codes and are possibly more robust than voluntary reports of infections.

The study results provide an important reference point for policymakers in concerting resources and strategies in tackling the main cost drivers for this condition. While it is the ideal scenario in aiming for complete identification and treatment of cellulitis, this is not always possible, but it may be appropriate to target those with repeated recurrence and ensure cellulitis education is readily available for primary care, especially as they see the majority of the cohort.

The coding issues relating to GP events/contacts are limitation, which was addressed in the assumptions made. However, by using the existing evidence,[21] a reasonable assumption was made of the types of contact that patients would have with either the GP or the Practice Nurse. It must be noted that this was before the COVID-19 pandemic, where virtual consultations have taken precedence. Another limitation is using a database to portray incidence. The incidence rates are not verified by case reviews and there could be a miscoding element.

Whilst the population of Wales and the respective Welsh NHS is smaller in comparison to England in the UK, with health being a devolved matter, we believe there is strength in the joined up of nature of services and data availability in Wales, which enables translatable findings which are reflective of the UK NHS as a whole since practice in relation to cellulitis and overall demographics are similar. Importantly, we did not cost other health-related costs, such as the antibiotics or analgesia or the potential impact on the patient, such as loss of employment or quality of life.

The perspective of the Welsh NHS costs shows just one picture of the economic burden cellulitis has on the UK healthcare system. A wider, more societal perspective would shed light on further substantial costs relating to caregivers, loss of productivity, and health-related quality of life.

6 | CONCLUSION

Cellulitis is a common and expensive problem for the NHS. This large data analysis showed that estimated annual direct costs for NHS Wales are substantial (over £28 million). Extrapolated for the UK, this amounts to over £571 million. In-Patient events and length of stay costs are the main cost drivers, with annual Welsh NHS estimates of £19 664 126 with primary care events costing

£8 890 212. Initiatives to identify early signs/risks of cellulitis, improving the accuracy of initial diagnosis, and improved evidence-based treatment pathways to reduce incidence and severity by even small percentages would result in major financial savings and reduce the burden on patients.

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DATA AVAILABILITY STATEMENT

The data used in this study are available in the SAIL Databank at Swansea University, Swansea, UK, but as restrictions apply they are not publicly available. All proposals to use SAIL data are subject to review by an IGRP. Before any data can be accessed, approval must be given by the IGRP. The IGRP gives careful consideration to each project to ensure proper and appropriate use of SAIL data. When access has been granted, it is gained through a privacy-protecting safe haven and remote access system referred to as the SAIL Gateway. SAIL has established an application process to be followed by anyone who would like to access data via SAIL at https://www.saildatabank.com/application-process.

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APPENDIX A

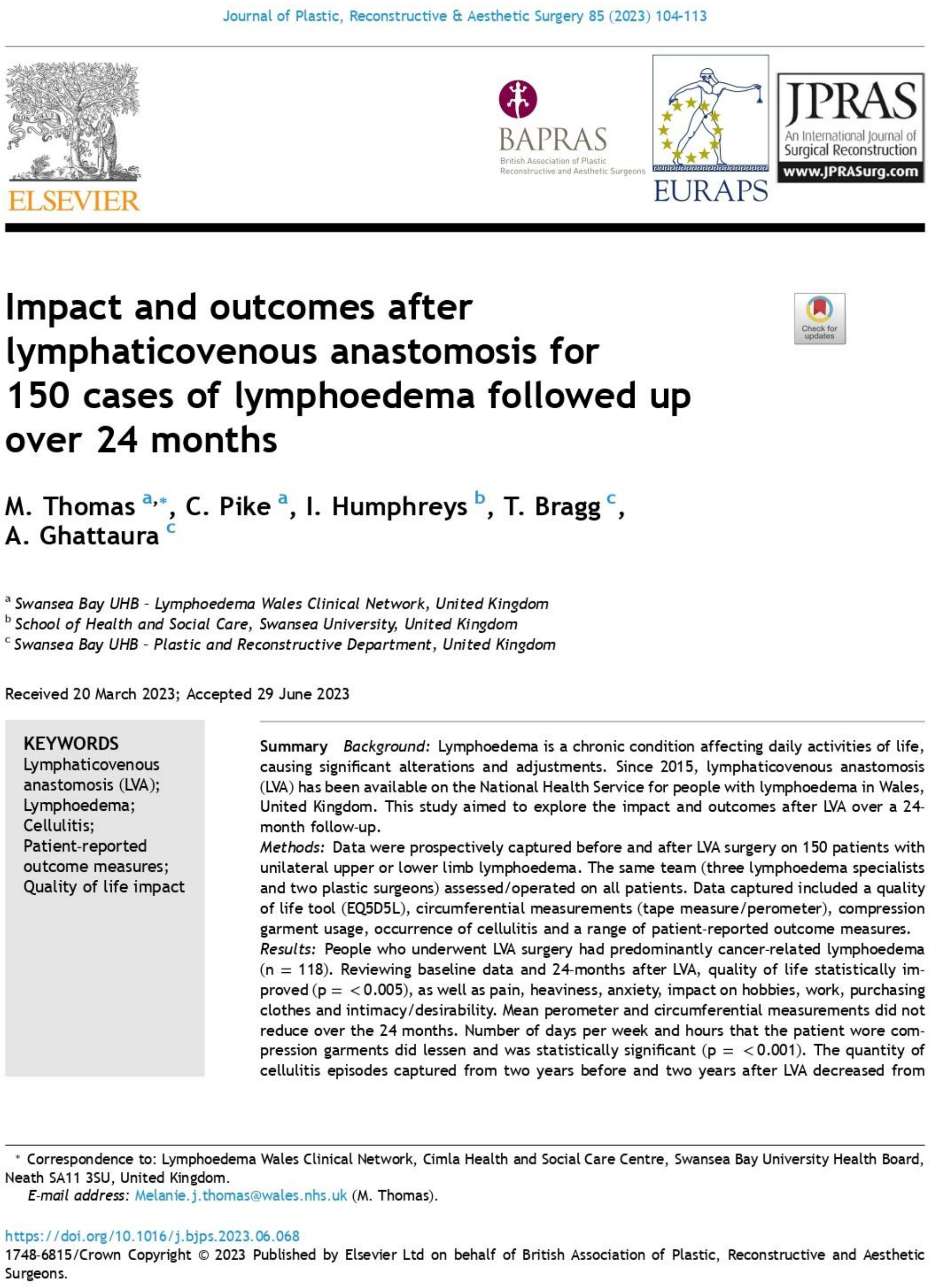
TABLE A1 Unit costs used in the analysis

GP costs	Unit cost (£)	Source
GP surgery visit (Per surgery consultation lasting 9.22 min)	£39	PSSRU 2020
GP home visit	£134	PSSRU 2013 (£114 inflated to 2020 prices using BOE calculator)
Practice nurse	£42	PSSRU 2020
GP telephone triage	£15.52	PSSRU 2020
Practice nurse telephone triage	£7.80	PSSRU 2020
District nurse	£88	PSSRU 2015 (£78 inflated to 2020 prices using BOE calculator)
Inpatient costs (Non-elective)	**Unit cost (£)**	**Source**
Mean daily In-patient costs	£416	Financial Delivery Unit 2019/20 Welsh Health Board Annual Costing Returns as submitted to Welsh Government Parameters: Diagnosis Code L03 (cellulitis and acute lymphangitis) & HRG codes for 'skin disorder'

TABLE A2 Clinical codes (ICD-10 codes in PEDW and read conditions codes in WLGP data)

Code list of ICD10 codes used to identify cellulitis in secondary care (PEDW) data	Modified read code list (conditions) for use in primary care (WLGP) events data
L03, cellulitis	M02z, cellulitis/abscess digit NOS (not otherwise specified)
L030, cellulitis of finger and toe	M036, cellulitis/abscess-leg ex. foot
L031, cellulitis of other parts of limb	M0363, cellulitis/abscess-lower leg
L032, cellulitis of face	M036z, cellulitis/abscess-leg NOS
L033, cellulitis of trunk	M03z, cellulitis/abscess NOS
L038, cellulitis of other sites	M03z0, cellulitis NOS
L039, cellulitis, unspecified	M08, Cutaneous cellulitis
	M085, cellulitis of leg
	M088, cellulitis of arm
	M08B, cellulitis of foot

Section 3:8 Paper 8 M Thomas, C Pike, I Humphreys, T Bragg and A Ghattaura, The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months journal of Plastic, Reconstructive &Aesthetic Surgery, (2023)



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Impact and outcomes after lymphaticovenous anastomosis for 150 cases of lymphoedema followed up over 24 months

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KEYWORDS
Lymphaticovenous anastomosis (LVA);
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Cellulitis;
Patient-reported outcome measures;
Quality of life impact

Summary *Background:* Lymphoedema is a chronic condition affecting daily activities of life, causing significant alterations and adjustments. Since 2015, lymphaticovenous anastomosis (LVA) has been available on the National Health Service for people with lymphoedema in Wales, United Kingdom. This study aimed to explore the impact and outcomes after LVA over a 24-month follow-up.

Methods: Data were prospectively captured before and after LVA surgery on 150 patients with unilateral upper or lower limb lymphoedema. The same team (three lymphoedema specialists and two plastic surgeons) assessed/operated on all patients. Data captured included a quality of life tool (EQ5D5L), circumferential measurements (tape measure/perometer), compression garment usage, occurrence of cellulitis and a range of patient-reported outcome measures.

Results: People who underwent LVA surgery had predominantly cancer-related lymphoedema ($n = 118$). Reviewing baseline data and 24-months after LVA, quality of life statistically improved ($p = < 0.005$), as well as pain, heaviness, anxiety, impact on hobbies, work, purchasing clothes and intimacy/desirability. Mean perometer and circumferential measurements did not reduce over the 24 months. Number of days per week and hours that the patient wore compression garments did lessen and was statistically significant ($p = < 0.001$). The quantity of cellulitis episodes captured from two years before and two years after LVA decreased from

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4.22 to 0.10 ($p = <0.001$). Significant results ($p = <0.001$) were also found in a reduction in patients taking days off work due to cellulitis (5.81 to 0.16).

Conclusion: LVA resulted in significant improvements in patient-reported outcome measures, cellulitis episodes and reduced need for compression garments. Limb circumference via tape measure/perometer did not alter, yet the patient's quality of life considerably improved.

Lymphoedema is a debilitating, progressive condition caused by an excess accumulation of protein-rich fluid within the subcutaneous tissues.[1,2] Lymphoedema occurs due to lymphatic system dysfunction from a reduced transport capacity or increased capillary filtrate. Lymphoedema is classified into two categories: primary and secondary. Primary lymphoedema is due to a genetic or lymphatic development dysfunction. In contrast, secondary lymphoedema is direct or indirect damage to the lymphatic system, such as cancer, venous disease, dermatological conditions, obesity, trauma or immobility.[3] Although lymphoedema can affect the young and old, it is more prevalent in the older population.[4] Those affected may experience skin changes, recurrent cellulitis, wounds, lymphorrhoea, pain, mobility issues and heaviness.[5] Evidence suggests that the impact of lymphoedema on an individual's health, well-being, sense of self, emotions and quality of life may be profound and even extends to work and family life.[1,4-7] As a chronic debilitating condition, lymphoedema can significantly impact health outcomes, and the consequences are a significant burden to the NHS.

The conservative treatment of lymphoedema focuses on skin care (moisturising daily), activity and movement, weight management, compression and massage. Subsequently, they aim to increase patients' knowledge and encourage self-management.[1,8] Nevertheless, the mainstay of treatment—wearing compression garments daily—causes substantial implications to life, affecting hobbies, work and intimacy and is a constant reminder that they have a chronic condition.[1] For some patients, complex decongestive therapy entailing manual lymphatic drainage massage and multi-layer lymphoedema bandaging is recommended, requiring daily attendance at a lymphoedema clinic for two to four weeks. Although time-consuming, this treatment can reduce limb volume and skin complaints; maintaining the improvements requires lifelong patient compliance in wearing compression garments.[1] Thus, although conservative treatment can relieve symptoms, the need for compression garments and self-management remains life-long.[9,10]

In recent years, surgery for lymphoedema has advanced from widespread tissue removal to physiological techniques such as lymph node transfers and lymphaticovenous anastomosis (LVA).[10] LVA is a minimally invasive super-micro-surgery that redirects fluid from inadequate lymph vessels to functioning veins by anastomoses.[9] LVA has a growing evidence base for patients with peripheral lymphoedema.[11,12] Research has demonstrated promising results, including decreased volume measurements, reduced cellulitis episodes and improved quality of life.[12-16] However, most studies have under 100 patients, and follow-up is less than two years.

Since 2015, LVA has been available annually on the Welsh NHS for forty people with lymphoedema. NHS Wales supported the service's inception in partnership with the Regional Plastic and Reconstructive Surgery Department and Lymphoedema Wales Clinical Network (LWCN). Capital funding from the local government supported the purchase of the indocyanine green (ICG) lymphography machines and microscope. Two years of prospective data have been captured on the clinical outcomes of the first 150 patients undergoing LVA.

Aims and objectives

This study aimed to explore the impact and outcomes after LVA over 24 months of follow-up of 150 patients.

The specific objectives were to:

- Measure changes in patient-reported outcomes before and after LVA;
- Determine limb volume change;
- Describe the health-related quality of life in service users before and after surgery.

Methods

The regional plastic and reconstructive surgery department and LWCN gained Wales NHS approval to deliver an LVA service for a small cohort of patients. Formal inclusion and exclusion criteria for referral were established (Table 1). The same team (three lymphoedema specialists and two surgeons) assessed and operated on all patients. All surgeries were undertaken at Swansea Bay University Health Board from September 2015 to February 2020 under local anaesthetic. All patients had LVA on unilateral limb lymphoedema performed by two surgeons over four hours. One hundred and fifty patients were prospectively followed up at three set points: 3, 12 and 24 months post-operatively. All patients consented to collect their data anonymously as part of the LVA service evaluation. The design was a prospective, interventional study with data collected before and after LVA surgery.

Table 1 Inclusion and exclusion criteria for LVA surgery based on ICG scanning assessment.

Inclusion Criteria	Exclusion Criteria
Lymphoedema diagnosed in upper or lower limbs with a maximum overall excess of 20% difference	Excess volume more than 21% difference
Skin tissues are soft	Skin folds, fibrosis, wounds or ulcers
Body mass index (BMI) 30 kg/m^2 or under at time of ICG scanning	BMI 31 kg/m^2 or over
Had cellulitis in last two years requiring two courses of antibiotics	Active cancer disease
International Society Lymphology staging I-II	Pregnant
ICG lymphography showing linear patterns and limited stardust patterns	Venous hypertension
Resides in Wales, United Kingdom	Smoker

Ethics and research governance

This study design was reviewed by the Joint Study Review Committee at Swansea Bay University Health Board, Wales and deemed a service evaluation/data audit. Swansea University College of Human and Health Sciences ethics committee provided permission to analyse the anonymised data (ethics application reference: 170419d).

Patient selection

All patients aged 18 or over, under the care of LWCN, were eligible to be referred for ICG lymphography with a view to LVA surgery if they met the inclusion criteria listed in Table 1. Patients with primary or secondary lymphoedema from cancer or non-cancer origin were included. All patients had to provide informed consent and comply with an existing lymphoedema management plan. No time limits were set on the onset of lymphoedema. In addition, all patients reported experiencing cellulitis requiring two courses of antibiotics in the previous two years.

Data collection measures

Data capture included soft and hard outcomes, including body mass index (BMI), the International Lymphedema Framework (ISL) staging, health-related quality of life tool (EQ5D5L), circumferential measurements using a tape measure and perometer, compression garment usage, the incidence of cellulitis and a range of patient-reported outcome measures (PROMs).

Health-related quality of life tools

The EQ-5D-5L is a generic health-related quality of life questionnaire capturing a descriptive classification consisting of five dimensions of health.[17] Patients completed EQ-5D-5L throughout the evaluation.

PROMs

Patients completed a range of PROMs pre- and post-operatively.[7] Items captured included the impact of lymphoedema on pain, heaviness, anxiety, intimacy/ desirability, hobbies, work, purchasing clothes/shoes and holidays on a scale of 0 (no impact) to 10 (highest impact).

Circumferential measurements

Tape measurements of the limbs were recorded, determining the size of the affected limb compared with the unaffected side. Upper limbs were measured every 4 cm from the wrist to the axilla and lower limbs from the ankle to the groyne. A standardised measuring protocol included the same patient position and a definitive starting point ('first mark'). The 'first mark' was identified as a set point from the fifth digit nail bed to 2 cm above the ulnar process in upper limbs; for lower limbs, the 'first mark' was measured from the base of the heel to 2 cm above the lateral malleolus. Calculations for limb volume utilised the cylinder method.[18]

The perometer is an optoelectrical device used to measure upper or lower limb volume. Its frame consists of photo sensors and infra-red light-emitting diodes, illuminating and scanning the limb. The limb is measured every 4 mm and limb volume is calculated using an elliptical disc (cylinder) method.[18,19]

Compression garments

Data capture included types of garments worn (made-to-measure or ready to wear at class 1, 2 or 3). Patients were asked at each visit how many days they wore the garment the previous week and the average number of hours worn per day. The patients also reported on the impact the compression garment had on their life, rated 0-10, with 0 being no impact and 10 maximum impact.

Occurrence of cellulitis

The number of episodes of cellulitis was recorded pre-LVA (two years before surgery) and then captured accumulatively until 24 months. This data also included whether the cellulitis required hospitalisation and cost calculated using national sources.[20,21] The numbers of patients prescribed prophylactic antibiotics and the impact of having cellulitis on life (rated 0-10) were also recorded throughout the evaluation.

Data and statistical analysis

Descriptive demographic statistics were captured by assigning each patient to a number in sequential order to maintain anonymity. All continuous variables were reported as mean with standard deviation and categorical data as frequency. Differences in pre- and post-operative means were analysed using paired-samples *t*-test with a 95% confidence interval to examine the effect of LVA. Linear regression analysis tested the differences in change scores (pre-operative minus post-operative) between the operated and 'normal' limb. To deal with loss to follow-up, post-operative outcome measurements were compared with the related pre-operative outcome measures for the specific number of patients.

Results were analysed using MS EXCEL and IBM SPSS Statistics for Windows, version 26 (INM corp. ®, Armonk, N.Y. USA). A *p*-value < 0.05 was considered statistically significant. The perspective taken was that of the UK NHS.[20]

Results

The first 150 patients eligible for LVA surgery were 19 male (13%) and 131 female (87%), with an average age of 53.8 years (range 21-79 years). Table 2 presents the demographic data captured, including age, BMI, lymphoedema classification, ISL staging and affected limb and side. The average BMI before surgery was 26.9 kg/m^2 (range 17.6-35.9 kg/m^2). The criteria for LVA was a BMI under 31 kg/m^2 on indocyanine green (ICG) scanning. However, the time lapse between ICG scanning and pre-operative assessment found 14 patients gaining weight resulting in a BMI in excess of 31 kg/m^2. One hundred and eighteen patients (79%) had secondary lymphoedema due to cancer, 21 (14%) had primary lymphoedema and the remaining 11 (7%) had secondary lymphoedema due to non-cancer causes. Pre-operatively, 150 patients were assessed, reducing to 147 at 3 and 12 months and 143 at 24 months. The attrition rate at 24 months was due to two patients who had died and five were lost to follow-up. All patients who did not complete the 24-months follow-up had upper limb cancer-related lymphoedema. Other missing data shown in the subsequent tables were due to unattended appointments or unable to capture the data objectively, as the appointment was virtual during the pandemic restrictions.

As detailed in Table 3, an increase in BMI was reported over time, with the difference from pre-op to 24 months being statistically significant ($p = 0.002$). The Obese Class 2 (35-39.9) changed from two people pre-operatively to 12 at 24 months. Nevertheless, the lymphoedema ISL staging reduced in severity, demonstrating significant improvement ($p < 0.001$) at each visit over the 24 months from the pre-operative baseline. For example, the ISL stage II b (non-pitting) was 73 people before LVA and 36 at 24 months post-LVA.

Table 2 Patient demographics.

Variable	Category	n	(%)
Age group	21-30	10	(7)
	31-40	12	(8)
	41-50	32	(21)
	51-60	51	(34)
	61-70	31	(21)
	70+	14	(9)
BMI	Underweight (<18.5)	1	(1)
	Normal weight (18.5-24.9)	49	(33)
	Overweight (25-29.9)	64	(43)
	Obese class 1 (30-34.9)	34	(23)
	Obese class 2 (35-39.9)	2	(1)
Lymphoedema classification	Primary	21	(14)
	Secondary cancer	118	(79)
	Secondary Non-cancer	11	(7)
ISL staging	Stage 0 (Latent)	1	(1)
	Stage I (Mild)	27	(17)
	Stage II a (Pitting)	49	(33)
	Stage II b (Non-pitting)	73	(49)
Affected limb	Upper limb	101	(67)
	Lower limb	49	(33)
Affected side	Right	69	(46)
	Left	81	(54)

Health-related quality of life and PROMs

Table 4 details the change over time for the EQ-5D-5L and PROM data. Both sections of the EQ-5D-5L form demonstrated statistically significant improvements in general health ($p < 0.04$). Using the visual analogue scale of 0-10 for pain and heaviness of the lymphoedema limb, there was a statistically significant reduction in pain ($p < 0.001$) and heaviness ($p < 0.001$). The patient-reported outcomes on anxiety, intimacy/desirability, hobbies, work and clothes also demonstrated a significant reduction in scores ($p < 0.001$).

In terms of the ability to work due to their lymphoedema, there were fewer people who had to modify their work, although, at 24 months, this was not statistically significant ($p = 0.051$). There was a statistically significant improvement in the impact lymphoedema had on patients' choice of holiday over the two years ($p < 0.05$). However, a reported increased financial burden was due to their lymphoedema over the 24 months ($p < 0.001$). For those whose lymphoedema was secondary to cancer, there was no change in how often they thought about their cancer journey following LVA surgery.

Circumferential measurements

The percentage difference between limbs using 4 cm tape circumferential measurements showed no statistically

Table 3 BMI and ISL staging over 24 months.

Variable	Pre-op		3-month				12-month				24-month			
	n = 150	(%)	n = 147	(%)	Mean Diff	p-value	n = 144[a]	(%)	Mean Diff	p-value	n = 140[a]	(%)	Mean Diff	p-value
BMI														
Underweight (< 18.5)	1	(1)	1	(1)	-0.36	0.278	1	(1)	-0.16	0.165	1	(1)	-0.47	***0.002***
Normal weight (18.5-24.9)	49	(33)	48	(32)			42	(29)			42	(30)		
Overweight (25-29.9)	64	(43)	63	(43)			67	(46)			58	(41)		
Obese class 1 (30-34.9)	34	(23)	32	(22)			29	(20)			27	(19)		
Obese class 2 (35-39.9)	2	(1)	3	(2)			5	(4)			12	(9)		
Lymphoedema staging														
Stage 0 (Latent)	1	(1)	20	(14)	0.33	< ***0.001***	29	(20)	0.48	< ***0.001***	39	(28)	0.75	< ***0.001***
Stage I (Mild)	27	(17)	20	(14)			24	(17)			27	(19)		
Stage II a (Pitting)	49	(33)	52	(35)			41	(28)			38	(27)		
Stage II b (Non-pitting)	73	(49)	55	(37)			50	(35)			36	(26)		

[a] Missing data due to virtual appointments as result of pandemic restrictions. All were upper limb lymphoedema related to breast cancer.

Table 4 Health-related quality of life and patient-reported outcome measures.

Paired-samples statistics	Pre-op		Pre-op to 3 months				Pre-op to 12 months				Pre-op to 24 months			
Health-related quality of life	Mean score	(SD)	Mean score	(SD)	Mean diff	p-value	Mean score	(SD)	Mean diff	p-value	Mean score	(SD)	Mean diff	p-value
EQ-5D-5 L index	0.74	(0.18)	0.79	(0.20)	0.05	< ***0.001***	0.80	(0.19)	0.06	< ***0.001***	0.80	(0.20)	0.06	< ***0.001***
Health score (0-100)	74.07	(19.50)	77.56	(18.35)	3.50	***0.032***	77.56	(17.98)	3.43	***0.032***	78.75	(18.89)	4.18	***0.003***
Patient-reported outcome measures (PROMs) (0-10)														
Pain	2.62	(2.39)	1.56	(1.74)	-1.06	< ***0.001***	1.36	(1.67)	-1.20	< ***0.001***	1.48	(1.92)	-1.04	< ***0.001***
Heaviness	4.55	(2.66)	2.65	(2.24)	-1.90	< ***0.001***	2.56	(2.53)	-1.90	< ***0.001***	2.79	(2.57)	-1.70	< ***0.001***
Anxiety	5.75	(2.66)	4.19	(2.72)	-1.56	< ***0.001***	3.77	(2.90)	-1.96	< ***0.001***	3.32	(2.82)	-2.42	< ***0.001***
Intimacy/desirability	6.34	(2.93)	4.64	(2.98)	-1.70	< ***0.001***	4.22	(3.22)	-2.11	< ***0.001***	3.74	(3.18)	-2.62	< ***0.001***
Hobbies	5.38	(2.69)	3.51	(2.79)	-1.87	< ***0.001***	3.05	(2.75)	-2.31	< ***0.001***	2.78	(2.73)	-2.52	< ***0.001***
Work	5.23	(2.93)	3.51	(2.81)	-1.72	< ***0.001***	3.07	(2.97)	-2.06	< ***0.001***	2.62	(2.80)	-2.45	< ***0.001***
Clothes	6.64	(3.04)	5.13	(3.31)	-1.51	< ***0.001***	4.46	(3.25)	-2.18	< ***0.001***	4.20	(3.57)	-2.48	< ***0.001***
Impact on general life														
Modify work	2.34	(1.37)	2.09	(1.42)	0.25	***0.014***	1.99	(1.47)	0.34	***0.004***	2.08	(1.56)	0.23	0.051
Impact on holidays	2.15	(1.06)	1.93	(1.15)	0.22	0.052	1.70	(1.08)	0.43	< ***0.001***	1.78	(1.18)	0.32	***0.002***
Financial status affected due to lymphoedema	1.80	(0.51)	1.85	(0.48)	-0.05	0.158	1.95	(0.45)	-0.15	< ***0.001***	1.98	(0.42)	-0.19	< ***0.001***
Think of cancer journey	3.00	(1.29)	3.18	(1.34)	-0.18	***0.037***	3.01	(1.42)	0.00	1.000	3.03	(1.46)	-0.01	0.952

Table 5 Limb volume percentage difference from tape circumferential measurement and perometer.

Measurement	Pre-op		3 months			12 months			24 months		
	Mean	(SD)	Mean	(SD)	p-value	Mean	(SD)	p-value	Mean	(SD)	p-value
Tape measure full limb	11%	(11%)	11%	(11%)	0.838	11%	(10%)	0.534	12%	(12%)	0.192
Tape measure distal[a]	15%	(15%)	0.16	(16%)	0.346	15%	(15%)	0.762	16%	(16%)	0.197
Tape measure proximal[b]	9%	(11%)	0.08	(11%)	0.443	8%	(10%)	0.294	9%	(11%)	0.326
Perometer full limb	11%	(12%)	11%	(12%)	0.748	12%	(11%)	0.317	13%	(12%)	***< 0.001***
Upper limb tape measure	14%	(11%)	13%	(11%)	0.531	13%	(9%)	0.184	14%	(11%)	0.587
Upper limb perometer	14%	(11%)	14%	(13%)	0.688	14%	(10%)	0.789	16%	(11%)	***0.010***
Lower limb tape measure	6%	(8%)	6%	(9%)	0.413	6%	(10%)	0.271	7%	(11%)	0.056
Lower limb perometer	5%	(9%)	6%	(10%)	0.061	6%	(10%)	0.079	7%	(11%)	***< 0.001***

[a] Distal includes the forearm from wrist to elbow or the lower leg from ankle to knee.

[b] Proximal includes the upper arm from elbow to axilla or the thigh from knee to groin.

significant change over the 24 months ($p > 0.05$). However, the percentage difference between limbs using the perometer showed a volume difference between limbs from 11% to 13% is statistically significant ($p < 0.001$).

When comparing the data for upper and lower limb lymphoedema, there were no statistically significant changes in limb volume difference with the 4 cm circumferential tape measurement ($p > 0.05$). However, the perometer showed a slight increase in volume and was statistically significant at both the upper ($p = 0.010$) and lower limbs ($p < 0.001$) (Table 5).

Compression garments

There was a statistically significant reduction in the number of days and total hours per day that patients wore their compression garments from before to 24 months after LVA surgery ($p < 0.001$). Positively, there was a reduction in the impact patients experienced in having to wear their compression garments at 3-, 12- and 24 months ($p < 0.001$) (Table 6). Conversely, the time patients wore their garments increased from pre-LVA to 3 months post-operatively but then reduced. The types of garments worn at pre-operative assessment were Made-to-Measure (n = 41 [27%]), Ready to Wear (RTW) Class 1 (n = 42 [28%]) and RTW Class 2 and above (n = 67 [45%]). At 24 months, the garments were reported as Made-to-Measure (n = 14 [10%]), RTW Class 1 (n = 63 [44%]), RTW Class 2 (n = 22 [15%]) and Nil (n = 44 [31%]).

Separating the upper and lower limb data also demonstrated a statistically significant reduction in usage. Further analysis into whether a patient had primary or secondary lymphoedema (Table 7) showed that those with secondary lymphoedema reduced garment wear more and was statistically significant ($p < 0.001$). Similarly, data analysis on lymphoedema ISL staging reported a higher reduction in garment wear for those patients with type I and IIa compared with IIb with statistically significant results (Table 8).

Occurrence of cellulitis

Episodes of cellulitis statistically reduced from pre- to post-LVA ($p < 0.001$), with costs reducing from £169.53 to £4.21

Table 6 Garment usage and impact pre- and post-LVA.

	Pre-op		3 months			12 months			24 months		
	Mean	(SD)	Mean	(SD)	p-value	Mean	(SD)	p-value	Mean	(SD)	p-value
Number of days per week wear garment	6.23	(1.54)	6.34	(1.62)	0.405	5.37	(2.60)	***< 0.001***	4.99	(2.76)	***< 0.001***
Number of hours per day wear garment multiplied by 7 days	101.17	(49.81)	103.89	(47.96)	0.488	81.95	(55.99)	***< 0.001***	78.90	(58.72)	***< 0.001***
Impact of garments on life (0-10)	6.90	(2.55)	5.40	(2.82)	***< 0.001***	4.45	(3.03)	***< 0.001***	3.87	(3.14)	***< 0.001***
Number of hours per day wear garment multiplied by 7 days for **upper limb**	97.85	(51.9)	99.45	(49.93)	0.721	78.23	(58.20)	***< 0.001***	74.32	(59.97)	***< 0.001***
Number of hours per day wear garment multiplied by 7 days for **lower limb**	107.94	(44.79)	112.94	(42.69)	0.519	89.47	(50.95)	***0.010***	89.47	(50.95)	***0.010***

Table 7 Garment usage comparing secondary and primary lymphoedema.

				95% confidence interval of the difference			
Secondary lymphoedema	Mean	N	Std. Dev.	Mean	Lower	Upper	p-value
Pre-LVA days wears garment	6.2	117	1.6	1.436	0.923	1.948	***< 0.001***
24-month days wearing garment	4.7	117	2.9				
Pre-LVA total hours per week	100.9	117	51.3	26.103	15.998	36.207	***< 0.001***
24-month total hours per week	74.8	117	61.4				
Primary lymphoedema							
Pre-LVA days wears garment	6.4	26	1.2	0.19	-0.5	0.9	0.563
24-month days wearing garment	6.2	26	1.5				
Pre-LVA total hours per week	101.1	26	46.1	3.54	-14.1	21.2	0.684
24-month total hours per week	97.5	26	40.9				

Table 8 Garment usage comparing ISL staging lymphoedema.

				95% confidence interval of the difference			
ISL stage I	Mean	N	Std. Dev.	Mean	Lower	Upper	p-value
Pre-LVA days wears garment	5.6	27	2.2	1.89	0.7	3.1	**0.003**
24-month days wearing garment	3.7	27	2.9				
Pre-LVA total hours per week	85.0	27	50.6	27.56	6.7	48.5	**0.012**
24-month total hours per week	57.4	27	57.9				
ISL stage II a							
Pre-LVA days wears garment	6.5	47	1.1	1.34	0.6	2.1	***< 0.001***
24-month days wearing garment	5.2	47	2.7				
Pre-LVA total hours per week	109.6	47	45.4	34.45	20.3	48.6	***< 0.001***
24-month total hours per week	75.1	47	52.5				
ISL stage II b							
Pre-LVA days wears garment	6.2	68	1.5	0.77	0.2	1.4	**0.014**
24-month days wearing garment	5.5	68	2.5				
Pre-LVA total hours per week	100.4	68	52.2	9.15	-3.9	22.2	0.165
24-month total hours per week	91.2	68	60.6				

per patient. The number of people hospitalised due to cellulitis also demonstrated significant improvements ($p < 0.001$), with average per-patient hospital costs reducing from £1421.68 to £107.34. Furthermore, the number of days patients had to take off work due to cellulitis reduced from 5.81-0.16 ($p < 0.001$). Surprisingly, even though the episodes of cellulitis had decreased, the patients reported impact of cellulitis worsened from 2.30 to 3.77 ($p < 0.001$) at 24 months. There was no change in the need for prophylactic antibiotics used over the 24 months (Table 9).

Discussion

This prospective, interventional study of 150 upper and lower limb lymphoedema patients followed up for 24 months demonstrated statistically significant improvements in quality of life from pre- to post-LVA. As well as general health improvements reported by the EQ-5D-5L, this study also captured PROMs via LYMPROM©. One of the strengths of this study is the largest population with the longest follow-up at 3, 12 and 24 months indicating improvements using validated tools. Other studies have used validated questionnaires; however, the sample sizes ranged from 10 to 100, with only one study following up for 24 months.[10,12-15,22,23] Surprisingly, improvements in quality of life did not correlate with a reduction in limb volume.

The volume circumference measurements did not improve for the upper or lower limbs using the perometer or the 4 cm tape measurement method. Neither did it improve when we only considered the proximal or distal volume measurements. Limited change of mean volume is similar to other studies.[12,15,24] All patients belonged to lymphoedema services and would have been treated with compression conservatively pre-LVA. However, despite no decrease in limb volumes, there was an overall reduction in the hours and days of compression garments used. Thus, lymphoedema progression may have been halted through the LVA surgery, as patients' limb volume differences were similar to the pre-operative, even though they were using less compression.

Compression greatly impacts patients with lymphoedema as they are cosmetically unappealing, negatively impacting their quality of life.[1] Highlighting and beginning to understand the psychological, physical and social implications compression garments have on life is important. In our study, the numbers of days and hours that compression garments were being worn reduced over the 24 months. Corroborating this, patient outcomes on the impact

Table 9 Cellulitis events, hospitalisation costs and impact.

	Pre-op		Pre-op to 3 month			Pre-op to 12 months			Pre-op to 24 months		
	Mean	(SD)	Mean	(SD)	p-value	Mean	(SD)	p-value	Mean	(SD)	p-value
Number of cellulitis events pre-LVA	4.22	-6.02	0.06	-0.24	< ***0.001***	0.20	0.44	< ***0.001***	0.31	0.61	< ***0.001***
Cellulitis cost (£) per patient	169.53	-241.72	2.43	-9.6	< ***0.001***	5.74	-14.1	< ***0.001***	4.21	-12.35	< ***0.001***
Hospitalised for cellulitis	0.46	-1.01	0.01	-0.08	< ***0.001***	0.02	0.14	< ***0.001***	0.06	0.31	< ***0.001***
Hospitalisation cost (£) per patient	1421.68	-3101.44	20.6	-251.5	< ***0.001***	41.77	-356.86	< ***0.001***	107.34	-847.64	< ***0.001***
Days off work due to cellulitis	5.81	-13.34	0.17	-1.29	< ***0.001***	0.34	1.59	< ***0.001***	0.5	1.8	< ***0.001***
Prophylactic antibiotics	0.12	-0.33	0.06	-0.24	***0.029***	0.1	-0.29	0.493	0.11	-0.32	0.656
Impact cellulitis on life	2.3	-0.69	3.92	-0.36	< ***0.001***	3.78	-0.59	< ***0.001***	3.77	-0.71	< ***0.001***

compression garments have on their life also improved. Interestingly, garment usage increased from baseline to 3-months post-LVA. Patients wanting the surgery to be successful may explain this increase in compression use by following the guidance provided post-operatively explicitly. Some studies have suggested that upper limb LVA patients reduced their need for compression garments more than the lower limb patients.[12] This was not seen in our study, as both upper and lower limb usage had reduced garment wear, which was statistically significant. A unique point of our study was delving deeper into the differences between primary and secondary lymphoedema, lymphoedema ISL staging and compression worn. Primary lymphoedema patients did not reduce the number of hours garments were worn per week (101.1-97.5 h; $p = 0.684$) compared with secondary lymphoedema patients (100.9-74.8 h; $p < 0.001$). Likewise, ISL stage IIb patients did also not reduce wear as much as stage 1 (100 h to 91 compared with 85 h to 57; $p = 0.165$ vs $p = 0.12$). This lack of reduction in hours may be related to a deeper lymphatic dysfunction and disease progression and questions patient selection.

Like many other LVA studies, most patients were female (131/150), with 118/150 diagnosed with lymphoedema secondary to cancer. Positively, a varying age group of young and older people were offered LVA (age 21-79 years) and 33% were for lower limb lymphoedema. The BMI data showed that over the 24 months of follow-up, the number of patients who were classified as Obese Class 2 (35-39.9) increased from 3/150-12/140, and this increase was statistically significant ($p = 0.002$). There is strong evidence that obesity is a cause and risk for exacerbating lymphoedema and increasing subcutaneous fibrosis.[25] All patients undergoing LVA are advised on the benefits of maintaining a healthy BMI to support effectiveness that is supported by leaflets and media films. As part of the LVA criteria, patients undergoing LVA had an ISL stage of II b (non-pitting) or less. Positively, there was a 50% reduction of those classified as stage II b (non-pitting) from baseline to 24 months post-LVA. ISL stage 0 (latent) rose from one patient pre-LVA to 39 patients recorded 24 months post-LVA. These improvements were statistically significant ($p < 0.001$). It is posited that the reduction in signs and symptoms of lymphoedema is due to patent LVAs having reversed the side effects of the previously dysfunctional lymphatics.

The PROMs on anxiety, intimacy/desirability, hobbies, work, clothes and holidays significantly improved scores from before and after LVA. From our previous study,[1] a possible explanation for the PROMs improvement may be due to the reduction in the believed need to wear compression garments or an effort justification by the high effort or cost of LVA elevating the perceived value of the benefit. Conversely, although most patients' outcomes improved, the financial burden did not. One reason given for this increase in financial burden was the need to travel to a central lymphoedema clinic for their appointments following LVA surgery, as opposed to their local lymphoedema services. However, further investigation is required to investigate the additional reasons that may be contributing to this increase in financial burden.

Cellulitis is a skin infection that causes pain, swelling and flu-like symptoms, with a life-threatening risk of sepsis if mismanaged. It represents a significant burden to the NHS, causing admission, antibiotic costs and appointments with primary care.[26] If the incidence of cellulitis reduces, so does the financial burden. Recurrent cellulitis has a detrimental effect on the lymphatic system exacerbating lymphoedema volume. Positively, the cellulitis episodes greatly reduced from 24 months pre-LVA to 24 months post-LVA, irrespective of upper or lower limb lymphoedema or primary or secondary causes. This is a very exciting finding as it has huge implications for financial savings and avoidance of NHS expenditure supporting future LVA services. Our study further supports the literature that LVA can reduce cellulitis episodes.[8,12] There remains a dearth of literature concerning the economic costs and benefits associated with LVA; our paper tentatively suggests cost avoidance related to cellulitis hospitalisation that should be expanded on in future research.

The LVA Service in Wales is run in partnership with surgeons and the National Lymphoedema Service specialists. This collaboration has many benefits in placing the patient at the centre, enabling treatment by the right person at the right time.

Limitations

Some patients were lost to follow-up. During the pandemic, we could not see some patients face-to-face, thus, key measurements were unable to be captured. Nevertheless, 143/150 (95%) patients' data at 24 months is extremely encouraging and is one of the largest prospective studies to date. While the population is limited to one area of the UK, the age of the patients did show some diversity; however, the cohort was mainly women with lymphoedema secondary to cancer. Ideally, a randomised controlled trial comparing costs of conservative versus LVA would be beneficial in the future.

Conclusion

This study highlights important improvements in patients' quality of life and PROMs after undergoing LVA. Irrespective of limited volume circumference change, patients were able to decrease their usage of compression garments. An important finding has been the significant reduction in cellulitis episodes that has a wider impact on supporting LVA services through cost avoidance and reduction.

Funding

No research funding was received; however, we did receive capital money from the Welsh Government Efficiency through Technology Fund to purchase the ICG Scanning equipment and the microscope.

Ethical approval

This study design was reviewed by the Joint Study Review Committee at Swansea Bay University Health Board, Wales and deemed a service evaluation/data audit. Swansea University College of Human and Health Sciences (CHHS) ethics committee provided permission to analyse the anonymised data (ethics application reference: 170419d).

Declaration of Competing Interest

None declared.

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Chapter 4

Critical review

This critical review of the previously published eight papers included in this thesis will include the chronology and timing of the research, and the subsequent reports and journal papers that were written based on the research undertaken. This review will also discuss how the research was developed by myself and LW, and also why they developed and written in such a way. The review will analyse how the papers fit into current research and how they relate to Health Economics and Economic Evaluation, HRQoL, PROMs, and concepts such as Prudent Healthcare and Value-Based Healthcare.

Throughout the critical review, the cumulative effect of these papers will be discussed and contextualised with Lymphoedema service practices in Wales and how they contributed to evidence-based practice and policy changes. The critical review will also discuss the research related limitations present across all the papers and will also discuss and contextualise my contribution to the papers, independent of the other co-authors. It will also discuss and analyse the impact of the research from the papers' inclusion in the REF 2021 exercise.

Finally, this review will consider research work currently underway with LW. This is both research in its infancy, and research currently under review with journal editors. And it will also discuss future work that is planned with LW with several partners including new collaborators (Cardiology services currently in the HWA) and existing collaborators like SAIL and STU.

The choice of papers included in this thesis was carefully considered and was very much informed by an earlier attempt to put together a PhD by publication. I previously tried to put a cohesive and coherent body of work together in 2018 with several papers that I put together in a decision matrix that I thought were linked by a common thread. In that case they all shared a common theme of having a cost effectiveness methodology, and that they were all concerned with public health. The reason I didn't proceed with the original PhD by publication during that time was that I was concerned that those two common threads linking the papers was too tenuous and would be open to criticism by examiners. The papers were very disparate and included conditions/diseases such as Multiple Sclerosis, Stroke, Falls Prevention, Dentistry, Wounds, as well as a couple of the early Lymphoedema papers. Therefore, I opted to wait and continue to collaborate and publish with Lymphoedema Wales to create a more robust and criticism free body of work.

When it came to choosing the papers for this current PhD by publication, I again, sought to address, potential criticism by examiners of 'watering down' both the quality and quantity of the papers by carefully selecting only papers that were directly linked to Lymphoedema and our research, and its relevant treatments. Therefore, there were three potential papers that were excluded from this thesis that I personally considered either not relevant and enough of a direct link to Lymphoedema and its related research, or a coupled paper in a different journal. These papers were Thomas M, et al (2017) paper which I though was excessive to the one I had already decided to include (Evaluation of the economic impact of a national lymphoedema service in Wales

I Humphreys, MJ Thomas - British Journal of Nursing, 2017) and therefore thought I could be open to criticism from examiners as duplication. Another two papers that I did initially considered to be part of the thesis, but eventually discarded, was Ceri J Phillips et al (2016) paper which was is mentioned in the thesis as informing the Lymphoedema research work we embarked on. I considered this paper to be too generic about wounds and the costs of wounds and not linked to Lymphoedema in enough of a meaningful way. Similarly, the other paper I omitted is CJ Phillips et al (2020) paper that again felt too linked to wounds and Venous insufficiency to be considered alongside the cost of Lymphoedema research.

One final paper that I also excluded was I Humphreys (2018) which was a short paper published in the British Journal of Community Nursing that was based upon a Hansard report of a Parliamentary debate that was raised by the MP Tonia Antoniazzi. The title of the debate was “Is there an inequity of UK lymphoedema service provision?” and even though it made for a nice short paper, it felt too much like an editorial or ‘letter to the editor’ to justify inclusion in this thesis.

Section 4:1 Publications presented in this thesis.

In 2016, Lymphoedema Network Wales (LNW) approached Swansea University (SU) and myself to help them evaluate their ‘All Wales’ lymphoedema service that had been set up some years previously. One of the reasons that LNW approached SU, was because of the universities’ record of research, publications, and experience of the economic burden of wound management (Phillips et al 2016). This work showed that wounds are a considerable economic burden on the NHS and chronic wounds relating to Lymphoedema are a major contributor to the problem. By 2016, I had already been involved in numerous health economic evaluations across both England and Wales. This had resulted in 20 journal publications, three of which were registered as first author. This initiated the collaboration between myself and LNW over the next eight years.

Section 4:2 Paper 1

Humphreys, I; Morgan, K; Thomas, M (2017) Managing chronic oedema in community settings. Wounds UK, Vol 13, 22-35 (Available on-line here: https://wounds-uk.com/journal-articles/managing-chronic-oedema-in-community-settings/)

This rapid assessment review showed that lifestyle choices, co-morbidities, demographics, deprivation, and current service provision were factors which affected Lymphoedema service providers and patients. The review highlighted the lack of research undertaken on the benefits of exercise promotion and diet for patients with chronic oedema. It also 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. One of the most important results to come out of this review was the lack of research undertaken to attempt to estimate the cost/economic burden of the treatment and management of chronic oedema in the community setting. These issues raised in this rapid assessment have a major impact on 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.

Section 4:3 Paper 2

Ioan Humphreys, Melanie J Thomas (2017) Evaluation of the economic impact of a national lymphoedema service in Wales British Journal of Nursing Vol. 26, No. 20 Clinical Focus doi.org/10.12968/bjon.2017.26.20.1093 (Available on-line here: https://www.magonlinelibrary.com/doi/abs/10.12968/bjon.2017.26.20.1093)

The aim of this evaluation was to estimate the economic impact of a national lymphoedema service on the NHS Wales budget. Work was undertaken to determine the care pathway within Lymphoedema Network Wales and develop a hypothetical 'world without' the service as a comparator.

Paper 1 highlighted the need to undertake further research and undertake an economic evaluation. Therefore, using economic evaluation methodologies, a 6-month pre and post intervention evaluation was designed to estimate the changes in resource usage between entry into the Lymphoedema service. Together with LNW we devised a resource utilisation questionnaire (RUQ) that would be administered to 50 patients currently on LNWs caseload. The first part if the RUQ asked patients for their resource utilisation 6 months prior to entering the LNW service. The second part of the RUQ asked the same questions 6 months after entering the LNW service. This enabled us to create a database of pre and post costs to compare and to generate a hypothetical 'world without' LNW service scenario. The patients were recruited from each of the seven Health Boards (HB) across Wales and were a mixture of patients from group 'types' devised by LNW. These types were:

- Group 0: 'at risk' - 11% of the annual case load of LNW at the time (n=9226)
- Group 1-2: 'uncomplicated' – 44% of the annual case load of LNW at the time (n=9226)
- Group 3: 'complicated/complex – 40% of the annual case load of LNW at the time (n=9226)
- Group 4: 'palliative care' – 5% of the annual case load of LNW at the time (n=9226)

The findings from this research estimated considerable costs associated with lymphoedema and its management in the Welsh NHS. The estimated mean cost per patient over 6 months was £4,859 (£9,718 for 12 months). When multiplied by the then caseload of LNW patients across the Welsh HBs (n=9226), the costs became considerable at an estimated annual cost of £89,658,268. This estimate is based purely on the 6 months prior to entering the service which we used as the basis of the 'world without' LNW comparator.

Analysis of the RUQ 6 months after entering the LNW service produced annual estimates of £40,908,084. In other words, a potential estimated annual per patient saving to the Welsh NHS of £2,642 (compared to the resource usage used in a hypothetical 'world without' a lymphoedema service). This paper presented various caveats and limitations to these estimates, but LNW were able to take the encouraging findings found in this initial evaluation and use them to ask the Welsh Government for further research funding to explore the economic burden of lymphoedema and start to develop the service as an exemplar of Prudent Healthcare and future Value Based Healthcare.

Lymphoedema Network Wales took the findings from the previous evaluation to leverage support and funding from the Welsh Government (WG) to develop an education programme of teaching and mentorship called the "On the Ground Education Programme" (OGEP). This was developed to enhance community nurses' management of chronic oedema prevention and management. LNW had been developing a series of innovative methods to support the management of chronic oedema within the community setting and OGEP was one of these. The innovation that was developed is a community-based education model involving the use of video prescription applications as well as an educator training programme to support community health professionals and patients in the management and care of chronic oedema.

Section 4:4 Paper 3

Ioan Humphreys, Melanie J Thomas, Karen M Morgan (2017) Pilot evaluation of the management of chronic oedema in community settings project British Journal of Community Nursing Vol. 22, No. 12 Long-term Conditions doi.org/10.12968/bjcn.2017.22.12.578 (Available on-line here: https://pubmed.ncbi.nlm.nih.gov/29189059/)

Lymphoedema Network Wales continued to work with myself on the economic analysis of OGEP, and part of OGEP was the development of the 'Wet Leg' Pathway. The main aim of the study was to evaluate the impact of OGEP on patients in one Health Board (Cardiff and Vale) in Wales. For this study LNW recruited 100 patients from the existing caseload within Cardiff and Vale's Lymphoedema service. Using lessons learned from the previous evaluation as detailed in Paper 2, as well as RUQs administered to patients, LNW also administered the EQ-5D-5L to the cohort. This addition enabled the research team to not only quantify any changes in resource utilisation over time but also to report changes in Health-Related Quality of Life (HRQoL) over time. This was an important element to any economic evaluation and an addition to the evaluation previous as described in Paper 2.

A single arm study (no comparator) based on an observational 'before-after' design was conducted using patients (n = 100) on OGEP. When reviewing the direct costs associated with the OGEP, it was identified that the largest differences between the outcomes at the baseline and the three-month follow-up review were in the reduction of the frequency of district nurse home visits (53%), resulting in a reduction in costs and a more effective service delivery. Furthermore, there was a large difference in the costs of dressings, which were £52,419 at baseline reduced to £19,667 (63%) once OGEP was utilised. There was also a statistically significant improvement noted in the patients' EQ-5D-5L utility score and the visual analogue scale which indicated an increase in the patient's perceived quality of life. As the evaluation was based on a service innovation delivered within routine clinical practice with budget and time constraints, the design of the evaluation was limited in several areas, particularly the lack of comparator, that is a matched control site or cohort of patients who did not receive the OGEP during the evaluation period and short time-horizon for the evaluation. Due to budget constraints, the OGEP team undertook data collection as part of their day-to-day project roles. However, this also allowed a first step in-depth examination of the potential impact of OGEP without manipulating or changing the service conditions, thus ensuring the evaluation remained grounded from the outset in a real-world context.

The OGEP programme of work (presented in paper 3) is now being used by District Nurses (DN)s in six of the seven Welsh HBs and is potentially treating 20,000 patients per year (based on the 2019 business case from LNW to WG). Based on the economic analysis outlines here, the research team were able to confirm to LNW that OGEP and the wet leg pathway reduced health care resource use and costs and health related quality of life scores increased where OGEP was trialled.

Moving forward, LNW were again able to take the results of the OGEP evaluation and leverage the WG for more monies to finance further independent economic evaluations conducted by myself in Swansea University.

Section 4:5 Paper 4

Melanie Thomas, Karen Morgan, Ioan Humphreys, Karl Hocking, Diane Jehu (2020) The benefits of raising awareness of lymphoedema among care home staff British Journal of Nursing Vol. 29, No. 4 Focus doi.org/10.12968/bjon.2020.29.4.190 (Available on-line here: https://pubmed.ncbi.nlm.nih.gov/32105542/)

Paper 4 is the first in-depth examination of the potential economic benefits of Lymphoedema education in a residential and nursing home setting. Working with the WG and the 1000 Lives Campaign, a pilot was developed to look at delivering the OGEP intervention within the care home setting of one Welsh health board (HB) with a population of around 290,000 people. A total of forty four out of the 47 care homes in the HB agreed to take part in the study. This meant a total of 960 care home residents taking part as a cohort.

The findings of the study indicated several benefits to raising awareness of lymphoedema in patients in a care home setting. The intervention developed by LNW was a visit that was mutually agreed with care home managers/staff to provide them with information on the lymphoedema education tool, which included e-learning, videos, a lymphoedema learning pack and a group teaching session. As well as the education tool, each home manager was asked to allow their care staff to accompany the lymphoedema specialist, who would carry out a scoping review of their residents to identify any person with limb swelling. The relatively inexpensive intervention (costed at approximately £2.65 per resident), was completely dwarfed by the findings that suggest that there are substantial costs incurred by residents with lymphoedema, and linked to cellulitis owing to hospitalisations, district nurse visits because of wounds and falls. These totalled £198 660 in 6 months.

Potential costs saved due to the avoidance of a GP home visit was estimated to be £29,213 over the same period. The evaluation also highlighted carer’s confidence in identifying lymphoedema early, therefore being able to refer to the lymphoedema service immediately and directly without having to contact the GP and have an expensive and time-consuming referral. The substantial costs potentially saved in just one HB indicated the need to conduct further research throughout the other 6 HBs in Wales. More than a quarter of residents in a care home setting in one health board area in Wales were identified as having lymphoedema and only 13% of those were known to the local lymphoedema service, portraying an unmet need. Many of the patients identified with lymphoedema had experienced falls, cellulitis and wounds. Additionally, this intervention demonstrated itself to be an inexpensive education tool on lymphoedema for care home staff raised awareness and knowledge, and this supported prompt and proactive care. This educational project has identified the prevalence of lymphoedema in care homes in one area and demonstrated potential value for money.

Section 4:6 Paper 5

Melanie Jayne Thomas, Karen Morgan, Ioan Humphreys, Rhian Newton (2021) Changing the process of prescribing to procuring lymphoedema compression garments: a service evaluation Journal of Prescribing Practice Vol. 3, No. 12 Clinical Focus doi.org/10.12968/jprp.2021.3.12.490 (Available on-line here: https://www.magonlinelibrary.com/doi/pdf/10.12968/jprp.2021.3.12.490)

For the next study, LNW and I looked to evaluate LNW's 'Project B' work. 'Project B' involved the LNW team working with Community Pharmacy Wales and Health Board Medicine Management teams to investigate a new way to procure garments instead of prescribing garments through a GP. Continuing to build on the philosophies behind Value-Based healthcare, LNW aimed to investigate whether compression bandages, stockings and garments can be procured more efficiently and cost effectively. Piloted in Cardiff and Vale University Health Board (C&VUHB) and Swansea Bay University Health Board (SBUHB), the study primarily looked at changing the way that garments lymphoedema patients are prescribed via a GP and investigate whether procuring them via the All-Wales Lymphoedema Compression Garment Formulary was a more efficient, and possibly more cost-effective way of procuring garments for patients. Also, delays in patients receiving the correct garments with which to treat and manage their lymphoedema can result in the patients' conditions worsening, HRQoL being detrimentally affected and potential substantial out of pocket expenses occurred by the patient.

The analysis was a first in-depth examination of the potential health and economic benefits of changing the process of accessing lymphoedema garments, from prescribing, to procuring. The analysis indicated substantial savings to the Welsh NHS (£71.10) per patient and also indicated the additional benefits to both the patient and the Welsh NHS through both timely and quality of care.

For the two lymphoedema services (C&VUHB & SBUHB) alone, potential costs avoided were estimated to be £383,371. There was also a big disparity in the time lag between ordering and fitting the garments. SBUHB's Primary Care Prescribing and Medicines Management reported costs of £343,916 for 2019/2020, but with a forecast of £214,452 for 2020/2021. The results from the 'Project B' analysis, and the real-time savings seen, have enabled this innovation to be rolled out across six of the seven Welsh HBs.

Section 4:7 Paper 6

Melanie Jane Thomas, Ioan Humphreys, Rhian Wyn Noble-Jones (2022) A spotlight on lymphoedema Did Not Attends: Demographics and workforce costs International Wound Journal doi.org/10.1111/iwj.13999 (Available on-line here: https://pubmed.ncbi.nlm.nih.gov/36411996/)

Next, LNW and I investigated the cost implications and impact of Did Not Attends (DNA) on lymphoedema services within the Welsh NHS. When a patient misses their appointment, it is referred to as a 'Did Not Attend' (DNA) or a 'Was Not Brought' (WNB) for children (NHS England 2023). During 2021/2022, nearly 7.5 million outpatient appointments were missed by patients, often for reasons outside of their control, and often linked to health inequalities (NHS England 2023).

In terms of GP Visits, there are around 307 million sessions scheduled with GPs, nurses, therapists and other practice staff every year and 5% – one in twenty – are missed without enough notice to invite other patients (NHS England 2023). That is estimated at around 15.4 million missed slots. Of these, around 7.2million are with busy family doctors, which adds up to more than 1.2 million GP hours wasted each year – the equivalent of over 600 GPs working full time for a year. Each appointment costs an average of £30, putting the total cost to the NHS at more than £216million pounds on top of the disruption for staff and fellow patients that would pay for. Reducing missed appointments helps to ensure that clinical time is used effectively and means patients on the waiting list can be seen more quickly (NHS England 2023). Additionally, the ability for the patient to let the service know in time that they are Unable to Attend (UTA), is a far less burdensome economic problem for the NHS. Unable to Attends allow healthcare professionals the time to reschedule an appointment to a different patient on the waiting list.

Using HB data between 2012 and 2022, the aims of the study were to estimate the financial costs and workforce implications involved with DNAs across all the lymphoedema services at a national level. The study also looked to gain more insight into the reasons for patients not attending an appointment without previous notice by analysing the notes from discharged patients (an anonymised subset of 870 patients) between 2019/2020.

The analysis indicated that approximately £1.1 million of 'lost' staffing hours were accounted for by patients missing and patients not attending an appointment without previous notice to Welsh lymphoedema clinics across 10 years. Most of the expenditure lost was from follow up (FU) patients (£716,000) despite being allocated half the time needed or offered to that of a new patient (NP). The NPs accounted for nearly £446,000 across 10 years.

The Covid-19 pandemic occurred during the period of this study, but interestingly, DNA rates actually fell (i.e. less patients failed to attend) from 6% to 2%. This was partly put down to the implementation of virtual consultations that were introduced during the pandemic. When the pandemic eased in 2021/2022, the DNA levels increased back to 5%, despite virtual consultations still being in place and offered.

Whilst our 2022 study showed that DNAs were incredibly costly and wasteful on resources and staff time, the reasons for DNAs still remain varied and nuanced, with health conditions and access to transport being two main causes. The introduction of more modernised processes such as digital and Patient initiated follow-up (PIFU) is potentially key to personalising outpatient care. PIFU enables patients to have more control over when they receive care and can lead to the reduction of unnecessary follow-up appointments and make best use of clinical time (NHS England 2023). This change in the dynamic of patient and healthcare needs may also lead to increases in patient HRQoL and therefore it may be possible that DNA rates, and the costs associated with them, can be minimised significantly.

Section 4:8 Paper 7

Ioan Humphreys, Ashley Akbari, Rowena Griffiths, Dave Graham-Woollard, Karen Morgan, Rhian Noble-Jones, Marie Gabe-Walters, Melanie Thomas Evaluating the cost of managing patients with cellulitis in Wales, UK: A 20-year population-scale study International Wound Journal doi.org/10.1111/iwj.14088 (Available on-line here: https://pubmed.ncbi.nlm.nih.gov/36648008/)

The next piece of research that LNW and I embarked on aimed to estimate the financial cost of cellulitis. Cellulitis is an acute bacterial infection of the dermis and subcutaneous tissue. The infected area, most commonly the lower limb, is characterized by pain, warmth, swelling, and erythema. Blisters and bullae may form. Fever, malaise, nausea, and rigors may accompany or precede the skin changes (NICE 2023). Cellulitis develops when microorganisms (most commonly *Streptococcus pyogenes* and *Staphylococcus aureus*) gain entry to the dermal and subcutaneous tissues via disruptions in the cutaneous barrier (NICE 2023). Risk factors include skin trauma, ulceration, and obesity. Complications of cellulitis include necrotizing fasciitis, sepsis, persistent leg ulceration, and recurrent cellulitis.

This study investigated the financial burden of cellulitis on the Welsh/UK NHS; to establish the numbers of cases attending primary care; and better understand the incidence of cellulitis in Wales and its implicated costs for UK NHS. The analysis was undertaken from data obtained through the SAIL Databank (SAIL Databank 2023). The Secured Anonymised Information Linkage (SAIL) databank is funded by Health and Care Research Wales and is based within Swansea University's School of Medicine. SAIL's work is supported by active partnerships with NHS and Welsh Government information agencies and brings together population-scale, individual-level anonymised linked data from a wide range of sources, including 80% of primary care GP practices within Wales (population coverage estimated 3.2 million). The study covered a 20-year period from 1999 to 2019 inclusive. All patients were linked via the relevant READ and ICD-10 codes, and were tracked through primary care settings, recording the number of GP events and number of in-patient stays. This was the first attempt to estimate the financial burden of cellulitis using routine data sources on a national scale from a big dataset. Patients with a cellulitis diagnosis in the Welsh Longitudinal General Practice (WLGP) dataset were included in the primary care cohort. Patients with a cellulitis diagnosis in the Patient Episode Database for Wales (PEDW) dataset were included in the secondary care cohort and the Welsh Demographic Service Dataset (WDSD) was utilised to gather basic demographics across both cohorts. The Welsh Index of Multiple Deprivation (WIMD) was utilised to measure deprivation and unit costs were applied from the Personal Social Services Research Unit (PSSRU) and NHS Reference Costs to attempt to estimate the financial burden of cellulitis in the Welsh and UK NHS across 20 years.

Our results indicated that the costs of GP and In-Patient events differed for female patients, accounting for £240,936,917, whereas for male patients this accounted for £170,887,185. And when the GP and In-Patient events were compared against age group, the biggest share of the expenditure was for the 76–90 age group at £132,217,75 (See Table 4 in paper 7 on page 6). Expenditure by WIMD quintiles indicated that the total resource usage was highest in WIMD Quintile area 1 (Most Deprived) with £97,939,134 (or a mean per-person cost of £1,614) This is in comparison to WIMD Quintile area 5 (Least Deprived) with £66,056,449 (or a mean per-person cost of £1,259) (See Table 5 in paper 7 on page 7). The study showed that cellulitis is a common and expensive problem for the NHS. This large data analysis revealed that estimated annual direct costs for NHS Wales are substantial (over £28 million) Extrapolated for the UK, this amounts to over £571 million annually. The study also indicated that in-Patient events and length of stay costs are the main cost drivers, with annual Welsh NHS estimates of

£19,664,126. This extensive study using big data from the SAIL Databank led the team to conclude that initiatives to identify early signs/risks of cellulitis and improved evidence-based treatment pathways to reduce incidence and severity by even small percentages would result in major financial savings and reduce the burden on patients.

Section 4:9 Paper 8

M Thomas, C Pike, I Humphreys, T Bragg and A Ghattaura, The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months, Journal of Plastic, Reconstructive &Aesthetic Surgery, (2023) doi:https://doi.org/10.1016/j.bjps.2023.06.068 (Available on-line here: https://pubmed.ncbi.nlm.nih.gov/37473641/)

The final piece of research that is included in this PhD is the study ‘The impact and outcomes following lymphaticovenous anastomosis (LVA) for 150 cases of lymphoedema followed up over 24-months’ (Paper 8: M.Thomas et al (2023). The study has generated two papers, and this is the first paper published.

The purpose of LVA surgery to make a connection between a lymphatic channel and a vein. This allows the lymph fluid to drain from the arm into a vein in the armpit, avoiding the channels that are no longer working. (LVA Overview 2023) LVA is carried out through a cut (incision) in the armpit. A plastic surgeon will identify the draining lymphatic channels in the armpit by using a dye that is injected. These lymphatics will then be bypassed. The wound will be closed with dissolvable stitches (sutures). This surgery is usually done as a day case, which means the patient does not need to stay in hospital overnight. (LVA Overview 2023)

This study had three main objectives; 1. To measure the change in patient-reported outcomes before and after LVA; 2. To determine limb volume change; and 3. To describe the health-related quality of life in patients before and after LVA surgery. The EQ-5D-5L results, indicated statistically significant improvements in general health (p-value = 0.04). The LYMPROM PROM analyses showed that statistically significant improvements in quality of life were observed in the reduction of pain and heaviness of the limb (both p-value <0.001). Furthermore, borderline statistically significant improvements (p-value - 0.051) in the patient's ability to work were observed. However, over the 24 months, there was an observed statistically significantly increase in reported financial burden to the patients themselves due to their lymphoedema (p-value <0.001).

In terms of costs relating to the LVA intervention, there was a statistically significant reduction in the number of days and total hours per day that the patients wore their compression garments pre and post the LVA intervention (p-value = <0.001). Further analysis indicated that those patients with secondary lymphoedema reduced garment wear over time, and that was also seen as being a statistically significant change (p-value = <0.001). This decrease in garment usage had an economic impact on the outcomes reported by the patients, but this is reported in paper 2 of this study (not yet published and not included in this PhD).

Additionally, the costs relating to cellulitis episodes reduced dramatically from £169.53 to £4.21per patient. Furthermore, the number of patients hospitalised due to cellulitis also showed a statistically significant decrease (p-value = <0.001) with mean patient costs reducing from £1,421.68 to £107.34. Finally, the number of days off work that patients had to take due to cellulitis also reduced from 5.81 to 0.16 (p-value = <0.001).

This study reports significant improvements in the health-related quality of life in patients after undergoing the LVA treatment. Patients were able to decrease their usage of compression garments and reported a statistically significant reduction in costly cellulitis episodes. There are statistically significant differences shown in what would be considered important and meaningful cost drivers in the management of chronic oedema such as reducing episodes of cellulitis and hospital stays due to cellulitis. These changes seen over time will have pronounced long term cost savings to the NHS, and related HRQoL effects for this cohort of patients.

Chapter 5

Discussion

The next chapter in this thesis will bring together all the previous analysis and synthesis of the eight papers presented here and will discuss such impacts as the contributions they have had on lymphoedema services in Wales, impact on HRQoL, government policy, government funding, etc. It will also include a reflection of current Health Economic practice and how the papers presented here have contributed to that body of knowledge.

Section 5:1 Contributing to evidence-based policy change

In terms of producing research evidence that has been employed to leverage economic support from policy makers within the WG, the impact of my work has been significant. Before OGEP, each health board in Wales had their own system of treating Lymphoedema, which was not Value-Based, evidence-based or cost effective. My research, showing the economic benefit of OGEP and the Wet Leg pathway across the Cardiff and Vale University Health Board (CVUHB) was used to underpin a significance policy and economic decision by the WG. The LNW successfully lobbied the Health and Social Care department in Wales to fund this pathway for rollout throughout Wales. Currently, £1.4 million has been permanently allocated by the WG to rollout the OGEP programme across all seven HBs for the benefit of all community nurses and District Nurses (DN)s.

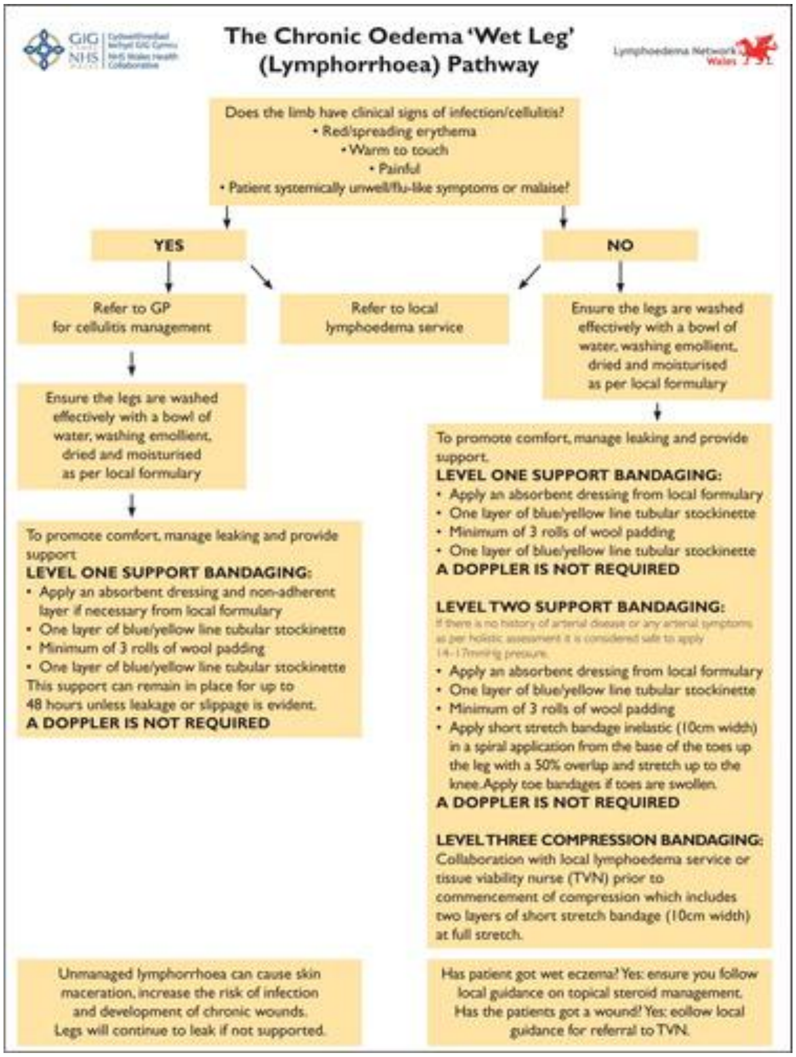
The ultimate goal of OGEP is to enhance community nurses' management of chronic oedema prevention and management. OGEP promotes more clinically effective 'Wet Leg' treatment among DNs. This education includes more appropriate use of compression bandaging and garment choice. This change in practice evidenced a significant reduction in the need for as many DN visits pre OGEP training. Chronic oedema can have a profound and long-term impact on patients and can impact on a range of health outcomes including health-related quality of life (HRQOL). Developing innovate solutions to the management of patients with chronic oedema has the potential to deliver prudent, cost-effective and high-quality care within NHS Wales. This Wet Leg pathway has been endorsed and supported by the Chief Nursing Officer of Wales, who noted that “The report makes sobering reading on the debilitating and distressing effects of this condition, and I believe it is therefore essential that community nurses take a leading role in delivering effective person-centred care, which minimises the complications of the condition and eliminates hospital admissions as far as possible.”

Section 5:2 The ‘Wet Leg Pathway’

The ‘Chronic Oedema Wet Leg pathway’ AKA The ‘Wet Leg’ Pathway was developed as an output from OGEP (Paper 2). This evidence-based pathway provides nurses and therapists with clear guidelines to support prompt, efficient and effective management of all patients with chronic oedema and wet legs (Morgan K & Thomas M 2018). The ‘Wet Leg’ pathway has proved incredibly useful to lymphoedema clinicians on a national scale and there is now international interest of the use of the pathway outside of UK. This is growing and currently includes Germany, USA, Belgium, Canada and Australia. The ‘Wet Leg’ pathway developed in Swansea has started to inform national practice where it is currently adopted by St George's Hospital, London. The Dermatology on-call doctors in South London have recommended the pathway and they have subsequently recommended it to the hospital ward staff asking for help in managing wet leaky legs. The pathway has also been requested by 10 Clinical commissioning groups community nurse teams in England. Lymphoedema Wales have also created a training video for all staff on the Pocket Medic platform.

Our research on the effectiveness of the ‘Wet Leg’ pathway has led to its implementation across primary and secondary care throughout Wales and is becoming a core treatment recommended in the successful treatment of chronic Lymphoedema in the community.

Figure 2. The ‘Wet Leg Pathway’



The Chronic Oedema 'Wet Leg' (Lymphorrhoea) Pathway

Does the limb have clinical signs of infection/cellulitis?
- Red/spreading erythema
- Warm to touch
- Painful
- Patient systemically unwell/flu-like symptoms or malaise?

YES

Refer to GP for cellulitis management

Refer to local lymphoedema service

Ensure the legs are washed effectively with a bowl of water, washing emollient, dried and moisturised as per local formulary

To promote comfort, manage leaking and provide support

LEVEL ONE SUPPORT BANDAGING:
- Apply an absorbent dressing and non-adherent layer if necessary from local formulary
- One layer of blue/yellow line tubular stockinette
- Minimum of 3 rolls of wool padding
- One layer of blue/yellow line tubular stockinette

This support can remain in place for up to 48 hours unless leakage or slippage is evident.

A DOPPLER IS NOT REQUIRED

NO

Ensure the legs are washed effectively with a bowl of water, washing emollient, dried and moisturised as per local formulary

To promote comfort, manage leaking and provide support.

LEVEL ONE SUPPORT BANDAGING:
- Apply an absorbent dressing from local formulary
- One layer of blue/yellow line tubular stockinette
- Minimum of 3 rolls of wool padding
- One layer of blue/yellow line tubular stockinette

A DOPPLER IS NOT REQUIRED

LEVEL TWO SUPPORT BANDAGING:

If there is no history of arterial disease or any arterial symptoms as per holistic assessment it is considered safe to apply 14-17mmHg pressure.
- Apply an absorbent dressing from local formulary
- One layer of blue/yellow line tubular stockinette
- Minimum of 3 rolls of wool padding
- Apply short stretch bandage inelastic (10cm width) in a spiral application from the base of the toes up the leg with a 50% overlap and stretch up to the knee. Apply toe bandages if toes are swollen.

A DOPPLER IS NOT REQUIRED

LEVEL THREE COMPRESSION BANDAGING:

Collaboration with local lymphoedema service or tissue viability nurse (TVN) prior to commencement of compression which includes two layers of short stretch bandage (10cm width) at full stretch.

Unmanaged lymphorrhoea can cause skin maceration, increase the risk of infection and development of chronic wounds. Legs will continue to leak if not supported.

Has patient got wet eczema? Yes: ensure you follow local guidance on topical steroid management. Has the patients got a wound? Yes: eollow local guidance for referral to TVN.

Section 5:3 Teaching and Higher Education

The 'Wet Leg' pathway is now being taught as an Agored Cymru accredited unit to Health Care Support Worker (HCSW) and Qualified Nurses and allied health care professionals. These education units are attended by approximately 283 people from all over the UK and are intended to be part of OGEP which is a community-based education model involving the use of video prescription applications as well as an educator training programme to support community health professionals and patients in the management and care of chronic oedema. Whilst the OGEP intervention requires additional investment in terms of resources to deliver, it could potentially result in more efficient use of health care resources including correct prescribing of dressing and garments from the All-Wales Lymphoedema Compression

Garment Formulary (All-Wales Lymphoedema Compression Garment Formulary 2017), alongside improving outcomes and experiences for people living with chronic oedema. In addition, the 'Wet Leg' pathway module on Lymphoedema for primary care health professionals has been introduced into the Nursing curriculum for Swansea University nursing student's intake since 2016. The Wet leg pathway is delivered in both pre graduate and post graduate courses such as Specialist Practitioner Qualification (SPQ) (district nursing). Based on our analysis of the benefits of OGEP, which helped LNW source funding from WG to roll it out across Wales, with the introduction of OGEP Lymphoedema Clinical Educators with a National Lymphoedema Community Clinical Educator, who will lead on the programme of work.

Section 5:4 Cost and efficiency savings

From the publications presented in this thesis, major cost savings and efficiency in utilising NHS resources was evidenced throughout. It was identified that the largest differences between the outcomes at the baseline and the three-month follow-up review were in the reduction of the frequency of district nurse home visits (53%), resulting in a reduction in costs. Furthermore, there was a large difference in the costs of dressings, which were £52,419 at baseline reduced to £19,667 (63%) once OGEP was utilised. Specifically, based on the potential cost savings seen in Cardiff &Vale HB, when these are extrapolated to all the Wales HBs, costs potentially saved by implementing the Wet Leg pathway/OGEP are estimated in the region of £24,460,000. (Avg. £1,223 per patient savings).

The analysis showed the difference in cost when using WP10 compared to Oracle was overwhelmingly more costly than using Oracle (mean per patient cost of £106 (SD £74) for WP10 and £77 (SD £54) for Oracle (p-value = <0.001)). SBUHB's Primary Care Prescribing and Medicines Management reported costs of £343,916 for 2019/2020, but with a forecast of £214,452 for 2020/2021. The results from the 'Project B' analysis, and the real-time savings seen, have enabled this innovation to be rolled out across six of the seven Welsh HBs.

Section 5:5 Health Related Quality of Life (HRQoL)

In papers 2 and 3, we showed that patients have a better-perceived quality of life when they are treated via OGEP, and our work prompted the WG to fund the rollout of OGEP. As discussed in Paper 2, the estimated Wales wide population caseload of lymphoedema at that time was 9226. Quality-adjusted life year (QALY) is a generic measure of disease burden, including both the quality and the quantity of life lived. It is used in economic evaluation to assess the value of medical interventions. One QALY equates to one year in perfect health. Therefore, extrapolated across the Wales wide caseload of 9226, then OGEP has the potential to generate 1254.7 Quality Adjusted Life Years (QALY) and increase patient outcomes.

Section 5:6 Welsh Government funding

LNW have recently secured annual rolling funding of approximately £120,000 to deliver their LVA treatment for the management of Lymphoedema in SBUHB. This was achieved from the results of the report delivered by myself (paper 8). The results from the 'Project B' analysis from Paper 5, and the real-time savings evidenced, have enabled this innovation to be rolled out across six of the seven Welsh HBs.

Section 5:7 LNW Research Staff (LYMPROM & CELLUPROM validation)

With funding secured from Welsh Government, LNW was able to secure from WG, LNW were able to increase their workforce and employ their own research staff. One of these Band 7 staff members took on the task of further developing the LYMPROM Patient Reported Outcome Measure (PROM) that LNW had begun to develop several years ago.

The LYMPROM is a 13-item patient reported outcome measure (PROM) for adults with lymphoedema. LYMPROM also has a free text box for patients to provide additional information. LYMPROM is made up of 3 dimensions; Physical Health; Social Health and Emotional Health. The 13 items are spread across the

3 dimensions and each item is scored using an 11-point scale, where zero indicates no impact and 10 indicates an extreme impact.

Lymphoedema Network Wales, myself and the Swansea University Trials Unit are currently in the process of validating the LYMPROM instrument and are finalising their statistical analysis for a forthcoming scientific paper.

After the validation of the LYMPROM is complete, myself and the research team will move on to validating the CELLUPROM (Figure 4). Under the auspices of LNW, the National Lymphoedema Cellulitis Improvement Programme (NLCIP) was established to offer all people given a diagnosis of cellulitis in Wales an evidenced based intervention to reduce their risk of recurrence and help to minimise pressure on unscheduled care. The main aim of the CELLUPROM instruments are to; Increase patient awareness in identifying and managing cellulitis; Improve patient understanding of risk factors for cellulitis; Reduce incidence of recurring cellulitis episodes through assessment, education, and treatment of identified risk factors; Increase healthcare professional awareness in identifying and managing cellulitis; and improve their understanding of risk factors for cellulitis. The CELLUPROM is very similar to the LYMPROM and is also based around the patients answering questions relating to their cellulitis, and the responses to those questions again feeding into 3 dimensions; Physical Health; Social Health and Emotional Health.

The current design of the CELLUPROM may change once the results of the LYMPROM validation process is completed. This is in part due to an issue that the statistician from the Swansea Trials Unit identified in the early stages of validation. This issue involved the variables that made up the three domains within the LYMPROM as mentioned above and in Paper 8. The three dimensions (Physical Health, Social Health and Emotional Health) include the following variables:

Physical Health: Pain, Heaviness, Home Life, Personal Care, Work, Shopping for clothes/Shoes and Walking

Social Health: Finances, Hobbies and Holidays

Emotional Health: Anxiety, Body Image, and Intimacy/Desirability

As shown in Figure 3, a LYMPROM respondent is able to select N/A (not applicable) for the ‘Work, ‘Finances’, ‘Intimacy/desirability’ and ‘Holidays’ variables. During the validation process, the statistician discovered that if a respondent selected N/A for both the ‘Finances’ and ‘Holidays’ variables, then the Social Health dimension would be represented by just one variable, that being ‘Hobbies’. Therefore, from advice given from the statistician, the LW team re-assessed what variables were represented in each domain and created a newer version of the LYMPROM. The domains are now as follows:

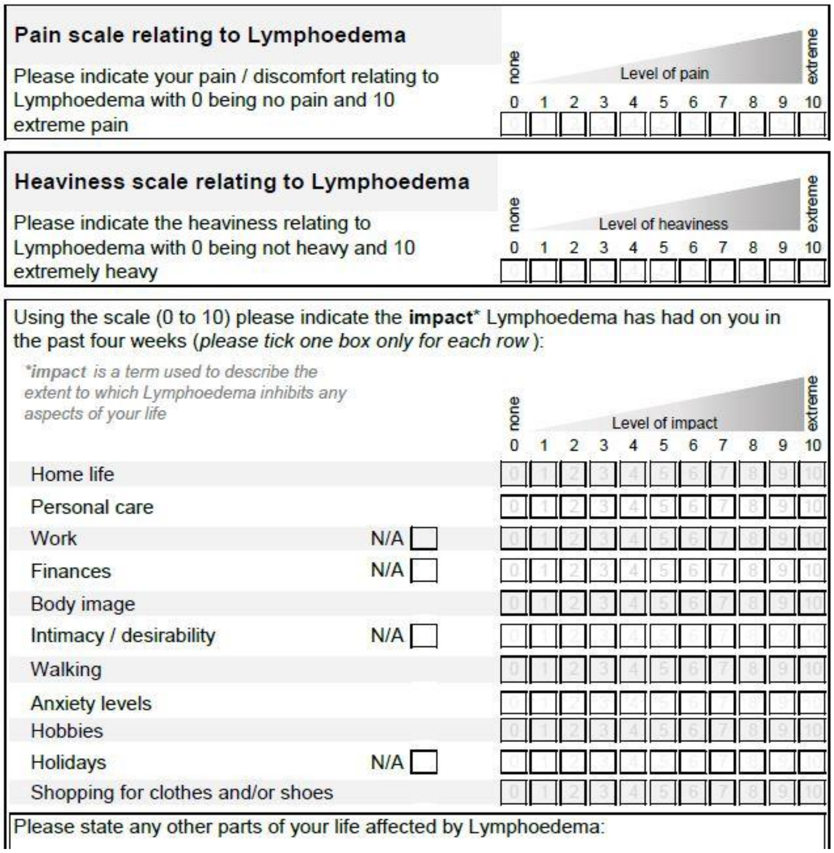
Physical Health: Pain, Heaviness, and Walking

Social Health: Finances, Hobbies, Holidays, Home Life, Personal Care, Work and Shopping for clothes/Shoes

Emotional Health: Anxiety, Body Image, and Intimacy/Desirability

This updated version of the LYMPROM will now be used going forward, and the validation process will feature these new variables within the domains.

<u>Figure 3. LYMPROM</u>



Pain scale relating to Lymphoedema

Please indicate your pain / discomfort relating to Lymphoedema with 0 being no pain and 10 extreme pain

none				Level of pain						extreme
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Heaviness scale relating to Lymphoedema

Please indicate the heaviness relating to Lymphoedema with 0 being not heavy and 10 extremely heavy

none				Level of heaviness						extreme
0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

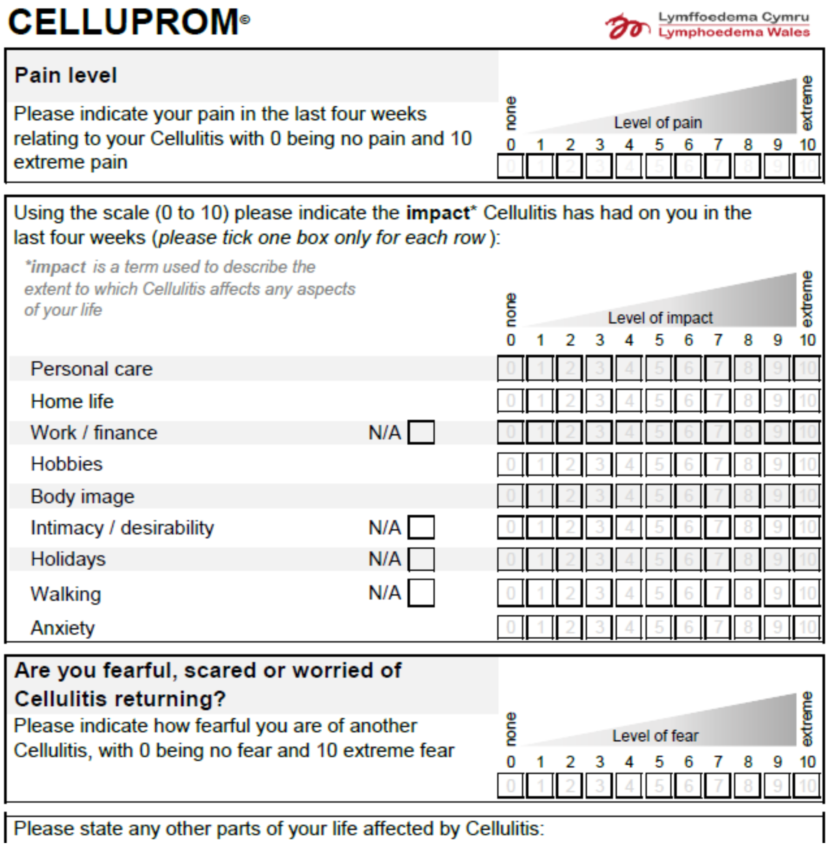
Using the scale (0 to 10) please indicate the **impact*** Lymphoedema has had on you in the past four weeks (*please tick one box only for each row*):

**impact is a term used to describe the extent to which Lymphoedema inhibits any aspects of your life*

		none				Level of impact						extreme
		0	1	2	3	4	5	6	7	8	9	10
Home life		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Personal care		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Work	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Finances	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Body image		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Intimacy / desirability	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Walking		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Anxiety levels		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Hobbies		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Holidays	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Shopping for clothes and/or shoes		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Please state any other parts of your life affected by Lymphoedema:

Figure 4. CELLUPROM



CELLUPROM®

Lymffoedema Cymru
Lymphoedema Wales

Pain level

Please indicate your pain in the last four weeks relating to your Cellulitis with 0 being no pain and 10 extreme pain

none — Level of pain — extreme

0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Using the scale (0 to 10) please indicate the **impact*** Cellulitis has had on you in the last four weeks (*please tick one box only for each row*):

**impact* is a term used to describe the extent to which Cellulitis affects any aspects of your life*

none — Level of impact — extreme

		0	1	2	3	4	5	6	7	8	9	10
Personal care		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Home life		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Work / finance	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Hobbies		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Body image		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Intimacy / desirability	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Holidays	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Walking	N/A ☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
Anxiety		☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Are you fearful, scared or worried of Cellulitis returning?

Please indicate how fearful you are of another Cellulitis, with 0 being no fear and 10 extreme fear

none — Level of fear — extreme

0	1	2	3	4	5	6	7	8	9	10
☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

Please state any other parts of your life affected by Cellulitis:

Additionally, LW have also employed 3 more researchers within the service to enable delivery and dissemination of further research projects and studies generated by the research undertaken by LW.

Section 5:8 Increase in LNW staff.

As a direct result of the body of evidence-based peer reviewed and published research that LNW and IH have built up since 2017, to date twenty-two new posts, including healthcare professionals and administrative posts have been employed by LNW. These healthcare professional posts ranged from through to allied healthcare and national lymphoedema specialists. These posts range from Band 4, through to Band 8a, and the majority of the posts were created to roll out of OGEP across all seven HBs in Wales. Nineteen posts were specifically created to deliver the OGEP programme throughout 6 HBs in Wales, with a pilot currently under way within the 7th HB. These posts work within the community workforce increasing confidence and competence in compression management and providing prompt and clinically optimum compression. The posts also relate to the roll out of the NLCIP, again a national programme and is continuing to expand and progress. Further posts include research posts, clinical psychologist, and data analyst positions.

Section 5:9 Academic partnership

In 2019, it was agreed that LNW, (now called Lymphoedema Wales (LW)) would rent accommodation from the University to house their expanding team and to be closer to IH and the Swansea University Trials Unit. Lymphoedema Wales took over a large unused teaching suite which became office space for several clinicians and admin staff. In addition, LW also rented two treatment rooms within the university's Health and Wellbeing Academy (HWA) which they used to treat both cellulitis and lymphoedema patients.

The proximity and visibility of LW on the Swansea University (SU) grounds has led to further research opportunities with both the Cardiology service within the HWA and also with the School of Psychology. The latter link has led to LW employing their first Psychologist Practitioner to start research into the psychological issues of both lymphoedema and cellulitis patients.

Another benefit of LW being housed in the SU is its proximity to the Nursing Department within the School of Health and Social Care (SHSC). Once LW located to the University, they immediately arranged with nursing programme leads to deliver sessions on compression bandaging, skin care, the Wet Leg pathway and wounds. This learning package was delivered not only to Swansea University year 2 student nurses but were also delivered in both Aberystwyth University and University of South Wales. Additionally, LW delivered webinars in both Cardiff and Bangor universities and also teach on the Swansea University Graduate Entry Medicine (GEM) Programme. LW also offer 'spoke and hub' placements for Nursing student, GEM, and Paramedic training too. Finally, LW also arrange for Tissue Viability Nurses (TVN) to come and teach sessions with the nursing students. This access to the LW teaching team means that the nursing students are being taught good practice right at the beginning of their careers, and this will benefit future patients and hopefully increase the quality of their care.

Section 5:10 Wound care

The innovative lymphoedema and cellulitis interventions and training programmes that LW have developed, and deliver are inextricably linked to the reduction of limb ulceration and hard to heal wounds in the Welsh population.

Complications resulting from both lymphoedema, and cellulitis can lead to hard-to-heal wounds that are chronic and costly to the NHS (Phillips et al 2016). If lymphoedema is left untreated, the build-up of fluid in the affected limb/s can cause a break in the skin (NHS 2023). This ulceration often allows colonisation of bacteria causing the wound to become infected and difficult to treat. The services and interventions that LW offer and have rolled out across Wales over the last seven to eight years, are helping to reduce the burden of hard to heal wounds on the Welsh NHS. As shown in Phillips et al (2016) the management of patients with chronic wounds and hard-to-heal wounds amounts to 5.5% of the total NHS expenditure in Wales. In a related wound burden study, Phillips et al (2020) estimated that the direct Welsh NHS cost of managing patients with Venus Leg Ulcers was £7,706 per patient per annum. When this is extrapolated to a UK population, the annual cost is estimated at over £2 billion.

Section 5:11 Common limitations across the papers

Across the 8 papers, similar limitations of the studies come up every time:

1. More sensitivity analyses to eliminate uncertainty around any estimates.
2. Length of the observational study
3. Narrow perspective taken

4. Limitations linked to the SAIL Databank

For Paper 1, there are no specific limitations related to study design as it’s a literature review. However, for Paper 2, we identified that more sensitivity analysis was needed around the cost savings estimates to try to eliminate uncertainty in the estimates provided. It was also noted that no HRQoL measures were utilised in this first evaluation, so therefore no QALY calculation could be conducted. It was also noted that a longer-term observational study could have been conducted. And finally, the HE perspective was narrow with using direct NHS costs, therefore negating the option to address societal costs.

With Paper 3 we addressed some of the limitations noted from Paper 2 and featured HRQoL analysis by using the EQ-5D-5L measure in the study. Again, the length of the observational study was a limitation, as was the relatively narrow perspective taken. The sensitivity and validity of using the EQ-5D-5L during this study became a limitation and a general talking point amongst the leading LW staff. Therefore, one of the most impactful and long-term positive effects of this study was the desire by LW to attempt to develop and validate their own PROM to use in future evaluations, alongside the EQ-5D-5L. This is when the development of the LYMPROM measure was instigated.

The limitations for Paper 4 were also aimed at the relatively narrow perspective of the direct NHS costs adopted, as well as the short observational study time adopted. The researchers again opined that a longer-term approach would garner more generalisable data. Similarly, with Paper 5, the authors offered that a wider, more societal perspective would also be useful for this piece of work going forward. Additionally, more qualitative data would be useful to garner what was more beneficial from a patients’ perspective. Interestingly, since the publication of Paper 5, two LW staff have looked at the qualitative results of this new way of procuring garments via their 2024/2025 Clinical Leadership projects.

The limitations laid out in Paper 6 again address the relatively narrow perspective of direct NHS costs. The paper also addresses some uncertainty raised by unit costs provided by the PSSRU. However, this uncertainty is addressed by sensitivity analysis within the analysis.

Limitations regarding Paper 7 are intrinsically linked to the SAIL Databank and are listed within the paper. However, the authors acknowledge that a wider, more societal attempt at estimating costs may have benefited the study.

Finally, Paper 8's limitations only address the suggestions that the LVA analysis may benefit from a more structured Randomised Controlled Trial (RCT) for future evaluations.

There are common limitations evident throughout the eight papers presented in this thesis. They include a need for a longer observational study time to collect more data. It also includes the lack of a QALY calculation in any of the studies. This is partly due to only one of the studies (Paper 8) exceeding the 12 months study period to enable a QALY calculation to be performed. However, no survival analysis was performed for this study to enable a formal QALY calculation to be conducted. Additionally, more rigorous sensitivity analysis could have been performed to eliminate uncertainty for several of the study results. And finally, a wider, more societal perspective would also have benefited several of these studies.

In terms of lessons learnt from the limitations acknowledged above, I would say that the need for survival analysis (to enable a formal QALY calculation) and the aim for a wider, more societal perspective, are the two key limitations that need to be addressed and eliminated as best they can in future health economic analyses relating to Lymphoedema research.

Additionally, one of the key emissions from all the papers, is something that is a seemingly recent concept, but one that has been prevalent for many years now. 'Caregiver burden' was originally defined in 1986 by (Zarit, Todd, & Zarit, 1986). They defined this issue as the extent to which caregivers perceive their emotional or physical health, social life, or financial status to be affected by their caring for an impaired relative (Zarit, Todd, & Zarit, 1986). This issue continues to today and is very much a 'hidden' economic burden for all conditions, diseases, etc prevalent in the UK today. In 2023, researchers from the University of Shefield estimated that the value of unpaid care in England and Wales was around £162 billion, which exceeded the then entire NHS budget in England for health service spending, which by comparison was £156 billion for 2020-21. Following analysis of the latest 2021 Census data, this research from the University of Sheffield and carers' charity, Carers UK, found that the value of unpaid care has increased by nearly a third since 2011. (https://www.sheffield.ac.uk/news/cost-unpaid-care-england-and-wales-now-exceeds-nhs-budget)

Section 5:12 A reflection on current Health Economic Research

ISPOR is the leading professional society for health economics and outcomes research (HEOR) globally. The Society's mission is to advance HEOR excellence to improve decision making for health globally. https://www.ispor.org/heor-explained As healthcare systems around the world struggle under the weight of rising costs and increasingly complex choices for decision makers, it's more important than ever to fully understand the value of treatments. Health Economics and Outcomes Research (HEOR) factors in effectiveness, cost, and patient quality of life to give decision makers a more accurate view so they can make choices that are good for patients and our healthcare systems.

As of 2024, ISPOR list the current top 10 HEOR trends as being Real-World Evidence; Drug Pricing; Artificial Intelligence; Fostering Innovation; Health Equity; Accelerated Drug Approvals; Value Measurement; Patient Centricity; Precision Medicine and Public Health. (https://www.ispor.org/heor-resources/top-10-heor-trends)

In terms of my research and the impact that the papers presented here (and in development as of writing), I feel that my research is linked to at least seven of the top 10 HEOR trends (Real-World

Evidence; Fostering Innovation; Health Equity; Value Measurement; Patient Centricity; and Public Health) and indeed have evidenced measurable impact in areas such as Value Measurement (Paper 3), Health Equity (Papers 3, 4 & 5) and Patient Centricity (Papers 2, 3, 4). Interestingly Caregiver Burden is not listed as one of the main HEOR priorities and is only given a small mention in the area of Patient Centricity. Personally, I think this under researched area should be become a bigger priority to public health (economic) research going forward.

As previously mentioned, the relatively narrow perspective taken throughout the course of these papers is a limitation when reflected upon and compared against the world stage of health economics and HEOR. However, studies that adopt a more societal perspective can be more research time intensive, and therefore expensive to the funder. Therefore, for funding, manpower and general practicality reasons alone, it was never possible to take a wider perspective than we did at the time.

Going forward, a wide perspective would be invaluable to capture all the hidden costs related to loss of income, employment and caregiver burden that is related to conditions such as lymphoedema. However, in terms of research practicalities and the design of questionnaires that are needed to collect this type of personal information, patient burden must also be considered. And it's this tension that I have always taken into account when designing questionnaires with extensive detail similar to a Client Service Receipt Inventory (CSRI) (https://www.pssru.ac.uk/csri/client-service-receipt-inventory/Patient). Throughout my research journey with LW, I have learnt that there is value in collecting as much data as possible, but there's also more value in being both pragmatic and realistic in what data is possible to collect, and what the researcher intends on doing with that data. That pragmatism appears to have been relatively successful with both journal publishers and policy makers throughout this body of work across all eight papers.

Finally, In the field of wound research, Julian Guest is undoubtedly the lead Health Economist in investigating the cost burden of wounds in the UK with numerous publications relating to wound treatment and overall burden (Guest et al (2022, 2023, 2023, 2020)) However, in terms of national or international leading authorities in Lymphoedema cost burden analysis, the field of research is very patchy and is not led by once notable researcher, or indeed, a Health Economist.

This really is where the work that I have undertaken with LW as a Health Economist is important as it becomes both a coherent and relevant body of work that can be referenced and built upon by other researchers and Health Economists. As can be seen in the 'Commissioning Guidance for Lymphoedema Services for Adults in the United Kingdom The National Lymphoedema Partnership March 2019', Paper 2 is cited as a reference, with Paper 3 referred to as a publication pending. My work is building upon Moffatt CJ et al's 2003 research and bringing it up to date with data collected from the LW Network that wasn't in place when Moffatt's initial study took place.

Section 5:13 A reflection on my contribution to the papers

Up until I started collaborating with LW In 2014, my role as a researcher and Health Economist was very much about being 'parachuted' into studies to look at the cost burden of the particular disease or health condition that was needed at by my line manager at that time. The unit I worked in then (Swansea Centre for Health Economics (SCHE)) was attached to many studies, but when I was asked to conduct an economic analysis of the All Wales Lymphoedema Service (Paper 2), this became the first time that I had worked closely alongside healthcare professionals, and designed the methodology of the study from scratch. This included the emphasis on the importance of collecting NHS resource utilisation from 6 months before entry, to 6 months after entry into the LW's national service.

This close working relationship continued with the economic analysis of the OGEP programme (Paper 3) which used a similar observational 'before-after' methodology. Once I left SCHE, I was able to work more closely, and primary solely as a researcher, with LW. We designed and conducted the study looking at raising the awareness of Lymphoedema in care homes (Paper 4) and also ‘Project B’ which looked at changing the process of garment procurement for health professionals (Paper 5). Again, I was included in the study designing process right from the start as LW knew that without having the correct methodology in place, then they may not collect the correct data, particularly the resource utilisation data that would be critical to the HE analysis. This continued with DNAs study (Paper 6) and Paper 8 where we evaluated the data collected by LW on lympho-venous anastomosis (LVA). Paper 7 where we collaborated with the SAIL Databank on the cost of managing patients with cellulitis in Wales was slightly different as the methodology, design and timeframe of the study data was, to a certain degree, steered by the SAIL researchers. However, as the Swansea University researcher, I was able to be the conduit for the discussions relating to methodology and what exactly was possible to get from SAIL. This proved to be a successful collaboration with which we built on and worked with SAIL again on the link to cancer surgery and Lymphoedema diagnosis (discussed on line 1605, page 165 in the thesis), which at the time of writing these corrections, this paper has been successfully accepted by International Wound Journal.

As a direct result of working with myself as a Swansea University’s researcher, LW moved part of their service headquarters to the Health and Wellbeing Academy at Swansea University. This move has already increased the visibility of LW’s research and has led to further collaboration between myself and researchers within the School of Medicine and the School of Health and Social Care. More importantly, this has led to LW’s access to teaching on the Nursing Programme.

My working relationship with LW is incredibly dynamic and fertile with many projects and collaborations being discussed well into the future. I am part of their quarterly Research meetings and have also been a key part of their Clinical Leadership Programme where all members of the LW network are encouraged to develop and present their own small study that has either a HE and/or NHS wastage eliminating element. I am part of the ‘panel of experts’ that help and guide the staff with advice on methodology, data collection and scope of their studies.

Section 5:14 REF 2021

In 2018 I was approached by the head of my then College of Human and Health Sciences to contribute an Impact Case Study (ICS) to REF 2021 under the Public Health, Health Services and Primary Care Unit of assessment (UoA2). This ICS would be solely based on the research work I had been working on with LW. The ‘Underpinning’ research included 5 of the papers that are presented in this thesis. These were Papers 1, 2, 3, 4 and 8. The Details of the impact were split into three main categories of impact. These were ‘Contributing to evidence-based policy change’, ‘Improving the management of chronic lymphoedema for patients and practitioners’ and ‘Increasing the economic impact of improved clinical care’.

This first part of the impact was concentrating on the work that we had done with OGEP and ‘The Wet Leg Pathway’ (WLP) and the work that LW had been doing developing innovative methods to support the management of chronic oedema within the community setting (discussed elsewhere in the thesis). The resulting report of my OGEP findings resulted in the Welsh Health and Social Care department funding the rollout of the WLP throughout Wales, and this impact was corroborated by testimonials by both Dr Melanie Thomas (National Clinical Lead/ Director for Lymphoedema in Wales) and Vaughan Gething MS (then Minister for Health and Social Services).

The £1.4 million permanently allocated funds by the WG for the rollout of OGEP and WLP across all seven HBs for the benefit of all community nurses and DNs was supported by further testimonials by Andrew Goodall (then Director General for Health and Social Services and the NHS Wales Chief Executive), Judith Paget (then Chief Executive, Aneurin Bevan University Health Board) and Professor Alan Brace (then Director of Finance, Finance Directorate, Health and Social Services Group, Welsh Government). Further, Again, the education on the WLP includes training on the appropriate use of compression bandaging and the resulting changes in practices have led to a significant reduction in the number of required DN visits was commented on in and email communication between Jean White (then Chief Nursing Officer for Wales (CNO)) and Timothy Kelland (then Assistant Director, Finance Delivery Unit, NHS Wales).

A survey of lymphoedema practitioners was conducted between January and March 2020 on the use of the WLP. Below are two example quotes from participants in the survey:

"[The WLP] hugely reduced the need for dressings and Multi-Layer Lymphoedema Bandaging (MLLB), more normal body image, patients able to socialise without stigma, reduction in washing and care needs for carers" (clinical nurse specialist, Wales);

"*[The WLP] reduces wet legs, improves healing, patients can return to work earlier, wounds heal quicker, improves mobility*" (lymphoedema clinical lead, Wales).

The analysis and report produced by myself on OGEP was also endorsed and supported by Jean White (then Chief Nursing Officer of Wales). She noted, "*The report makes sobering reading on the debilitating and distressing effects of this condition and I believe it is therefore essential that community nurses take a leading role in delivering effective person-centred care (OGEP), which minimises the complications of the condition and eliminates hospital admissions as far as possible*". In addition, OGEP was supported by all Directors of Nurses in Wales, across all seven Welsh Health Boards (WHB)s, and crucially, my work was cited as the only economic analysis source in the updated clinical guidelines for the WLP created by the LW.

Our review of the direct costs associated with the OGEP showed that the largest differences between the outcomes at the baseline and the three-month follow-up review were in reduced frequency of DN home visits (53%). A DN at Hywel Dda University Health Board (HDUHB) noted the effectiveness and cost-efficiency of the WLP in a comment on Twitter in 2019 when replying to a comment by a LW staff member): The DN said” *Introducing the ‘Wet Leg’ Pathway was one of the most clinically effective, cost-saving and labour saving innovations I have ever seen in District Nursing!!”*

My ICS was submitted to the REF 2021 panel (REF 2021), along with one other ICS in UoA2. The results of those two ICSs were 2.5* and 3* rated. The name of the author or the title of the ICA is never made public, but depending on what star rating my ICS was awarded, my ICS and the research that myself and LW conducted was between 2* (“*Quality that is recognised internationally in terms of originality, significance and rigour*”) and 3* (“*Quality that is internationally excellent in terms of originality, significance and rigour but which falls short of the highest standards of excellence.”)* (REF 2021)

In terms of what is happening in the service currently as a result of my work, and what has been changed, the following statement is from Dr Melanie Thomas:

“The impact of the health economic studies has added to the overall success of the Lymphoedema Wales Clinical Network. In times of great austerity, financial benefits evidence is of vital importance. Using real time action research especially before and after intervention can prove to those who hold the NHS Wales purse strings that change is essential. The results for lymphatic venous anastomosis, on the ground clinical education programme and nursing home education proved that the changes implemented by the network made a difference not only to avoiding funds but improved quality of life for their patients. Gaining evidence from both patients and savings for the NHS supported the ongoing permanent changes such as the successful implementations of the Value Based Business case for permanent staff. One could tentatively suggest that had the network not used the vast amount of evidence gathered permanent funding would not have been released. Using real time before and after design data utilising resource utilisation has enabled the network to be one of the most successful examples for Value Based healthcare in NHS Wales and the research that I have been involved in has certainly supported this impact”.

Chapter 6

Conclusion (including current and Future research)

This last section of my thesis will draw on all the previous discussed papers, research and analysis into a concluding discussion regarding both the current research that is underway with LW, and the future research that is currently being planned.

Section 6:1 OGEP

Due to the success of the OGEP pilot in C&VUHB, the OGEP innovation was slowly rolled out across the health boards in Wales. Five hundred and sixty-one patients were recruited into the OGEP programme over the evaluation period across five health boards. Data, including resource use, EQ-5D-5L and LYMPROM was collected, and the analysis of the participant data (n = 561) is complete and the subsequent paper is currently being finalised. The results of the study are very positive. At baseline, there was considerable health care input over the 2 months recall period prior to receiving the OGEP; with community nurse visits showing the highest frequency of resource inputs with 8.53 (SD 11.1) mean number of visits per patient. At 2 months, these remained as the highest frequency of resource inputs 5.56 (SD 8.41) mean number of visits per patient. This was a 2.97 (CIs 2.11, 3.83) mean difference of which was statistically significant (p-value <0.001). Overall, the total mean per patient costs were £1,457.1 at baseline and £964.4 (including the addition of intervention costs) at follow up. This indicates a statistically significant (p-value <0.001) per patient cost difference of £492.7 across 561 patients (CIs -£621.5, -£363.9).

This economic analysis has provided an examination of the patient related impact with the OGEP intervention. It evidences its importance in providing an innovative solution to delivering best standard of care practice. The findings suggest an observed trend in reductions in resource utilisation costs to NHS Wales and an increased HRQoL in relation to the LYMPROM and EQ-5D-5L outcome measures.

These findings can be used to inform the direction of any further development and evaluation of the OGEP model of care across NHS Wales and beyond.

Section 6:2 Lymphaticovenous Anastomosis (LVA)

The second paper from the LVA study is currently being finalised ready for submission. The focus of this paper is on the cost elements of the LVA intervention and not the health-related quality of life outcomes presented in Paper 8. The results obtained so far indicate that mean overall costs for the two cost elements of the analysis (cellulitis infections and hospital cellulitis admissions) were estimated at £1,575.30 (SD £3,222.50) pre LVA and £185.40 (SD £960.70 24) post LVA. The mean difference was -£1,389.85 (CIs -£893.95, -£1,885.76) p-value = <0.001. The mean per patient reduction in costs across the 24 month are estimated at -£1,405.62 (CIs -£1,878.09, -£933.16). However, when the cost of the treatment (£4,551) is included, the reductions are offset by the treatment cost (£3,145.37 (CIs £2,672.90, £3,617.84). The mean EQ-5D-5L utility score (n = 143) increased from 0.743 (SD 0.168) to 0.800 (SD 0.196 which was again statistically significant (p-value = <0.001). The results provide an indication that health care costs are significantly reduced in cellulitis incidence and cellulitis admission/hospital stays. Whilst the analysis provides no indication of whether such of costs are sustained beyond the 24-month follow up, this analysis provides an indication of the potential for LVA treatment in significantly reducing resource use and its associated costs.

Section 6:3 Collaboration with industry

LW and I have recently started to collaborate with industry with both the ‘Lymphassist at home’ (Huntleigh Healthcare Ltd) and ‘Ready Wrap’ (Lohmann & Rausher) economic evaluation studies being funded by industry partners and collaborators. The Lymphassist study is a longitudinal study looking at the economic and health related benefits of using a Lymphassist machine at home, compared to attending a clinic once a week. The LymphAssist™ Homecare compression system has been designed and developed from the Cardiff based company Huntleigh. It is the only product which has been designed specifically to compliment the patient's traditional treatment in the comfort of the patient's home (Huntleigh Website 2023). The clinically proven and effective LymphAssist™ Homecare system comprises two parts, a simple to use, single key press pump and a range of twelve chamber inflatable garments or sleeves (Huntleigh Website 2023). The study is using a RUQ and the LYMPROM tool to measure the HRQoL of the patients, along with any changes in resource utilisation. Data collection has just finished and analyse of the data will begin shortly.

The smaller 'Ready Wrap' study is investigating the cost and health related implications of lymphoedema patients who wear the new Ready Wrap adjustable compression garments. ReadyWrap® adjustable compression garments are designed as an easy-to-use compression therapy for patients with venous and lymphatic disease (Lohmann Rauscher website 2023). Enabling patients to manage their condition from home. ReadyWrap® makes it easy to achieve clinically effective compression and offers the support patients need to manage their condition (Lohmann Rauscher website 2023). As well as cost data relating to the procurement and nursing time relating to the garments, the LYMPROM was also administered to look for any change in the HRQoL of the patient. The data is still being analysed and will be written for publication in the near future.

Section 6:4 Link between lymphoedema and cancer

LW and I are currently working with the analysts in the SAIL Databank team on a study examining the occurrences of cancer within the SAIL cohort and investigating the onset of lymphoedema after cancer diagnosis and surgery. The study is investigating ways of predicting when lymphoedema will occur after the cancer related surgery. The factors we are examining are the different types of cancer types diagnosed and time from diagnosis to surgery. We will be utilising regression analysis and survival analysis to detect any relationships in the data that we could highlight as predictors for lymphoedema occurrences.

Section 6:5 Collaboration with cardiology specialists

LW and I are in the process of initiating a collaboration with the cardiology specialist in the HWA with regards to collaborating on a study looking at lymphoedema and heart failure. In a recent echocardiogram pilot study conducted by the cardiology specialists and myself in the HWA, a significant proportion of participants presented with lymphoedema and potential heart failure. Since HWA is continuing to see these types of patients on a regular basis, it has been suggested that some sort of intervention or type of monitoring would be beneficial to both the patients, and the specialist treating them.

Section 6:6 LYMPROM and CELLUPROM validation

This work is ongoing with the first paper currently being written. See above on pages 34 - 38.

This body of work consists of eight peer reviewed published papers and spans work undertaken between 2017 and 2023. The research undertaken by myself and LW has had a profound impact on LW services and helped them collect evidence of their service being an exemplar of good Value-Based healthcare in practice. The evidence of these economic evaluations/economic burden calculations presented in this thesis, has given LW the opportunity to present to the WG independent costing analysis of the work they are undertaking throughout Wales. This body of evidence and the demonstrably positive impact it is having on people living with lymphoedema in Wales has enabled WG to permanently allocate the funds needed for both the LVA and OGEP programme. This has enabled LW to continue to implement their interventions and innovative practice. This has given LW the time and resources to bring their experiences and evidence of best practice to other health practitioners. This includes the nursing students within Swansea University, ensuring the education of cohorts of future healthcare practitioners throughout Wales, and other practitioners that have received training and education via the Agored and OGEP programmes.

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Appendices

Appendix 1.

Table 2. 5 Important issues relating to lymphoedema (Advice and General Information on Lymphoedema after Assessment 2023)

	If you have been assessed and diagnosed with lymphoedema, it is important to manage your condition by following the treatment plan.
Skin Care	**Skin care is very important to the management of your lymphoedema as it reduces the risk of having a cellulitis which in turn causes more damage to the lymphatic system. Things to do to take care of your skin include:**
	• Wash your skin daily with warm water and soap/soap substitute
	• Dry your skin thoroughly especially between the fingers and toes to avoid getting a fungal infection
	• Apply moisturising cream to your skin to prevent it from drying and cracking. Creams or lotions are ok
	• Jointly, you and your therapist will decide which soaps and creams are best for you
	• Daily skin care will help you to reduce your risk of cellulitis (skin infection)
	• When cutting your nails, take care not to damage the cuticles as it could cause cellulitis
	• Avoid sunburn by using a high factor sun cream. Be aware that you can also burn through your compression garment if you are given one
	• On your affected limb, try to avoid repeated needle-stick procedures such as blood tests, immunisations, or tattoos, to reduce the risk of cellulitis. However, please do not worry in medical emergencies

Movement, Exercise, Activity	**Movement, exercise, and activity will help muscles pump lymph fluid more effectively through your body. It helps you improve and maintain flexibility and keeps your bones strong. Being more active helps you achieve and maintain a healthy weight and may improve your emotional wellbeing. Activities such as walking are ideal, as are swimming, cycling, bowling, gardening, housework and even shopping. Basically, move your body. If you tend to sit a lot, then try getting up and moving around a little bit more every day. If your mobility is limited, you can still move your body. Try lifting your arms and legs while sitting in the chair. Some people have found that dancing when music comes on or walking on the spot while waiting for the kettle to boil can help. Some watches and mobile phones can capture the numbers of steps you take every day. Keep a note of the amount you do. Ultimately the more you move your body, the more you move your lymph. Agree your activity programme with your therapist.**
	• We suggest that you wear your garment when exercising to improve the muscle pump action
	• Try to do a little every day and build up to about 30 minutes of activity 5 days a week
	• Pacing yourself is important. Use your common sense - listen to your body when it says I am tired
	• Work jointly with your therapist to set personal and achievable goals
	• Sometimes keeping a record of how much activity you do can be beneficial i.e. walked to the corner shop and back, walked upstairs 4 times
Compression	**There are different types of compression:**

	1. Compression garments which are the main treatment in controlling and managing your lymphoedema
	2.Multi-layer Lymphoedema Bandaging is often used to reshape the swollen area, reduce the swelling, and stop leaking fluid
	3. Compression wraps may also be used to reduce swelling in combination with compression garments
	• A compression garment lasts for 6 months of daily wear. You will receive two garments per limb to allow you to wear one whilst the other is being washed. Therefore, your compression garments should last for 9-12 months. Compression garments can be hand or machine washed but you should not use fabric conditioner as part of the wash cycle. All garments come with manufacturer's guidance, and it is important you follow their instructions. Please do not keep one garment for best as this will not help to manage your swelling.
	• Compression garments should be worn every day. You can start off using them for a few hours and the building up to all day. Your therapist will tell you how many hours but there are no rules. You are in charge of your lymphoedema and garments but the more you wear them, the more your swelling will reduce
	• Compression garments need to be applied evenly over the limb so that there are no creases.
	• Rubber or gardening gloves will help smooth out the garment preventing creases or folds as well as preventing ladders and pulls in the fabric
	• Do not fold your garment over at the top as this will double the compression making it more difficult for the fluid to move off your limb

	• There are aids available to help get a garment on and off. Ask your therapist for advice
	• Remember there are many different types of garments so if one is not suitable you can try another style. If you develop increasing pain or discolouration when wearing your compression garment, remove it immediately and contact your therapist.
Weight Management	• It is vitally important to maintain a healthy body weight i.e. a BMI (Body Mass Index) around 25.
	• It is known that being overweight will make your lymphoedema worse as fat cells obstruct the flow of lymph. Therefore, if you put on weight, you risk making your swelling worse.
	• Ask your therapist for support in managing your weight.
	• Weight management is extremely important.
	• Some patients who lose weight find that their lymphoedema greatly improves.
Lymphatic Drainage	**There are two types of lymphatic drainage:**
	• Simple Lymphatic Drainage (SLD) is a lymphatic drainage massage which you are able to do for yourself. SLD aims to move the fluid from swollen areas. It is a very slow, gentle series of stroking techniques, exercises and breathing exercises. For best results, SLD needs to be done at least once a day and should take you between 15-20 minutes to complete. Your therapist will teach you SLD and provide h with a written information leaflet and link to a film.
	• Manual Lymphatic Drainage (MLD) is a specialised Lymphatic Drainage massage for patients with severe or complex lymphoedema. Only qualified therapists can deliver this treatment in combination with skin care, exercise, SLD and compression.