University of Southampton

Unknown Stories: Biographies of Adults with Primary Lymphoedema

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ABSTRACT

Lymphoedema is a chronic, debilitating and disfiguring medical condition caused by lymphatic insufficiency. This insufficiency may be characterised as primary, when there is congenital or hereditary cause; or secondary, when the cause is related to trauma or illness. It can lead to extreme swelling, most usually in the limbs, and creates increased susceptibility to recurrent, and sometimes life-threatening, infection. Physical, psychological and social effects of the condition can have a significant impact on an individual's life and their levels of participation. Lymphoedema has attracted a very small amount of research activity by comparison to other chronic conditions and almost all research has focused on the views of medical experts and relates to their management of the condition. What qualitative research that exists concerning the experience of living with lymphoedema has focused primarily on women who survive breast cancer. The voice of individuals with primary lymphoedema has not been heard.

This study investigated the life stories of eight people using an auto/biographical approach to illuminate their experiences of living with advanced and complicated primary lymphoedema. The specific aims of the study were to: record the stories of those who live with primary lymphoedema in order to better understand their lived experience; analyse those stories in the context of what is known about the condition and reflect on the management of lymphoedema; draw conclusions and make recommendations that might make a useful contribution to the lives of those who experience lymphoedema.

Each person took part in an interview with the researcher in which they were invited to talk about their lives. These interviews were

transcribed and then reviewed by the participants. Narrative and biographical analysis was employed to explore the meaning in the data.

Findings reveal the extensive impact that primary lymphoedema has on individual lives depriving participants of relationships, employment, leisure and self esteem. Although each person provided a unique and individual point of view of their life, there were certain common themes that emerged. Participants spoke of their difficulty in finding a correct diagnosis and access to effective treatment; the importance of information and education and the challenges of daily self management.

Finally, recommendations are made to increase lymphoedema awareness amongst providers of healthcare and those who commission it; promote intensive and community-based treatment programmes; teach and monitor effective self-management techniques.

In memory of 'Angela' who died during the production of this study and who gave of her time to be a part of it

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To 'Joe' who inspired my interest in lymphoedema

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To Daniel Lambert whose story is so similar to those in this thesis

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Chapter 1
Introduction

In common with probably every other doctoral student, I have struggled with questions related to the theory of knowledge (epistemology), the nature and essence of being (ontology) and the manner and means of researching (methodology). I have been particularly challenged to undertake reflexive analysis of my own role in the research I have conducted and to acknowledge its situated nature. As a consequence, this chapter intends to set the scene for the thesis by introducing my own story of 'discovering' lymphoedema through my work as an occupational therapist. In doing so, I explain my interest in the narratives of those individuals who live with lymphoedema and why I felt it important to hear their stories. The chapter closes by outlining the path that the research took and providing a summary of the chapters that follow.

My Story of 'Discovering' Lymphoedema

I had wanted to become part of the caring community since pre-teens and occupational therapy combined, for me, the ideal of working in a variety of settings with a wide range of people who faced physical and psychosocial challenges related to ill-health and trauma, mental health problems and disability. My ambition was not to care for these people, in the sense of looking after them as a nurse might, but to work alongside them to create a sense of independence, or self reliance, so that they could identify and overcome their obstacles and fears and live the lives that were important to them. In this sense, occupational therapy is closer to social work than other health professions.

When I was a student occupational therapist in 1978, I was placed for three months into the hand therapy unit of a physical rehabilitation service at the Royal National Orthopaedic Hospital (RNOH) in Stanmore, North London. As the name implies, this was a resolutely physically orientated hospital but one which received patients with interesting or complex cases from all around the country. At that time, there was little attention paid to the psychological impact of the illnesses or disabilities that patients experienced apart, perhaps, from within the occupational therapy unit where I was placed. Even there, the skills of working in mental health (which every qualified occupational therapist would have acquired as part of the basic training) were secondary to the requirements of physical rehabilitation. There were no formalised counselling sessions nor any input from clinical psychologists or psychiatrists.

When I arrived, I had just spent the previous three months in a therapeutic community (mental health) setting. This was a day hospital for people who had just been discharged from acute psychiatry services and were being assisted in their transition to community living. Many of the patients had still been very ill and I had built up some skill in dealing with challenging behaviours and in channelling those behaviours into positive activity. This was known to my new supervisors at the hand therapy unit in RNOH and when I arrived they immediately allocated a patient with an unusual case history to my care.

Joe (a pseudonym) was in his mid-thirties and had a particularly gross swelling of his left forearm and hand. The swelling stopped abruptly at his elbow and the rest of his body was completely normal. The swelling was so large that he had no movement at all in his hand and wrist and this obviously created difficulty; he even

had to cut his shirt and jacket sleeves to get them over the swelling. He suffered recurrent infections on the skin of his hand and the soft tissues in his arm were hardened and fibrotic. On reading Joe's notes, it became clear that healthcare workers felt that he was responsible for the swelling: In a self-abusive act, over many years since a teenager, Joe had used a ligature intermittently to stop the flow of blood into his left arm, stopping each time only when the arm became grossly engorged. He used this form of self harm in the same way that other young people who cut themselves do - as a way of relieving emotional anguish. Joe clearly needed mental health services – but astonishingly had never received them (this form of self abuse was little researched or understood at that time). Each time he compromised his hand and arm he would be referred to a local hand therapy unit where clinicians would work to resolve the swelling and restore function and movement. Over the years the abuse took its toll until, despite the best efforts of local therapists, Joe's arm remained constantly engorged and swollen.

As a student therapist I found myself in a dilemma: I knew that Joe's physical problems would not resolve until his mental health needs were attended to. But I was so junior in the team that I did not have the courage to question the treatment regime that had been decided for him. I decided to take a middle route in working with him. I would not pretend during our treatment sessions (as others apparently had) that I was unaware of how he came by this disability and I would talk to him about it and why he self harmed. I would also, however, do my best to focus on the hand and arm to see if we could reduce the swelling since there seemed to be no reason why we could not assist in some measure. I did report to my supervisor and to the medical consultant in charge of his care that I felt he needed referring to a clinical psychologist in the hospital or mental health services in his home town.

Joe and I entered a positive therapeutic relationship and strove to restore his hand function by traditional hand therapy methods. The hand, however, did not respond to treatment for simple oedema as I had hoped it might - and one would normally expect. I began to suspect that there was some more complex physiological cause and determined to find out what that could be.

So began my introduction to lymphoedema. After much searching I came across lymphoedema as a diagnosis. There was very little written about the condition, but what did exist confirmed for me that this was not a problem of venous supply – rather one of lymphatic flow. I deduced that Joe had damaged the lymphatic vessels in the area of the ligature so profoundly that there was now very little transport of lymph fluid across the affected area. This created a backlog of protein-rich fluid which was causing his physical symptoms. He and I were both elated at my discovery; it seemed that whilst the condition could not be cured, it could be managed. During the time remaining on my placement we focused on massage and compression and began to see some results. When I left the placement, I never forgot Joe or his interesting story. I have always hoped that he found some resolution to his personal dilemmas and that he found others able to help him manage his lymphoedema.

In later years, I continued to notice articles written about lymphoedema and to seek them out to read them. I was discomfited when I began to come across reports of women who experienced iatrogenic lymphoedema following surgery and radiotherapy for breast cancer – even more so when close friends or colleagues experienced this unwanted and unanticipated side-effect of the life saving treatment they needed. Increasingly, I became

impassioned about the injustice implicit in the fact that there were ways of managing this condition but that ignorance and lack of resource were preventing people from receiving the help they required. In 1999 (some 21 years after my first exposure to lymphoedema) I undertook specialist training in Australia as a lymphoedema therapist. Gradually, I came to the conclusion that individuals who experience primary lymphoedema were the most disadvantaged of all since they were so often ignored or marginalised when it came to treatment opportunities. My contribution to reducing that disadvantage could be in educating and informing a younger generation of therapists, nurses and medics about lymphoedema and its prevention and management. As a consequence I have developed and taught lymphoedema programmes for undergraduates since that time and engaged in modest research activities linked to the condition. On embarking on this doctoral programme it felt only natural to use lymphoedema as a focus for research – especially if that research could mirror my interest in holistic approaches and work to expose the injustices that people with lymphoedema experience in accessing services that might assist them to lead more fulfilling lives.

Structure of the Thesis and Research Aims

Chapter 2 reviews the literature related to lymphoedema and provides the background to the later discussion of the issues raised in the biographies of individuals with lymphoedema. It begins with a background to lymphoedema including its physiology and aetiology and continues with a discussion of the available treatments for lymphoedema. A section on body-image concerns follows with a discussion of the psychosocial effects likely to attend the condition. Coping with chronic conditions is also discussed with reference to theories related to loss and biographical disruption and,

finally, access to information, education and services is considered. The chapter closes with a summary and an explanation of the research aims which are to: record the stories of those who live with primary lymphoedema in order to better understand their lived experience; analyse those stories in the context of what is known about the condition and reflect on the management of lymphoedema; draw conclusions and make recommendations that might make a useful contribution to the lives of those who experience lymphoedema.

Chapter 3 offers a rationale for the auto/biographical approach used in this study and an explanation of why it is particularly useful to adopt for this research study. The plan of investigation is described providing details of selection procedures and data gathering methods. Further, the methods of data analysis are discussed along with the importance of reflexivity in qualitative research. The chapter ends with a section on ethical considerations in research of this nature.

Chapter 4 contains a brief biographical portrait of each of the eight individuals who took part in the study whose pseudonyms are: Susan; Belinda; Gerry; Philip; Louise; Angela; Martyn and Marie. In doing so, it outlines how their primary lymphoedema manifests itself and what effect it has on their everyday lives. It comments on their family and employment circumstances, and on the nature of the conversation between us.

Chapter 5 presents a synthesis of the findings and discussion. It focuses on the themes that emerged from the biographical interviews. Even though each participant provided a unique and individual point of view of their life, a number of common themes emerged across all stories. These include the impact that

lymphoedema has had on individual lives by depriving participants of relationships, employment, leisure and self esteem. It chronicles the difficulty in finding a correct diagnosis and access to effective treatment. It comments on the shock and fear of cellulitis - and its financial cost to the NHS. It reveals the importance of information and education – both for themselves, and for the healthcare professionals who work with them, and the challenges of daily self management.

Chapter 6 concludes the thesis and reflects on the outcomes of the research including the efficacy of its methodology and methods. In addition, the potential for future research in this area is explored and finally, some recommendations and conclusions are drawn about improving the circumstances of those who live with primary lymphoedema.

An **Epilogue** is included which chronicles the life of Daniel Lambert, who lived in Leicester between 1770- 1809. Mr Lambert was noteworthy because of his great weight and girth on his death despite reports of his abstemiousness. In the epilogue, I present a hypothesis that he was, in fact, living with lymphoedema – a condition that was not known about at that time. In many respects, Daniel's life-story was very similar to the stories voiced in this study and provides a poignant reminder that things have not moved on as much as one might have hoped in 200 years.

The **Reference Section** contains references used throughout the study and is presented in the Harvard style.

The **Appendices** contain evidence of permission for the project including ethical approval and documents used in the research.

Chapter 2
Review of Literature

Introduction

This study seeks to reveal the life stories of adults with primary lymphoedema. It seeks to uncover the particular challenges of living with a condition for which there is no cure and in which self-motivation and self-care are fundamentally important to the long-term management of the condition (Twycross *et al.* 2000).

This chapter provides an overview of the challenges presented to those who live with primary lymphoedema and initially introduces the reader to the condition itself, its aetiology and incidence. It summarises the physical and psychosocial difficulties, including body image and lowered self esteem concerns that confront individuals who live with the condition and, in doing so, makes reference to theories on loss and biographical and occupational disruption. This is followed by a brief discussion of the current management techniques employed by the National Health Service (NHS) to maintain the health of individuals living with this chronic condition. Since the condition is largely self-managed, theories of motivation related to medical self-management are explored. Comment is made on the issues surrounding access to healthcare services and highlights factors that might have impeded progress in widening access to information on the condition for individuals who have lymphoedema and to the healthcare professionals who will deal with them. This leads into issues surrounding the education of healthcare professionals who deal with those affected by lymphoedema. The chapter closes with a summary of the literature review and a rationale for the research along with a statement of the research aims.

Further information on the lymphatic system and treatment of primary lymphoedema is included in Appendix 1.

Lymphoedema – aetiology and incidence

The lymphatic system is a one-way drainage system which transports lymph fluid from the tissues of the body back to the blood circulation. Lymph is a clear, watery fluid that contains protein molecules, electrolytes, fats and cell debris (Browse *et al.* 2003). Lymphatic vessels are present in all tissues except the brain and bone marrow (Woods 2007). Tiny lymph vessels converge to form larger vessels which pass through a series of lymph nodes to drain into two specialised ducts (the thoracic duct and the right lymphatic duct) and from there into the venous system. Lymphatic return is the only means by which fluid and protein can be returned to the bloodstream and the lymphatic system acts as a waste disposal unit for the body. As the lymphatic system extracts excess fluid and protein from the tissues and back to the blood (Berne & Levy 1996), it additionally acts as a filter - removing foreign particles such as bacteria and cancer cells. Lymph nodes contain many lymphocytes which trap and break down such offending material. Thus the lymphatic system plays an important role in the body's immune system.

Lymphoedema is a chronic form of oedema or water retention. It is an unstable, debilitating and disfiguring condition that results from lymphatic obstruction or insufficiency (Berne & Levy 1996). It manifests in the body as chronic and progressive swelling of the soft tissue caused by a failure in lymph drainage. This results in disfigured, heavy and painful body parts – most often in one or more limbs - and is associated with life-threatening infections. If lymphoedema is left untreated, it will result in swelling that is

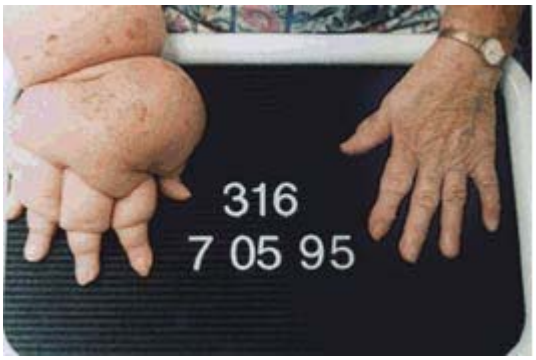
irreversible and skin tissues that become increasingly hardened and fibrotic (Twycross *et al.* 2000).

If the initial lymphatic vessels cannot open (for whatever reason) to allow large protein molecules to pass out of the interstitium, those proteins rapidly build up and lead to a change in colloidal osmotic pressures. When those pressures increase, a concomitant build up of excess fluid and swelling occurs as the body seeks homeostasis (Casley-Smith & Casley-Smith 1997). This is the basis for the characteristic high protein swelling in lymphoedema.

These high levels of protein also provide an environment on which bacteria and other mutant cells can feed, and therefore a constant risk of infection - both fungal and bacterial - is present (Browse *et al.* 2003). A frequently reported feature of lymphoedema is the development of acute bacterial infection known as cellulitis.

Photographs

The following graphic photographs demonstrate the typical appearance of swelling in lymphoedema and give an impression of the challenge of living with swollen and disfigured limbs. Some readers may prefer not to look at them since the images may be shocking to those who have never seen the condition. My rationale in presenting them is not to exploit or sensationalise but, rather, to aid understanding that this is not a condition with mild swelling (which to some may not seem too malign) but actually one that has a profound impact on everyday lives. In that sense, my aim in presenting the images is to empower those with lymphoedema, by helping to have the seriousness of this condition acknowledged.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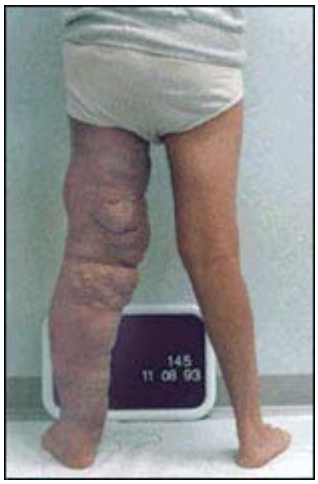
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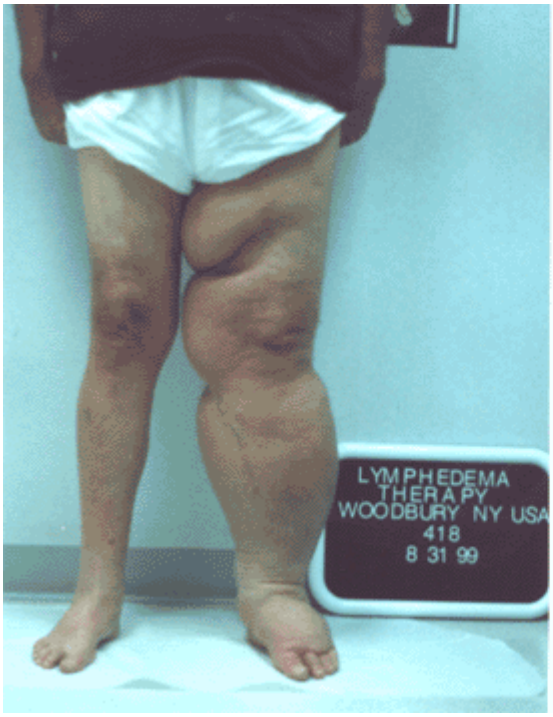
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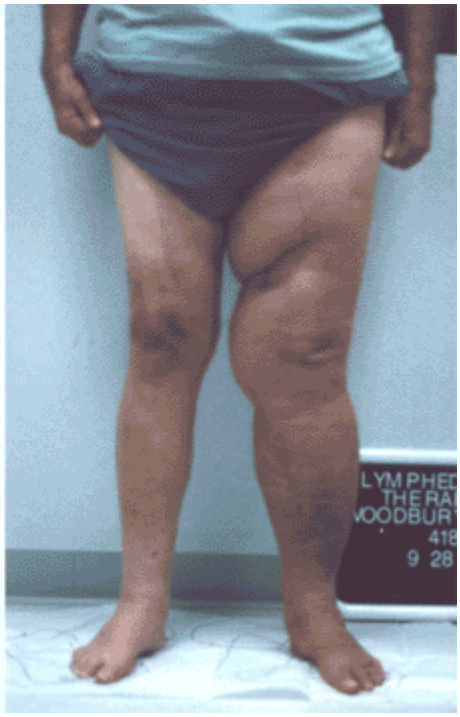

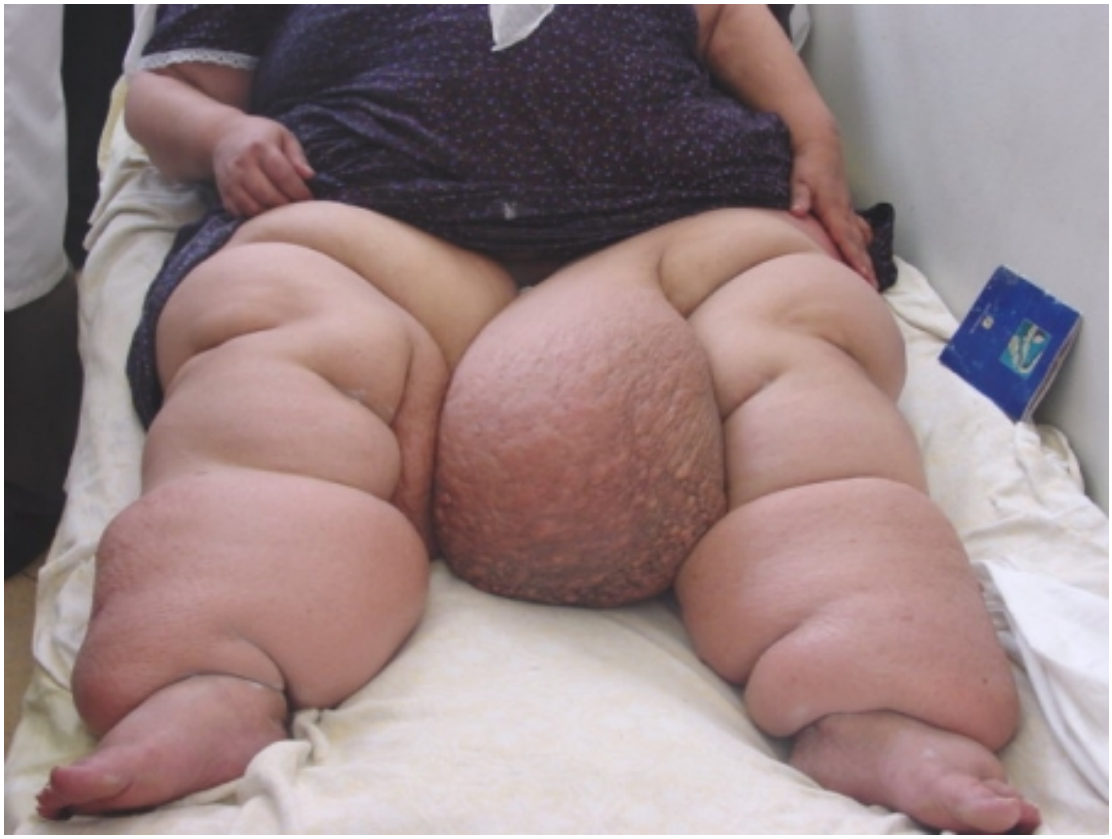

Episodes of cellulitis are characterised by a sudden onset of systemic symptoms such as fever, shivering, headaches and nausea which are associated with localised symptoms of pain, rash, inflammation and increased swelling (Woods 2007). If not recognised and treated rapidly, cellulitis can quickly lead to extreme illness and hospitalisation. Recovery can take several days or weeks – at great financial cost to the NHS and personal cost to the individual – and if the episode is not fully resolved can lead to frequent recurrence (Woods 2007).

Lymphoedema may be **secondary** to a known cause, such as:

- Trauma – perhaps following severe burns, surgical or radiological interventions (usually following treatment for cancer). In this case the lymphatic tissue is actually damaged and although capable of some regeneration, cannot function effectively and is easily overwhelmed (Foldi 1994, Browse *et al.* 2003). This is the most common cause of lymphoedema in developed nations where survival from these traumas is more common (Berne & Levy 1996).
- Infestation – most often Filariasis in which a parasitic worm enters the lymph system through a mosquito bite and then obstructs the flow of lymph and destroys the lymph vessels. This is the leading cause of lymphoedema in tropical/subtropical nations where mosquitoes are unchecked. In these nations only malaria is a more frequent cause of illness than lymphoedema (Casley-Smith & Casley-Smith 1997, Browse *et al.* 2003, World Health Organisation (WHO) 2008).
- Infection, such as the cellulitis mentioned above, further destroys localised lymphatics and a non-virtuous cycle is set up (Woods 2007).
- Neoplasm – perhaps secondary to cancers in other sites, e.g. breast, lung or bowel, or a cancer originating in the lymph nodes

themselves e.g. Lymphoma – which invades the lymphatic space and/or destroys lymphatic vessels (Berne & Levy 1996).

The lymphoedema may also be of unknown cause, i.e. be a **primary lymphoedema**, and arise from:

- Genetic factors – linked with Milroy's Disease (present from birth) or Miege's Disease (appearing later, between puberty and fifty years) in which certain chromosomal abnormalities have been identified (Browse *et al.* 2003, Evans *et al.* 1999, Mangion *et al.* 1999).
- Congenital factors – perhaps where development of the lymphatic system is interrupted in its development (usually in the womb) and no, or very few, lymphatics can be found in affected areas, e.g. Connatal Lymphoedema (Browse *et al.* 2003, Burrow & Kauffman 1996).

In the UK, the number of people who have lymphoedema is estimated at approximately 2 per cent of the population (Lymphoedema Support Network 2009). This figure may be hard to substantiate given that many people with lymphoedema – particularly those with primary lymphoedema - are cared for in the primary healthcare sector and there is considerable potential for under-reporting or misdiagnosis in this condition because of confusion with more common oedemas or obesity (Twycross *et al.* 2000, Mortimer *et al.* 1995). If fully accurate, these figures suggest that lymphoedema may be more prevalent than conditions as well known as Parkinson's disease (Aminoff *et al.* 1996). Lymphoedema may also be under-recognised and under-treated because of: its tendency to be insidious in onset, developing gradually over several years; a lack of standard criteria for measuring and diagnosing lymphoedema; a lack of education for key health professionals; a view that lymphoedema is not a life-threatening condition –

although that view is challenged by experts (Armer 2004). Following treatment for breast cancer it has been estimated that as many as 28 per cent of women may be affected by secondary lymphoedema because of the removal and irradiation of lymph nodes (Mortimer *et al.* 1995) although a more conservative 20-25 per cent is currently agreed (Woods 2007, Medical Education Partnership 2006).

Physical and psychosocial difficulties

The affected limbs are grossly swollen and have characteristic skin changes. Hair follicles and sebaceous glands stop functioning, warty type protuberances, called papillomatosis, may appear and the skin becomes thickened and discoloured, often with associated fungal infection. Affected limbs may feel heavy and painful (with 'aching' or 'dragging' being the terms most often used to describe this pain) and mobility will be restricted over any intermediate joints (Twycross *et al.* 2000, Browse *et al.* 2003). In extreme swelling, where pressure has built up considerably, fistulae may occur in over-stretched skin and lymphorrhoea (a leaking of lymph fluid from the affected limb or body part) may occur (Woods 2007, Browse *et al.* 2003).

Functional problems will be likely - particularly (in lower limb involvement) with movements involving leg and ankle bend such as: sitting to standing; climbing stairs; driving; kneeling; bending and walking - especially over a distance or on steep slopes (Twycross *et al.* 2000). Moving a very heavy leg can create fatigue and the leg may be dragged. Asymmetry in the body can cause a loss of balance and sometimes results in falls. Individuals with lower limb lymphoedema may not undertake normal levels of movement and exercise and this may lead to a downward spiral of immobility and obesity (Woods 2007). Locating footwear, hosiery and clothing to fit will also be problematic (Disabled Living Foundation 2008).

Lymphorrhoea, which occurs most often in the lower limbs, will cause distressing problems such as soggy footwear or even puddle formation where the patient is sitting or standing (Todd 1996, Casley-Smith & Casley-Smith 1997, Woods 2007). With upper limb involvement, finger, hand, wrist, elbow and shoulder movements will be restricted and manipulation of small items will be very difficult. Raising the heavy arm will also be difficult and grip and carrying strength will be reduced. Self-care activities such as shopping, household cleaning, cooking and eating, bathing, dressing and grooming are all likely to be compromised. As with the lower limb, asymmetrical limbs will create difficulties in purchasing clothing which will fit from normal retail outlets (Disabled Living Foundation 2008).

There are likely to be important social and psychological implications for individuals with lymphoedema which can drastically reduce their quality of life. It is well documented that individuals may experience pain, disfigurement and altered body image, low self-esteem & confidence and low mood and motivation (Tobin *et al.* 1993, Woods 1995, Carter 1997, Allam & Waters 2003, Medical Education Partnership 2006, Woods 2007). There is a dearth of research, however, into the psychosocial effects of lymphoedema and that which exists is predominantly concerned with post breast-cancer treatments. To date, there is only one published tool for the assessment of quality of life which is specific to those with lymphoedema (Launois *et al.* 2001). In a validation study involving over 300 patients, physical and social-withdrawal dimensions, but not psychological dimensions, were found to be correlated to the severity of the condition. The findings also showed that the volume of swelling was poorly correlated with the impact on the patient. This reflects earlier research with breast cancer survivors (Carter 1997) where even a mild swelling was shown to cause great

distress amongst some participants whilst others with much greater swelling were relatively sanguine about their situation. It may be that pre-morbid character traits play a part in this outcome (Carter 1997).

Dewing (1997) asserts that, in western society, the body ideal is based around images of youth, vigour, beauty and good health. All of these attributes carry physical implications. There are social and cultural norms about what is acceptable and what is considered ideal in terms of body image and these are reinforced strongly by the media, clothing and fashion industries. They are also reinforced by many in the non-disabled, majority community who may have their own, negative, assumptions about body changes related to illness or disability (Dewing 1997). 'Body image' as a term is difficult to define and has been linked to body concern, sexuality and self esteem (Shearsmith–Farthing 2001) but what is accepted is that body image develops from childhood and is determined and/or influenced by socio-cultural environments. Body image consists of subjective expectations and self perceptions and is inextricably linked to how an individual feels about 'self'. This, in turn, is linked to psychodynamic and psychosocial factors (White 2000). The impact of an illness or disability may result in physical change that threatens the 'self' and the body image. Body appearance, beliefs about the strengths and limitations of the body and how it functions within its environment may all undergo change (Shearsmith-Farthing 2001). These changes may be concealed by the individual or made apparent to those around them and the extent of the loss and grief associated with them will be linked to the value that the individual ascribes to the 'ideal' body concept. People with lymphoedema have an ever-present, visible reminder of their disfigurement and difference – either in the appearance of the swollen limbs themselves, or in the bandages and garments used to

manage the condition. It is a reasonable assumption that they will experience some distress at their altered body image – although there is very little research (or even commentary) which deals directly with body image concerns and lymphoedema and, once again, where it exists, it is concentrated in the field of post-cancer survivors (Carter 1997, Twycross *et al.* 2000, Woods 2007). It does, however, remain the case that there may be a considerable residue of distress for those with lymphoedema. There is some earlier research which contests this assumption of distress, however, and instead posits the notion that people with disability – even those with acquired disability – will not automatically suffer body image concerns (Samonds & Cammermayer 1989, Stensman 1989). Further, advocates of the social model of disability, in which impairment is seen as a social construction as opposed to a medical problem, have argued that people with disabilities are put under pressure to conform to non-disabled norms around body image - even when those body concerns are not owned by them (Barnes *et al.* 2002). It may be that non-disabled people, including those working in health and social care, should consider this different view which challenges the current, predominant thinking based on an able-bodied perspective. This reformulation of disability as a form of social oppression as opposed to a purely medical or welfare concern began in the 1970s: "The social modellist idea that disability is the outcome of social arrangements that work to restrict the activities of people with impairments through the erection of social barriers has become the leitmotif of disability studies in Britain. The social model of disability is the rallying call for disability organisations identifying with the disabled people's movement" (Thomas 2002:40). What is interesting is that this emergence of the social model of disability has had little or no impact on the 'medical model' perspective which continues to equate disability with impairment and continues to put its efforts into avoidance, elimination,

treatment and classification of ‘deviations’ of body and mind from the social norms (Thomas 2002). Although I will argue that in lymphoedema (as with other chronic and degenerative diseases) a stubborn refusal to respond to medical inputs leads to the condition – and those suffering with it – being marginalised by the medical profession. Mike Bury attempted to provide an early challenge to the medical model through his work on the International Classification of Impairments, Disabilities and Handicaps (ICIDH) for the World Health Organisation (WHO 1980). His stance reflected a shift away from a strictly biomedical/rehabilitative perspective and towards one in which there was recognition of social disadvantage resulting from impairment or disability - but he has never totally embraced the social model of disability arguing that it is ‘oversocialised’ in its denial of any causal link between impairment and disability (Bury 2005). Notwithstanding this debate concerning approaches to disability issues, a major problem for people with chronic illness or disability is that attitudes towards them are frequently overtly negative. Disability is constructed as essentially problematic (Barnes *et al.* 2002) and a host of technologies and interventions have been developed to eradicate its effects. Some diagnostic tests have even been developed to eradicate foetuses which are shown to have impairments which will lead to disability – arguably, a form of eugenics. Whilst these may be undertaken with good intentions (i.e. to reduce the pool of disability along with the challenge it presents), they none-the-less stigmatise people with disability and leave them with feelings of low self worth (Shakespeare & Watson 1998). As such, people living with disability or disfigurement will often have to come to terms with pre-conceived, negative, attitudes: they may be considered pitiable, dependent, non-sexual and incapable of participating in everyday life (Shakespeare & Watson, 1998). If their disability is an acquired one, like late onset primary lymphoedema or secondary

lymphoedema, they may hold these negative attitudes themselves. Further, they may be denied the same opportunities available to others without impairment and, as a consequence, experience physical, psychological, cognitive and emotional deprivation. The loss of self esteem – indeed loss of self and one's imagined future self – that occurs with chronic or disruptive illness makes the reconstruction of a valued self a daunting project (Charmaz 2000). Arthur Frank, following up on his (1995) seminal work on the impact of serious illness or disability 'The Wounded Storyteller', described the loss of an expected life-course: "The destination and map I had used to navigate before were no longer useful. [I] needed … to think differently and construct new perceptions of my relationship with the world" (Frank 2004:304). Low mood and motivation are likely to follow and this is particularly problematic in a condition such as lymphoedema where high levels of continuous self management are critical to the maintenance of the condition (Medical Education Partnership 2006, Lymphoedema Support Network 2008b).

In the last 25 years, there has been an explosion of illness narratives and in particular those which deal with the experience of disability (Campling 1981, Good 1992, Crossley 2000, Salmon 2002, Greenhalgh & Collard 2003, Smith & Sparkes 2008). Research has demonstrated the significance of subjective realities in the abilities of people to adapt to illness and disability (Salmon 2002, Finlay 2004). For instance, numerous qualitative studies on chronic illness have demonstrated the potentially devastating impact that illness can have on individuals' lives when it threatens to disrupt their world (Bury 1982, Carter 1997, Crossley 2000). Crossley (2000: 539) notes that 'The underlying existential assumptions that people hold about themselves and the world are thrown into disarray' when they experience disability or chronic

illness. This impact of illness has been characterised as an ontological assault and as narrative wreckage (Frank 1995). Despite this plethora of exploratory, narrative work, the stories of those with primary lymphoedema have not been heard and as a consequence it is difficult to ascertain to what extent their experiences might mirror those of other, studied, groups. Bury (1982), in his seminal work on biographical disruption, described three elements of the process that an individual undergoes in responding to chronic illness: coping – involving the development of self-worth by coming to terms with an altered bodily state and situation; strategy – referring to the ways in which people manage the chronic condition itself and its impact on their life. This would include how they manage getting access to sources of support and how they balance their wish for independence against the benefits of being actively supported by others; style – refers to the extent to which individuals are prepared to disclose their condition to others in various social situations.

Erikson, an influential developmental psychologist, wrote in his seminal studies about the eight expected life stages through which an individual passes (Erikson 1950). He posits that each life stage is concerned with the resolution of a particular crisis, or a task that must be completed satisfactorily. Failure to do this would, according to Erikson; result in an individual having ongoing problems in life in relation to the issues that task is concerned with. Illness or disability could be considered an obvious antecedent to difficulty in resolving these life tensions. According to Erikson, each task or crisis involves a conflict between two polarised possibilities, with the successful resolution of the crisis occurring when the individual is able to incorporate into their sense of self mostly the positive aspect of the crisis, but balanced with a little of the negative too. Erikson thought that if an individual incorporates only the positive element

of the crisis this is a maladaptive result, whereas if an individual incorporates mostly the negative element of the crisis then this is a malignant result. An individual who experiences lymphoedema as a young person – as many of the participants of this research have done – may well have been unable to resolve certain of the life stages leaving them vulnerable to identity crisis in later life. Kubler-Ross (1973) has written of the stages that individuals go through as they struggle to deal with their own end of life and these stages (denial, anger, bargaining, depression and acceptance) have been extrapolated out to those who suffer catastrophic loss – including chronic illness or sudden disability.

As well as personal loss, many individuals may suffer occupational deprivation or loss and may be unable to continue with, or pursue, activities which are important to them. 'Occupation', in this sense, is not simply related to paid employment but rather all the activities that occupy people's time and give meaning to their lives (Neistadt & Blesedell-Crepeau 1998). Since human beings are essentially defined by their engagement with activity, anything which interrupts their ability to perform in what they consider to be normal ways will potentially create great distress (Hagerdorn 2000). For those with primary lymphoedema, as has already been suggested, there is the potential for great functional and psycho-social disruption and these difficulties may well lead to problems in self-care, employment and leisure activities. The result will likely be occupational deprivation and reduced self reliance leading to a downward spiral of depression and despair (Wilcock 1998).

Current interventions

While there is a distinct frailty in the evidence base for the basis of treatment in lymphoedema and a dearth of research into the efficacy of interventions for its management, there is a body of

literature on best practice in this area (Twycross *et al.* 2000, Browse *et al.* 2003, Medical Education Partnership 2006, Woods 2007).

Medical interventions include the use of drugs, particularly antibiotics, which are used to control and prevent cellulitis in lymphoedema and to prevent further skin damage that can predispose to recurrent attacks (British Lymphology Society & Lymphoedema Support Network 2006). Diuretics, which act to remove water from the tissues, are also commonly prescribed although their efficacy is rejected by experts in the field (Medical Education Partnership 2006) and they are only suggested for mixed aetiology oedemas or where they might ease cardiac complications of lymphatic therapy. It has been suggested that benzo-pyrones act to remove excess protein and therefore enhance homeostasis and lead to reductions in swelling (Browse *et al.*2003, Casley Smith & Casley Smith 1986). There is, however, debate about the efficacy of these drugs, with the Cochrane review of their use concluding that they were not of benefit (Badger *et al.* 2004). It is noteworthy that there has been relatively little investigation by pharmaceutical companies of the use and effectiveness of drugs in the treatment of lymphoedema and this may be linked to the fact that the major 'market' is in palliative care settings or third world countries, where profit margins are likely to be low. It will be interesting to observe whether this stance is maintained as the number of people in the developed world who are surviving cancers grows but experience secondary lymphoedema as a result of oncology treatments. Other medical treatments include low-level laser therapy, which is a relatively new intervention and one in which very little serious, scientific research has been done (Piller & Thelander 1996). It is unclear whether the improvements reported from this treatment are from new lymphatic growth stimulated by the laser treatment, new

lymphatic growth occurring spontaneously, a reduction in fibrosis such as that claimed for ultrasound treatment, some other form of concurrent treatment or some other, unknown, effect. Liposuction treatments to remove excess fluid accumulations and other forms of surgery to remove dermal flaps are also used by some surgeons although there is a good deal of controversy in the medical literature concerning their potential for further harming compromised lymphatic systems (Casley Smith & Casley smith 1997, Medical Education Partnership 2006, Woods 2007). For this reason they are advocated by experts in this condition only as a last resort to alleviate very distressing symptoms such as eyelid or genital swelling (Twycross *et al.* 2000, Browse *et al.* 2003, Woods 2007).

Therapy interventions include the use of compression therapy which involves the application of gentle external pressure to the body by the use of multi-layer bandaging or specifically designed low-stretch pressure garments. The compression provides resistance against normal muscle action and so aids in the pumping of lymph fluid into the initial lymphatics (Twycross *et al.* 2000). Another important intervention is manual lymphatic drainage (MLD) which encompasses a special form of lymphatic massage that evacuates lymph from the central regions of the body to the vascular system and thus gives the accumulated lymph in the periphery a place to drain to (Foldi 1994, Casley Smith & Casley Smith 1997). A consensus currently exists amongst the experts in the field of lymphoedema that MLD should be an integral part of the treatment offered to those who have lymphoedema (Medical Education Partnership 2006, Mortimer & Todd 2007, Woods 2007) although Browse *et al.* (2003) has questioned its importance, proffering the opinion that compression bandaging alone will yield similar results. It is important to note here that neither view is

supported by robust evidence since research in this area has been minimal. Pneumatic compression therapy involves the use of an air filled sleeve with the air pressure controlled by means of an attached pump (Allenby *et al.* 1973). The pumps apply either static or intermittent compression and the effect is a massage-like compression of the superficial tissues (Disabled Living Foundation 2008). There is some debate in the literature concerning the use of these compression pumps, with suggestions of potential for harm expressed quite forcefully in the Australian literature (Lymphoedema Association of Australia 2008, Casley-Smith & Casley-Smith 1997). Skin care is important in the treatment of lymphoedema as the skin undergoes significant changes and skin care is critical to prevent the deterioration of the condition and to prevent cellulitis (Browse *et al.* 2003). Oil based emollients soothe and hydrate the skin and should be used as part of a self management programme (Casley-Smith & Casley-Smith 1997). When skin scale is excessive, it may need to be treated with salicylic ointments and antiseptic agents. Topical steroids can also be used where skin is suppurating and smelly but have a thinning effect on skin and need to be used sparingly (Browse *et al.* 2003). Exercise is used in both the active and maintenance phases of lymphoedema management as it increases the uptake of fluid by the initial lymphatics, encourages the collecting lymphatics to pump more strongly, keeps joints mobile, strengthens muscles and prevents muscle wastage and may help to prevent skin from stiffening (Goddard *et al.* 2008). Aerobic exercise increases intra-abdominal pressure which facilitates pumping of lymph into the thoracic ducts (Medical Education Partnership 2006). Although acknowledged to be useful, advice on exercise regimes are non-specific and vary between therapists and regions (Medical Education Partnership 2006, Twycross *et al.* 2000, Wessex Cancer Care 2008). Additionally, there is little research evidence to indicate which

types, intensities or frequencies of exercise may be safely used in the management of primary lymphoedema (Goddard *et al.*2008). For more information on all of the treatments described above, please see Appendix 1.

It may be that the weaknesses of the evidence base for successful lymphoedema therapy, and the paucity of knowledge and expertise amongst healthcare professions about the condition and the treatments generally available in lymphoedema, are factors in the lack of attention paid to this profoundly debilitating condition by UK healthcare professions. It is not surprising that a concomitant lack of access to treatment exists for those who might otherwise benefit. The cost of both professionals' time and of the resource required is high and many patients are simply denied access to these forms of therapy (British Lymphology Society 2003, Lymphoedema Support Network 2008b).

Self management and motivation

Profoundly important and inherently problematic, effective and sustained self management provides for the best long term prognosis for this condition (Petrek *et al.* 2000, Medical Education Partnership 2006, Woods 2007). It is the most important means to maintain treatment outcomes, ensure ongoing good health and avoid the complications that can have such a deleterious effect on functional abilities and quality of life. The most promising lymphoedema self-management techniques are time consuming to perform, however, and are sometimes difficult to accomplish without support from family or friends. Self management involves undertaking a modified form of self-massage (taught to the person as part of active treatment), regular skin care to prevent fungal or bacterial infection, bandaging or the wearing of pressure garments and gentle exercise to promote lymph drainage (Twycross *et al.*

2000). The massage and skin care alone can take up to half an hour twice daily and compliance with exercise regimes is notoriously difficult to sustain (Goddard *et al.* 2008). Perhaps the most difficult aspect is the application of bandages or garments. It is very difficult to bandage one's own limbs and so most people only attempt to do so on a regular basis if their limbs cannot be accommodated in garments because of huge bulk or irregular shapes. Most people fare better with pressure garments which are especially fitted to conform to the unique shape of the swollen limb (Woods 2007). These too have their challenges: because of the nature of the fabric they do not stretch easily and actually getting them on and off is very difficult. To do so requires good hand and grip strength and an ability to stretch the torso to the furthest extent of the affected limb. Many individuals can only manage with the help of another, stronger, person (Armer 2004). Although some design modifications have been made by manufacturers, these have not solved the problems and many people simply cannot manage to continue wearing the garments. It can take considerable time, perhaps half an hour, to correctly apply and remove the garments. The garments have to be worn all day and can only be worn once before they need hand washing to restore their shape and compression properties. Additionally, once the garments are in place, they are hot and uncomfortable to wear and are rather unsightly in appearance. In hot weather especially, many people abandon them altogether (Armer 2004, Woods 2007). Compliance with self-care regimes, therefore, is an important issue and is further complicated by the fact that many people do not have the information on the physiological basis for why they are asked to perform these tasks (Armer 2004). A study which focussed on the under-recognition of the impact of un-managed lymphoedema has indicated that (post breast cancer) women demonstrate inconsistency in their knowledge of what regimes benefit outcomes

(Radina *et al.* 2005) and are consequently not in a strong position to understand the implications of non-compliance with self care. This finding is likely to be reproducible in those with primary lymphoedema and may be even more marked since many have been unable to access treatment that would include such advice. Radina's (2005) study in the United States highlights the need for improved patient education. Motivation to consistently take part in self management activity is influenced by whether a person understands why s/he needs to undertake this action; what effort s/he is prepared to put into it; and how long the individual is prepared to continue trying (Arnold *et al.* 1998). People with primary lymphoedema may not have easy access to sources of advice and treatment and often lack the information required for them to understand the importance of making a positive decision to self-manage; further, they may not be able to commit to the effort required for effective management for practical reasons; and they may lose heart to continue when the task itself is so difficult and time-consuming. In order to overcome these difficulties, it may be advantageous to engage with a self-help group for mutual support and information-sharing and such groups do exist in the UK – the Lymphoedema Support Network (LSN) has a network of regional groups which meet on a regular basis (Lymphoedema Support Network 2008). Additionally, and ideally, regular contact with therapy services that can monitor and encourage is also likely to develop and maintain a positive and active approach to self-care (Medical Education Partnership 2006).

Access to accurate information about lymphoedema

Despite the high level of incidence of lymphoedema in the UK, access to specialist information and intervention is, according to the Lymphoedema Support Network (2008), difficult to obtain. This mirrors the findings of research undertaken with other groups of

people living with unstable and progressive conditions (Neurological Charities 1992, Baker *et al.* 1998, Disabled Living Foundation 2009). People with deteriorating or disabling conditions - such as lymphoedema - want access to information about the variety of services and benefits that could make their quality of life improve and enhance their independence (Cale & McCahan 1993). This desire reflects a general change in service users' attitudes over the past 40 years: they have moved from being passive recipients of services towards active engagement in decisions affecting their lifestyle and health (Ovretveit 1992, Hewison 2004) and their information needs may concern health and social care services; voluntary services; assistive equipment; and support groups. Service users may wish to examine the evidence base for therapy and uncover any contra-indications (Lymphoedema Support Network 2008b). However, there is evidence to suggest that many people are unaware of the range of services that exist and that, furthermore, they do not know how they may gain access to them (Disabled Living Foundation 2008, Bradby 2009, Barnes *et al.* 2002). An assumption is often made that when an individual with a disability or chronic illness comes under the auspices of health, social or educational professionals they will automatically be furnished with information on the services and benefits that they are entitled to and referred appropriately to these services. In practice this frequently does not happen and there is a concern that many people may be suffering a diminished quality of life because of patchy and inappropriate services, exacerbated by the attitudes of those delivering them (Neurological Charities 1992, Barnes *et al.* 1999). Barnes *et al.* (1999), in a survey of elderly people with physical impairments who were living in rural environments, discovered that only 55 per cent of those questioned knew of relevant support systems and of those only 16 per cent had been given that information by health and social care professionals

(Barnes *et al.* 1999). This may be due in part to the fact that professional education in disability and rehabilitation issues (and particularly in relation to lymphoedema) appears to be haphazard and inadequate (Baker *et al.* 1998, Greene & Waters 2003). The Disabled Living Foundation (a national charity that collates and disseminates information on equipment and services designed for people with physical impairments) believes that numerous people may be slipping through the information net entirely (Disabled Living Foundation, 2009).

Service users' views on the skills, knowledge and educational needs of professionals can differ from those of the professionals themselves: In the outcomes of a Disability & Rehabilitation Open Learning Project (Baker *et al.* 1998) it was demonstrated that people with chronic illness or disability such as lymphoedema want an investment in education in such areas as communication skills in order to underpin their wish to be respected and understood by professionals providing a service. They also want to be involved in the planning and decision-making about their care and services. In the same survey, it was revealed that professionals felt that they had good communication skills and thought that their continuing professional education should focus on developing clinical knowledge and skills. But they did, however, admit to difficulties in communicating, negotiating and coping with the feelings of failure or inadequacy experienced in coping with some of the more complex problems posed by chronic disability, such as that produced in lymphoedema (Baker *et al.* 1998). Poor communication is an all too familiar area of dissatisfaction among patients. The traditional doctor's role as the expert and decision-maker is, perhaps surprisingly, still omnipotent: Patients are expected to co-operate fully with their doctors, who then, in turn, are expected to apply their specialist knowledge for the benefit of the patient (Souza

2007). But this can leave the patient feeling disempowered and sceptical – with a residual feeling that they are not being listened to. It is reported that patients frequently comment that they are not listened to (Mercer *et al.* 2008) whilst a common complaint from professionals is that they have insufficient time with their patients, blaming economic pressures for this inadequacy (Coulehan *et al.*2001). Langewitz *et al.* (2002) undertook a US based study which demonstrated that a patient visiting a doctor gets only 22 seconds on average for his/her initial statement before the doctor takes the lead. Maguire & Pitceathly (2002) concur that this is a problem in the UK also and describe this 'taking over' as a blocking technique, enabling the doctor to take control of the discussion and bring the appointment to a swift conclusion within the time limit (usually 5-8 minutes) allowable within a busy GP practice. It is easy to see that this pattern might well be replicated amongst other healthcare practitioners. In this effort to take control, however, the professional not only fails to obtain sufficient information about the patient's problem – in this study, lymphoedema – but also ignores the functional, emotional and social impact of the problem. Maguire & Pitceathly (2002) further comment that doctors may fail to recognise their patients' psychological problems, but instead explain their distress as normal and focus on physical aspects of the presenting condition.

Access to healthcare and rationing of services

Any person informed of the scale of disability, distress and disruption to life created by the condition of lymphoedema would surely question why access to information and services is so difficult – and why so few healthcare professionals take an active interest in this condition. For those whose lymphoedema is secondary to cancer, there are services provided by both health and charitable sectors (British Lymphology Society 2008). But the provision of

these services is patchy and is often based in palliative care settings – potentially very off-putting to those who feel that they have 'survived' their cancer episode (British Lymphology Society 2008, Medical Education Partnership 2006). The situation for those who have primary lymphoedema is even worse with reports of difficulty in getting accurate diagnosis and effective treatment (Lymphoedema Support Network 2008b, Medical Education Partnership 2006, Twycross *et al.* 2000. Once again, they may only be able to access services through palliative care settings - a slightly bizarre situation if you do not have (and have never had) a terminal illness. Despite great need for and use of health services, people with primary lymphoedema face considerable barriers to access. There is compelling evidence that those disabled people who have the greatest need for service are the very ones who have greatest difficulty obtaining them (Basnett 2004). The reasons are likely to be located in the fact that healthcare services in the United Kingdom are stretched beyond the limits ever envisioned by the architects of the National Health Service (Walshe & Smith 2006). In line with other developed nations, the UK's healthcare sector is subject to certain inexorable and challenging social trends: The demographic shift in which people are living longer and the numbers of elderly and very elderly people are rising fast. Secondly, there is a rise in the numbers of individuals living with chronic long-term conditions – many of which are lifestyle related (World Health Organisation 2005). Both sets of people are making much heavier use of the health service and are very time and resource intensive with their complex, chronic health care needs. The pace of technological innovation allows for an increased ability to control chronic disease and extend life. Most obvious in pharmaceuticals, but also in surgery, diagnostics and other treatment areas, health professionals keep finding new ways to cure or manage disease (Walshe & Smith 2006). Previously fatal conditions become

treatable, and interventions to slow the progress of disease or manage its impact become more available. Sophisticated organ transplant procedures – such as heart-lung transplants; pharmaceutical treatment for cancer and Human Immuno-deficiency Virus (HIV); and enhancements to diagnostic Magnetic Resonance Imaging (MRI)/Computer Tomography (CT) scanning technology are just some examples of these technological changes. Changing user and consumer expectations mean that people want more from health services than previous generations did. Not content to be passive recipients of health services, as indicated above, they want choice and sovereignty in decisions affecting their health and are much more likely to demand new and more expensive treatments (Hewison 2004). Rising costs are an inevitable corollary to those challenges identified above. Each of them contributes to the constant upward pressure on healthcare funding and, despite increases in the proportion of Gross Domestic Product (GDP) allocated to healthcare in most developed nations; spending never seems to be enough (Walshe & Smith 2006). Those who manage healthcare services and attempt to resolve the tension between high demand and low resource, do so under constant public and media scrutiny.

In this climate, 'Cinderella' conditions - such as lymphoedema – will struggle to gain resource from already overstretched budgets and manpower and there may be an implicit rationing of service by actively deterring uptake of services in order to alleviate pressure on budgets. Primary Care Trusts, who would fund lymphoedema treatments, are very unlikely to create new services until such time as it is demonstrated that not to do so would, in the long-term, cost more money. General Practitioners (GPs) provide the bulk of health care; they support those with conditions that need to be self managed; they act as gatekeepers to more specialist services and

are central figures in facilitating access to continuing, community-based care. It is unfortunate, then, that their knowledge of lymphoedema is weak and their ability to successfully diagnose and treat the condition is thus compromised (Medical Education Partnership 2006, Lymphoedema Support Network 2008b). When they were fund-holders, GPs may even have had a perverse incentive to ignore long-term conditions such as lymphoedema as they would increase financial drain on a practice (Walshe & Smith 2006). It is interesting that those individuals with lymphoedema (or groups representing them) do not challenge more vigorously the lack of services – perhaps through an action in the courts. But although the Disability Discrimination Act (Her Majesty's Stationary Office 1995) applies to access to buildings, employment and the provision of goods and services; it excludes decisions on the allocation of health resources or whether to treat an individual and so any such challenge would founder at the first hurdle.

Properly funded academic research into the condition will be depressed for similar reasons: firstly, a lack of knowledge about lymphoedema amongst those qualified to undertake appropriate research; secondly, funding agencies (often the same organisations that provide healthcare services, e.g. the Department of Health) may be concerned that finding effective treatments will inevitably lead to greater pressure for those treatments and a concomitant pressure on budgets.

Education about lymphoedema amongst healthcare professions

The professional groups who appear to have the most obvious potential input in the treatment of lymphoedema are: Medical doctors – with expertise in diagnosis and prescription of medical or surgical treatments; nurses – with expertise in wound care, skin

care and bandaging; occupational therapists – with expertise in mental health issues, pressure therapy, functional activities and assistive equipment & techniques; physiotherapists – with expertise in massage, movement and exercise and podiatrists – with expertise in the treatment of bacterial and fungal infections and in lower leg wound care. Despite the potential for these groups to support the management of primary lymphoedema, small scale investigation revealed that pre-registration programmes in these professional groupings deal with the pathology and treatment of lymphoedema in a cursory fashion, if at all (Greene & Waters 2003). This is remarkable given the contribution that could be made for a sizeable number of patients (Waters 2003). In a study of UK therapy students, it was demonstrated that despite 33 per cent of students meeting patients with lymphoedema on practice placement, 57 per cent claimed to know nothing about the condition (Waters 2003). An exploration of the literature used by pre-registration students in the UK reveals scant information on this condition and its treatment, with most attention focussed on secondary lymphoedema but very little paid to primary lymphoedema and no referencing onward to specialist literature or information sources (Turner *et al.*1998, Bloom 1999). What little evidence there is suggests that very few student therapists believe that they are equipped to undertake a role in the amelioration of lymphoedema (Waters 2003). Lymphoedema is mentioned more frequently in post-registration nursing texts, particularly those that deal with oncology or palliative care (Carter 1997, Hassey Dow 2004). However, some of the information presented in the literature is erroneous or does not match with current approaches, e.g. the use of compression pumps alone (Turner *et al.* 1998). Further, the literature does not reflect the fact that there are important contra-indications in certain situations, and that the misapplication of some procedures can cause real harm. While it is acknowledged that pre-

registration programmes, already packed, cannot hope to deliver detailed information on every pathology or disabling condition, it is remarkable that a condition which is as prevalent as Parkinson's disease (which is well attended to in basic training) receives such little attention in general health education.

Conclusion

In conclusion, this chapter has set out to establish the aetiology and background to lymphoedema and its treatment and the factors that have influenced access to information and treatment for those who have lymphoedema. In so doing, reference to the relatively high incidence of lymphoedema has been made and set against the paucity of information and education available both to those who experience lymphoedema and those responsible for treating it. It has been demonstrated that there is a great deal more research that needs to be undertaken before we can fully articulate - using robust evidence - what interventions are likely to be most useful to those with lymphoedema. Further, it has also alluded to the fact that professional education may need to have a greater focus on this topic.

The stories of those with primary lymphoedema have not been heard to date and consequently it is difficult to ascertain to what extent their experiences might mirror those of other, studied groups. In order to allow individuals to come to an accommodation with their personal experiences and challenges, and to assist them in recovering what they value, it would undoubtedly be productive for people with lymphoedema to share their stories as a method of linking relations between events, motives, desires, wishes and hopes into meaningful configurations. In this sense, relating stories could be a form of therapy in itself. But, importantly, in telling their

stories, individuals with lymphoedema can also assist others to identify their own potential for an effective, enhanced response to the challenges arising from living with this condition.

The aim of this research, therefore, is to:

- Record the stories of those who live with primary lymphoedema in order to better understand their lived experience;
- Analyse those stories in the context of what is known about the condition and reflect on the management of lymphoedema;
- Draw conclusions and make recommendations that might make a useful contribution to the lives of those who experience lymphoedema;
- Disseminate the findings to those who live with lymphoedema and to those healthcare care professionals and managers who may have a positive role in enhancing services.

Chapter 3
Methodology and Method

Introduction

In order to elicit the voices of individuals with primary lymphoedema and to hear their views and opinions about the impact their condition has had on their lives, research located within the interpretive paradigm was conducted over a two year period commencing in October 2005. The intention was to gain an insight into the nature and meaning of living with primary lymphoedema.

Primary lymphoedema is a little known, chronic and disfiguring disability, which is unstable and debilitating (Lymphoedema Support Network 2008a). It impacts on the lives of individuals by both overt and subtle means related to physical illness, impairment and appearance; to the ways in which those individuals are viewed and valued within society and, importantly, to the ways in which they view and value themselves (Williams 2003).

The qualitative methodology employed sought to reveal the counter-narratives of those who experience the condition rather than focusing on the, usually dominant, narrative of the medical or social care professionals who work with these individuals (Kitchin 2000). Counter-narratives are designed to repair damage inflicted on individuals (and their identities) by power systems which are, or have been, abusive (Nelson 2001). Further, the auto/biographical method is recognised as having considerable potential for self-representation amongst research participants (Atkinson & Walmsley 1999) and evidence suggests that it may be empowering for groups whose voices have hitherto been unheard (Clarke 1998). Therefore,

an auto/biographical study of people with Primary Lymphoedema was conducted in order to give these individuals a voice.

This chapter outlines the rationale for the methodology used in this research, followed by a description of the investigation method and an explanation of the data analysis procedures. Finally, the ethical issues that arose from the research study are discussed.

Rationale

Qualitative approaches

In determining a methodology for this research, it was felt important to examine the two major approaches, qualitative and quantitative, which emerged as a result of the nineteenth century debates about the source of 'truth' in research (Grbich 1999).

Despite challenges, quantitative orientations have been persistent and have dominated twentieth century research activity. These presume that there is a single reality or truth that exists independently and that this can be revealed by careful and rigorous research (Denzin & Lincoln 2005). In the last half century, however, qualitative researchers have become increasingly sceptical about the capacity that quantitative methods have for describing the human condition in any depth or meaning (Denzin & Lincoln 2005). Terms such as 'reductionist' and 'mechanistic' tend to be applied to quantitative research and are used in a pejorative way by many health and social care researchers (Greenhaigh & Taylor 1997, Finlay 2004, Nayar *et al.* 2007, Denzin & Lincoln 2005). The implication is that reducing something to numbers reflects a sterile philosophy that refuses to acknowledge that people are more than biomechanical systems. To quantify may be to ignore the spiritual, inner dimensions of human life. The use of quantitative methods in this study, therefore, may not effectively realise its' ambitions.

By contrast, qualitative researchers accept that there is a pluralistic view of knowledge and that there are multiple realities and truths. They explore phenomena in their natural contexts and seek to discover complexity and meaning. Their chosen research designs are fluid and evolve throughout a period of study in response to what the data reveals. Methods of analysis are thematic and inductive and they seek to generate theory that will be useful in explaining lived experience. The relationship to participants is close, ethical and egalitarian (Clarke 2004). But qualitative research can, in turn, be criticised by those with a positivist perspective (Bird *et al.* 1995) as being unscientific – producing subjective results which cannot be applied on a wider stage. It is claimed by some (Lincoln & Canella 2004) that in qualitative methods there is an over dependence on the researcher's interpretation and outcomes are therefore subject to personal and cultural bias. Furthermore, qualitative research is accused of having such very small sample groups that findings cannot be generalised to others (Bird *et al.* 1995). The contrasting concerns of positivist and qualitative researchers are summarised in Figures 1 & 2.

Figure 1: The Concerns of Positivist Research

- Validity – the extent to which findings are an accurate reflection of the underlying purpose and/or reality of the study
- Reliability – the extent to which findings can be replicated and measurements are consistent
- Objectivity – the extent to which a researcher is able to remove him/herself from the research study
- Generalisability – the extent to which the findings derived from the study are applicable to a larger population (Finlay 1998).

Figure 2: The Concerns of Qualitative Research

- Trustworthiness – in which the findings are a true reflection of the individual's lived experience
- Transferability – the applicability, on a modest scale, of the findings to other similar conditions or situations
- Reflexivity – the extent to which the researcher has engaged in a self-conscious, self-critical, systematic and analytical approach in capturing more subjective dimensions. It places the researcher as central to the construction of knowledge following careful analysis and interpretation (Finlay 1998).

Qualitative approaches themselves have evolved considerably over the last century: In the era before the 1960s qualitative research was dominated by anthropological research on tribal groups and on urban cultures. Ethnographic studies were prevalent with researchers observing behaviours and recording outcomes but making no attempt to intervene or become participants (Kelly 2003, Grbich 1999). The 1960s saw a social revolution in its challenge to power and authority – particularly evidenced by the women's movement, the racial equality movement and (in the US) the disability equality movement. There was much striving towards a more egalitarian culture at this time and this was reflected in a pre-occupation with more collaborative approaches in research which has persisted over the past forty years. The challenge to power and authority was mirrored in the world of qualitative research and the emphasis shifted to the joint construction of social meaning between the researcher and their research participants (Denzin & Lincoln 2005). People and their experiences were studied in order to attempt an understanding of their experiences and the effects they have on individuals (Grbich 1999). Researchers who base their studies within the naturalistic paradigm are sometimes referred to as interpretive researchers (Bird *et al.* 1995) and they generally become closely involved with their participants and

employ egalitarian data collection tools such as participant-observation, case studies and interviews. This was an important influence on this study where a concern to hear the hitherto unheard voice of the participants was uppermost.

A major limitation of much existing research focusing on people with sickness or disability is that it has not allowed the voice of the disabled individual to be easily heard (Finlay 2004). Rather, researchers have tended to defer to the voice of the 'experts' – medics; nurses; occupational therapists; social workers and social scientists (Karp 1996). Inevitably, the research conducted has tended to reflect the views of those experts rather than listening to, and learning from, the personal experiences of those living with chronic disability. The auto/biographical method provides an effective vehicle for this study in that it empowers the participants and places their narratives at the centre of the research.

Auto/biographical method

The rise of the biographical method is grounded in a desire to illuminate the complexity of individual lives and, through analysis, to provide greater insight into the social and cultural networks within which that individual exists. In parallel, it develops the capacity in the researcher for ontological enquiry (i.e. questioning the nature of human beings), reflexivity and moral reasoning (Erben 1998). It differs from purely narrative approaches in its focus on life stories told against a backcloth of the passage of time.

The biographical method involves the collection and study of life documents or spoken histories – especially when these describe, or relate to, turning points (so-called 'epiphanies') in peoples' lives (Denzin 1989). When these stories or documents are analysed and recorded by the subject themselves, they are 'autobiographies';

when observed by another, they are ‘biographies’. Post-modern approaches to qualitative research methods (which would encompass biographical method) invite us to consider seriously the role of the biographer/interpreter and their influence on the represented story: hence the adapted term ‘Auto/biography’ first used by Liz Stanley in 1992 (Merrill & West 2009). This is utilised to signal awareness of the power of the interpreter/writer in bioographical research and the extent to which we use our own stories to make sense of other’s lives and experiences (Merrill & West 2009, Grbich 1999). We also, of course, use the stories of others to make sense of our own biographies. Biography is therefore always written with a double perspective – the author’s and the subject’s.

Narratives are stories – symbolic accounts of the actions of human beings which are held together by recognisable plots. They usually involve some sort of human predicament and an attempted resolution (Sarbin 1986). They introduce heroes and villains, quests and sufferings, relationships and dilemmas, triumph and failures. Through narratives, we make sense of our lives. “Culturally ubiquitous, stories are an important vehicle for creating meaning. Whether at home or work, stories are used to construct our lived realities” (Fraser 2004:196). Ricoeur (1986) goes further in arguing that the self comes into being in the process of telling one’s story. In narrating one’s life an individual can make sense of that life and allow a new self to emerge (Ricoeur 1986). Smith & Sparkes (2008:18) concur in their discussion of the contribution of narrative to disability studies, saying: “We organise our experiences into narratives and assign meaning to them through storytelling. Narratives thereby help constitute and construct our realities and our modes of being.” Narrative gives a uniquely rich and subtle understanding of life situations, and the story provides a very

feasible way of collecting data because it is such a common device in everyday interactions (Punch 2005). Carr (1986), building on the work of Ricoeur (1986) outlines the role of narrative in organising temporality and giving coherence to our experiences as we are having them. In a reflexive act, we attempt to make sense of the flow of experiences by "gathering them together in the forward-backward grasp of the narrative act" (Carr 1986:62). Carr brings the twin concepts of narrative and temporality together in his observation: "Lives are being told in being lived and lived in being told." (Carr 1986:62). It is this attention to time and the way it reshapes or reframes our narratives that distinguishes biography from other qualitative methods.

Narratives also help listeners/readers to empathise and attempt an understanding of the lives of others. They offer the biographical researcher the opportunity to begin to see from another's perspective (Erben 1998). In the shaping of a biography, through the narration of a life, that life is given meaning as the self is located in different stories. They allow others to appreciate not just the impact of the particular story, but also something of the person's life, struggles and relationships (Finlay and McKay, 2004). Hearing such stories helps one to deal with others in a more constructive way and to gain insights into their coping strategies. Such insights may subsequently be helpful in understanding other individuals with similar experiences: indeed, whilst the person's unique story cannot be generalised, it can serve as a vignette with some transferability to others in similar circumstances (Grbich, 1999). Inviting people with primary lymphoedema to share their life-stories is a way of allowing them to describe their experiences in their own words and to ascribe their own meanings to these experiences. Barnes *et al*. (2002) assert that biographical and narrative methods are appropriate methods in researching about

disability issues. They allow a challenge to common assumptions about truth and reality and can develop alternative realities from the viewpoint of the hitherto marginalized group - such as people with primary lymphoedema**.**

In other forms of research, it is often the case that personal accounts are not given the prominence they warrant, and we hear only the 'outside' voices from battalions of experts (Karp 1996). This is particularly so in the field of medicine where, all too often, the research and literature on illness is written from the perspective of the medical and health professions. As Karp (1996) noted in his book on depression and mental illness *'Speaking of Sadness'*, the essential problem with nearly all studies of depression is that the voices heard are those of mental health experts – and rarely those of depressed people themselves. Much research into disability issues has been researcher-orientated and based around the agendas of research and funding bodies, rather than around the issues that are important to people with disabilities (Kitchin 2000). This can be characterised as an exploitative approach and has led to calls for research strategies to be both emancipatory and empowering, with researchers acting as respectful participants who continually assess their own impact on the research and defer to the issues raised by the participants (Kitchin 2000). Meaningful research actively seeks out the opportunity to allow counter-narratives to come forward: these may reject the dominant voices – in this case medical narratives – and instead allow fresh perspectives to be considered. Auto/biographical research therefore has considerable potential for self-representation and is a method that allows marginalized people to speak for themselves. Auto/biography offers the possibility of conveying an authoritative account of lives without stereotyping or misrepresenting.

It is important to acknowledge some of the limitations in the autobiographical approach and the changes, over time, in the conceptualisation of people whose lives are being recorded (Merrill & West 2009). An early view was that 'truth' lay in their self reflective, subjective accounts which were distinct from the social world (Grbich 1999). Later views proposed that the individuals being researched were the products of their social experiences and norms and that the 'truth' was harder to capture (Denzin 1989). Atkinson and Silverman (1997) have been particularly critical of the narrative turn in social science research – especially where it focuses on illness narrative - and feel that it presents an overly sentimental, romanticised and hyperauthentic view of participants' lives. They cede, however, that it can be useful if subject to thorough analysis, categorisation and theorising. Finally, of course, the researcher's representation of the stories may overlay or distort the original. Where a researcher presents large volumes of unedited voices in an attempt to present 'pure' stories, they may risk problems of ownership and also, importantly, fail to give the space for sufficient attention to their own motivations, position, views and interactions with participants (Merrill & West 2009).

A researcher can wittingly or unwittingly bias the data collected by avoiding certain relevant issues or by being from a different social background (e.g. class, culture, status) from the storyteller and so have quite distinct or different perceptions of the stories told. Further, the researcher might present him/herself, or interact, in such a way that the participant censors or omits large parts of their lives - presuming that the researcher would not understand or approve of these aspects of their stories.

The participant also has the potential to influence data in their recollection and representation of life events. Their motivations in

reconstructing certain stories and the extent to which they dramatise or downplay those events needs some scrutiny, although it may be difficult to determine the degree of any such distortion.

Reflexivity is now a defining feature of qualitative research (Finlay & Gough 2003). Qualitative researchers have accepted that they are central figures in the process and it is they who actively construct the collection, selection and interpretation of data. Notwithstanding their understanding and appreciation of partnership with participants in research endeavours (alluded to above), the researcher's presence in the work has the power to transform that research in subjective ways. The issue now is not so much whether there is a need for reflexivity rather than how it is best achieved (Finlay & Gough 2003). This study embraces reflexivity as 'hermeneutic reflection' as Finlay (2004) adroitly puts it, in an attempt to move beyond partial understandings and the investment in particular research outcomes.

In summary, the use of auto/biographical method allows a focus on how each life – past and current, as well as future aspirations and goals (Denzin 1989) – has, in this particular research, been affected by the experience of living with the chronic disability endowed by lymphoedema. By exploring these narratives the intention was to uncover realities and expose and challenge stereotypes (Kohler Reissman 1993). It was also intended that this exploration could be helpful to the research participants in that their views of the management of their condition would be revealed and debated (Denzin 1989). It was expected that critical self-awareness through reflexivity would lend credence to the outcomes (Finlay 2004).

Plan of Investigation

The aim of this cross sectional, qualitative research was to write the biographies of adults who have primary lymphoedema and in so doing, reveal how their lives had been affected by their experiences of living with such a condition. The overarching goal was to reveal the voices of the subjects, to listen to their stories recounted in their own words and to 'weave social context and individual life together' (Erben 1998:13) through narrative analysis.

Study Population

The study population is comprised of eight adults aged between 41 and 72 years with a diagnosis of primary lymphoedema and who are members of the Lymphoedema Support Network (LSN).

Assumptions

This research rests on the premise that individual participants would feel able to share their biographies subject to the expected conventions of ethics, confidentiality and collaboration (Morris 1997; Davis 2000).

Limitations

My role and presence as a researcher may have had an impact on the biographies offered by the participants of this study (Denzin 1989) and I made no attempt to conceal the fact of my background as an occupational therapist. My professional role may have located me within the medical model but my experience in dealing with people and listening to their life stories as part of a therapeutic milieu was, I argue, advantageous in the conduct of the interviews and may therefore strengthen the study. Because of the small sample size, the study may have limited generalisability but it might reasonably be expected to have potential for transferability to others in a similar situation (Erben 1998).

Ethics Permission

Ethics permission for the study was granted in October 2005 by the School of Education, University of Southampton (see Appendix 2). In addition, and because of the nature of the research population, a further, research proposal was submitted in October 2005 to the Ethics Committee of the School of Health Professions & Rehabilitation Sciences, University of Southampton, for full ethical approval before any data collection began (see Appendix 2). Full permission was granted in January 2006 and insurance confirmation was received from the University's Research Governance Office in February 2006 (see Appendix 3). Data collection could then begin.

Sample selection and recruitment

Eight, self-selecting, adults were involved in the study – three male and five female. These participants were recruited in two ways: firstly, through the quarterly newsletter of the Lymphoedema Support Network (LSN), which is a national self-care group set up by people with lymphoedema to offer support and counsel on matters pertaining to this chronic condition. The Chairwoman of the LSN was approached in writing beforehand to seek permission to access the network members. She agreed to this request (see Appendix 4) and suggested that a short article on the study might be a helpful means of informing the intended target group and this article was prepared and published in June 2006 (see Appendix 4). The LSN also has a web-site with information on current research into lymphoedema and details of the study were posted there (see www.lsn.org.uk/research).

Secondly, the Wessex sub-group of the British Lymphology Society (a national special interest society for health care professionals) offered to distribute copies of the article to those attending self-help groups and clinics in the region. The researcher is a long-standing

member of both the Lymphoedema Support Network and the British Lymphology Society.

By the end of August 2006 over one hundred people from across the country (and from overseas) had contacted the researcher to indicate their willingness to take part in the study. People continued to respond until February 2009. The first eight people who responded from the geographically local area were selected to take part. This was to ensure that no further selection process distorted the study. This was an unfunded study, so it was impractical to travel to other parts of the UK because of the costs involved.

Every person who volunteered to take part was written to, thanking them for their interest, and either invited to join the study or offered an explanation of the reason why they had not been selected (see Appendix 5). Several individuals responded for a second time to wish the researcher well with the study and to offer to take part in any future research. One woman, however, was very frustrated and angry at not being invited to join the study and I wrote to her again to explain the ethical and practical issues and to apologise for disappointing her by not being able to include her in the study.

The eight participants who had been invited to join the study were sent an information sheet describing the study along with a consent form (see Appendix 6). They were asked to complete this form and return it with their full contact details to indicate their continued willingness to be involved in the study. On receipt of the completed forms, I telephoned participants and made appointments to visit them between August and October 2006.

Apart from the diagnosis of lymphoedema, those involved in the research did not have any other common traits formalised as inclusion criteria. They are not, therefore, a homogenous group and have different social, economic and educational backgrounds.

Data collection procedures

Interviews were the primary means of data collection to assist in the development of each person's biography. The interviews were not scripted because I was actively seeking the participants' stories; it was felt that any list of topics or prompts would contravene the spirit of the research. The interview therefore took the form of a conversation about the individual's life and specifically their experiences of living with lymphoedema. They will be referred to as both interviews and conversations in the following account.

Conversations were long enough to enable the participant to feel that their story had been fully captured. This took some time, as each participant required enough time to tell, dwell on and then recast his or her story. No specific time limit was placed on each interview - although I had predicted that each one would last between one and two hours. In fact, most lasted two hours and some a little longer.

I anticipated that my professional background as an occupational therapist could prove helpful in conducting the interviews and encouraging the participants to tell their stories. This is because occupational therapists habitually construct their interventions with patients/clients around their personal stories and activities. In most cases this proved to be so: a rapport was quickly established and it was not difficult to encourage the participants to unfold their narratives. Nonetheless, I recognised the possibility that my professional background, rooted as it is in the medical model of

health delivery, may have been a barrier to the freedom of some of the participants to comment on certain aspects of their life (Grbich 1999).

Appointments were made at a time and in a location that was convenient for each person. For six of the eight interviewees, this meant visiting the individuals in their homes. For the other two, the interviews were conducted in their place of work.

The conversations started with a brief review of the consent form and a short, social, exchange to set the participants and myself at ease. The focus of each interview (i.e. that this was to be the participants' own accounts of their lives) was also clarified at the outset of each interview. Further, it was explained that each person would have an opportunity to clarify issues and confirm stories told in the interviews by reviewing and editing a transcript.

Each conversation was audio-recorded (after consent was granted) and transcribed as soon as possible after the completion of the interview. In reality, this took several months due to the pressures of full-time work and because of the time it actually takes to transcribe such long conversations. As each transcript was prepared, it was sent to the participant for them to 'clean up' the text and to allow them to judge whether their story had been expressed clearly. Finlay (2004) suggests that this cleaning up process is essential to allow the participants the opportunity to reflect on their story, as told so far, and to reconsider its meanings. Once the participants had sent their amendments, the revised, final, transcripts were prepared and once again, they were sent to the participants.

Although the intention was that the final biographies would represent a genuine partnership between the researcher and the participants, I acknowledge that the final decision on the presentation of the text of the thesis following analysis lay with myself as researcher. This is because in order to complete this thesis the researcher needs an authoritative voice and should be able to make final decisions about the presentation of the work.

Brief field notes, made after the interview, and comprising impressions on such things as (for instance) non-verbal behaviours, emotions and use of humour, provided another dimension to the analysis of the data gathered (Finlay 2004). I also made notes on my own reactions to the conversations and the impressions received and reactions felt. The intention was to aim for reflexivity (Finlay & Gough 2003), as discussed further in the data analysis section that follows, and to assist in determining the researcher's role in the research process.

Data analysis

Glaser & Strauss, writing in the 1960s, sought to elevate qualitative approaches above the criticisms of positivist researchers by describing ways in which data could be analysed using the same reproducible rigour that would be applied in quantitative methodologies. They introduced the notion of grounded theory which emphasises the conceptualisation of data and generates abstract data grounded in the data (Glaser & Strauss 1967). The inherent difficulty, however, with grounded theory is that it requires the fragmentation of data and a subsequent decontextualisation (Coffey & Atkinson 1992). Although grounded theory writers are aware of these problems and have suggested ways of recombining and recontextualising data (Miles & Huberman 1994) there remain some tensions between attempting to be subjective and interpretive

at the same time as being scientific. This inevitably led to the development of other, more holistic, approaches - notably the narrative and life-story approaches to which I turned (Punch 2005).

Narrative analysis and biographical interpretation seek to deal with data in a much more holistic way: There is a storied quality in much qualitative data, and thinking about stories in the data enables a creative and potentially empowering approach to data analysis (Punch 2005). This holistic approach has the potential to give voice to the participants/narrators. Narrative analysis seeks to understand the actions of an individual through hermeneutic processes. Hermeneutics being the theory or science of interpretation as applied to the analysis of cultural life (Riceour 1986). Narrative analysis approaches were therefore referred to in analysing the data gathered in this study using the phases of analysis suggested by Fraser (2004) which itself builds on the work of Catherine Kohler-Reissman (1993). Formal narrative analysis requires a focus on identifying the structural features of narrative and their arrangement and the use of language and symbols are considered important. As a novice researcher, I found this challenging and was more focused on the function of the stories, their underlying meanings and messages, but I also considered the social and cultural context in which the stories were told. Murray (2000), writing specifically about the challenges of narrative analysis in health research, describes the layers of narrative analysis as personal, where narratives are portrayed as expressions of the lived experience of the narrator; interpersonal, where the narrative is co-created in dialogue. He also refers to positional analysis, which considers the differences in social position between the narrator and listener; and finally to societal analysis, which is concerned with the socially shared stories that are characteristic of certain communities or societies. All of this is important in the

analysis of this study and is referred to in the section on findings and discussion.

There are echoes of this approach in Erben's advice on auto/biographical analysis (1998). He outlines stages that the auto/biographical researcher must attend to for the analysis of life stories, and suggests a schema that might prove helpful in biographical analysis.

Figure 3: Stages in Auto/biographical Research

- specific events – which provided the initial incentive for the research aims
- local context of specific events – significant people, places, experiences and emotions
- societal context – relating to the characteristics of the wider society
- documentary sources – personal and public accounts, both oral and written

(Erben 1998)

Figure 4: Schema for the Analysis of Auto/biographical Data

- cultural system – the personal value-sets that shape the stages above
- chronology – the important and public events that shape them
- rehearsals – refer to the process of reconsidering and refining of all the assembled data

(Erben 1998)

Erben (1998) postulates that differing parts of the schema will receive different emphasis, but that all will need to be addressed in some measure. These frameworks were also referred to in conducting the data analysis.

The biographies created were unique to the individuals concerned and although there may be some similarities, the intention was to present divergent stories - not to present convergent issues to reinforce any hypothesis. That is because the purpose of the study

was not to prove or disprove a hypothesis in order to generalise from that to a wider population, rather my intention was, as stated, to give participants the opportunity to present their stories about their lymphoedema and for those stories to be explored in depth.

Ethical Considerations

The responsibility of the writer to represent the story of another person's life in an ethical and authentic manner is fundamentally important, since the power the writer wields is immense (Denzin & Lincoln 2005). An auto/biographical study such as this must be handled with great responsibility because, essentially, the research will be making private-lives public (Denzin 1989). Writing about another person's life is a complex endeavour and will eventually become a representation of the life in question. It turns the subject (and the self) into an object that is quite concrete. The account can be physically held and read about and will become "a commoditised extension and representation of the life in question" (Denzin 1989:30).

Such research renders participants vulnerable in that the research is potentially intrusive and may create uncertainty and dissonance as participants reflect on their lives and reinterpret their stories. Narrating their memories and stories about their lymphoedema may also bring about catharsis – and this can be both positive and negative. As Tillman-Rogers points out (1995:119): "The researcher, simply by being there, causes a form of 'knowing' an event differently (reflection). Many people survive, or indeed endure, by deliberately not being aware of all the complexities and dangers…" In recognising this, and if this research had precipitated issues in this way, there was a predetermined plan to deal with the aftermath, which involved an empathic approach and referring the participants to appropriate advice/counselling services if necessary.

The researcher-participant relationship and the importance of establishing an ethical boundary to the work were also important ethical considerations. My aim was that the participants should feel fairly treated and not exploited or unduly misrepresented in the researched account. The telling or exposing of a life renders the subject vulnerable and the researcher has a duty of care to those individuals to ensure that they are treated fairly and with respect. Given these concerns, I took care to adhere to the code of ethics for researchers adopted by the British Sociological Society (2003) that draws attention to issues such as the reduction of harm to a subject; to protecting the interests, sensitivities and privacy of a subject; and to the issue of power imbalance between the researched and the researcher.

Finally, confidentiality is a major issue. In a biographical study such as this it was not possible to guarantee complete confidentiality for participants, although, obviously, there was an attempt to disguise the identity of participants and provide anonymity by using initials instead of names on the transcripts and by not referring to the location in which they lived or worked. This issue was fully discussed with participants prior to them giving consent to participate. In the discussion section of this thesis, pseudonyms (rather than initials) are used to enhance the flow of the text.

Participation in the research was entirely voluntary and there was no coercion to become involved. The participants were not asked to join the study directly since there is an implied pressure to do so when this occurs (Punch 2005). As explained earlier, the participants self-selected by responding to an article in a newsletter or through discussion with a third party, *viz.* their lymphoedema specialist practitioner. All who agreed to be involved were advised

that they could withdraw at any time and without giving any reason. They were also advised that they could change what they had said at any point until submission of the research thesis.

Summary

This research study used an auto/biographical methodology to give voice to adults who have primary lymphoedema. The participants were encouraged to describe their life experiences using their own words and to ascribe their own personal meanings to those experiences. The research aimed to provide a counterpoint to common assumptions about people with chronic disability by constructing an alternative account based on the viewpoint of those with primary lymphoedema.

Chapter 4
Profiles of Participants

Introduction

This chapter provides a short profile of each of the eight participants and a summary of the narrative provided by them. It also includes a brief personal reflection on how the interview went and the impressions gained. This is a prelude for an in-depth exploration of the findings and a detailed examination of the extensive impact that lymphoedema has had on their individual lives in the next chapter. To protect the anonymity of participants, pseudonyms have been used throughout this text.

Susan

Susan is a 67 year old woman who has bi-lateral lower limb lymphoedema with pronounced swelling and repeated, severe cellulitis. She was the only child of elderly parents and was educated privately after failing her 11+ examination. She attributes this early educational disappointment to her ongoing ill-health during her primary school years but felt that the private education enabled her to catch up academically with her peer group. Susan had childhood experience of radiotherapy when she was suffering from fluid drainage problems which affected her hearing and this sparked her interest in becoming a Diagnostic Radiographer. Susan received a diagnosis of Miege's type lymphoedema at 35 years of age, although she had experienced full-blown symptoms for at least six years before that. Further, she believes that she may have had lymphoedema since childhood but no other members of her family had ever exhibited symptoms or been known to have lymphoedema and the possibility was never mentioned by the various doctors who dealt with her health problems. Susan is single with no children and

has lived independently from her parents for much of her adult life until a particularly serious bout of cellulitis (lasting for more than six months) resulted in her moving back to her parental home so that her mother could look after her. She continued to have gross swelling and repeated bouts of cellulitis which impacted on her ability to do her job and, despite adjustments to her role, she had to take early retirement (at 57 years of age) from her NHS post. This experience of early retirement on medical grounds was clearly very important to Susan, as she had to deal with an occupational health physician who knew nothing of lymphoedema and acted in a derisory and insulting manner before he would finally acknowledge the chronic nature of her condition and its potential for life-threatening infections. During one of her hospitalisations for this problem, her situation was exacerbated by contracting MRSA. It is Susan's belief that the NHS Trust for which she worked finally intervened with the occupational health physician to allow her to take early retirement in order to prevent her from taking legal action over the cause of her MRSA. Susan has quite severe mobility problems which create difficulty in moving about in an external environment and she also continues to experience frequent episodes of cellulitis. Her parents have both died in the last decade and she now lives alone in their adapted bungalow. She does not have a busy social life as she cannot cope with her lymphoedema well outside her home, but Susan is a regular churchgoer.

The interview was very cordial although we quickly slipped into 'patient/professional' roles. Although Susan told her whole story, in the main it was devoid of emotion or great detail except when she spoke of her struggle to take early retirement. I was left with a sense of frustration that it was not possible, within the time constraints of this study, to explore the emotional dimensions of Susan's life in follow-up conversations. I certainly could not expect

her to disclose personal thoughts and feelings on such short acquaintance but I formed a residual sense that Susan's life had not been fully grasped and lived: Susan was still living in her parents' house surrounded by their possessions and there was no mention of any relationships – or even close friendships. My judgement in this regard reflects my own sense of the importance of family and friends but may, of course, have little relevance for Susan. Susan said that her single regret was that her father did not live long enough to see her win the victory in her battle with the occupational health physician over her early retirement. This was a somewhat unexpected expression of regret, since people often use this expression with reference to something much more intensely personal. This work-related narrative, however, might have been an allegory for her battle with lymphoedema.

Belinda

Belinda is a 69 year old woman with severe and extensive lymphoedema in both legs and arms. She also suffers from trunk and facial oedema. Belinda first noticed the signs of lymphoedema when she was 10 years old and although she did have investigations she was not diagnosed with Miege's type lymphoedema until she was in her twenties and pregnant with her first child. Belinda has a family history of lymphoedema – although it was not recognised as such – with her grandfather and several aunts exhibiting undiagnosed symptoms. The most recent 'victim' (Belinda's word) to the condition is an aunt of Belinda's who committed suicide a year ago because she could not cope with her condition any longer. Belinda has had repeated, radical de-bulking surgical procedures including non-specialist liposuction and has also been encouraged to use a pneumatic compression pump extensively. These interventions excite controversy in the field and are largely considered damaging, if not administered with extreme caution, in the current literature (see chapter 2). Belinda's swelling has increased over the past 40 years and she has experienced repeated and lengthy periods of hospitalisation for cellulitis. This picture has been further complicated by hospital acquired infections (MRSA) and by other non-related conditions such as diabetes. Indeed, Belinda took several months to confirm that she was happy with the transcript of this interview because she was hospitalised soon after our conversation, due to a bout of cellulitis, for some five months. Belinda lives with her husband, who is her devoted carer, in an adapted house of which she is obviously proud. She was very happy to show me around and demonstrate all her aids and equipment. Her gross swelling has restricted her mobility to a great extent and she walks a few yards only with the assistance of a walking frame. For external journeys, her husband drives her in a car that has been adapted to take her wheelchair.

Belinda talked of her physical self in a somewhat deprecating manner and seemed to have low self esteem. She recounted story after story about her various medical crises and interventions for her lymphoedema, and I had a sense of unease that she was relishing the attention she received because of her condition in a rather unhealthy way. I speculated that these two things were linked in some way. Belinda has had a multitude of interventions for her lymphoedema, paid for by her husband's private health insurance, but most were inappropriate and delivered by non-specialists in the lymphoedema field. Again, I was left with a sense of unease that she had been exploited in some way by being subject to these privately paid for interventions which were not remotely best practice as defined by the Lymphoedema Framework Project (Medical Education Partnership 2006). Belinda was complicit to some degree in that she begged a young medic, who was expressing some disquiet about a procedure that he was about to embark on, to continue with the procedure. What was interesting is that Belinda's husband, who joined us for lunch, was apparently very well read on the topic and a frequent visitor to the LSN website. It was difficult to see how the couple could not have been aware of some of the contraindications of the treatments they were receiving. I did not feel able to demonstrate my dismay at the treatment Belinda had received for fear of upsetting her and her husband and undermining her medical care, but left my meeting with them with substantial concern for their situation. Subsequent to my visit, I sent them literature which outlined both best practice and outlined the reasons why some treatments were considered controversial.

Gerry

Gerry is a 44 year old schoolteacher who has bilateral lymphoedema of the lower limbs but with most of his swelling and cellulitis confined to his left leg. The youngest of three children, he suspects there may have been a familial tendency for lymphoedema since old photographs reveal that his fraternal grandmother had one leg that was noticeably larger than the other. None of his living relatives have any manifestation of the condition and Gerry has deliberately chosen to remain childless in view of the risk of passing on the condition to subsequent generations. Privately educated, Gerry was destined to follow family tradition and enter the military academy at Sandhurst for officer training. But at the selection stage he changed his mind and followed his early and passionate interest in the theatre where he rose quickly through performance to become a very youthful (19 year old) stage manager. Always athletic and interested in both dance and mime, Gerry was devastated when the onset of his lymphoedema threatened his burgeoning career. Gerry experienced his first episode of lower limb swelling when he was just 18 years old whilst taking 'A' levels and he had abdominal disturbance – possibly truncal lymphoedema – when he was just 22 years. He was also constantly unwell during this time with low-grade fevers and frequent infections. It was following subsequent investigation of these symptoms of swelling, and his history of ill-health in one so young, that he received a diagnosis. Gerry is convinced that the attempt to diagnose his condition through the use of a lymphangiogram actually destroyed the lymph system in his left leg and made his situation much more serious. The subsequent 'loss' of his patient notes is interpreted by Gerry as a cynical attempt to cover up medical incompetence and he still feels very raw about this. He described a feeling of foreboding before the lymphangiogram procedure and his battle to resist having both legs assessed at the same time. His subsequent

struggle to forgive the surgeon who carried out the procedure is framed by his strong Christian ethic but, despite his protestations that he has now forgiven, I was left with a sense of his residual anger and bitterness about how much was stolen from him. He lost his first career choice along with his sense of athletic, aesthetic self; he lost valuable relationships and self esteem; he even lost the chance to father children. In short, he lost his future narrative, the story did not unfold as he expected. Gerry's description of his struggle to overcome depression and despair is particularly poignant and he has a powerful message about the need to mourn and to be supported in that need. Once again, he has not received regular, active treatment and his swelling has gradually increased over the years. His fear of the recurrent cellulitis is palpable and he is outspoken about his resentment at both the scarcity and costs of treatment. Gerry feels angry that the medical profession know so little about the condition and yet can be so scathing about its impact. He fears for the future and worries that his need to work long hours may further compromise his health. Despite all this, Gerry has a job which gives him great satisfaction and a religious faith that brings him great comfort. His wife and stepson provide a loving family environment and he believes that he has much to be grateful for.

The interview itself was interesting in that Gerry was very conscious of not being alone with me in his house because he was concerned that neither of us should be accused of any inappropriate behaviour. I have no idea why he should have been particularly sensitive on this issue. We had therefore made arrangements for his wife to be present during the time I was in his home. In the event, his wife was held up and his stepson let me in to the house and then disappeared. Despite his earlier anxieties about being alone with me, Gerry was very relaxed and talking fluently about his early

relationships until his wife arrived and he became much more reticent and guarded in what he was saying. This reminded me of just how fickle interviewing is as a means of extracting narratives – especially personal narratives - and how these can change subtly (or even radically) when the audience is different.

Another interesting feature of the conversation I had with Gerry was the way in which the verbal and non-verbal aspects of his storytelling were often in direct contrast: Gerry was sometimes talking of Christian forgiveness when his body language revealed anger and frustration. For example, when he talked of having a dialogue with God and coming to a point when he could forgive a medic whom he felt had mismanaged his condition, Gerry was holding his hands in a fist and beating them against each other. The anger was clearly not yet put away completely - although of course the retelling of the story may have re-opened old wounds.

Philip

Philip is a 72 year old retired businessman who has had bilateral lower limb lymphoedema since he was 33 years of age. He lives in an adapted bungalow with his wife who has also experienced some ill-health. His story is characterised by a lack of diagnosis for an inordinate number of years and by concomitant mismanagement. Philip first noticed the unusual swelling in his left leg when he was still a single man and it took 31 years and 10 episodes of cellulitis (some requiring hospitalisation for weeks) before he received an accurate diagnosis and a referral to a lymphoedema management service. During those intervening 31 years, it had been assumed that Philip was suffering from a cardio-vascular problem which caused the swelling and from a separate, unconnected, immunological problem which was responsible for his recurrent skin infections. The fact that the leg swelling was unilateral at first, and that no reason for congestive heart failure (a common cause of oedema in older people) in one so young had been established, did not deter the medical profession from its decision to pursue treatment for cardiac and immunological problems which did not, in fact, exist. Philip was prescribed and took diuretics for that entire period. These would have been helpful for cardiac-related oedema but were not helpful at all for lymphoedema. It was also suggested that he took prophylactic antibiotics over the long term but he resisted this treatment. Only one junior doctor, during the course of a routine, private, medical check-up, had suggested that this constellation of symptoms could be lymphoedema – but this was immediately, and arrogantly, dismissed by a senior medic without further consideration. The attitude of medical staff was a source of concern to Philip who felt marginalised by their sometimes off-hand, sometimes arrogant, treatment. Eventually, it was his own doggedness which led to a diagnosis when he challenged his GP having discovered through the internet information about

lymphoedema. Philip was disappointed that the trail of mismanagement and misdiagnosis was covered when the NHS lost his notes. He feels certain that this was not an accident but an attempt to prevent any action over mistakes made. Even after his correct diagnosis was made it took several more years for Philip to be referred to specialist services. During the intervening period, he was advised to wear inappropriate, elastic hosiery to control the swelling and to use a compression pump. Both of these can have a deleterious effect on the lymphoedema (see chapter 2). Philip's overarching feeling is one of sadness at the length of time it took for the medical profession to recognise and treat his condition and a fervent hope that other people can be spared this by better education and training for healthcare professionals and greater access to information for those with primary lymphoedema.

The interview with Philip took place over two occasions as on the first meeting, following an interruption, the tape recorder did not record. Even the second meeting was dogged with bad luck as I had travel difficulties which made me late. Philip and his wife were very patient and gracious but I felt that I should record his story as quickly as possible in order not to waste any more of their time. Philip told his diagnosis story slightly differently on each of my visits: The first time he was very humorous in the telling, almost brushing it aside; but on the second telling, he seemed much more raw and angry about his situation. It may have been that talking of his difficulty in securing a diagnosis to me had made him re-consider issues which he had kept submerged and made him acknowledge his distress. Or, of course, it could have been that he was in a different mood on my second visit. Philip's story was almost exclusively about his condition and the diagnosis narrative and it would perhaps have been more illuminating if we had had time to explore more personal issues and the emotional and social

impact the lymphoedema had had on his life history and future plans. It seemed to me, though, that his gentle good humour masked a great deal of anger and frustration at the experiences created by his lymphoedema.

Louise

Louise is a 41 year old woman who has had unilateral lower limb lymphoedema in her left leg for the last 13 years. She has no history of lymphoedema in her family and so the onset of her symptoms in 1993/4 during her second pregnancy was a complete surprise. At first, when her left leg became massively swollen, it was assumed that she was suffering from a Deep Vein Thrombosis (DVT) as a complication of pregnancy. She was put on bed-rest and had many venous injections to discover the whereabouts of the clot. She was also given Warfarin and Heparin, two drugs used in the treatment of DVT, for 8 months. The presence of a clot was never firmly established but after 8 months the swelling had resolved and the leg was back to normal. Louise was an athletic person, being a keen runner who took part in road races. She noticed over the next two years that she would sometimes get swelling after running but it resolved itself fairly quickly and she ignored it. In 1996, after a 10km competition, her left leg blew up again massively from toes to groin and she was admitted to hospital. No firm diagnosis was made at that point either and it took another two years of intermittent and gradually increasing swelling until a lymphoscintography revealed the hypoplasia (lack of lymphatic vessels) and the diagnosis could be made.

Louise' story was distinctive for three reasons. Firstly, she was diagnosed relatively quickly by comparison to others (four years after the swelling first occurred) and then referred fairly immediately for correct treatment and is managed for compression garments by a lymphoedema specialist. Although she has to pay for her Manual Lymphatic Drainage treatment herself, having had it paid for only once through her PCT, she finds it very beneficial: The reductions in her swelling following each episodic treatment is marked and backed up by measurement evidence. Louise maintains

her reductions in swelling by constant wearing of her compression garments. She describes the pain she experiences as a dragging pain but explained that the pain resolves following massage therapy. Louise is the only person whom I have interviewed who has had regular treatment and she has had no episodes of cellulitis. This could be coincidence but is noteworthy all the same. Secondly, the relationship with her 12 year old daughter is complex as this child believes it to be her fault that her mother suffers this intractable problem. She believes this because the swelling was originally defined as pregnancy related and first occurred when she was in her mother's womb. It has placed a strain on their relationship at times. This is an interesting reversal of the guilt often expressed by those with primary lymphoedema who believe that they might be responsible of passing it onto their own children. Louise did not mention the hereditary aspect of the condition and I sensed that this was not a conversation that she (or I) would be happy to embark on during this one-off interview. Finally, perhaps because of her age and gender, Louise was more focused on body image issues than other participants. She was grateful for a very supportive husband who understood her condition and did not make any issue of it, but was upset by the cruel comments that friends and acquaintances sometimes make. One friend in particular, whom she considered had his own issues about body image because of obesity, was very scathing about her wearing compression garments and made her feel humiliated in his public comments. She does very occasionally remove her garment (perhaps for a special occasion or on the beach) but is strong enough to wear them continuously otherwise despite their unflattering look. Again, it is notable that this level of adherence to treatment is rare with long-term chronic conditions but in Louise's case it has brought obvious benefit.

Louise is an education administrator who elected to meet me at her place of work. This gave an interesting and different tone to the conversation which was much more like a work-related interview than most of the other meetings which took place in people's homes. I had a sense in which Louise was more comfortable on this neutral territory than she would have been at her home. Although she did not say so, I felt that to take up too much time would have been inappropriate and this framed the conversation we had. Louise was rather reserved and I had to initiate disclosures about life-stories by prompting her about life events. That being said, it was very easy to forge a rapport with Louise: She and I have a lot in common – both busy working parents with daughters the same age and from a close geographic and social background. It almost felt that we would have schools, friends and acquaintances in common if we allowed ourselves to fall into 'social' conversation and I have wondered whether perhaps Louise and I were engaged in a defence to prevent probing too much lest we inadvertently revealed our informal selves.

Angela

Angela is 72 years old and has had bilateral lower limb lymphoedema since she was 14 years old. Angela is a retired piano teacher and has three children, one of whom has mild cerebral palsy following a difficult childbirth. Angela grew up in idyllic rural circumstances during the Second World War. She had a quiet, peaceful life on a farm and learned to play the piano to a high standard from a very young age. She wonders whether her sedentary childhood contributed to the early development of her lymphoedema which has always been quite severe. Her father appears also to have had lymphoedema but it affected him mainly in old age as he had a very active life as a farmer and this held the symptoms at bay. Angela went to London to train as a concert pianist and then became a music teacher. She describes her bohemian lifestyle where music was more important than fashion and relationships, but where her inability to either sit or stand for long periods (because of her pronounced swelling) impeded her ambitions in the music world. She felt that she 'compensated' (Angela's word) in her life for what she couldn't have and closed off from things that were difficult for her. Becoming a music teacher instead of a concert pianist and forging a relationship with a man – later to become her husband – who also had disabilities were the examples she gave. Nonetheless, she revealed that her years as a mother were happy and fulfilling and a period when her lymphoedema mattered little. When she tried to re-enter the world of teaching in her early forties she found she could not cope physically and she resorted to playing the piano as a volunteer and then taking private lessons. Recently, Angela has been unable even to attend concerts as sitting for lengthy periods increases her swelling and pain. She had still had no formal diagnosis although she had tried to interest her GP in her condition having discovered it herself. The GP was dismissive and just advised that she put her

feet up and take diuretics. Angela felt that women are marginalised by the medical profession and that she was probably too passive and undemanding. She paid for manual lymphatic drainage (MLD) herself but, although she found it helped greatly with the pain associated with lymphoedema she could not afford to continue to pay for the treatment privately and it was not available through the NHS. Angela was finally referred to a lymphoedema clinic when she was 71 years old (57 years after her first symptoms appeared) and this was because of an extreme bout of cellulitis and swelling and because an MLD therapist whom she had consulted wrote to her GP suggesting she be referred to a specific clinic. She now has compression stockings – although she finds them very hard to put on – and is monitored every six months. She has no active treatment, bandaging or MLD, but finds the LSN literature informative and helpful. At the end of this conversation, Angela expressed her anger at the fact that she had not been adequately managed for so long and her belief that a male dominated medical profession saw this condition as a minor thing which wouldn't kill you and one that they perceived as a 'women's issue'. She re-iterated her opinion that women, especially older women, were not pro-active in having their illnesses attended to in the same way that other groups were.

Reflecting on our conversation, I felt that Angela had decided to settle for second best with regard to many important life issues and that her early onset lymphoedema had framed and shaped her life in ways that did not always content her. Angela was a very easy person to have a conversation with and needed little encouragement to share her life stories. She had an almost respectful attitude to my research and therapy background (which she enquired about) and I was surprised at the sense of power which this engendered and I felt both positive and negative about it.

I have always been very sceptical and negative about hierarchical approaches in the health care world but surprised myself at being quite buoyed by her deference to my knowledge. In retrospect, of course, this is unsettling especially in the light of her negative comments about women's experiences with medical doctors and I now wonder whether I was inadvertently reinforcing a sense that she was not the 'expert' in her own condition.

I am saddened to record here that Angela died some two months after our meeting following hospitalisation for cellulitis. Her husband approved the transcript of our discussion and gave permission for its continued use in this study.

Martyn

Martyn is a 56 year old man who has unilateral lymphoedema affecting his left leg - which is considerably swollen by comparison to his right leg and with his very slight frame. He first experienced the swelling when he was 39 years of age and was immediately referred for an opinion since his GP did not know what the possible cause could be. After initial misdiagnosis, Martyn received a diagnosis within six months. Despite this he was told that there was no treatment apart from 'general measures'. He does now have compression therapy (albeit not especially well fitting and only a half leg sock) but no active treatment has ever been offered to him. He did have advice from a lymphoedema specialist nurse in a cancer unit but felt uncomfortable about using a resource intended for people with cancer. Martyn has had two serious bouts of cellulitis requiring hospitalisation for several weeks, and has fibrosis along with papillomatosis-type changes in his skin (see appendix 1). Married with two children, Martyn tries not to allow the lymphoedema to impact too greatly on his life, although he is occasionally self conscious of his limb's appearance, and is generally fit and healthy. On first diagnosis he was very upset by the incurable nature of the condition but feels that he has come to terms with the psychological impact after 16 years of coping.

Martyn and I met at his place of work and I feel that this influenced the conversation by giving it a work-related feel. Martyn was quite a formal person and, although very pleasant, it would not, I sensed, have been appropriate to broach more sensitive issues during our fairly brief conversation. He engendered in me a sense that I needed to project a 'professional' image and I found myself offering healthcare advice which in retrospect was tangential to my ambition to have his voice heard and may have further caused him to be reticent. I left the meeting with a sense that, although he was an

active member of the LSN, he did not seem to fully appreciate how beneficial active treatment could be. On my advice, he stated that he intended to seek out a referral to an excellent specialist clinic close to his home - but I had an impression that he might have been humouring me to some extent. He seemed, if not complacent, then certainly unaware of the future risks of half-hearted management.

Marie

Marie is a 58 year old teacher and lecturer who has bilateral lower limb lymphoedema. She grew up in comfortable surroundings and attended teacher training college in Leicester where she met her husband who was studying sports science. Marie is a very vibrant person and describes herself in those early years as an '8.5 stone, perfect princess'. She married very young and was pregnant at 22 years of age. It was during this first pregnancy that her symptoms began to manifest. Her lower legs and feet became swollen to an extreme and she gained four stones in weight. Although the swelling reduced between her two pregnancies, Marie never regained her original body shape, remaining slim above the waist but with obvious swelling in her lower limbs. Her husband began to be very negative about her physical appearance and this had a very debilitating and long-lasting effect on Marie's self esteem. She describes the twenty seven years of their marriage as unhappy and abusive, with her husband constantly humiliating her and telling her that she was unattractive – even to the extent of not wanting to attend social gatherings with her. He provided no support to her in her struggle with lymphoedema. Marie received an accurate diagnosis after fifteen years but had a lymphangiogram as part of the diagnostic testing which is likely to have created further harm in her already compromised lymph system. Marie was never offered any advice or treatment being simply told that there was nothing that could be done and that she should not stand on her feet all day. As a mother and a school-teacher this was advice that she found difficult to follow. She was given diuretics which she took for 27 years. Because her husband was a pilot with the military, she lived in many different locations and did not have a network of support from friends and family. Marie has had several episodes of severe cellulitis with swelling that has caused her skin to, literally, burst and peel off, but during a particularly bad bout of cellulitis,

associated with other ill-health, Marie's two children had to be sent away while she was hospitalised for over a year: Her daughter to boarding school, even though she was very young, and her son to stay with his uncle (Marie's brother) whom he hardly knew at all. Marie's husband was away on active service at the time. The sadness and pain of this separation has remained with Marie ever since, although she seems to have a good relationship with her children. One of her fears is that her grandchildren will inherit the gene for lymphoedema because she now knows that her form of primary lymphoedema is hereditary. Marie and her husband finally divorced after 27 years, and she has subsequently gone on to marry for a second time. Her second husband is much more supportive and it has been with his encouragement and assistance (and private health insurance) that Marie has sought out specialist help from an expert in the field and finally discovered that treatment is available 30 years after her symptoms first appeared.

The conversation with Marie was very relaxed and she did find it quite easy to tell her stories. I did not need to prompt her as she was very talkative and outgoing and to 'get a word in' would have made me feel over-assertive. I was content to simply listen and let her take the story where she chose. After the interview, Marie wrote to me to tell me that she had discussed things with me that she had never spoken of before and that she found telling her life story cathartic. She has promised herself to be more rigorous in managing her lymphoedema in order to keep as well as possible.

Chapter 5
Findings and Discussion

Introduction

The aim of this research was to write the biographies of adults who have primary lymphoedema with the specific objective of demonstrating how their lives had been affected by the experiences they have had. The overarching goal was, as previously stated, to reveal the voices of the participants, to listen to their stories recounted in their own words and to "weave social context and individual life together through narrative analysis" (Erben 1998:13). In this chapter, I explore their biographies and examine what my participants had to say about their lives.

It might be considered ideal to analyse each person's story discretely and separately since the aim of this research was to highlight the individual voices of participants and not to produce a homogenised view of their lives just because they have a similar medical diagnosis. However, due to the word restriction of the thesis, themes which arose from one or more participant were followed in the discussion and the topics raised in the biographies have been used as a framework for the structure of this chapter. This yielded both rich individual perspectives but also some transferable themes which have the potential to resonate with others with primary lymphoedema and those who live and work with them.

Participants in research about living with disability have commented in the past that much health-related research is paternalistic in that it purports to act on behalf of people with disability and claims to promote their best interest, but without ever seeking their views or

asking their permission (Kitchin 2000). Indeed, people with chronic illness or disability are not always afforded an opportunity to speak for themselves. This research has actively sought to avoid this and in order to allow the voices of the participants to be heard throughout this work, the text is illustrated with quotes from their biographical stories as suggested by Fraser (2004) Murray (2000) and Hatch & Wisniewski (1995).

The process of data analysis effectively began with the start of the data collection in my first interview and there is no doubt that each subsequent conversation was shaped by those that had preceded it. I looked for significant events or epiphanies (Denzin 1989) and began to identify recurring experiences that seemed to be important for the whole group of participants. I was aware that I was filtering the data through my own lens of experience and using my own imagination in interpreting it, but as Erben (1998:9-10) observes:

> …imagination very often fills the gaps within, and develops architecture for, the research data.

As Mason (2002) has put it, a transcript is an incomplete record of the interview and data presentation and analysis seeks to convey what the interviewer *interprets* as the interviewee's *intended* meanings. I believe that I have made reasonable inferences, being mindful both that I have been influential in shaping the data and that those participants may have told me what they thought I wanted to hear.

The study has revealed the extensive impact that primary lymphoedema has had on individual lives. Even though each person's story provided a uniquely individual view of their life, there were, in places, certain common themes and sub-themes that emerged in their narratives and I have given weight to these issues

in the following discussion. However, I have tried not to ignore the singular or minority responses as these are also central to the research focus. Frequency of assertion may not always correlate to importance and meaningful insights can emerge from small amounts of data (Denzin & Lincoln 2004).

Emergent themes identified included:

Difficulty in accessing healthcare support and services - particularly securing an accurate diagnosis and effective treatment, finding information about their condition and the need to be pro-active in seeking out information, treatment and support. Financial issues related to the requirement and ability to pay for services also created distress;

Inadequate levels of professional education and knowledge about lymphoedema and negative attitudes of medics or other healthcare professionals. It was even the case that their condition was worsened by treatment or assessment offered;

Psychological concerns included low self-esteem and body image concerns, with distress, stress or depression being evident among the group;

Relationship issues with partners, spouses, children and friends coupled with a fear of passing on the condition to children or grandchildren;

Loss of career and leisure opportunities and, for some, their condition was worsened by their working environment;

The challenges of daily self-management, including coping with the pain and discomfort of the condition and complying with daily routines. There were fears for the future as their condition worsens and they are less able to cope and particularly about the life-threatening nature of cellulitis and the time & costs of treating it.

These issues are the focus of the discussion that follows and I now turn to each theme to explore the data fully.

Difficulty in accessing healthcare support and services

The extent of difficulty that individuals have had in accessing services was truly shocking. The average length of time it took to find an accurate diagnosis was 17 years with one participant, Philip, waiting 31 years before lymphoedema was confirmed as the cause of his ongoing problems of lower-limb swelling and recurrent, life-threatening, cellulitis (see Figure 5, at the end of this chapter). This reflects much recent research which reports the potential for underestimating the prevalence of this condition because of under-reporting or misdiagnosis (Woods 2007, Twycross *et al.* 2000, Mortimer *et al.* 1997).

Many participants had been wrongly prescribed diuretics or antibiotics for decades before the correct diagnosis was made. Philip recalled his first consultation after he had noted his swelling as a young man:

I remember the vascular specialist saying in 1970 "I am sure that somewhere in the world there is somebody who understands swollen ankles- it might mean cutting off a bit here for testing and a bit off there – but let's just put you on a diuretic". I stayed on diuretics for 31 years. In the early to mid 80s, I got so fed up with symptomatic treatment… for

cellulitis, because they didn't associate cellulitis with lymphoedema. Please stop treating the symptoms – what's causing them? Lymphoedema wasn't diagnosed until 2-3 years ago - 31 years after I was put on diuretics.

Belinda had a similar experience of being prescribed diuretics after misdiagnosis:

I was about 10 years old and doing a lot of athletics, high jump and running – I used to run for the school – and they kept saying "you must've strained your ankle" because my ankle was always swollen. They sent me to an osteopath who bandaged it. And it was fine all the time it was bandaged but when he took the bandages off the swelling came back. I started to play tennis afterward but the swelling gradually got worse and it just wasn't practical. Once I left school it started in the other leg and after we got married in 1959 and I had a baby, I spent most of that time in hospital as I was swollen in both legs and at the base of my back. …They just called it oedema and that my kidneys had been damaged during pregnancy and I took diuretics.

Angela went for many years without proper diagnosis despite her GP finding her condition interesting:

But then around the time of my 14th birthday, um, my left leg – I had a bad pain in my left foot one day and the next day there was a little egged shaped thing on the top, just below my toes – a little egg shaped thing. The pain had gone, and um I took a feel and I found if I could press my finger into it, it would sink in – didn't really hurt – made a dent and then after a little while it would just rise up again. And um, I didn't take much notice of it – but um, I found that this egg thing came up every day and it was a bit of a nuisance in the way

of shoes, but not too much, I just squashed it in and forgot about it. Jumping forward now, I went to visit my sister in Massachusetts and my legs just ballooned… and when I was pregnant that was a funny looking time because the swelling came up my legs and sort of stopped at a certain level and it looked as if I was wearing Wellington boots. I was given a diuretic pill. That got me through that first pregnancy, but I don't think they've worked since. My GP always said "Oh well, that's interesting" but they never suggested anything.

Both Browse *et al*. (2003) and Twycross *et al.* (2001) have described the ineffectiveness and dangers of using diuretics in the treatment of lymphoedema and have pointed out that there may be a causal link between the prolonged use of diuretics and the frequency of cellulitis in people wrongly prescribed them. All of the participants, with the exception of Louise, were given diuretics or antibiotics over many years, or even decades, without routine monitoring, when it is known that these drugs can create higher protein levels and increase the risk of infection and may also lead to resistance to mainstream antibiotics (Browse *et al.* 2003, Mortimer *et al.* 1999). Diuretics are sometimes prescribed when a medical doctor believes the oedema to be uncomplicated or has not come across lymphoedema before (Twycross *et al.* 2000). They work by removing fluids from the interstitial spaces and returning it to the capillary (venous) system rather than through the lymphatic system. Patients will usually experience a reduction in the swelling and may be pleased with the results of the use of diuretics in the short term (Browse *et al.* 2003). However, what diuretics cannot do is filter the fluid and reduce the protein level in the interstitium. This can quickly lead to complications in the guise of bacterial infection. High protein levels create a perfect environment for bacteria to grow and spread and cellulitis is dangerous and

sometimes life-threatening (Twycross *et al.* 2000). Antibiotics are necessary to control and prevent cellulitis in lymphoedema and are essential to prevent further skin damage that can predispose to recurrent attacks (British Lymphology Society & Lymphoedema Support Network 2006) but they should always be used under supervision and as sparingly as possible.

With reference to diagnosis, there has been a shift in emphasis in the last 30 years from a subjective approach, in which a patient's symptoms are considered by a GP who knows an individual's health history, life-style and personal identity, towards an objective approach, where (with the rise of hospital medicine and diagnostic technology) the identity of the disease rather than the patient comes to the fore. The doctor's expertise no longer lies in recognising the significance of symptoms for an individual patient but rather in a detailed knowledge of the disease itself (Bradby 2009). While this may be fine if the disease is recognised by the doctor but in the case of lymphoedema, where the disease is little studied or understood and where a neat 'cure' is not available, it creates the type of difficulty described earlier by Philip, Belinda and Angela.

Doctors still act as gatekeepers to healthcare services and diagnosis is crucial if access to services is to be gained. Those in this study who suffered symptoms without a recognizable diagnosis found their journey through the system severely hindered. Further, it is likely that doctors faced with an 'untidy' set of symptoms and signs will attempt to shoe-horn individuals into their pre-conceived diagnosis categories and treatments even in the face of obviously unsuccessful outcomes for the patient. This would account, for instance, for doctors prescribing diuretics when they patently had no long-lasting impact on the swelling. These doctors assumed that

swelling in the lower limb must be linked to congestive heart failure where diuretics are a first course of action.

Participants were sometimes (but by no means always) referred by their GP's to 'specialists' in the search for a diagnosis, but these were usually not actually specialists in lymphoedema, most often being consultant cardiologists or immunologists. It is obvious that these would be the first choice for a busy GP confronted by chronic oedema or repeated infection, but it is less easy to understand the extent to which those consultants were failing to make accurate diagnosis in the face of distinct symptoms that refused to conform to their pre-conceived schema. Philip explained that:

> *A consultant haematologist told me to stop using NSAIDs* [non steroidal anti-inflammatory drugs] *and drinking alcohol (not that I was a great drinker). A consultant physician told me to dry between my toes with a hairdryer! The immunologist sat and gabbled for a long time about IGGs and IPPs* [immune system clinical indicators]*. She then put me on Erythromycin* [antibiotic] *and said "you will be on this for the rest of your life". She was investigating why I had had several bouts of cellulitis and noted that I had swollen legs. But she didn't make the connection!*

There appears to be a disturbing tendency for luck to play a part in eventual diagnosis, with junior medics only occasionally recognising the condition because of a rotation they had completed rather than because of any systematic education and knowledge about the condition. This supports the claim by Greene & Waters (2003) that medical education is not dealing effectively with lymphoedema and Belinda's experience illustrates this well:

> *When I was 40 my GP retired and a young doctor came straight from his hospital year and directly he saw me he said*

"I know what's wrong with you. You've got lymphoedema and my old boss who I trained under devised an operation for that. I'll send you straight to him". So, 30 years later…

Philip also had the experience of a young doctor recognising the condition only to be told by another, more senior medic that this had been wrong:

When I moved to the south of England, I began to have regular medicals, which were organised by my company. BUPA medical, not full blown ones because the company was too mean to pay! But they all commented on the swelling, one doctor after another. One young doctor said one year that I suffered from lymphoedema, but the next year this exceedingly arrogant doctor said "it's a load of rubbish; he wouldn't know what lymphoedema was if it hit him in the face – you've just got swelling of the ankles, which is called oedema". But I hadn't a clue what it was and in subsequent years it was ignored. As simple as that.

Alternatively, participants had sometimes made their own diagnosis once they had accessed information about lymphoedema. Many spoke of finding information not through healthcare practitioners but via websites after searching under swelling. Angela recalls finding information on a website which eventually led to her being seen in a specialist clinic 57 years after the onset of her first symptoms:

After Vienna, I was frightened of the summer and I went on the internet, LSN, and found an MLD person and she said "this won't do – and you need help" and she wrote to my doctor and told him what she had diagnosed and he wrote to the special clinic for lymphoedema.

Even when a correct diagnosis had been established, participants found that accessing effective treatment services and information about their condition was very difficult – a factor apparent in the findings of Barnes *et al.* (2002) and Swain *et al*. (1993) who showed that people were not being effectively directed to services and information by their healthcare professionals. Louise was fortunate to get a reasonably early diagnosis (after only four years) but still struggled to get further information:

> *I had to go back and they said "yes, it was definitely lymphoedema". …So I was given leaflets on how to look after my own skin, not to gain any weight and to wear my stocking. Otherwise it was "bye, bye" and off I went into the world with no more information than that.*

Martyn had similar difficulty in that he was given cursory information about 'general measures'.

> *So I did see a specialist who diagnosed the condition and just said there is no treatment – well, he did refer to surgery but didn't recommend it, said it didn't work. In fact he was quite emphatic about that. And he just prescribed what he called 'general measures' which amounted to wearing a support sock.*

Referral to specialist clinics seemed to be dependent on a crisis emerging, as Angela describes:

> *I got caught in the summer heat-wave in 2003 on the continent and we'd already had a 5 hour wait at the airport and my legs swelled. Well, I've never seen anything like it – I thought it was going to burst it was so tremendous – I can't describe how it was! …and they took me on at 71 years. And it had started when I was 14 years. 57 years later.*

None of my participants had experienced what has recently been defined as best practice in the treatment of lymphoedema through the NHS (Medical Education Partnership 2006). Most had been told that there was nothing that could be done and were left to cope with the condition as best they could for very many years. Some were given rather eccentric advice such as that given to Gerry:

> *So the immediate advice was to try and elevate the leg. The fact that the wound would never heal in the foot - it suppurated the whole time - um, I used to sleep with it up – I was given the most bizarre advice about sleeping with it at 45 degrees to bring the swelling down. Now I give the challenge to anyone who can sleep with their legs at 45 degrees! I rigged something up, I was staying with my parents at the time and I rigged something up in my bedroom there, with an old guitar strap, and twice nearly dislocated my hip rolling out of bed in the night. You know, ankle the wrong way round! Leg up there, body on the floor! (Laughter)*

In some cases, coping without treatment might have been the best option since there was undoubtedly some inappropriate (even damaging) treatment suggested. Examples included recommendations to Susan for the use of elastic stockings (as opposed to lymphoedema compression hose) which damage the remaining lymphatics by compressing them (Woods 2007). Many patients and health care practitioners may not understand the differences between low stretch and elastic compression in the treatment of lymphoedema and damage can be caused to an already compromised lymph system by careless prescribing or application of inappropriate elastic hosiery. Many GPs, community nurses and podiatrists prescribe or apply elastic stockings for the control of conventional oedema and for leg ulcer control but this can be disastrous if used on a lymphoedematous limb as it can lead to

further damage to delicate lymph vessels, an unsightly and painful swelling at the margin of the elastic sleeve/stocking, development of lymphoedema in an adjacent area (e.g. genital or facial areas) and an increased risk of cellulitis as proteins build up in the over-compressed areas (Twycross *et al.* 2000, Medical Education Partnership 2006, Woods 2007). When the elastic garment is removed, the limb will become even more swollen than it had been previously.

For Philip, Belinda and Marie there was also an issue with the unregulated use of compression pumps which Casley-Smith and Casley-Smith (1997) have condemned as being a source of infection and likely to move lymph into an area previously unaffected such as the face or genitalia. Complications reported by Casley-Smith & Casley-Smith include: raising of protein levels still further in the lymphoedematous limbs by forcing water only into the blood whilst protein remains trapped; creating an area of fibrous tissue around the region proximal to the sleeve of the pump if fluids cannot drain further along the system; development of lymphoedema in a previously unaffected region e.g. typically genital or facial regions; lymphatic vessels – already vulnerable – may be damaged by the high pressures exerted by the pumps and this excess pressure can cause small fistulae to form at the surface of the skin through which lymph fluid leaks creating a risk for infection; pumps themselves may spread infection from one patient to another if they are not cleaned thoroughly between uses. These negative findings should be balanced against some evidence for their successful use (Twycross *et al.* 2000) and it is certainly the case that many patients anecdotally report an easing of symptoms and improvements in limb volumes (Lymphoedema Support Network 2008b). Philip and Belinda did talk of the temporary relief from pain afforded by the pumps and Marie referred to the reductions in

swelling – although these did not last. Philip and Marie had abandoned the use of the pumps but Belinda continued to use hers until very recently. What is concerning is that patients may be experiencing short term relief only to find that they are confronted by long-term problems associated with the use of these pumps. In the UK, despite the negative evidence that exists, pneumatic compression pumps are recommended and used with little reference to this debate (British Lymphology Society 2003, Turner *et al.* 1998).

Non-specialist liposuction was administered on Belinda even when research has shown this can remove remaining lymph tissue and introduce infection (Brorson 2000). Brorson (2000), writing in the context of lymphoedema of the arm following breast cancer surgery, has claimed that specialist surgical liposuction has the potential to reduce lymphoedematous swelling. Conventional liposuction usually seeks to remove adipose (fatty) tissue - but some delicate lymphatic tissue will always be removed at the same time by large, imprecise needles. Brorson uses specially designed fine needles which he claims, when used skilfully, can remove lymph accumulations in the interstitium without damaging the remaining lymphatics (Brorson 2000). This is contested by the Lymphoedema Association of Australia (LAA) (2008) who have queried its safety - claiming that in untrained hands it could prove damaging to those with lymphoedema since it is likely to also remove remaining healthy lymph tissue thus worsening the condition. Belinda had experienced very many episodes of cellulitis and her lower limbs were mis-shapen and scarred.

Radical and repeated de-bulking surgery was undertaken on Belinda when this is shown clearly to be contraindicated for the vast majority of lymphoedema patients (Medical Education Partnership

2006, Woods 2007, Browse *et al.* 2003). Surgery is advocated as a last choice for those with specific and distressing symptoms, e.g. eyelid or genital swelling. It may also be used to de-bulk grossly swollen and infected limbs. Swollen and engorged areas are removed and the surface areas remodelled - sometimes using skin grafts. Although it may seem persuasive, surgery does not seem to have a substantially greater effect than conservative methods and it is claimed that it can further damage an already overwhelmed lymphatic system and create even greater disability and disfigurement in the years following the surgery (Casley-Smith & Casley-Smith 1997, Mortimer 1997). Belinda was particularly unfortunate to be offered many of these treatments despite these known contraindications:

> *By that time both legs were very swollen and still hadn't had any bandaging or any conservative treatment - they didn't do that sort of thing then. So he did the first leg op and it took five hours and that was from the toe to the groin. I've kept the bandage off so that you can see what they look like. That's the dermal flaps, one on each of side of both legs, and that's how the skin goes when lymphoedema gets established because of virtually chronic cellulitis on the legs. …They gave me elastic stockings but they were too tight. I can't wear them now because the drain hole on the second upper flap keeps opening up. …After the first op they suggested I use a pump… I used that pump up until last year when the cellulitis and swelling got too sore. I used to have it on for about an hour and a half every day and it did put a bit of pressure on your kidneys same as the diuretics. If I do get very swollen and the skin starts opening up, I take diuretics and lose about 7lbs! …He was scared to do the other leg – because he thought I might stop breathing – and he said "well we'll try liposuction, we needn't give you an anaesthetic for that, that's*

not a five hour op. So he did that six times - sometimes on one leg, sometimes on both.

Such interventions were clearly inappropriate and it is interesting to note that Belinda was paying privately for her health care. It is a matter of speculation whether a lymphoedema specialist working within the parameters both of best practice and the budgetary constraints of the NHS would have undertaken such treatments. To suggest mal-practice for profit is probably excessive but to suggest ill-informed practice would certainly not be so.

Only Louise has had regular, appropriate, active treatment for her lymphoedema (albeit not through the NHS) and, tellingly, she is the only person with no history of cellulitis. Louise had received one course of treatment that was paid for by her Primary Care Trust but has subsequently had to pay privately for her MLD treatment. She explained:

We did try – the NHS and um, nobody, the surgery wouldn't help – they wouldn't even pay half and half and my bill, well it was something like £1200 by the time I had finished my 6 weeks treatment. I was a wee bit cross… because I couldn't get it reimbursed… I just let it go in the end and I had to pay for the whole lot.

All participants were angry at the inequity which exists in relation to the treatment of their lymphoedema compared to other chronic conditions and were sceptical about the concept of a National Health Service when services were so patchy and inadequate or had to be paid for privately. Susan, who has worked in the NHS all her life, commented:

In my letter to the PCT – one of the things I said, you know, that I felt as a retired radiographer, I felt it was a poor thing

you have this post code lottery to get treatment for the lymphoedema.

Angela was very distressed at the costs of private treatment when she had a small income and was angry that lymphoedema was not properly treated by the NHS.

She [MLD therapist] *got it down with bandaging and massage, you know, and um, but it was £30 a time and I was only making something like £10 a week. Ridiculous that everything else can be treated on the NHS but not, you know, and it makes us really ill and immobile. Why? Why?*

This notion of 'postcode lottery' has been well-rehearsed in the media in recent years and also much debated within medical sociology texts (Bradby 2009, White 2002). It is identified as being a profound problem in a national system which is predicated on equity of service and free at the point of delivery. Interestingly, Philip sagely commented that if there were some financial incentive for GPs to refer individuals with lymphoedema, services might improve:

There is another issue there, GPs are very, very happy to dish out Statins now against high cholesterol, even against average cholesterol levels, because they are financially encouraged to do so. If they were financially encouraged to refer possible lymphoedema patients to hospital, you would find that you could not move at the… hospital with potential lymphoedema patients. And it does annoy me…

It has been known since the publication of the Black Report (Department of Health 1980, Townsend *et al.* 1988) that there are distinct inequalities of health between British social classes and ethnic minority groups, but interestingly, in this study all

participants were white British with occupations that would be defined in that report as ‘professional’ or ‘managerial’: they were well educated and had successful and reasonably well paid employment histories but, nonetheless, found it difficult to negotiate effective treatment regimes. All had presented themselves for treatment but had been unable to access it. It can only be presumed that this, once again, was due more to a lack of understanding of what might constitute useful intervention rather than any overt or subliminal attempt to ration services. That said, chronic illnesses like lymphoedema provide particular challenges to the healthcare services in that they cost a great deal to manage over an individual’s lifetime and are obstinate to curative processes (Bradby 2009). Because of this they often represent no opportunity for the accumulation of profit (fiscal and intellectual) for pharmaceutical companies or healthcare researchers and so investment research is minimal and treatment innovations remain in stasis. The absence of a profit motive for multi-national pharmaceutical and medical technology companies means that any reduction in inequality for people with primary lymphoedema is unlikely in the foreseeable future.

Despite the seeming affluence of this group of participants, all struggled to pay privately unless they had private health insurance. Gerry, employed as a school teacher, captured this difficulty in his comment:

> *The only* [private, MLD] *person I have found in this area wants £70 a pot and it’s like, ‘don’t be silly’… The lady I spoke to… said ‘well aren’t you prepared to pay for it? And I said ‘lady if you had the condition you probably would be prepared to pay for it, but I can’t, it’s not a question of being prepared – I can’t’.*

It is interesting to speculate on the extent of inequity which might exist for others with more limited means: those with less well paid employment, those out of work or those on a pension would all find paying for private treatment even more difficult. It is likely that the difficulty experienced by my participants represents a microcosm of that which exists in the wider population who live with lymphoedema and underlines the need for the NHS to deliver treatment based on best practice, free at the point of delivery, to all those with lymphoedema.

Participants also felt frustrated by the length of time it took to find useful and accurate information about their condition. It is disturbing to note that despite this study taking place more than a decade after Cale & McCahen (1993) Barnes *et al.* (1999) and Baker *et al.* (1997) highlighted this issue in their research, my participants still found that healthcare professionals did not automatically give them the information which could prevent them living with a diminished quality of life and prevent further deterioration of their condition. Marie felt angry that she was not offered any meaningful advice following her diagnosis. She said:

> *…they told me I had got Milroy's Disease… But, I actually don't have Milroy's disease, but that was their interpretation of it and there was no real advice given except 'don't stand on your feet all day'. I'm a teacher - what else was I going to do? They also gave me diuretics which I took for 27 years. …Which is not good really, because it didn't help, and I wasn't given any garments at all. …Never, no, there was nothing. That was it, into the fog, um, and just left really.* [B: Just asked to kind of get on with it basically?] *Yes, and you know that was it.*

Where participants had found information on their condition, most sourced that information independently of the healthcare professions – often through the Lymphoedema Support Network after a web search. This reflects the findings of Barnes *et al.* (1999) who demonstrated that only 16 per cent of elderly people with physical impairment had received information on their condition and on support services from health and social care professionals. Again, it is interesting to note that my participants were well-educated, resourceful people but there may be many others who struggle with this condition who have not been able to access useful information. All participants had been alerted to this study through the LSN Newsletter or Website and so it can be assumed in theory that all had access to the robust sources of information contained in these information sources. It remains a matter of conjecture as to how many people with undiagnosed or un-managed primary lymphoedema are struggling to cope alone. Angela had experience of a friend who had received no information despite having severe lymphoedema.

> *I've got an old friend I've just come in contact with again, I saw her on Sunday, and she's coming here to work and she has had some terrible problems – she's got terrible legs. So I got out the 'Lymphline' papers, you know, and all this and the literature and things. She was fascinated with it, and um, and um, as much as I could find – and gave it to P – she's taken it to her GP and waved it under his nose – she also has a first appointment in September. And she wouldn't have done, unless I printed all the stuff out.*

Philip described how he had introduced a neighbour to the local LSN group. He explained that she had never been formally diagnosed (even though she shared a GP with him) but was immediately grateful for the opportunity to meet with others experiencing the

same challenges and to discover that there was a body of information that could be useful to her.

> *I went round to see a neighbour and was wearing shorts and she said 'what's wrong with your legs' and I told her. And I said 'what's wrong with your legs?' because she also had swollen legs. 'Oh, I don't know, I've just been to see my GP and I'm supposed to have 2 knee replacements, but I can't face it, my legs are so swollen now' and I said 'what did the GP say?' 'Oh, he gave me some diuretics and said they might or might not do some good'. I said 'J, you've got lymphoedema; I'm going next week to a lymphoedema support group - why don't you come with me? Then afterwards you will…. see your GP and get a proper referral for lymphoedema management'.*

Although all individuals in this study spoke very positively about the benefits of LSN literature and advice, relatively few of them were completely up-to-date with current best practice as defined by the International Consensus document (Medical Education Partnership 2006). Only Philip attended self-help meetings regularly, whilst Angela and Belinda had attended sporadically. This was partly due to their perception that the meetings were focussed more on lymphoedema which was secondary to breast cancer, also due to time restrictions if people were in full-time work, but additionally, there was some general reticence about attending such meetings. Some perceived them to be more appropriate for elderly people and many coped best with their condition when they could define it as an irritation as opposed to something that dominated their lives. Louise hoped to find someone her own age to share her experience with.

> *C has a support group, which we all lobbied for, but it is such a shame it is in the week on a weekday and very often I can't*

get there – in fact I haven't been a lot. But they always have a speaker and they always come with new stockings or with new footwear. …it's just about touching base with other people with primary lymphoedema. It is, it is and um, finding somebody younger, because I haven't found anybody the same age or younger than me yet to actually speak to, um I haven't looked or delved into you know that sort of thing, or I'd love to talk to somebody my age.

All participants commented that they would welcome greater access to 'experts' through the self-help groups they attended and did not seem to acknowledge that, in many ways, they were the most expert about their own condition (White 2002). This was, of course, wrapped up in the issue about paucity of services which is a legitimate concern. It seems unlikely, however, that clinicians - already so stretched in their clinical work and battling against a system which does not appear to value the service they offer - will be able to make any more time to set up and attend self-help groups. Arguably, though, their impact could be considerable and positive if they were able to do so. I remain deeply concerned that my participants did not always seem to understand the difference that regular, appropriate care could make and often found myself torn between acting as 'researcher' and as 'therapist'. This is an issue I return to in my final reflection on the study.

Perceived inadequacy in levels of knowledge amongst medics and other healthcare professions

All participants were surprised at the lack of education and knowledge about their condition amongst all healthcare workers. Angela found this surprising given the substantive education required to work in health care.

They don't seem to know about it, do they? And even if they've heard of it they don't know what to do about it. Surprising when you think how much education you need to be in these jobs.

Marie expressed her surprise at the paucity of advice she was offered:

And it wasn't anything that anybody really helped me with, despite these outbreaks of cellulitis and septicaemia there was still no advice, there was still no [pause], well you know – you should actually take some prophylactic drugs in case it happens again, or let you know that you should bind your legs up or do something. There was nothing.

This reflects the concern expressed by Greene & Waters (2003) that healthcare professionals and particularly medical doctors were not well informed about lymphoedema. Mortimer (1997), a dermatologist specialising in the treatment of lymphoedema, claims that this is because GPs define it as a rare condition for which there is no cure. But in reality, most GPs have had no education on this topic and the real concern is with the Royal Colleges, Deaneries and Medical schools who have not ensured its inclusion in undergraduate or postgraduate programmes or training. This general lack of knowledge was thrown into relief by the expertise of lymphoedema specialists once these had been accessed. I was questioned frequently about levels of education at initial training and the reasons why the condition had become such a 'Cinderella' condition. Angela was angered by this lack of interest and knowledge.

I feel a bit cross about the fact that it was just brushed under the carpet, when I did actually say in my young married years that it was beginning to start to hurt and annoy me and when I was working and um, but nobody was interested. They

didn't know anything about it – they didn't even want to think about how to deal with it. It didn't come – it wasn't on their agenda. They didn't pick it up on their agenda – they had no experience – it was a minor thing – it wasn't going to kill me. It didn't come into their training or consciousness.

Even though he received a diagnosis within a much shorter time-frame than other participants, Martyn was surprised at the capacity for misdiagnosis amongst the medical profession.

I went to the GP who was very interested and didn't know what it was. I was referred to other hospitals… and there was some misdiagnosis. A couple of the doctors there thought it was some kind of phlebitis condition.

Gerry had similar experiences of misdiagnosis and, again, found levels of ignorance very unsettling.

I got these amazing cramps by the left knee, and I thought I had been sitting still for too long in the exam, but I could barely put my heel to the ground all the way home. The next day the leg swelled up and I went to the GP – and he said 'I've no idea, go to [names hospital]*'. And they prodded and poked and did all sorts of things and said 'it's a ruptured cyst, you've ruptured a cyst behind your knee'. And I've subsequently been told that basically they hadn't got a clue – they didn't know.*

Philip reflects on the extent to which his experience is mirrored by others in the self-help group that he attends and on their resigned attitude. He also expresses his disquiet that even after the diagnosis had been made; he was still not referred for appropriate treatment.

Yes when I tell them [LSN group] *that I have been 31 years on diuretics - so often I get the response 'oh yes, the same*

old story'. They all have some experience of not getting a correct diagnosis or treatment. What is so disappointing is that basic information is not being acted on. The link between cellulitis and lymphoedema has been around so long and yet the medical people are not making the connection. Dr T, who finally diagnosed lymphoedema, didn't even refer me to the lymphoedema clinic. He obviously didn't know that I could get treatment there. I don't understand why. They never made the connection. No, lymphoedema wasn't diagnosed until… it's only about 2-3 years ago, when I was diagnosed at the clinic that my swollen ankles were due to lymphoedema.

One of the revealing facets of my conversations with the participants was the extent to which they still focus on the senior doctor as the arbiter of care. Although many had had positive experiences with nurses, junior doctors and Manual Lymphatic Drainage (MLD) practitioners they nonetheless continued to view the GP or consultant as the lynch pin of diagnosis and treatment. An MLD practitioner or junior doctor may have mentioned the word lymphoedema but until the senior medical doctor had officially afforded a diagnostic label, participants had not 'owned' the condition. This resonates with Frank's (1995) view that medical narrative is the master narrative that supersedes all other stories and becomes the story against which others are judged. Nonetheless, this is surprising given that the status of other healthcare professions has grown in recent decades and many nurses, physiotherapists and occupational therapists have developed specialism in the area of primary lymphoedema - although without medical referral, these specialists cannot be accessed through the NHS. In terms of improving access to service, this means that making lymphoedema more visible in medical education – and thus educating a new generation of doctors - is still

the most promising way of improving access to lymphoedema services.

Four of the participants (Susan, Philip, Angela and Gerry) had been subjected to what they felt was arrogant and demeaning behaviour from GPs or consultants at the time their initial symptoms developed or on diagnosis. Susan, who was working at the time as an NHS Radiographer, had an unpleasant and distressing encounter with a consultant in vascular medicine:

> *Then I went back to see the doctor and he asked 'are you having a venogram?' and I said 'why no! It's not my circulation!' but he said 'I'll ask the questions!' he was a bit arrogant you know. I tried to explain to him – I wanted to explain to him that the underlying problem was lymphoedema. But he wouldn't listen to me, they don't sometimes, they think they always know best… So that really upset me at the time - he was so harsh.*

It is surprising and dismaying that this doctor did not allow her to speak let alone consider her personal situation and professional knowledge. Philip also experienced disdainful and ignorant behaviour from an occupational health doctor:

> *After my 3rd or 4th bout of cellulitis, my company (who were possibly pushing for me to take early retirement due to ill-health) referred me to the company doctor who was basically the old military doctor. You know the type: 'You are skiving. There is nothing wrong with you and I am going to find out that there is nothing wrong with you. 'Why have you been referred to me?' 'Well, I've had 3 or 4 episodes of cellulitis.' 'Huh, I've seen more episodes of cellulitis than you have had hot dinners, there's nothing wrong with that and it's just one of those things. You've just got oedema of the ankles, and*

you've had 3 or 4 bouts of cellulitis.' What can you say? When I did see him several years later and said my cellulitis count was now up to 8 he was a little taken aback and said 'this is more serious than I thought'.

Gerry's GP was insensitive and intolerant to a quite unacceptable level:

The GP …um well he would best be described as insensitive I think – until he got cancer of the throat and went through a transformation. But prior to him having the cancer himself, he was notoriously intolerant and he did a classic of you know 'you're not going to die from this – they're bound to have given you some exercises, just do those, don't come whingeing to me' you know, that sort of attitude.

Quite rightly, we do have very high expectations of doctors and expect them to work within an up-to-date evidence-base as well as acting with humanity and compassion (White 2002, Bradby 2009). In this study, however, there was much evidence of healthcare professions falling below that expectation. It is my belief that respectful care should be afforded to all regardless of the 'tidiness' or 'untidiness' of their conditions. By 'tidy' I mean those symptoms that fit neatly into an existing schema of knowledge. Participants described GPs and vascular specialists (in particular) as dismissive of symptoms and advising that weight loss would solve the problem. This reveals a lack of understanding about the condition and its aetiology and a cavalier approach to the distress of others. Many participants were told simply to 'get on with it' since no cure was available. GPs and occupational health physicians were singled out for criticism because of their ignorance of the day-to-day restrictions and discomfort imposed by the condition and because of their unwillingness to acknowledge its effects on daily life or

working practices. It is possible that some less skilled doctors, when faced with uncertainty or when discombobulated by their own ignorance of a condition such as lymphoedema, may blame the patient for being difficult - rather than question their own knowledge base.

Assessment techniques used to aid diagnosis (especially in the 1970s and 1980s) had, in some cases, been extremely damaging and some participants had experienced at first hand the medical establishment covering its tracks when iatrogenic mistakes had been made. Gerry described with great emotion his experience of an assessment procedure going wrong and causing further damage to his already compromised lymphatic system.

> *When I went into his room he was angry, and fretting, and I thought this is odd, um, I was only young so I wasn't used to professional men behaving like this. And he said 'I don't understand how to have this conversation – I'm just really sorry' and he said 'look, this is your chart' and he put it up on the light-board and you could see this single lymphatic vessel going down my leg until it got to my calf, and then there was just this thread – there was no more vessel! He said 'basically the lymphangiogram has told us we should have not done the lymphangiogram – you have a hypo-plastic system and what vessels you have, we have just damaged – and there's no retrieving what we've done'.*

Later, Gerry saw another doctor and was very upset to find that his hospital notes had gone missing. He believed that this was deliberate and was to cover mistakes made and to prohibit any accusation of blame.

> *…so I went into the appointment and he said 'I'm very sorry, but your notes have not turned up yet'. Um, picked up the*

phone, the sister came in, she just stood there in the doorway, and he sat back in his chair and said 'no, no, don't tell me that sister', and she went 'yes, they're not there. Mr [Gerry's] *name was on the file, but there was nothing in it'. And all the notes, notes all gone, and have never been found. Well, I mean at the time, he said 'I'm not going to tell you why I think your notes have gone, Mr G, but you're an adult and I am sure you can probably guess'. And he showed me and there was nothing. So I was like 'well here we go, I've scored again'. Um hey.*

As a profession, medicine has striven to become a scientifically-based, concrete-world of 'knowable facts' (Bradby 2009). If an individual medic fails to live up to the ideal of being 'all-knowing' both s/he and his/her professional peers will be harsh in their judgement of this lapse. It appears to be difficult for the profession to readily admit to uncertainty and this creates difficulty in its relationship with patients and other healthcare professionals when errors or omissions occur. When things go wrong (as they inevitably will in such a complex arena) doctors may self-construe this error or ineffectiveness as personal and culpable ignorance and this perhaps leads to their sometimes defensive behaviour and the 'covering of tracks'.

Gerry struggled for several years to forgive the medic who had exacerbated his lymphoedema by using an inappropriate assessment technique and, after much soul searching, finally decided to speak with the medic concerned as an act of forgiveness and a form of closure. He recalled that when he contacted the doctor some years later he met with a very cool response:

I went to a Christian Conference in Canada and somebody challenged me about my attitude towards the whole incident,

in a friendly way not in any aggressive way, and said 'well have you forgiven the people who did it?' I said 'well…' And he said 'come on, let's have a prayer', and I found I couldn't even say the words. So I had to do some business with God to get over that - to try and forgive. When I came back to the UK, I phoned [the hospital) *having returned thinking he probably would've moved on, he would've done other things, la, la, la etc. And I asked 'could I talk to* [the doctor] *please?' They put me through and he remembered me. And I said 'you may think this is kind of weird' and I said that 'I may be doing this more for me than for you, but I want you to know that I forgive you and that I appreciate that you were just doing your job'. He said 'thank you, I appreciate that very much, it's very decent of you to phone'.* [The tone of voice adopted by Gerry intimates that the medic concerned was rather abrupt]

It is interesting to note that at the time of my conversation with Gerry, I recorded in my field notes that he did not seem to have truly forgiven the medic concerned. His body language was very tense with balled fists clenching and unclenching as he was talking of forgiveness. His paralinguistic signals implied anger with his speech emphasis abrupt and staccato. Much has been written about the importance of non-verbal communication and Sugarman *et al.* (2005) have stressed that where there is an apparent contradiction between what is being verbalised and what is being observed non-verbally, it is usual that the non-verbal behaviour is communicating the true feelings. It could be that Gerry's apparent act of forgiveness was actually an act of confrontation and blame and that the doctor concerned picked up this aggression and responded coolly. They may have been complicit in an elaborate show of civility when, in reality, both felt very aggrieved.

Psychological concerns

The extent to which individuals were willing to disclose their innermost feelings was undoubtedly limited in a first meeting with a stranger and I had a sense that all participants were aware at some level of what was 'tellable'. Smith & Sparkes (2008) discuss this as being on a gradient, bounded on one side by narratives that were insufficient to warrant listener interest and on the other by stories that would be 'untellable' because they are too personal, too embarrassing, or too frightening. Nonetheless, my participants did all share their thoughts on how the condition had impacted on their personal lives and levels of psycho-social distress varied in the group from mild feelings of distress and occasional embarrassment to clinical depression treated by medication. Martyn describes a delayed reaction to the diagnosis:

> *…but then of course I had the problem of struggling with the psychological effects. I think in the first 18 months or so they were quite mild until the realisation, you know. I felt that the shock was realising that it was incurable, I think.*

For many people following a diagnosis of chronic illness, they will nonetheless stick to a restitution narrative (Frank 1995) in which there is a belief that though they may currently be experiencing a period of ill-health, this is temporary and well-health will be restored in the future. For Martyn, after 18 months, this restitution narrative could not be sustained in the obdurate face of his continuing lymphoedematous swelling. Frank (1995) describes this period as one of narrative wreckage as there is no other story to fall back on. A new and undesirable body-self relationship ensued for Martyn and he descended into depression. But his story does go on to reveal that he did cope eventually by adopting a strategy in which the condition is not allowed to disrupt his life and by adopting a style which minimised the challenges and kept the details of his

impairment hidden from friends and colleagues. This reflects the three elements of the process that Bury (1982) described in an individual responding to chronic illness. It is also possible to observe the stage process described by Elizabeth Kubler-Ross in her 1973 work on death and dying which has come to be adapted and applied to any who experience catastrophic loss. For the first 18 months Martyn could be described as being in denial, and once this could no longer be sustained he fell into depression and then, only latterly, acceptance (Kubler-Ross 1973). Marie has also experienced many periods of depression linked with coping with her lymphoedema.

> *Well, I mean that's another side to it and one of the other things that I have suffered from* [depression]*. Mainly possibly because of the way I don't cope with things and I don't let things go. I have suffered from depression and, you know, there are times when I'm great, and times when I'm, you know, not so great.*

Even close family members may not pick up on how profoundly depressing such a diagnosis can be: Gerry expressed this very clearly in his description of a particularly distressing exchange with his parents.

> *… I mean famously on one occasion, when I had a bad day with the leg, I broke down at the meal table in the evening and stomped off. Parents came after me, because that's not normal, acceptable, expected behaviour, you know, all ex-army. My mother's family were all ex-army too – you just don't do that sort of thing. And I actually said 'I wish I was dying, because at least I would know there was an end to it'. And that <u>really</u> rocked them most profoundly. Profoundly. …I was depressed but nobody picked up on it.*

Of course, the family and friends of those with lymphoedema will also be clinging to the narrative wreckage (Frank 1995) hoping for restitution and normalisation and there is a powerful imperative in Western societies for people to transcend despair and to continue to hope even in the face of incurable, life changing illness (Smith & Sparkes 2008). Lupton (2003:70) has said "To despair, to lose hope, are frowned upon as strategies for dealing with diseases such as cancer." Psychological treatment or support was reported as non-existent and no participant had been offered any professional or formal psychological support at or after their initial diagnosis. Any support that was provided was as a side-effect of general lymphoedema care by dedicated and empathetic lymphoedema practitioners. When Gerry was finally seen by lymphoedema specialists, he was able to let go of his internal grief and anguish:

> *And I was thinking, because at this point since 1987 through to, where are we now? 1991, I've had nothing. Nothing. I've had nobody coming alongside and ask 'how do you feel?' Nothing.* [Bernadette: And during all that time, that experience, you'd still had no psychological counselling offered to you, had you?] *No, none at all, absolutely. Well, I cried more with J in those first two or three sessions that I had probably done in the whole four years, because the first time I ever cried – I mean my GP who was my friend from school - um he said 'this isn't going to kill you – you're not going to die from this – you can still walk, you can still run if you <u>need</u> to, there's so much worse than this'. Humph. I thought 'Oh all right, I have to be humble; I have to be good and say, yes, there are people worse off than me'. … I had a really bad few weeks, …and he* [locum GP] *phoned C* (lymphoedema practitioner) *on the spot and got her ansaphone as we all do, and um, said 'I'm going to write to her and get an appointment for you, because this is*

ridiculous, that you haven't had any help – have you had any help with depression?' And literally that was out of left field and I said 'well no, but I mean it's been kind of – I mean I'm not as happy as I used to be – I just put it down to the fact that I was rather kind of cheesed off'. He said 'you're kidding, that should've been picked up, oh well, I'm only here for another week, um, pick that up when… Dr K comes back', I said 'yes, right' (but thought) no chance'. And then I met C and it was kind of like being with J all over again, … I sat with her and we took off the bandage and it smelled by this point cause it fell apart by the time I washed it and all that. And I took all the bandaging off and the leg was just a complete mess and the knee was undefined and it was all just… And I just sat there with big old tears running down my cheeks and she said 'it's all right, it's absolutely fine. If you want to cry boy, you cry, it's absolutely fine' and that was phenomenal and obviously working with C as I have done, um.[pause] *I had a bit of a time.*

This final statement is clearly an understatement of the psychological challenges that Gerry was coping with and this quote is powerful in the way that it creates empathy for his situation and a frustration about the inadequacies of psychological support offered to people diagnosed with chronic, incurable disease. Individuals experiencing illness or disability will naturally want answers to questions which go to the soul and are not just about treatment techniques and curative processes: 'why me? Why now? What next?' The urgency of these questions is very real and the impotence of medicine to answer them may be frustrating and result in depression for the patient and discomfort for the physician (Bradby 2009). Medicine aims to alleviate symptoms but is so much less equipped to deal with the distress caused by disruption to

everyday life, and the uncertainty and fears that chronic illness or disability presents. Cheryl Mattingly (1998:79) suggests that therapists and other healthcare professionals work to construct 'success' stories: "They presume that patients will not be committed to therapy without success, for success breeds hope, and hope is essential". She goes on to argue that certain story outcomes are welcomed and others feared: "When a story is told, if that storytelling is successful, it creates in the listener a hope that some endings (generally the endings the hero also cares about) will transpire. We hope for certain endings; others we dread. We act in order to bring certain endings about, to realise certain futures, and to avoid others." (Mattingly 1998:93) This could well be a reason for reticence on the part of healthcare professionals in proffering psychological support as they feel helpless in the face of these narratives with unwelcome and uncomfortable endings. It is interesting that although clinical psychologists are best placed (given appropriate training and experience) to address the depression associated with chronic illness or physical disability, they were not called on by any of the individuals in this study nor did any access professional counsellors of any description. Again, this may have been due to the GP or consultant not suggesting any such referrals and, given the benefit that might have ensued, it is hard to avoid the conclusion that this must have been due either to insensitivity to the psychological needs of patients or to an implicit rationing of scarce NHS resources. No individual self-referred to private psychological care, but this could be due to a lack of financial resource to do so; or to a feeling that it would not be beneficial; or even that one should simply cope with these issues privately. Nursing prides itself on being a holistic profession and it is certainly heartening that amongst the experience of some participants, attention was paid by lymphoedema specialist nurses

to the psychological issues – albeit not by any active intervention but rather by an empathetic and supportive manner.

All participants were aware of their own body/self image to varying degrees and some, like Martyn, were anxious not to be defined as disabled.

> *I don't call it a disability – but more a condition that is disabling. …I don't find people looking at me a lot really (laughs) – I'm not desperately attractive (laughs) – so um, although I think that most people just don't notice – I'm sure most people don't notice – most people don't spend their time looking at my legs – um, occasionally I will see that someone has noticed – children actually – more observant – perhaps it's because they are nearer the ground (laughs). …I don't wear shorts very often, um, partly because I don't want to advertise the condition um, but the occasion doesn't arise, um, but no I'm – it's not – you know. If I was young and rather good looking – well then maybe you know (laughs). …Yes. It doesn't effect my relationship with my wife or my family, um no.*

There is a sense here that Martyn is trying hard to minimise this issue of body image whilst actually relaying a very strong sense of embarrassment and self-consciousness. I formed an impression that it was, at a subliminal or subconscious level, an issue of real importance to him despite his actual words. His language during much of the rest of the interview was quite crisp but I recorded in my field notes that at this point in his story, his paralinguistic patterns were different: he seemed to pause, make faces, laugh and use language fillers much more. It was these linguistic pointers that led me to suspect he harboured a paradoxical attitude to his body image. Body image consists of subjective expectations and

self perceptions and is inextricably linked to how an individual feels about 'self'. This, in turn, is linked to psychodynamic and psychosocial factors (White 2000). The impact of an illness or disability may result in physical change that threatens the 'self' and the body image. Body appearance, beliefs about the strengths and limitations of the body and how it functions within its environment may all undergo change (Shearsmith-Farthing 2001). These changes may be concealed by the individual or made apparent to those around them and the extent of the loss and grief associated with them will be linked to the value that the individual ascribes to the 'ideal' body concept. It may be that there are gender differences in the way that individuals deal with these issues: Martyn appeared to be trying to underplay these issues while Angela displayed a more overtly negative view of the impact on her life (especially as a young woman) but still reveals some ambivalence.

> *I didn't think much about it, except not being able to wear the same things that the other girls did. Couldn't wear stockings or high heel shoes and things like that. … I didn't wear any of that – I would've liked to but um, it wasn't an option – my mother wouldn't have bought them! I was used to the old socks and shoes and so I went the other way – sensible shoes with heavy socks on. The socks would keep the swelling down. You know they were quite heavy so you wouldn't have known whether there were ankles in there or not! …If I could get in those little shoes, I would. But I did have double socks in some shoes to keep them down, yes I did, and there was a fashion of bobby socks – do you remember bobby socks? Right, that was for me! … I wrapped them in things so they were kept down. … First of all it was just my foot then it came up to just the ankle. When I was 19 (I was a student by then) the other foot joined it. …So um, again, but then, I was more aware of it at that age. …Well, yes* [training to be] *a*

music teacher actually, because you have to be practical. In that atmosphere [the conservatoire in London, late fifties] – *the only thing anybody thinks about is your music – you forget about your ankles and the rest of it because you are in a more – should I say bohemian – you know the duffle coats and patchwork skirts, sweaters, earrings and so on. … I didn't try to be up with fashion – it weren't considered that smart - if anything we went the other way and dressed as a gypsy. … Oh small waisted, yes, small ankles – well they were absolutely the thing then. But we leaned heavily on what we were – we're pianists and musicians so our voices or talents – that was the thing that mattered – and physical appearance, although desirable, came in second. I don't know - that's how it was – I can remember one girl being very pretty and much sought after, but she was a rotten pianist, so we didn't bother very much! Compensation. I suspect I used it as compensation for what I couldn't have.*

There are contradictions in the above accounts which come through in the telling of the stories: on the one hand participants minimised the physical appearance of the condition but on the other they displayed or expressed a quiet yearning not to be different. A person's self image is the mental picture that depicts not only details that are potentially available to objective investigation by others, but also items that have been learned by that person about himself or herself, either from personal experiences or by internalizing the judgments of others (Shearsmith–Farthing 2001). Louise was affected by the hectoring and negative attitude of a 'friend'.

Yes, um there were 3 couples and we are very, very close the 6 of us – we get on very well and we go on holiday together and our children have all grown up together and they've got

on well for years, but one of my friends – he doesn't like my stocking – he doesn't cope with it. But …he gets very, you know, funny about it and if I put my leg up, if we're having an evening in or something, and I just get a stool so that my legs are elevated, he'll say 'oh you've got that horrid stocking thing on again', I said 'yes B I have actually', I um take the mickey, but um. … It's his problem and my problem – that's right, yes, and he doesn't seem to cope with things like that, but um. And he is a very huge, very overweight guy, you know (laughs) um, and he's got problems of his own really and so, but he just has to pick on someone sometimes, you know – I can be pick of the evening sometimes and … it's his problem – that's it, but it doesn't bother me.

Once again, I formed an impression that this was actually much more important, and hurtful, than Louise's words conveyed. Philip was also stung by the apparently humorous but actually rather harsh comments of others.

We mentioned at our earlier meeting that during the summer time I like to wear shorts and friends will comment, and say 'ooh, that's putting me off – can't you cover those up?' They are bantering, but it can still be hurtful.

These hurtful comments (particularly by people who are considered friends or close family members) are a clear threat to positive self-esteem and body image and consequently a threat to selfhood. The narratives of these participants reflect how chronic illness or disability is an assault on the self (Crossley 2000, Kleinman 1988, Frank 1995). Marie captured its impact in her comment about her 'infiltrated consciousness' (Smith & Sparkes 2008:227)

… the low self esteem has never really gone on a personal basis. It's gone on a professional basis, because if I am

> *standing up at* [name] *University giving a lecture I have got my professional persona and I am fine, I am strong in what I know and I can project myself in what I'm talking about and hopefully with humour but with insight as well. But as a person, that humiliation has always stayed there, you know, with me, underlying, all the time.*

Smith & Sparkes (2008:227) describe the damage done to an individual's emotional well-being in their comment on infiltrated consciousness: "…damage to a person's body, identity, and emotions, is created when a person internalises, as part of their understanding, other people's oppressive, dismissive or exploitative understandings of them, and then lose or fail to acquire a sense of themselves as worthy of full moral self-respect."

One participant, Belinda, showed signs of self-loathing in relation to her own body form which was, for me, reminiscent of the condition known as body dysmorphia. This label is usually applied to those who have an obsessive pre-occupation with their appearance and often loathe things about themselves that others do not notice or attach importance to (Arthur & Monnell 2007). Such people often seek out medical – even surgical – interventions to change their body features and, in this regard, they may suck clinicians into their own self-loathing. In this case, Belinda appeared to encourage and relish, to an unhealthy level, the many, and often radical, interventions suggested by ill-informed medics. On one occasion, when a surgeon was reluctant to undertake further surgery, she pleaded with him to continue:

> *And then a new surgeon said he would do the liposuction again and when he got my details "I'm not doing both legs – I'll just do one" and I said "Well, it's not much good just doing one" and he said "no – I'll only do one". I got right down to*

theatre and they were just about to give me the injection and he came running in "Stop, stop, stop!" He said "I can't do it!" And I said to him "Please do it" because I was there, you know, and he said "no I can't do it" he said, "I'm so frightened that I'll put the knife in and your leg will just explode".

Another way of characterising this desire for intervention might be to infer a deep wish to gain attention. Munchausen's syndrome has been described as a call for attention by seeking medical treatment for spurious illnesses or fabricated symptoms (Hamilton & Feldman 2006). It is clear that although Belinda's symptoms are not fabricated, she and her husband are very well read on the subject of lymphoedema treatment which makes their motives for agreeing to these potentially damaging treatments worthy of suspicion. Belinda also recounted a plethora of other conditions to which she had fallen prey and reeled off a lengthy list of healthcare professionals with whom she had had dealings. Hamilton & Feldman (2006) have suggested, in the context of Munchausen's, that the patient (or her carer) may have difficulty with self identity. The hospital is seen as a safe environment or a way to escape from everyday life and they hope to strengthen their own identity or get sympathy for themselves or the person that they care for by frequent healthcare interventions. The individual may have a very low self-image and be subjecting themselves to painful and intrusive medical procedures as a form of self-punishment. I recognise that I could, of course, be guilty of 'medicalising' behaviours which are not sinister at all and this may say more about my position in the medical model than anything else.

Most certainly, participants talked of the impact the lymphoedema had on their self esteem and the 'spill over' that this had into their

personal relationships – a factor that I discuss in the section that follows.

Relationship issues

Although the participants largely had very positive and supportive spouses or partners, there were reports of negative and abusive relationships and occasions when close personal relationships had foundered when confronted with the reality of this chronic and sometimes disfiguring condition. Louise felt particularly fortunate in having a very supportive partner.

> *…my partner's been fantastic – he was brilliant from day one and he has always supported me and I don't think he realises I'm wearing the stocking anymore, he just doesn't take any notice (laughs), um it doesn't bother him and I think it's probably because my legs never looked that awful. It doesn't look huge so it doesn't look as though I have a deformity of any description really, but, um it's never bothered him.*

Marie, by contrast, endured an abusive and corrosive relationship with her husband of 27 years.

> *…although it wasn't a happy marriage, it actually became abusive when my lymphoedema set in. That was because I wasn't the 8.5 stone perfect princess any more. You know, I was somebody who had put 4 stone on, had enormous legs and enormous feet and that was not attractive, or so I was constantly told. Um, so life changed. … He was always away and having fun and was very physically fit and here was I, the woman he had started calling, you know, the fat cow and you know, not wanting to go anywhere with me socially, and 'why can't you do this?' and 'you weren't like this when I married you' and 'why are you like this now?' and it was really silly,*

really silly I suppose. … that humiliation has always stayed there, you know, with me, underlying, all the time.

Gerry found that the diagnosis put an intolerable strain on his first important relationship.

I had got engaged to my first (another) girlfriend at university. But it didn't happen, we didn't get married, because she had real issues with it – because she thought a) I wasn't physically perfect as… you know. I couldn't go out and rave and the rest of it um, and b) what happens if I pass it on to our children? So that was an issue.

The responsibility of chronic illness is often shared with partners, parents, children and other carers and when the impairments (physical and social) that attend lymphoedema impinges on those peoples' lives it can elicit a variety of responses. For instance, there may be a positive and genuine support as shown by Louise's partner, above, or a rejection or even punishment as demonstrated by Gerry and Marie's former partners. Here there is evidence of stigmatising the person with the condition when they fail to conform and this resonates with Dewing's work (1997) on the socially acceptable body image in western societies. Even in Louise's case, we can discern a sense in which she explains her partner's acceptance as contingent on the fact that her legs were not 'awful' or 'deformed' in any way.

All participants, with the exception of Susan and Gerry, were parents and expressed concerns for their children and grandchildren and there was an element of guilt if the condition were to be passed through them to the next generation. Gerry's distress at his own situation led him to make a very difficult decision about having children of his own. He said:

I reached a point where I wasn't going to have kids, because if this was hereditary in any way, shape or form… Because when they started suggesting that it was hypoplastic then I would probably pass it on. So I was like, 'Right, that's that. Not going to father children'.

To be a parent is, for the majority of people, an expected part of a life narrative and to feel forced to decide not to be a parent when one would otherwise wish to be is, if not life shattering then certainly life changing. Erikson (1950) wrote in his seminal studies of the developmental life stages through which an individual could expect to pass and believed that each life stage is concerned with the resolution of a particular crisis, or a task that must be completed satisfactorily. Failure to do so would, according to Erikson; result in an individual having ongoing problems in life in relation to the issues that task is concerned with. For young adults, 'generativity versus stagnation' is the stage concerned with giving, with selfless care for others. It may, for many people, include becoming a parent and be given expression in caring for children. For Gerry then, his decision not to have children – forced upon him by his hypo-plastic lymphoedema and his fear of passing it to on to his children – may cause unresolved internal conflict.

For Marie, there was a suggestion of guilt as she had had her children before her lymphoedema was diagnosed and therefore the future die was cast. She turns this into a positive by using awareness and information as weapons in the battle with the condition and is confident that her experiences will act to remediate those of her grandchildren if they should develop the condition.

My sister who's a medic at A's hospital. She's got it just in her feet. My mother did too. My grandmother, my mother's mother – she had, you know, …I remember her having swollen legs and swollen feet. …Well, I think so, because a

concern I have had um, these grandchildren coming along, because looking at one of them, two of them actually, they are the same shape as I was at the same age. In fact if you show them black and white photographs they think – there are 3 siblings – one of them thought that the picture I had shown him of me as a child was actually his baby sister. So I worry that they might develop it. Obviously I'll be long gone, so at least what we have tried to do, both my brother [who also has lymphoedema] *and myself is channel all the information that we have got, give it to the kids and say 'be aware' if anything occurs.*

For Louise this guilt was turned backwards as her child shouldered the guilt for 'causing' the lymphoedema since her mother had first exhibited symptoms during pregnancy. She admitted that her daughter could be difficult:

I must admit my daughter – she is 12 and she always pushes things - but I think, hopefully, that's just the phase she is going through in her life and she's going through puberty and that sort of thing. But she gets very down in herself and sometimes she'll say 'I caused that, mum – it's me that did that'. And I say 'no you didn't' – you know. …and sometimes she just gets like 'well you didn't have to wear that stocking – it was only because I was in your womb' – and I think 'well no, it's not you' – and she does get upset sometimes, then she just keeps saying it – then I say 'yes it was you, fat baby!' (laughs). But no, it wasn't her fault and I don't blame her in the slightest, it could've been the other way round – it could've been him [Louise's first child, a son]*, so it's not – it just happens. …I have tried to make her think positive about that sort of thing.*

There is an implied contradiction within this narrative, in that whilst Louise seeks to reassure her daughter that she is not to blame; she cedes under pressure that it was caused by the fact that her daughter was a 'fat baby'. An impression that she does feel some subliminal resentment, at least at an emotional level if not on a rational level, is created by her admission.

Loss of career or leisure opportunities with resultant occupational deprivation

The occupational deprivation (Wilcock 2001) created by the physical and psychological limitations imposed by primary lymphoedema were profound, particularly for those participants with a creative, musical or dramatic orientation. Certainly for Gerry and Angela this created an enormous gap in their lives and a sense of under-achievement or lack of fulfilment. This reflects the research undertaken by Hagerdorn (2000) who demonstrated the impact of disability on occupation and the concomitant loss of self-esteem amongst those who could no longer pursue their previous activities. In the context of paid employment, Priestley (2003:133) has put it "Employment is significant in the construction of disability as a social category, since 'ability to work' is the primary criterion that states use to define who is disabled." The stories told here reveal that primary lymphoedema has deprived participants of preferred activities, employment and leisure. Many participants, including Gerry, Louise and Belinda had been very athletic before their lymphoedema manifested itself and there was a sadness that they could no longer engage in sport at their previous level. Gerry explained:

> *I had to stop playing sport because the last game I have ever played competitively was a doubles match of tennis, um, which happened about 3 days before this, and um, I used to run 5 miles a day, because I was fit, I was a dancer, for*

crying out loud, so I was pretty fit. I used to do workouts almost every day.

Louise had been a keen and competitive runner before her leg became swollen and effectively stopped her in her tracks.

So my leg was back to normal and I was always a very sporty person. I've always played netball and I've been a runner and all sorts of things like that. So I went back to my running and I probably had about 2 years of getting back into my sport and then one day, I did a road race, 10k, and when I came woke up the following morning, my leg was swollen absolutely huge – it had just blown from top to bottom. Yes, all the way down from groin to toes. So, I had to go to the GP and the hospital.

Belinda had had to stop engaging in sporting activity at quite a young age even though she enjoyed it tremendously.

I was doing a lot of athletics, high jump and running, I used to run for the school and they kept saying "you must've sprained your ankle" because my ankle was always swollen. …And it was fine all the time it was bandaged but when he took the bandages off and of course the swelling came back. … Well I just kept going like that, on and on, and when it got too swollen then we used that to get it down a bit. By bandaging. But they didn't know what was wrong with me! (laughs) … but as I say, they just thought it was sprained ankles, that I had a weak ankle and I wore an ankle bandage, the elastic ones you pull on. … I did [carry on with sport] *until I left school, started to play tennis afterwards, but the swelling gradually got worse and it just wasn't practical.*

It is interesting to note that Gerry remained very sad at what he saw as a diminution in his sense of self created by an inability to engage in sporting activity, while Angela and Louise were apparently more pragmatic: It could be that there is a gender issue at play here, since research demonstrates that women very often give up sporting activity once they take on gender role obligations, particularly those around childcare (Grovaerts 1985, Hagerdorn 2000). This may mean that they cope better than men with this occupational disruption in that it does not interfere with their sense of self. However, women are not a homogenous group and therefore generalisations about leisure and sport cannot be applied to all.

Whilst there was a sense of regret at this loss of sporting activity, Angela and Gerry felt a deep sadness at not being able to engage in creative activity. Smith & Sparkes (2008:226) suggest that disability creates "a form of social oppression involving the deprivation of opportunities connected to socially imposed structural barriers. …Furthermore, being deprived of such opportunities positions [an individual with acquired disability] in ways that help undermine his emotional, expressive lived body, thus helping to maintain the cycle of chaos and adding to the damage to his identities and psycho-emotional well-being". Angela, a talented pianist, is unable to sit at the piano (or anywhere) for any length of time since her legs ache and become painful and the swelling increases. She expressed her sadness and frustration.

> *Oh yes, well it was still bothering me. I could use nasty words but I won't. But it did restrict things – I mean I could not go to things, do things that I would've liked to go to. … Like concerts, like supper parties, in other words I retreated. … had to stop playing the piano you see, cut it out of my life, so that really closed the door on my music. …I didn't play for*

how many years. … I do think it's sad that you can't go to concerts - that's kind of sad.

The relationship of creativity to health and well-being is proposed in the literature as a strong one (Risteen Hasselkus 2002). "Creativity has the power to alter the darkness in our lives, whether we paint with it, draw with it, write with it, sing with it, work or play with it, or even just think with it" (Cohen 2000:200). Most people would wish to be active enough to stay fit and healthy and to undertake necessary self-care activities of life at a reasonable level of independence. But human beings also desire to engage in non-essential, creative, activity because these occupations make us feel good about ourselves and the world around us and help us to interpret the experiences we have had and translate them into entities that can be shared with, or explained to others (Hagerdorn 2000). Humans are the only animals who make things or do things which have no purpose other than a symbolic or aesthetic one. The dancing, running and piano-playing described in the quotes above are not essential activities but they are ones which have profound meaning for Gerry, Louise and Angela. Loss of the ability to do what we want, independently and effectively, damages our perceptions of the self and adversely affects our roles and relationships (Hagerdorn 2000). The previous vignettes demonstrate how Angela, Gerry and Louise feel impoverished by their reduced ability to engage in activities that are meaningful for them. Gerry encapsulated the powerful emotion of loss that attends occupational deprivation in his comment:

And he said 'how has it affected your life?' And the only way I can give it to you is as an image, because that is what it was like at the time, it was like I turned around and I looked down a corridor and all these doors in the corridor opened and I just looked in at all the things I had lost: I couldn't swim anymore,

swimming was a great delight. I couldn't run, running was so important, I mean if somebody said to me 'if we could restore your leg, what would be the first thing you would do?' I would say 'run' – I used to do 5 miles a day and most people don't do 5 miles unless they train for it, I used to do 5 miles a day as my thinking time. Being able to move properly as a dancer and all those sorts of things and all of the…. and all the sort of opportunities that the theatre had given me and the potential, which was gone. All those doors closed. And I broke down - which wasn't the intention - and I hadn't been feeling emotional up to that point.

An inability to follow a chosen career path or even to work at all may create a situation whereby individuals feel not only deprived but also their selfhood is challenged (Wilcock 2001). Individuals may wish to work for many reasons: to be part of a specific group which gives identity and status; to be self-supporting financially and thus gain security; to conform and contribute to society. Alternatively, individuals may feel that their self esteem is wrapped up in being in particular employment which utilises their individual skills and talents to the full. Work in its various forms is seen as being fundamental to the way society is structured and it plays an essential part in the lives of most people (College of Occupational Therapists 2007). Three of my participants, Susan, Angela and Gerry found that they could no longer pursue their preferred career options because they were simply impractical in that they exacerbated their physical symptoms. Susan explained how her working regime made her lymphoedema worse.

I carried on working – there was no question of me not working you know… the problems started because I then had another bad bout of cellulitis in the May of that year. …and it was so bad that I went into hospital and had intravenous

therapy – I was in hospital for a fortnight – and then I came out – and of course it all became all infected and swollen and I think there was some question of the possibility of MRSA, you know. …I went to see my GP when I came out of hospital …and when I saw him he said "you're in the wrong place and the wrong job with this sort of thing"… And on one occasion when he came up to visit my parents afterwards he'd say to my parents "S needs to stop working now for the sake of her leg. S will not work again if I have anything to do with it". I think in some ways it was a relief to me because I'd always been in small units and then by the time I got the cellulitis I was working in a big department. Working there I found it a bit of a problem getting around. …but you know walking around the big department on my feet all day it was making it worse.

Angela found that standing as a music teacher created difficulty and that even sitting for long periods became impossible.

I tried to go back to teaching aged about forty or forty-one – um I forget why. Part-time because I was a musician you see, but it just didn't work … it didn't work very well. The standing, you know. You have to stand to conduct and so on. Having realised I couldn't do that, I started into playing for various, no money involved, but getting involved in adult education where a piano was needed. And from that I gradually built up my private practice and peripatetic work in the school. And again, once you're doing that you're sitting long hours, and it was from then my lymphoedema started to bother me.

Gerry talked with considerable pride about his great achievement in becoming a stage manager at a very young age but his voice

changed tone (from high excited to slow disappointed) to reflect his feelings when he went on to explain that he had had to change career path.

> *So I went off at 19 to just be a lackey in a theatre company and never realised how much experience I got – then they said 'my gosh – we would like you to be an assistant stage manager'. A few weeks into being an assistant stage manager the producer came to me and said 'you'd better be stage manager because we can't find one'. So I was a stage manager aged nineteen in the theatre. Which was great. And the real, well, you say that some professional who is 20 years my junior and they say 'no, it's not possible!' So it's a thing of yesteryear – even though it was only 20 years ago…. I have tried lots of different things: I tried to get into film; I tried to get into TV; lots of jobs where you don't have to stand all day – whereas in theatre you have to stand up all day – but nothing came good.*

For some, compromises had to be made in career choice or trajectory because of the physical limitations imposed by their condition. Gerry exhibits considerable stoicism in explaining how he moved from his beloved theatre work into teaching.

> *So anyway, 1988 came around – went in as a chaplain on a youth camp for kids who had social disadvantages etc and um, somebody turned around to me and said 'you would be a brilliant secondary school teacher'. And um… I went and sat on the hill and had a chat with God, as is my way, and um, said 'oh alright, if this is a view'. …So I said 'well if this is a view – open the doors.' And I didn't even tell my family I was going to the interviews because I thought if this is something that crashes and burns then it is one more disappointment to them. And I went to the college, did the interviews, got*

offered the place on the spot… And, I said 'alright Lord, I assume this is your view' and I tottered off and did 4 years teacher training.

Half the participants - Marie, Gerry, Susan and Angela - were disconcerted by uncomfortable working environments (physical or attitudinal). Gerry's experiences encapsulated the difficulty others experienced when their employers failed to recognise or understand the challenges of living with lymphoedema. He revealed that:

Not all the kids at school know about it because I teach sitting down. Which my Head, who is new as of two years ago, he watched me teach one day and was like 'oh, you haven't got out of the chair once' – he didn't say that to my face, but he went back to the senior tutor and said 'we'll have to tell G, we can't have a teacher sit down for the entire lesson'. And it was like, aahh, we need to tell you.

In Susan's case, the intolerance of her limitations is especially surprising given that she was working in an NHS setting where more enlightened attitudes would be expected. She said:

*I was working in a big [*Radiography*] department. Working there I found it a bit of a problem getting around. And they knew this [*managers*] but it was well, like, the job has to be done.*

Occupational deprivation – an inability to realise one's creative, employment or social aspirations - was apparent among the group, with half the participants (Gerry, Susan, Angela and Marie) having to make a substantive career change or retire early on the grounds of ill-health, or to withdraw from an area of occupation which they had valued highly and which defined the essence of their being in an important manner. This was particularly frustrating when none of

those participants had received adequate treatment and, if they had been managed appropriately, might have been able to pursue their original ambitions and orientations. The participants all spoke of being fortunate in having interesting and engaging work which was important for self identity in that it allowed for self-development (for one this was as a mother and wife), and all had managed to adapt and compensate to the changed circumstances imposed by their condition. There were, however, still at least two (Gerry and Angela) who clearly mourned for the life that might have been and although not alienated from their current work, may have become estranged from their inner selves in the way described by Burkitt (2008:141) in his work on social selves. “Because most of us have to work to earn money and spend most of our time at work, the activities we are employed in determine to a large degree our capacities, skills, abilities, knowledge and self-identities. It is not simply falling under the category of some general form of individuality such as being a worker that is important for self-identity, but it is mainly the scope for self-development (or the lack of it) afforded by a person’s work that is important.”

Challenges in daily self management

The response to the impairments created by lymphoedema, and the extent to which it interfered with participants’ lives, depended on the meaning and significance that they attributed to the diagnosis and prognosis and their own particular familial, social and economic environment (Bradby 2009). Belinda, Gerry and Angela characterised having lymphoedema as a bleak and confounding situation while others placed less emphasis on the negative diagnosis and conceptualised it as of minor significance to their quality of life. Gerry clearly struggles to come to terms with his situation and expresses his anger quite forcefully.

It is a very grim thought – I would say still probably, er, I wouldn't say I have completely come to terms with it. …Am I at peaceful acceptance? Um, I couldn't, probably not say absolutely – I think there are still elements of denial. The worst time of all is first thing in the morning – that is the worst time – the first thing in the morning and when I'm lying in bed… I used to be a morning person, …but there is a choice every day, you know there's that 'got to get up, got to get up', but there is that little moment before I put on my toe sock, then the stocking, then the ankle support, then the 'giraffe' – that some days is harder than others, but there is always that moment of 'here we go'. And I would like to be mature enough and adult enough not to be angry and to just get on with it. But there are still problems and anger. And there are times especially at night when, you know, if the leg is not doing too well, because it does go up and down depending on busy-ness. There are times when I will take all the support kit off. OK. – yes, um, and I still think, still: [whispers] *'dear God, bloody hell!' …It'll be 20 years next year.*

Martyn, by contrast, seems to have accepted the constraints placed on him and characterises them as an inconvenience rather than a disaster. He admitted that he was:

… more inclined to use measures which recognise that the condition is not going to fundamentally change, and simply make my life easier by minimising the inconvenience it causes me. As I was saying, my main concern is to avoid cellulitis. My second concern is just, you know, to make the thing as unimportant (laughs) as possible. I don't want to be picky about lymphoedema all the time – it was only when my condition was first diagnosed that I was, you know, I used to

be quite pre-occupied by it. But I hardly give it a second thought now. (Martyn)

Cellulitis was a source of much fear to all participants who lived with constant risk of infection. As Woods (2007) has shown, if cellulitis is not recognised and treated rapidly, it can quickly lead to extreme illness and lengthy hospitalisation. Recovery from cellulitis can take several weeks and if it is not fully resolved can lead to frequent recurrence (Browse *et al.* 2003). All but one (Louise) of the participants had experienced severe episodes. Martyn felt that he had not been sufficiently prepared for the possibility of contracting cellulitis and its potential severity.

Um, you mentioned cellulitis – I have had that a couple of times. And I think if I've got a significant complaint about the doctors, the consultant – it's that they didn't warn me – perhaps because they didn't know – of the risk from cellulitis. But I'm prone to in-growing toe nails – and I twice contracted cellulitis because of in-growing toe nails and that was no joke. …I was very ill – it was very serious the first time round. I was kept in hospital for several weeks and pumped full of antibiotics – so that was a very nasty experience – and it's much worse than the lymphoedema itself. Um, the second attack was also brought on by the in-growing toe nails – not quite so severe, but I was still hospitalised.

Marie was unfortunate in having a bout of cellulitis which was resistant to treatment and she was hospitalised for a very long time at great personal and financial cost.

I had a very bad attack of cellulitis and septicaemia and was back in hospital, um, obviously I couldn't walk and it was a very difficult time. …It was quite a tough time. On and off for about a year. I suppose in and out for about a year.

The average number of episodes of cellulitis amongst participants was five, with one, Belinda, experiencing cellulitis two or three times a year. The cellulitis was severe enough to warrant several weeks in hospital for almost all participants and some had contracted Methicillin-resistant Staphylococcus aureas (MRSA) whilst in-patients. Susan experienced hospital-induced infection on top of an attack of cellulitis.

> *I carried on working right up till 1997 in actual fact and, then, as far as I was concerned, were when the problems started because I then had another bad bout of cellulitis in the May of that year. Same leg again, yes that's right – and it was so bad that I went into hospital and had intravenous therapy – I was in hospital for a fortnight – and then I came out – and of course it all became all infected and swollen and I think there was some question of the possibility of MRSA, you know.*

Belinda managed to avoid MRSA for many years. She explained:

> *I was there for about another month after that and they said 'really you'd be better off at home – because you'll only catch horrible things here. But then I got another very, very bad cellulitis and felt really ill. …I had had several attacks, but this was a really bad one and I just couldn't get out of bed I felt so ill… He said 'I hope it's a bacterial thing, but it might not be'. And it wasn't, it was MRSA. So after all those years that I'd been going, there it was, MRSA.*

The cost (in human terms, as well as time and finance) of treating cellulitis is difficult to estimate, but it is undoubtedly considerable. All participants who had been hospitalised (that is all except Louise) had been an in-patient for weeks (sometimes months) and this is extraordinary when most patients – even following complex surgical or medical interventions are currently routinely discharged within

24-48 hours (Bradby 2009). The NHS could actually save money by treating lymphoedema more effectively, and the savings could provide funding for preventative healthcare.

The pain and discomfort caused by lymphoedema varied in the whole group but some participants were severely restricted in their social and or working lives because of their heavy, swollen and painful limbs. Marie recalled a particularly distressing time:

> *Christmas 2004, we were going shopping in C and I couldn't walk from – you know the car park near the Theatre in the High Street? I just couldn't walk, and I cried because the pain was so bad in my right leg. I – this was awful, this business of not being able to walk, and it got worse to the point that on New Year, on New Year's Eve I had these terrible pains in the night.*

Angela described her dragging, aching pain and explained that it affected her ability both to walk and to sit for any length of time. This in turn, affected her leisure opportunities.

> *Because I hadn't been able to go for walks it bothered me more from then on. I'd like to walk still, and I try so much to walk, but 15 minutes is my absolute limit. …There are concerts to go to – supper parties things like that - and you know my legs start to hurt and it's agony sometimes to go out to dinner – to get my legs around the table, and they ache and they swell and they swell and they ache – it can be very nasty that can be. …It tingles and aches before I get off to sleep. …it's dragging when you are sitting with your legs under the table or in the concert hall – it's a dragging ache.*

Gerry was relieved that although the pain was very intense on occasions, it was at least intermittent.

> *Yes, it* [the pain] *was totally unique – completely – I still get it once in a blue moon in the groin, the upper thigh. Once in a blue moon I get it and I think 'oh it can't get any worse', but it passes.*

This pain and discomfort was reported as under or un-acknowledged amongst the medical profession and no participant had been referred to a pain clinic or given pain-relieving medication. One can only assume that GPs felt helpless to treat these symptoms or did not acknowledge the profound impact on everyday living created by constant pain. Chronic pain may destroy both the body and the mind, pressurising the person into changes in lifestyle (as shown in the narratives of Angela and Marie above), relationships and even personality (Turner *et al.* 1998). Those with chronic pain may lose the motivation to perform essential personal activities – as with Gerry who struggles to get out of bed in the morning. Further, they may also be unable to continue in domestic, work or social roles and may avoid close personal or sexual relationships which might cause further pain. In extreme cases, pain can alter behaviour with individuals becoming withdrawn, tearful, angry, depressed or demanding (Turner *et al.* 1998). The narratives clearly show that my participants experienced these challenges. People with other forms of chronic pain – such as Rheumatoid Arthritis – are routinely referred for specialist care but, once again, lymphoedema seems to have fallen below the 'sight line' of the healthcare professions – at least in the experience of those participants in this study.

Despite the importance of self-care in positive long-term prognosis (Woods 2007), all participants commented about the demands of self-care – often with little or no support from healthcare professionals – and many confessed that they were unable to keep

strictly to the recommended regime. Philip was typical in finding any excuse not to wear his uncomfortable garments.

> *But many, many sufferers of the primary lymphoedema do not like wearing the hosiery on their legs. Once you have had cellulitis – then at that time, you cannot put a stocking on there. That is a lovely excuse 'I shan't have to wear my stocking again will I?'*

A particular problem was putting on the compression garments since it can take considerable time, perhaps half an hour, to correctly apply and remove the garments. Doing so requires considerable hand strength, to grip and pull on the low-stretch fabric. The garments have to be worn all day and can only be worn once before they need hand washing to restore their shape and pressure-properties. They are expensive and most individuals are only prescribed, or can afford, two sets of garment which then only last for 6-8 months before they need replacing. Keeping the garments on during warm weather was also problematic as the discomfort created by wearing pressure garments or bandages can be considerable since they are very hot and can constrict movement and manipulation (Medical Education Partnership 2006).This supports the findings of a US study (Armer 2004) which found that individuals abandoned their pressure garments during summer weather. Angela understood that her legs were more swollen when she failed to wear her garments but could not bear to do so for the whole day. She explained:

> *They are so heavy and hot – especially in this weather. Um, I don't and I can't wear them all day, or very seldom, and eventually they will go in that crease and they have to come off, but if you wear them for half a day or as long as you can bear them, then the other half of the day, your legs will not*

come up so much. If you don't put them on in the morning, I mean this will come up by the evening.

There are also well documented problems with continued patient compliance since the garments must be worn all day and, in some cases all night, to optimise the therapeutic effect (Casley-Smith & Casley-Smith 1997, Twycross *et al.* 2000, Medical Education Partnership 2006, Woods 2007). Although some design modifications have been made by manufacturers, these have not solved the problems and many people simply cannot manage to continue wearing compression garments.

Many also found the skin care and exercise regimes taxing and commented on the time available for this level of self-care. This was particularly so for those participants who worked full-time as Gerry outlines in his comment:

I mean in my profession of work, I don't have time to do exercises, just too busy, you know, I hardly have time to eat at night.

Angela expressed her frustration at the sheer amount of care required to keep her skin healthy.

It's all so much really, the creaming and massaging and the exercise and that. It takes over your life. And the time to get on the stockings and you're supposed to do that before you get out of bed in the morning and it's all too much.

The skin undergoes significant changes in lymphoedema, and skin care is critical to prevent the deterioration of the condition. Skin becomes thicker and harder (fibrotic) with increased scaling (hyperkeratosis) and warty changes (papillomatosis). Compromised skin contributes to both psychological challenges, because of its

disfiguring appearance, and to physical problems, because of its reduced elasticity and tendency to infection. Skin with reduced elasticity will compromise lymph flow further and is more likely to suffer damage as stiff skin will not withstand pressure or shearing forces so easily as resilient healthy skin. Scaly skin tends to harbour greater amounts of micro-organisms – particularly fungal and bacterial agents which inevitably lead to associated infections (Browse *et al.* 2003). Oil based emollients soothe and hydrate the skin and should be used at least twice daily as part of a self management programme. They do, however, lead to a shortened life for pressure garments (and other clothing) as the oil affects the fabric by staining and stretching fibres (Casley-Smith & Casley-Smith 1997). When skin scale is excessive, it may need to be treated with salicylic ointments and antiseptic agents. Topical steroids can be used where skin is suppurating and smelly but have a thinning effect on skin and need to be used sparingly (Browse *et al.* 2003). Typically, an individual would need to spend at least twenty minutes at the beginning and end of each day gently massaging the emollients into the skin before donning pressure garments. For most people this represents a real challenge in self management and in reality many patients report non-compliance with the regime (Lymphoedema Support Network 2008b, British Lymphology Society 2008). Marie described the necessary routine as constant drudge and recalled how easy things were before her lymphoedema.

> *…and the other thing is the constant drudge associated with it. I remember, as a kid, thinking 'God, I'm going to miss that lecture' – jumping out of bed, splashing on the old face, putting on the jeans and clearing off!! Well now it's very sort of slow and steady, you know. Out comes the aqueous cream, on comes the sock and my grandchildren are fascinated you know, they come in and watch 'Grandma, why are you doing*

that?' and I said 'well, because my legs aren't quite what they should be'.

All were fearful to some extent of the future and what it would hold for them. Belinda, Angela, Marie and Louise could not put on their garments without the help of another, stronger, person and this reflects Armer's finding in her study (2004) which explored the grip strength required to position properly fitting garments. Participants, particularly those who were elderly like Angela, Philip and Susan were concerned that their physical condition might worsen and worried that when their supportive spouses or partners were no longer able to help them they may become dependent on a faceless or uncaring health and social services. This was a source of real anxiety and distress as Angela shows in her comment:

I don't want to be a burden and my husband has his own problems but if he couldn't help me I don't know how I would cope. The district nurses won't come so it'll be some untrained person or off into a home for me. I dread it, really I do.

Counter to these concerns, I found that the participants and their families were, in the main, resourceful and demonstrably quite able to take the initiative in relation to their condition and its management. They left me with a strong sense of their potential for pro-activity in demanding services once they are fully aware of what these can be. To assist them in this process, I purchased some newly published literature on best practice in lymphoedema and gave this to each participant as a thank you gift. My intention was both to express my gratitude at the giving of their time and also to ensure that they had access to up-to-date information on their condition and its management. I did not send this gift until several months had elapsed after they had approved the transcripts as I did

not wish to create any ethical issues by seeming to exert pressure on participants for their participation.

Summary

This chapter has explored the issues uncovered in this research and it has revealed some really startling findings about the challenge of living with primary lymphoedema. The study has graphically shown how individual lives are affected by chronic illness and how important it is to secure correct diagnosis and effective treatment. Difficulty in accessing healthcare support and services for those with lymphoedema has been highlighted as have the inadequate levels of professional education and knowledge about this condition. The physical and psychological challenges for participants have been detailed and the importance of the support of families and friends in coping has been confirmed. Loss of career or leisure opportunities with resultant occupational deprivation was a key issue for some participants while all face challenges in daily self management.

The next chapter will reflect on the research process and set out some conclusions and recommendations.

Figure 5 Summary of Information (age onset; diagnosis; treatment; cellulitis)

LO = Lymphoedema

Participant	Susan	Belinda	Gerry	Philip	Louise	Angela	Martyn	Marie
Current age	66yrs	69yrs	42yrs	72yrs	41yrs	72yrs deceased	56yrs	58yrs
Age at onset of Lymphoedema (LO)	21yrs	10yrs	18yrs	33yrs	28yrs	8yrs	39yrs	22yrs
Length of time until firm diagnosis made	25yrs	30yrs	4yrs	31yrs	4yrs	25yrs	1yr	15yrs
Has seen LO specialist for treatment	No	No	Yes	Yes	Yes	No	No	Yes
Is currently seeing LO specialist for active treatment	No	No	No	No	Yes	No	No	No
Number of episodes of cellulitis requiring hospitalisation	6	10	3	10	0	6	2	3

Notes:

Average time for accurate diagnosis = 17yrs post onset

Average number of hospital admissions for cellulitis = 5 per participant

*Only Louise is engaged in active treatment and has no history of cellulitis

Chapter 6
Reflections, Conclusions and Recommendations

Introduction

This study has employed biographical methods to reveal the devastating impact that primary lymphoedema has had on the lives of Susan, Belinda, Angela, Louise, Martyn, Gerry and Philip. Such an approach has allowed their stories of living with this condition and the challenges it represents to be heard for the first time. As such, new and revealing insights have been provided into the ways in which those with primary lymphoedema view the issues related to their condition. The literature review revealed the dearth of research which focuses on quality of life issues for people with primary lymphoedema and showed that much of what had been undertaken was from the perspective of healthcare professionals – not the individuals themselves. My overarching goal, therefore, was to allow the participants to have a voice and platform to speak from (Clarke 1998, Denzin 1989).

This chapter reflects on the research process and in so doing, includes comments on the strengths and limitations of the study; its methodology; the participants' experience of the process and my own personal experiences as a novice researcher. In connection with this, I acknowledge that the field researcher needs to be self-critical and self-aware, superimposing an 'outsider's' perspective on an 'insider's' view (Finlay 2003) in order to recognise my own subjective thoughts and feelings about the experiences of those with primary lymphoedema. I then move on to bring the strands of the study together and to draw some conclusions and recommendations for future practice and research.

Reflections

> *Maybe the lesson here was that Narcissus wasn't reflexive enough. Had he been more self-aware as opposed to mesmerised by the reflection of himself as an 'other', perhaps he would not have fallen in love with himself… (Finlay 2003:112)*

The qualitative paradigm holds as a central assumption that a profound understanding of the world can be gained through conversation and observation in natural settings (Denzin & Lincoln 2005). Certainly the approach proved to be an appropriate choice in the light of my findings where the previously unheard stories of those with primary lymphoedema were given voice. The study resonated for those who live with primary lymphoedema – both for those who took part in the study and the wider lymphoedema community. I was fortunate to gain the support of the Lymphoedema Support Network (LSN) in publicising my proposed study and following the publication of a short article in the LSN newsletter asking for participants, found that the response was quite overwhelming and unexpected. As stated, I received over 150 responses from individuals who wished to participate in the study to have the opportunity for their stories to be told. Many of them sent highly personal and emotional letters or emails thanking me for taking an interest in their plight by engaging with research in this area. Those who contacted me came from across the UK and also from Europe, United States, South Africa and Australia. Subsequent to this, the LSN then placed information about the study on their website and I wrote a follow-up article for their newsletter containing interim results. Again, I received many responses and indeed, continue to do so at the point of writing (more than 2 years after the study's recruitment phase closed). In addition, I have spoken at two LSN support group meetings and have been asked to

speak at another in the summer of 2009. The level of interest amongst healthcare professions was also heartening, with the professional journal '*Lymphoedema'* asking to reproduce my interim findings in a 2008 editorial; the British Lymphology Society printing an article in their 2009 Spring newsletter and most recently a television production company have asked for my assistance in the making of a documentary on primary lymphoedema. Such moves are indicative of the interest in the life-stories of those with primary lymphoedema once they are revealed. It is my hope that the study might serve as a pilot to seek out funding for a national, larger scale study to capture the experiences of a greater number of those with primary lymphoedema. This response from people who live with lymphoedema and the professionals who work most closely with them, lends considerable weight to my belief that this was a worthwhile and significant topic for research.

Although a notable strength of this study lies in the depth of detail captured, it is acknowledged that only small number of participants were included. Sample size in qualitative work has no predetermined formula and no 'power calculation' can advise the exact number. Instead, as Erben (1998:5) has pointed out the purpose of the study should dictate the sample size:

> *The exact size of any sample in qualitative work cannot be ascertained through quantitative methods. It is for this reason that it is all the more important that the consciously chosen sample must correspond to the overall aims of the study.*

Notwithstanding, eight is a very tiny proportion of those with primary lymphoedema and critics of the study might well argue that its findings are rendered marginal by the lack of generalisability to the wider population of those with primary lymphoedema. It was not my intention, however, to produce quantitative, generalisable findings - rather to allow the voices of those with lymphoedema to

speak for themselves. If there was some transferability (which does seem to be the case given the response I have had since publication of interim findings) then that is sufficient. For a study framed by the demands of an educational programme, there was a need to be pragmatic in planning for time available to undertake the work and finances available to underpin the activity. Although desirable, it would have been quite impractical, even unethical, to recruit a larger number of participants when I did not have the time available to undertake the massive tasks of transcription and analysis required. Even transcribing and analysing eight conversations proved a very time-consuming task. While it was advantageous to have recruited participants who lived within a reasonable travelling distance of my own base (i.e. the southeast of England), on reflection, this is something that I have regrets about insofar that I should have made it clear in my recruitment article that I was seeking participants from a fairly constrained geographical area (because of the time involved and cost of travelling). I failed to do so – not deliberately but thoughtlessly. Hence, there may be many people from other parts of the UK who would feel cheated if they knew that in reality they were never likely to be recruited to the study. Such an experience has led me to seek out funding for a further study in order to remedy this situation.

Biographical research has two particular strengths that apply to health care practice: firstly, it sets out an individual's story simply and straightforwardly which acts at an emotional level on the reader/listener and encourages empathy for the storyteller and their situation. Secondly, the use of auto/biography as a method was intended to be liberating for the participants and to allow their unique perspective on their lives to be shared by others.

Many who read the life stories are likely to feel some resonance with their own lives or those of their friends and families. For those who do not find a reflection of their own experience in the narratives, it is still the case that life histories may engage them in a greater understanding of lives that are very different to their own. Erben (1998) and Smith (2008) have written of the role of imagination in developing empathy and reading these accounts does allow imagination to be harnessed to this end. As Erben (1998) has argued, these stories can develop the capacity in the researcher for moral reasoning. Further, for me, the powerful emotional connection engendered by reading a 'real' biography has more affect than reading a fictional account of an eventful life. In this particular research, my intention was to use this study as a particularly powerful tool for lobbying for better services for people with primary lymphoedema. Statistical or number-driven accounts of the challenge of lymphoedema would probably not impact on the consciousness of those with the power to change and improve services (e.g. politicians, NHS managers, GPs, educationalists, nurses or therapists) but an account which affected them at an emotional level might have the ability to engender empathy. While the response from those healthcare professionals who have read of my study has been heartening, the real test lies in accessing powerful others who do not yet understand the issues facing those with primary lymphoedema but may be pivotal in improving services.

An ambition of the study was to give a platform for the participants to present their counter-narratives which might challenge the dominant medical perspective, but it was interesting to observe that the stories recounted were very much situated in the medical model and it may be the case that participants are so familiar with this dominant view that they did not recognise other possibilities. None,

for instance, gave any sense that this was anything other than a personal burden which medicine could and should be more able to 'cure'. No participant explored a social model view of their impairment and its attendant disability (Bury 2003).

The intention was to collaborate with the participants to produce and analyse their biographies. In reality, whilst I was able to reproduce their stories, none of the participants chose to make further comment or offer any interpretation on their stories to any large degree although the theme of catharsis was evident in some of the conversations after the tape had been turned off, when participants did offer that they had rarely been asked about their condition and that it was helpful to have been able to discuss it with someone who had an interest in their situation. Nonetheless, only one participant, Marie, gave form to her feelings by writing to me of her view that the process had been transformative. She commented in her letter on her past and present view:

> *Your research study has done several things: a) it was cathartic, I have never discussed my situation/feelings with anyone before; b) a weird determination to look after myself better has set in; c) I will do what I can for the lymphoedema cause. (Marie, personal correspondence, May 2007)*

An interesting point to note here concerns the power of researchers who are also therapists: I acknowledge that I have been actively trained to listen attentively as an occupational therapist to the narratives of patients in order to extract the maximum amount of assessment information that I can then use in formulating intervention approaches. It could perhaps be argued that my level of skill in this regard could render me overly powerful in participatory research, in that I could unwittingly use these skills to extract information which the participant would not normally reveal

of themselves or is intensely personal. They may later regret that they had allowed such sensitive information to be heard. In an extreme view, this could be construed as a form of symbolic violence (Smith 2008) in its creation of a selfish empathy.

Since there was no analysis from the participants, it could be argued that my voice took precedence and this raises an issue about ethical representation (Clarke 1998). However, I felt strongly the responsibility to represent the participants' life stories in an authentic and ethical way and to ensure that I treated them and their accounts with respect (Denzin & Lincoln 2005). It was disappointing, then, to have no formalised commentary from participants and I did consider whether I had inadvertently created an impression that they should merely confirm the conversation rather than collaborate in shaping the biographical account. On reflection, I feel confident that I gave all participants the opportunity and encouragement to add, amend, interpret and edit their stories. In seeking informed consent, I attempted (although perhaps rather inexpertly) to explain the philosophical values upon which biographical research is predicated: the storied nature of our lives; that we reveal ourselves through the stories we tell; that telling our stories can be transformative – even cathartic; that stories can and do change with each re-telling; and that it would be difficult to recapture the exact experience of the storytelling in the research text. I did, however, suggest that I would be responsible for the final choices about presentation of the text following my analysis and justified this by suggesting that in order to complete the thesis I would need an authoritative voice. In doing so, I may have set up a power relationship which negated my earlier urging for them to feel that they could contribute in an equal fashion. This may have been compounded by their knowledge of my background as a member of the healthcare professions. Nonetheless, I remain

slightly surprised at the somewhat unexpected willingness on the part of my participants to allow me free rein in presenting their stories. Apart from removing conversational oil (i.e. 'ums' and 'ahs'), correcting grammar and slightly re-phrasing clumsy sentences, none chose to do so and none questioned the presentation in any way. It may be that had I chosen to meet with the participants on other, subsequent occasions to discuss the biographies, they might have felt more able to become full collaborators. The simple act of being with someone on more than one occasion could create a bond which could enhance trust and an ability to share more information. I had not done this because I did not feel I had the time or skill to do so, but it is something that would, I feel, be worthwhile if repeating the study. Arvay (2003) has described a particularly collaborative approach to narrative analysis, describing her participants as co-investigators and working to make the relationship between researcher and participants absolutely equal in order to deal with these issues of voice and representation. My own study limited itself to asking participants to collaborate with me in the production of agreed biographies whereas Arvay provided her co-investigators with copies of their transcripts with a reading guide and asked them to analyse the transcripts independently ahead of an interpretive interview during which they share their analyses and, based on this, produce one blended text (Arvay 2003). There may be some disadvantage in this method, however, in that researchers may feel constrained in their analysis and the final text could become less powerful as a result. I might, for instance, have felt uncomfortable making comment on Belinda's apparent relish for medical intervention if I felt that it might produce distress for her and a potentially confrontational situation for me. Arvay (2003) was using her method with a group of women who were survivors and now protagonists in the battle against domestic violence. The method may be less powerful with

different groups – although I accept that my position as 'therapist' may be constraining my potential as 'researcher'. As my skills and experience as a researcher grow, I may feel more equipped to emulate this method in some measure and thus increase the participation and representation in subsequent interpretive studies.

When the participants narrated their biographical experiences, they offered one version of their stories that seemed to work in that moment of telling. Philip (whom I visited twice) told his diagnosis story slightly differently on each occasion. The first time he employed much humour in the telling, almost brushing it aside, but on the second telling, he seemed much more raw and angry about his situation. It may have been that talking about it to me on the first occasion had made him re-evaluate issues which he had hitherto kept submerged and made him acknowledge his distress. Or, of course, it could have been that he was in a different mood on my second visit.

Five of the eight participants (Belinda, Susan, Louise, Martyn and Philip) shared narratives about their condition without offering too much information about the rest of their lives. It could be argued that these accounts are illness narratives (Smith & Sparkes 2008) or autopathographies (Bradby 2009) which represented epiphanies – important transformative moments – in their lives (Denzin 1989). There seemed to be a great need to have these stories told as a way of redressing a situation which most saw as unnecessarily difficult. Whilst these accounts yielded very useful insights into living with lymphoedema, they were perhaps not what one might describe as 'true' auto/biographical accounts. Although all did deal with life issues and so I may be unnecessarily rigid in my interpretation of what constitutes an auto/biographical account. There were a few individuals who actually recounted their whole life

story (Angela, Gerry and Marie) and these accounts which deal with lives over time have given me a much greater opportunity to interpret their reactions to the challenge of living with, and adapting to primary lymphoedema - albeit at an inexpert level.

In seeking to understand each person's story more completely, it may have been preferable to present and analyse each biography separately within the text. But the word limitation made this impossible and as a result, I have written the discussion in relation to themes as suggested by Grbich (1999) and Denzin & Lincoln (2005). Inevitably, I have had to be selective and some information has been omitted. In doing so, I may have submerged individual voices into a more general voice – particularly since it was not possible to provide quotes from all eight participants when commenting on each theme.

My own journey through the research process has been an eventful one since during the period following the meetings with the participants; I became unwell with a condition which may predispose me to secondary lymphoedema. The poignancy of the stories took on a new meaning when viewed through this lens and I feel sure my own illness experience will have impacted in different ways on my relationship with the research. In one way, I have lost my status (if that is the correct word) as an impartial observer: I can no any longer claim to have a solely professional relationship with those who have lymphoedema. I am no longer 'other' but now the same. In another way, the experience has made me feel less powerful in my ambition to lobby on behalf of those with lymphoedema because, within my personal frame of reference, it is acceptable to do battle on behalf of others but not for oneself. Both these observations give an impression of a diminished role or self – and that is a very telling accompaniment to illness – but the

experience may also make my research activity stronger and more credible because I am now an 'insider'.

Despite these perceived shortcomings, the biographical narratives I have collected in the study tell a powerful story of the marginalisation of those with primary lymphoedema. Whilst simply giving a voice to their stories will not lead to an immediate transformation of services – albeit long overdue - it does challenge the privilege given to other conditions over lymphoedema and paves the way for further research.

Conclusions and recommendations

The study has demonstrated that people with primary lymphoedema have unacceptable difficulties in securing accurate and timely diagnosis and effective treatment within the NHS. The research has also revealed that levels of professional knowledge amongst those who should be able to diagnose and treat the condition are woefully inadequate. The participants' life stories vividly demonstrate psychological distress, challenges to personal relationships and disruption of chosen careers and lifestyle activities. In their telling, the participants have shared their concerns for the future and revealed their diminishing ability to cope with the continuing management of their condition. Moreover, their stories illustrate that individuals with primary lymphoedema are living with unacknowledged physical pain and constant fear of cellulitis.

Given these conclusions, the following recommendations are intended both to enhance the life experience of those with primary lymphoedema and to advance research methods and future research activity in the field. Many of these recommendations

revolve around an increase in education concerning lymphoedema which is, I believe, pivotal to the enhancement of services.

1. The autobiographical nature of this study has been successful in revealing the voices of those with primary lymphoedema and is therefore recommended as a useful method in future qualitative healthcare research which has, in the main, tended previously to ignore this important issue.

2. Medical students require better education concerning lymphoedema during their initial training so as to enable them to recognise and diagnose the condition with greater facility. Post-graduate training for GP's should include lymphoedema awareness since they have a particular role as gatekeepers to services.

3. Consultant dermatologists, it is argued, should be the first point of referral from GP's for management advice and long-term supervision of people with lymphoedema. To enable this to happen, dermatologists should be able to access suitable post-qualifying education.

4. Rehabilitation therapists (such as occupational therapists and physiotherapists) and nurses should be equipped to routinely manage those with lymphoedema on a long-term basis by incorporating manual lymphatic drainage (MLD) training into both pre and post-qualifying education programmes.

5. Individuals with lymphoedema should be routinely referred to community rehabilitation and nursing services so that they can be supported in their self-management processes. Where people experience difficulty in physically managing their condition (e.g.

donning and doffing compression garments) they should be referred for home care services to prevent their condition worsening.

6. The psychological concerns of people with primary lymphoedema should be better acknowledged and referral to counselling and support agencies should be facilitated where necessary and desired. In connection with this, individuals with primary lymphoedema should be able to access genetic counselling services and, where necessary, pain management clinics.

7. Occupational therapists should be equipped to give information and advice on work environments and leisure activities including how they can be adapted to enable those with primary lymphoedema to continue with their chosen lifestyle and career choices.

8. Commissioners of healthcare services and education should be targeted to increase their awareness of lymphoedema and the challenges it provides. Since there is no formalised training for those who commission services in the NHS, this recommendation might be best achieved by publishing information pieces in the 'Health Services Journal' or equivalent.

9. Information and education – both for those who live with primary lymphoedema and for those involved in their care and support – needs to be more readily available and accessible through the medium of the GP's surgery or local pharmacies which are the first port of call for most with healthcare queries.

10. The prevention of cellulitis should be the focus of greater attention from healthcare providers both to decrease the pain and

suffering of those with primary lymphoedema and to reduce the costs of treating it.

Future Research

The study outcomes – through the stories shared – have immediate relevance for those who live with primary lymphoedema and for those who work in healthcare professions. Indeed it would be valuable for the study to be replicated with a larger, national group to provide an even more robust exploration of the challenges for the many who live with primary lymphoedema and who have not, to date, been able to share their experiences through the telling of their life stories. This could establish the extent to which the findings of this study reflect the life experiences of the wider primary lymphoedema population.

Given that this study has revealed that cellulitis rates were very high – especially for those who were not actively managed - a study which explores the correlation between active management of lymphoedema and the incidence of cellulitis should be undertaken. This could provide useful information for those responsible for commissioning health services about the opportunity cost of cellulitis care.

Epilogue

Daniel Lambert

Introduction

This thesis has been about unknown stories and has sought to cast light on the hidden lives of those with primary lymphoedema. Two of its central findings were that individuals with lymphoedema struggle to find a diagnosis and that they often experience problems around self and body image.

During the writing of the thesis, I came across the fascinating story of a man, Daniel Lambert, who lived in the 18th century and whose life story had been recorded because of his enormous proportions and the undignified manner of his death. Whilst staying in the George Hotel in Stamford, Lincolnshire, my attention was captured by a portrait of a man with a body shape that looked remarkably like those of the people whom I knew to have lymphoedema. Alongside the portrait was a short piece about Mr Lambert which further intrigued me. I came to realise, after undertaking some research into his life history, that his unknown story was that he suffered (in all likelihood) from primary lymphoedema and very probably died from cellulitis. As an adjunct to the other unknown stories which this thesis has chronicled, I would like to give a summary here of Daniel Lambert's life - seen through the lens of primary lymphoedema.

Daniel's story

Daniel Lambert was born in Leicester in 1770 and was the son of the city's 'Bridewell' gaol-keeper. As a young man, he was very fit and was possessed of an "uncommonly healthy disposition" (Dixon,

1809:19). He enjoyed walking and field sports including horse-riding and taught children in Leicester to swim. He bred cocks and dogs throughout his life and this activity supplemented his income. At fourteen years of age, he undertook an apprenticeship in engraving with a Birmingham jewellery company, Taylor & Co., but after the usual seven years (now aged twenty-one years) he returned to Leicester to assist in running the gaol with his father and then, on his father's death, took over the role. It was said by the chief inspector of prisons, James Neale, in his 1803 annual report that Daniel Lambert was 'improper' for this role being excessively kind to the inmates and being held in very high regard by them (Courtney 2001).

It was at this time that his normal weight began to increase substantially even though it is recorded that he did not drink alcohol and ate only one dish at mealtimes. At the time, and in later accounts of his life, this weight gain was explained by the more sedentary lifestyle associated with becoming the gaol-keeper – although this seems no less sedentary than jewellery engraving and Daniel continued to engage in sports activities until five years before his death at thirty-nine years of age. Along with the pictorial representations of Daniel, this timing of the onset of symptoms immediately led me to suspect primary lymphoedema: Daniel's onset of weight gain (swelling) occurred at a typical time for one who has Miege's syndrome, which is between puberty and fifty years (but most typically in the late teens and early twenties) and which leads most often to swelling of the lower limbs but can affect upper limbs, face, neck, genitalia and torso. Miege's syndrome is sometimes linked with autosomal dominant pattern of inheritance, which means that it can occur in either gender and only needs one parent who carries the gene to pass it on. In one account, the author notes that Daniel had an aunt and an uncle who were very

heavy, although his own parents and siblings were not overweight (Courtney 2001). This reinforces my suspicion of a chromosomal abnormality leading to primary lymphoedema. In 1793, aged twenty-three years, Daniel weighed 32 stone. A little over ten years later in 1804 - and despite many accounts of his abstemiousness and being on a strict diet - Daniel had increased in weight to 49 stone. Primary lymphoedema that remains untreated can lead to immense proportions known as elephantiasis – even when the diet is not excessive and, again, there are clues in these accounts which suggest it as a contributory factor in Daniel's great bulk.

By this time, his size caused him considerable difficulties in mobility and in practical matters such as clothing and furniture. He had to have several servants to assist him in daily self-care and even to have reinforced carriages built to allow him to travel. The financial costs of maintaining this lifestyle led him to the practice of exhibiting himself as a natural curiosity – something that Daniel found difficult and most undignified. When not actually showing himself for money, he resented people who were plainly just curious about his astonishing size and there are several accounts of him rejecting visitors and keeping himself very private. He took up residence in London (and later toured the country) and charged a shilling (five pence) for visitors to look at his body. This was a great amount of money at that time and it is suggested that Daniel charged a large amount in order to deter the vulgar (although that probably just means he attracted wealthy vulgar people).

Sometimes his friends would offend him as in one instance when, knowing that he disliked being weighed, they contrived to squeeze him sideways into a carriage and lead him over a weighing machine. This insensitive act apparently caused him great mortification and

reveals difficulty with body image and perhaps relationships. He never married nor had children despite his very good nature and kind disposition and, again, I would speculate that this might have been linked with negative body image and low self esteem.

One account is very interesting in that it describes Daniel's general good health in some detail but mentions briefly that the only ill-health he endured was inflammatory attacks:

> *During his residence in London, Mr Lambert found himself in no wise affected by the change of air. Unless we ought to attribute to that cause an occasional momentary trifling depression of spirits in a morning, such as he felt on his recovery from inflammatory attacks, which were the only kind of indisposition he ever remembered to have experienced.* (Dixon 1809:21)

These inflammatory attacks are very likely to have been lymphoedema-related cellulitis. Untreated by antibiotics, these attacks would lead to greater damage to the lymphatics and a downward spiral of swelling. In the end it could have been cellulitis which killed Daniel as his demise was sudden and unexpected coming on over two days when he was in Stamford intending to show himself to the wealthy visitors at the race meeting held there every summer. This would be a typical presentation of cellulitis as described by those with primary lymphoedema and left untreated, even today, would be life-threatening in causing systemic septicaemia.

There had to be special arrangements to remove Daniel's corpse from the Inn where he was lodging before he died by demolishing a whole wall. This was because his body could not be taken through the door or window without turning him sideways which was felt to

be inappropriate. It is interesting to note this courteous attention to Daniel's dignity. A purpose built coffin with wheels was used. At the burial site, a gentle slope was dug in order to wheel the bulky coffin into position. It took twenty men to complete this burial process. His gravestone, in St Martin's churchyard, was paid for by Daniel's friends in Leicester and it is recorded there that he weighed 52 stone and 11 pounds and that his body and leg had a circumference of 9' 4" and 3' 1" respectively.

A genial, gentle person; Daniel Lambert was also weary of his great proportions and his size undoubtedly shortened and probably blighted his life. The similarities in the narratives about Daniel to some of those unfolded by my participants in this study are marked: Daniel may, therefore, perhaps have been another person who struggled with undiagnosed and untreated primary lymphoedema.

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Appendix 1

Primary Lymphoedema: Lymphatics and the Management of the Condition

The Lymphatic System

The initial lymphatics are highly permeable lymph capillaries similar to venous capillaries but they have opening junctions between the endothelial cells. They are anchored to surrounding connective tissue by fine filaments. During the contraction of skeletal muscle, these fine strands pull open the lymph capillaries allowing protein, other large particles and cell debris to pass through into the lymph system (Berne & Levy 1996). The openings allow the initial lymphatics to perform as inlet valves - filling when total tissue pressures are low. The initial lymphatics discharge into the collecting lymphatics which have smooth muscle and move with a peristaltic action. This action forces the lymph centrally and through a series of lymph nodes. The nodes contain high concentrations of lymphocytes and macrophage cells which destroy protein, bacteria and other cells which are potentially injurious to the body (Woods 2007). The filtered lymph fluid is finally returned to the blood via the lympho-venous communications. These are many, and have not been fully described by physiologists (Foldi 1977, Casley-Smith & Casley-Smith 1997, Woods 2007) but the major junctions occur at the thoracic duct, which drains the lower extremities and the upper left quadrant of the body; and the right lymphatic duct, which drains the upper right quadrant of the body. Both these ducts pump the lymph into the great veins of the neck (Browse *et al.* 2003).

Lymphoedema is characterised by high protein levels. If the initial lymphatic vessels cannot open (for whatever reason) to allow the large protein molecules to pass out of the interstitium, these proteins rapidly build up in the interstitium and lead to a change in colloidal osmotic pressures. When those pressures increase, a concomitant build up of excess fluid and swelling occurs as the body seeks homeostasis (Casley-Smith & Casley-Smith 1997). This is the basis for the characteristic swelling in lymphoedema. Because

these rich levels of protein also provide an environment on which bacteria and other mutant cells can feed, a constant risk of infection - both fungal and bacterial - is present (Browse *et al.* 2003). A frequently reported feature of lymphoedema is the development of acute bacterial infection known as cellulitis. Episodes are characterised by a sudden onset of systemic symptoms such as fever, shivering, headaches and nausea which are associated with localised symptoms of pain, rash, inflammation and increased swelling (Woods 2007). If not recognised and treated rapidly, cellulitis can quickly lead to extreme illness and hospitalisation. Recovery can take several days or weeks – at great financial cost to the NHS and personal cost to the individual – and if the episode is not fully resolved can lead to frequent recurrence (Woods 2007). Lymphoedema is characterised by high protein levels. If the initial lymphatic vessels cannot open (for whatever reason) to allow the large protein molecules to pass out of the interstitium, these proteins rapidly build up in the interstitium and lead to a change in colloidal osmotic pressures. When those pressures increase, a concomitant build up of excess fluid and swelling occurs as the body seeks homeostasis (Casley-Smith & Casley-Smith 1997). This is the basis for the characteristic swelling in lymphoedema. Because these rich levels of protein also provide an environment on which bacteria and other mutant cells can feed, a constant risk of infection - both fungal and bacterial - is present (Browse *et al.* 2003). A frequently reported feature of lymphoedema is the development of acute bacterial infection known as cellulitis. Episodes are characterised by a sudden onset of systemic symptoms such as fever, shivering, headaches and nausea which are associated with localised symptoms of pain, rash, inflammation and increased swelling (Woods 2007). If not recognised and treated rapidly, cellulitis can quickly lead to extreme illness and hospitalisation. Recovery can take several days or weeks – at great financial cost to

the NHS and personal cost to the individual – and if the episode is not fully resolved can lead to frequent recurrence (Woods 2007).

Current interventions and the evidence base for their use

While there is a distinct frailty in the evidence base for the basis of treatment in lymphoedema and a dearth of research into the efficacy of interventions for its management, there is a body of literature on best practice in this area (Twycross *et al.* 2000, Browse *et al.* 2003, Medical Education Partnership 2006, Woods 2007).

Medical interventions include the use of drugs, particularly antibiotics, diuretics and benzo-pyrones; low-level laser therapy; liposuction and surgery.

Antibiotics are used to control and prevent cellulitis in lymphoedema and are essential to prevent further skin damage that can predispose to recurrent attacks (British Lymphology Society & Lymphoedema Support Network 2006) Diuretics are sometimes prescribed when a medical doctor believes the oedema to be uncomplicated or has not come across lymphoedema before (Twycross *et al.* 2001). Diuretics work by removing fluids from the interstitial spaces and returning it to the capillary (venous) system rather than through the lymphatic system. Patients will usually experience a reduction in the swelling and may be pleased with the results of the use of diuretics in the short term (Browse *et al.* 2003). However, what diuretics cannot do is filter the fluid and reduce the protein level in the interstitium. This can quickly lead to complications in the guise of bacterial infection. High protein levels create a perfect environment for bacteria to grow and spread and cellulitis is dangerous and sometimes life-threatening (Twycross *et al.* 2001). In primary lymphoedema, recurring episodes of cellulitis

will, in themselves, further damage the lymphatics and will compound the situation for the individual (Medical Education Partnership 2006). In some limited studies, Benzo-pyrones have been shown to reduce swelling; reduce pain; reverse loss of function; increase oxygenation levels in tissues; increase rates of wound healing; reduce chronic inflammation and reduce excess fibrosis (Casley-Smith & Casley-Smith 1986). They act by creating an increase in the numbers of macrophage cells in the body which act to ingest excess protein in the interstitium. It is suggested by the Casley-Smiths (1986, 1997) that once protein levels have been brought under control, a reduction in the colloidal osmotic pressure occurs as the body moves towards homeostasis. This leads to a parallel reduction in the abnormal accumulation of fluid and hence swelling. Benzo-pyrones do not offer a cure for the underlying condition but they ameliorate it by providing an alternative for the removal of protein and water from the interstitial tissues when the lymphatic system has been overwhelmed. However, the literature reveals that there are hepatic risks (albeit small) associated with the use of these drugs and they have been withdrawn in their oral form in most developed nations although the topical form is still widely available (Plumley & Waters 2003). There is some evidence which contradicts the claims made for the benzo-pyrones: Mortimer *et al.* (1995) and Taylor *et al.* (1993) conducted double-blind, randomised testing which sought to replicate the findings of the Casley-Smith's Australian study (1986) but neither found that the benzo-pyrones had a reducing effect. The methodology, however, for these two more recent studies was not identical to the original study which calls into question their ability to qualify the earlier research. There is certainly a need for a greater evidence base – especially as the original research is now more than twenty years old. It is noteworthy that there has been relatively little investigation by pharmaceutical companies of the efficacy of these

drugs and this may be linked to the fact that the major ‘market’ is in palliative care settings or third world countries where profit margins are likely to be low. It will be interesting to observe whether this stance is maintained as the number of people living in the developed world who are surviving cancer grows along with the number experiencing secondary lymphoedema as a result of oncology treatments.

It has been suggested by Piller & Thelander (1996) that low-level laser treatment might stimulate the development of new lymphatic tissue. This may be particularly useful where the lymphoedema is secondary to trauma where tissue damage or scarring has occurred. In an Australian pilot study, 10 patients were reported to show significant improvements with a 41 per cent reduction of oedema at 6 months (Piller & Thelander, 1996). However, this study is too small to be conclusive and a larger scale research trial is necessary before any conclusions can be drawn. Further, it is unclear whether the improvements are from new lymphatic growth stimulated by the laser treatment, new lymphatic growth occurring spontaneously, a reduction in fibrosis such as that claimed for ultrasound treatment, some other form of concurrent treatment or some other, unknown, effect.

Surgery is advocated as a last choice for those with specific and distressing symptoms, e.g. eyelid or genital swelling. It may also be used to de-bulk grossly swollen and infected limbs. Swollen and engorged areas are removed and the surface areas remodelled - sometimes using skin grafts. Although it may seem persuasive, surgery does not seem to have a substantially greater effect than conservative methods and it is claimed that it can further damage an already overwhelmed lymphatic system and create even greater disability and disfigurement in the years following the surgery

(Casley-Smith & Casley-Smith 1997, Mortimer 1997). Brorson (2000), writing in the context of lymphoedema of the arm following breast cancer surgery, has claimed that specialist surgical liposuction has the potential to reduce lymphoedematous swelling. Conventional liposuction usually seeks to remove adipose (fatty) tissue - but some delicate lymphatic tissue will always be removed at the same time by large, imprecise needles. Brorson uses specially designed fine needles which he claims, when used skilfully, can remove lymph accumulations in the interstitium without damaging the remaining lymphatics (Brorson 2000). This is contested by the Lymphoedema Association of Australia (LAA) (2003) who have queried its safety - claiming that in untrained hands it could prove damaging to those with lymphoedema since it is likely to also remove remaining lymph tissue thus worsening the condition. The LAA (2003) have also suggested that conventional liposuction – when used for cosmetic purposes - may even cause lymphoedema in previously healthy people by removing healthy lymph tissue.

Therapy interventions offer the best means of managing lymphoedema and include the use of compression therapy; manual lymphatic drainage (MLD); skin care; exercise; kinesio taping and hyperbaric therapy (medical Education Partnership 2006).

Compression therapy is the application of gentle external pressure to the body by the use of multi-layer bandaging or specifically designed low-stretch pressure garments. It provides resistance against muscle action and so aids in the pumping of lymph fluid into the initial lymphatics (Twycross *et al.* 2000). The pressure used should not be so strong that it actually flattens the delicate lymphatic structures otherwise it is counterproductive. Therefore elastic or high-pressure garments are contraindicated (King 1997)

and healthcare practitioners should have specialist training before using this treatment method (MLD UK 2008, British Lymphology Society 2008, Medical Education Partnership 2006). Bandages or garments are used to prevent swelling from occurring in the first place in a vulnerable person, e.g. one who has just had surgery or radiation therapy for cancer (Medical Education Partnership 2006, Woods 2007). It can also be used to maintain the reductions provided by other treatment methods (Medical Education Partnership 2006, Woods 2007). The choice of compression bandages or garments is often to do with comfort and convenience. Bandages have an advantage in that they are often more comfortable to wear (not so hot or constricting) but require considerable time and expertise to put them on properly (King 1997). With extra padding they can be more appropriate for grossly swollen limbs of irregular shape or with bulging deformities (Casley-Smith & Casley-Smith 1997, Twycross *et al.* 2000, Woods 2007). Garments are easier for patients to apply once they have been measured for accurately, and they provide uniform pressure when new and clean. The pressure of fluid (hydrostatic pressure) in both venous and lymphatic vessels is greatest distally in the limbs and gradually reduces towards the proximal end of a limb. Compression garments can replicate this graduated pressure and thus prevent the build up of lymphatic fluid – leading to a bulging and fibrotic cuff - at the proximal end of the compression garment (King 1997, Medical Education Partnership 2006). Although this intervention appears straightforward and effective, in fact, the discomfort created by wearing pressure garments or bandages can be considerable since they are very hot and can constrict movement and manipulation (Medical Education Partnership 2006). There are also well documented problems with continued patient compliance since the garments must be worn all day and, in some cases all night, to optimise the therapeutic effect (Casley-Smith & Casley-

Smith 1997, Twycross *et al.* 2000, Medical Education Partnership 2006, Woods 2007). Although some design modifications have been made by manufacturers, these have not solved the problems and many people simply cannot manage to continue wearing the garments. It can take considerable time, perhaps half an hour, to correctly apply and remove the garments. The garments have to be worn all day and can only be worn once before they need hand washing to restore their shape and pressure-properties. They are expensive and most individuals are only prescribed, or can afford, two sets of garment which then only last for 6-8 months before they need replacing. The use of pressure garments is also complicated in that many patients and health care practitioners may not understand the differences between low stretch and elastic compression in the treatment of lymphoedema and damage can be caused to an already compromised lymph system by careless prescribing or application of inappropriate elastic hosiery. Many GPs, community nurses and podiatrists prescribe or apply elastic stockings for the control of conventional oedema and for leg ulcer control but this can be disastrous if used on a lymphoedematous limb as it can lead to further damage to delicate lymph vessels, an unsightly and painful swelling at the margin of the elastic sleeve/stocking, development of lymphoedema in an adjacent area (e.g. genital or facial areas) and an increased risk of cellulitis as proteins build up in the over-compressed areas (Twycross *et al.* 2000, Medical Education Partnership 2006, Woods 2007). When the elastic garment is removed, the limb will become even more swollen than it had been previously.

Manual lymphatic drainage (MLD) was introduced as early as 1892 by Winiwater and refined by Vodder and Leduc in the 1930s (Casley-Smith & Casley-Smith 1997), but fell out of use for many decades because of interruption of the war years, which tended to

stop therapy developments, and because of an emphasis on more scientific, often pharmacological treatments as these were newly developed. Since the 1970s, it has gradually regained recognition for its effectiveness, particularly in mainland Europe, Australia and the US (Foldi 1994, Casley-Smith & Casley-Smith 1997). MLD encompasses a special form of lymphatic massage that seeks to evacuate lymph accumulations from the central regions of the body first and thus to give the accumulated lymph in the periphery a place to drain to. The massage uses the application of gentle, stroking pressure over the affected limb. It takes into account the complex anatomical structure of the lymph system to direct lymph fluid and large molecules, such as proteins, from the periphery to the centre. Typically, and ideally, the patient will have daily massage treatments, of about 1-2 hours duration, over a period of 3-4 weeks in order to achieve maximum reductions in swelling. Each massage is followed with special bandaging techniques (as outlined above) and exercise regimes in order to maintain these reductions day to day. Assessment for, and prescription of, permanent pressure garments follows up the active phase of treatment so as to maintain the reductions achieved, and to re-model the limb. This re-modelling process can take six months or more. A consensus currently exists amongst the experts in the field of lymphoedema that MLD should be an integral part of the treatment offered to those who have lymphoedema (Twycross *et al.* 2001, Franks & Moffatt 2003, Piller 2004, Medical Education Partnership 2006, Mortimer & Todd 2007, Woods 2007) although Browse (2003) has questioned its importance, proffering the opinion that compression bandaging alone will yield similar results. Those who believe in the efficacy of MLD base their belief on an emerging evidence base and several successful, documented case studies (Casley-Smith & Casley-Smith 1997, Mortimer 1997, Twycross *et al.* 2001, Medical Education Partnership 2006, Woods 2007) but it is

important to note here that both these counter claims are unsubstantiated by robust evidence, since research in this field has been minimal. Although the costs of setting up MLD services in mainstream therapy settings may initially be high, if the claims for its efficacy are substantiated, the benefits for patients could be enormous. Nonetheless, the current ability of healthcare professionals to deliver this gold-standard treatment is questionable with less than 500 fully trained and registered MLD Therapists based in the UK (MLD UK 2009).

Another form of mechanical massage therapy, Pneumatic Compression Therapy, was introduced in the 1970s as an adjunct – or possibly even an alternative – to therapist intervention (Allenby *et al.* 1973). It involves the use of an air filled sleeve with the air pressure controlled by means of an attached pump. The pumps apply either static or intermittent compression and the total pressure applied can be regulated (Disabled Living Foundation 2009). The effect is a massage-like compression of the superficial tissues. There is some debate in the literature concerning the use of pneumatic compression pumps, with suggestions of potential for harm expressed forcefully in the Australian literature (Lymphoedema Association of Australia 2008, Casley-Smith & Casley-Smith 1997). Complications reported by Casley-Smith & Casley-Smith include:

- Raising of protein levels still further in the lymphoedematous limbs by forcing water only into the blood whilst protein remains trapped.
- Creating an area of fibrous tissue around the region proximal to the sleeve of the pump if fluids cannot drain further along the system.
- This may be further compounded if surrounding lymph tissue is overwhelmed and could lead to development of lymphoedema in

a previously unaffected region e.g. typically genital or facial regions.

- The lymphatic vessels – already vulnerable – may be damaged by the high pressures exerted by the pumps.
- Excess pressure can cause small fistulae to form at the surface of the skin through which lymph fluid leaks creating a risk for infection.
- The pumps themselves may spread infection from one patient to another.

These negative findings should be balanced against some evidence for their successful use (Bunce *et al.* 1994) and it is certainly the case that many patients anecdotally report an easing of symptoms and improvements in limb volumes (Lymphoedema Support Network 2002). What is concerning is that patients may be experiencing short term relief only to find that they are confronted by long-term problems associated with the use of these pumps. In the UK, despite the negative evidence that exists, pneumatic compression pumps are recommended and used with little reference to this debate (British Lymphology Society 2003, Turner *et al.* 1998). It may be that the use of pneumatic compression pumps is more economically viable than the intensive therapist involvement required for successful MLD, and this alone might create an imperative for their use in the UK. However, the absence of an evidence base on which to base clinical judgements creates a situation in which people with lymphoedema may not be receiving optimal treatment and advice – and as a consequence be directed towards a treatment which could cause them harm. Further, there are very few longitudinal and reliable studies of their use. Nonetheless, pneumatic pumps may not have been rigorously and fairly assessed against other interventions and it may be that if a base line of intervention were employed, a meaningful comparison of other treatments versus compression pumps could be made.

The skin undergoes significant changes in lymphoedema, and skin care is critical to prevent the deterioration of the condition. Skin becomes thicker and harder (fibrotic) with increased scaling (hyperkeratosis) and warty changes (papillomatosis). Compromised skin contributes to both psychological challenges, because of its disfiguring appearance, and to physical problems, because of its reduced elasticity and tendency to infection. Skin with reduced elasticity will compromise lymph flow further and is more likely to suffer damage as stiff skin will not withstand pressure or shearing forces so easily as resilient healthy skin. Scaly skin tends to harbour greater amounts of micro-organisms – particularly fungal and bacterial agents which inevitably lead to associated infections (Browse *et al.* 2003). Oil based emollients soothe and hydrate the skin and should be used at least twice daily as part of a self management programme. They do, however, lead to a shortened life for pressure garments (and other clothing) as the oil affects the fabric by staining and stretching fibres (Casley-Smith & Casley-Smith 1997). When skin scale is excessive, it may need to be treated with salicylic ointments and antiseptic agents. Topical steroids can be used where skin is suppurating and smelly but have a thinning effect on skin and need to be used sparingly (Browse *et al.* 2003). Typically, an individual would need to spend at least twenty minutes at the beginning and end of each day gently massaging the emollients into the skin before donning pressure garments. For most people this represents a real challenge in self management and in reality many patients report non-compliance with the regime (Lymphoedema Support Network 2008, British Lymphology Society 2008).

Exercise is used in both the active and maintenance phases of lymphoedema management as it increases the uptake by the initial

lymphatics, encourages the collecting lymphatics to pump more strongly, keeps joints mobile, strengthens muscles and prevents muscle wastage and may help to prevent skin from stiffening (Goddard *et al.* 2008). Aerobic exercise increases intra-abdominal pressure which facilitates pumping of lymph into the thoracic ducts (Medical Education Partnership 2006). It is particularly useful in conjunction with compression bandaging or garments (see 'compression garments' above) (Twycross *et al.* 2003, Casley-Smith & Casley-Smith 1997). Although acknowledged to be useful, advice on exercise regimes are non-specific and vary between therapists and regions (Medical Education Partnership 2006, Twycross *et al.* 2003, Wessex Cancer Care 2008). There is also debate in the literature about the intensity of exercise which is likely to be most beneficial (Lane *et al.* 2005, Harris *et al.* 2000) with some advocates for gentle, non-impact exercise routines to prevent increases in swelling (Casley-Smith & Casley-Smith 1997) and others asserting that even vigorous exercise such as Dragon Racing (rowing) has no deleterious effect on arm limb volumes (Lane *et al.* 2005). At present, there is little evidence to indicate which types, intensities or frequencies of exercise may be safely used in the management of primary lymphoedema. That said, there is a consensus (Twycross *et al.* 2000, Casley-Smith & Casley-Smith 1997, Woods 2007) that leisure/sporting exercise regimens should be undertaken regularly but not too vigorously in order to achieve maximum benefit. Low impact sports such as walking, cycling, swimming, badminton and rowing are said to be helpful, whilst high impact sports such as running, football, rugby, tennis might be harmful (Johansson *et al.* 2005, Goddard *et al. 2008)*. Scuba diving as a leisure activity may also be beneficial to those with lymphoedema because of gradient water pressures which are greater under water and increase with depth. It is interesting to note that high altitude flight will, conversely, exacerbate the

problem of lymphoedema because low air pressure will permit greater lymph flow outwards into the peripheral tissues. Some experts have cautioned against long-haul flying for those who are vulnerable (Casley-Smith & Casley-Smith 1997). Hydrotherapy may be a particularly useful form of therapeutic exercise since water provides a gentle resistant force which can encourage lymph drainage (Medical Education Partnership 2006, Goddard *et al.* 2008, Woods 2007) but barrier creams may need to be used to prevent cross-infection in water.

Kinesio-taping which originated in Japan, has been used for many decades for orthopaedic and sporting injuries (Stockheimer & Kase 2004). In recent years, it has been suggested that it may be useful in the treatment of lymphoedema if used in a very specific manner following dedicated training in its use. Specialist adhesive tape is applied to the skin following the same anatomical patterns as would be used in MLD. It is claimed to act by lifting the skin slightly and assisting the superficial lymphatics, which lie just under the skin, to open up. This, in turn, encourages lymph flow along the anatomical pathways and assists with lymph drainage (Stockheimer & Kase 2004).

A recent trial conducted by the Royal Marsden Hospital in London (Gothard *et al.* 2004) looked at hyperbaric oxygen therapy (where patients are placed in a specialised chamber and have oxygen delivered at high pressure) and demonstrated a significant improvement (average 20 per cent reduction in volume) for a small number (3 out of 19 patients) of the study group (Gothard *et al.* 2004). Again, this is inconclusive and a larger, randomised and controlled trial is being undertaken before any conclusions can be drawn. The proposal is that the benefit is accrued from the pressure created and works rather like deep-sea diving to compress

lymphatic vessels and encourage lymph flow. Since Hyperbaric chambers are few in number and situated close to naval/diving centres, it is difficult to envisage any large-scale treatment using this method even if results are conclusive and positive.

References

All references for this appendix are included in the main reference section.

Appendix 2

Ethics Approval

Appendix 3

Insurance Confirmation from

University of Southampton

and

Risk Assessment

Appendix 4

Permission to Recruit through

Lymphoedema Support Network

and

Recruitment Article and Web Entry

Appendix 5

Letters to Participants and Non-Participants

Appendix 6

Information Sheet and Consent Form

Appendix 7

Information on Daniel Lambert