Genital Oedema

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Contents

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Foreword—Genital Oedema

Isabelle Quéré

Genital oedema is gaining increased recognition internationally. This condition occurs as an acute swelling after an infection or inflammation of the pelvic organs and usually disappears with treatment of the underlying disease. It may also present as a chronic swelling due to either acquired or congenital dysfunction of the lymphatic system that is draining the genital area.

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Lymphoedema with genital involvement has been recognised by the World Health Organization (WHO) as a ‘painful and profoundly disfiguring visible manifestation of the disease.’ Lymphoedema, elephantiasis and scrotal swelling occur later in life and can lead to permanent disability, and people with genital oedema are ‘not only physically disabled, but suffer mental, social and financial losses contributing to stigma and poverty.’ The WHO estimated that 36 million people are affected by lymphatic filariasis, with 25 million men suffering from a hydrocele and lymphoedema of the genitals (WHO, 2021). In countries described as high or medium high income, lymphoedema of the genitals is a frequent complication of pelvic cancers treated with surgery and radiation, as well as of lymphoedema affecting the lower limbs. It can occur in those affected with primary lymphoedema, often in conjunction with swelling in other areas of the body. It can start early in life and can badly affect relationships and sexuality in adolescents and young people.

Lymphoedema of the genitals following treatment of pelvic cancer seems to occur less frequently than lower limb lymphoedema, but the epidemiology is relatively scarce, leading to the conclusion that the true size of the problem is likely to be underestimated. Evidence from the literature using a PubMed Central quick research, with the Mesh terms ‘pelvic cancer’ and ‘lymphedema’ identified 1568 references (30 June 2021), with this falling to only 383 after introduction of the term ‘genital.’

Literature on lymphoedema affecting the genitals is more clearly described in men, while it is rarely described in the literature on women. This can be seen in the reporting of recent high-quality studies focusing on the epidemiology or management of lymphoedema after gynaecological cancers. These studies did not explore genital lymphoedema as a potential consequence of the cancer or its treatment or consider outcomes measures for the condition. These factors support the argument that it is likely the problem is under-reported (Hayes et al, 2017; Shallwani et al, 2021). The reasons for low reporting are likely to be complex and multifactorial and include cultural, religious and gender issues in both sexes. It is recognised that women do not find it easy to discuss with clinicians potentially embarrassing problems. This is likely to be compounded by other factors, including lack of awareness and education among health professionals. Lack of knowledge and professional embarrassment will result in clinicians not asking relevant questions or clinically examining the genitals as part of the assessment process.

This *Journal of Wound Care* supplement on genital lymphoedema addresses this under-reporting of genital oedema by using a holistic framework through three papers written by authors from five different parts of the world and presenting research from different professions working in varied specialist areas. The first paper describes five cases of male genital lymphoedema and allows the authors to compare the different approaches to assessment and treatment that they are using. This is particularly valuable at present due to the dearth of evidence-based protocols to inform practice (article 1 of this supplement). The second paper reports on a survey to investigate the education requirements of health professionals working with patients with genital lymphoedema.

Respondents from 31 countries completed the survey which allowed an exploration of what they thought was the best way to manage these patients and the developments that should be focused on to improve care (article 2 of this supplement). The aim of the final articlef was to explore the contextual factors surrounding the treatment of patients with genital oedema, how these affect the education requirements of lymphoedema professionals and what developments might be designed and implemented to support both the health professional and patient.

Overall, the supplement encompasses identification of the needs and barriers to be overcome to improve the diagnosis of genital lymphoedema and its management.

The subject will be extensively discussed during the International Lymphoedema Framework conference in Copenhagen, on 18–20 November 2021. **JWC**

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Conservative and integrated management of genital lymphoedema: case reports for men

Pinar Borman, Rhian Noble-Jones, Melanie J Thomas, Thomas Bragg, Kristiana Gordon

ABSTRACT

Genital oedema of primary and secondary aetiology is gaining increased recognition; it occurs in people of all ages, although accurate prevalence remains unknown. While reports of surgical intervention are quite common, clear description of conservative management is rare, and, overall, diagnosis and management remain largely delayed. Conservative management of genital oedema requires imaginative use of compression bandages and support garments, adapted manual lymphatic drainage, enablement of exercise and activities required for school/employment and diligent skin care to avoid cellulitis.

The aim of this article is to present case reports from two European countries discussing five males of differing ages and with genital oedema of different aetiology as examples of conservative management of male genital oedema.

The case reports include an adolescent boy with genital oedema since age 1 year, a 49-year-old man treated for penile cancer, a 60-year-old man treated for primary rectal lymphoma, a 48-year-old man with morbid obesity and a 49-year-old with obesity and recurrent infections. The latter two cases integrate surgery and conservative care.

Case reports can be a useful means of addressing education needs related to rarer conditions and sharing practice where diagnostic criteria and specific protocols of care are lacking.

INTRODUCTION

Genital lymphoedema is a progressive and chronic oedematous condition of the genital organs due to disrupted lymphatic circulation. It is often associated with lower extremity primary or secondary lymphoedema. This condition can affect adults or children, both males and females, but is more common in males due to greater tissue elasticity of the scrotum and penis combined with the effects of gravity (Murillo and Fishman, 2015; Norton, 2018; Vignes, 2018).

The exact incidence and prevalence of genital lymphoedema are not known, and estimates are based on specific populations mainly from service audits (Rockson et al, 2008; Schook et al, 2014; Pike et al, 2021). This could be due to the lack of awareness, set diagnostic criteria and/or delayed diagnosis, as well as the intimate nature of the condition and consideration of this topic as unpleasant or taboo (Noble-Jones et al, 2019; article 3). The co-existence of genital swelling in patients presenting with lower extremity lymphoedema has previously been estimated as 10% (Garaffa et al, 2008; Watt et al, 2017).

Primary lymphoedema affecting the genitalia occurs as a result of a genetically determined developmental abnormality of the lymphatic system. The swelling is not always present at birth and can develop later in life. Primary lymphoedema of the genitalia almost always occurs in association with lower limb swelling (unilateral or bilateral). An underlying gene mutation may be detected in 25% of patients with primary lymphoedema, but the remainder have an as-yet-undiscovered causal gene mistake (Gordon et al, 2020). Primary lymphoedema is not one disease, but an 'umbrella term' for several different conditions that present with lymphoedema. To date, there are four subtypes that cause genital lymphoedema for which mutations have been discovered: Emberger syndrome (associated with myelodysplasia and progression to leukaemia) due to mutations in the GATA2 gene; lymphoedema distichiasis syndrome

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(associated with extra eyelashes, varicose veins and a risk of cardiac valve anomalies) due to FOXC2 mutations; generalised lymphatic dysplasia (e.g. due to CCBE1 or PIEZO1); and Noonan syndrome, in which the association of the PTPN11 gene is indicated in over half of all cases (associated with a central conducting lymphatic anomaly and chylous reflux affecting the genital tissues) (Gordon et al, 2020). Pelvic lymphatic malformations may also present with genital and/or lower limb lymphoedema.

Minor surgical interventions, such as circumcision, or perineal and anal region infections may trigger the onset of paediatric primary lymphoedema (Norton, 2018). However, cancer surgery, particularly with radiotherapy and pelvic lymph node resections, trauma, infections and inflammatory conditions, such as anogenital granulomatosis and hidradenitis suppurativa, are common causes of secondary lymphoedema. Globally, the most common cause of hydrocele (oedema within the lining of the testis) is filariasis (World Health Organization (WHO), 2021). Treatment of gynaecological, testicular, penile, urological, abdominal, intestinal or prostatic malignancies, as well as melanomas in inguinal lymphatic areas may cause genital lymphoedema (Lockwood-Rayermann, 2007; Dessources et al, 2020; National Institute for Health and Care Excellence (NICE), 2021). The swelling may occur immediately after surgery or years later (Executive Committee of the International Society of Lymphology, 2020).

Various presentations of genital lymphoedema may occur. Combined penile and scrotal swelling is more common than isolated penile swelling in males (Garaffa et al, 2008; Otsuki et al, 2012; Shim et al, 2019). In females, labia majora and minora and pubic triangle areas may be included in the swelling (Lockwood-Rayermann, 2007; Fadare et al, 2011; Silva et al, 2015; Dessources et al, 2020). Genital oedema frequently causes urination and sexual activity problems, depending on the extent of the involvement. The affected tissue initially swells from accumulation of lymph, and the resultant inflammation stimulates adipose deposition and fibrosis, which may lead to papillomas and warty growths. Hyperkeratosis, lymphatic leakage from the skin and bleeding from the vesicules is commonly seen. Complications associated with genital lymphoedema include lymphorrhoea, chyluria, haematuria and cellulitis; the last may lead to sepsis and be life-threatening and, therefore, should be treated early and aggressively (Whitaker, 2007; Torio-Padron et al, 2015; Borman, 2018).

Genital lymphoedema is a challenging condition that causes long-lasting and debilitating physical, emotional and social problems, leading to impaired quality of life for affected patients and families (Moffatt et al, 2019a; Noble-Jones and Thomas, 2021; Voznesensky et al, 2017). Due to the many causes of genital oedema in women, men and children, an accurate diagnosis is imperative to providing appropriate counselling and treatment (Azadgoli et al, 2019; Gordon et al, 2020). Diagnosis depends on the differential diagnosis of other conditions, history and physical examination. On diagnosis of genital lymphatic disease, complex decongestive treatment (CDT), also known as decongestive lymphatic therapy (DLT), should be initiated (Whitaker, 2007; Silva et al, 2015; Torio-Padron et al, 2015; Fadare et al, 2011; Borman, 2018), unless there is clear indication for immediate surgery, for example, testicular torsion (Azadgoli et al, 2019; NICE, 2021).

Patient and/or family education and infection prevention, as components of CDT, play a key role in management. Daily washing of the genitalia and application of moisturising agents to prevent skin breakdown should be emphasised, with information about signs and symptoms of cellulitis (British Medical Journal Best Practice, 2021). Education about healthy nutrition and weight control is needed in all patients (Zvonik et al, 2011; Borman, 2018). Manual lymphatic drainage (MLD) and compression therapies are the mainstay of management in the majority of cases. Compression is performed by bandaging in the active treatment phase and compression garments in the maintenance phase. Due to the irregular shaping and the location of genital lymphoedema, bandaging or wrapping and garment material can be difficult to apply and require advanced skills from the lymphoedema specialist and patient/family (Whitaker, 2007; Silva et al, 2015; Noble-Jones

FIGURE 1. **Adolescent boy before treatment (case 1)**

et al, 2021a). Surgical treatment of genital lymphoedema may be elective or urgent, for example, in the case of testicular torsion (NICE, 2021). Non-urgent surgery can improve disfigurement and dysfunction beyond that possible with conservative approaches and may be used in combination with or integrated with CDT (see case report 5) (Zvonik et al, 2011; Torio-Padron et al, 2015).

Much of the existing literature on the treatment of male genital lymphoedema focuses on surgical techniques, including treating hydrocele from filariasis (Lim et al, 2015; Smith-Harrison et al, 2020; WHO, 2021). In contrast, there are very few reports on the usefulness of conservative measures (CDT), in these patients, particularly in females (Whitaker, 2007; Silva et al, 2015; Watt et al, 2017). In the following cases, from two different European countries, the authors confine their remit to males. Case reports for females will be presented in a future paper, and the specific techniques involved will be presented in greater detail in a forthcoming Genital Oedema Position Paper. All the patients reported here were treated conservatively, but one received a combination of CDT and surgical intervention. The case reports include:

- An adolescent boy with genital oedema since age 1 year
- A 49-year-old man treated for penile cancer
- A 60-year-old man treated for primary rectal lymphoma
- A 48-year-old man with morbid obesity
- A 49-year-old man with obesity and recurrent infections.

BOX 1. **GLS system based on subjective symptoms of genital lymphoedema**

	No	Yes
Sensation of heaviness	0	1
Sensation of tension	0	1
Swelling	0	1
Urinary troubles due to genital oedema	0	2
Cutaneous lymphatic cyst	0	2
Genital lymphorrhoea	0	2
Total from range of 0–9		

Case reports of male genital oedema

Case 1

A 16-year-old adolescent with bilateral lower extremity and genital lymphoedema was admitted to a hospital-based lymphoedema unit 2 years ago. According to his medical history, the oedema in the legs and genitalia had been present since he was 1 year old, but become more apparent with the beginning of puberty. Although he consulted different physicians (paediatrician, paediatric urologist, vascular surgeon, dermatologist) in the small city where he lived, a differential diagnosis was not made. However, recently, a vascular surgeon from another centre made the differential diagnosis of lymphoedema based on the clinical picture and lymphoscintigraphy and referred the adolescent to the lymphoedema unit. Lymphoscintigraphy indicated obstruction of lymphatic flow, dermal flow-back and lack of transition to the inguinal lymph nodes in the lower extremities. There was no history of trauma, comorbidity or family history, and review of the systems yielded unremarkable findings.

On physical examination, his vital signs were in the normal range, and his BMI was 22.9 kg/

m² (weight: 64 kg, height: 167 cm). He had bilateral lower extremity lymphoedema (more prominent on the left) with a positive Stemmer sign and the presence of skin folds. He had a hard, non-pitting, painless oedema of the scrotum and, to a lesser extent, penile oedema, which disfigured the genital contours; he also had slight redness of the scrotum. Further, he had a small cutaneous lymphatic cyst at the back of the left scrotum. There were no signs of infection or active inflammation. Based on these characteristic findings, he was diagnosed with stage 3 bilateral lower extremity lymphoedema and genital lymphoedema (ISL, 2016) (Figure 1). According to the genital lymphoedema severity scoring system (Yamamato et al, 2016), he had a score of 5 (Box 1).

After his acceptance to the lymphoedema unit, he was educated on self-management techniques, and circumferential measurements of the legs were recorded. The cone formula was used to obtain limb volumes from the circumferential measurements on both extremities (Sitzia, 1995). Photographs of the patient were taken as routine. The patient received 15 sessions of CDT (5 days/week for 3 weeks), which comprises skin care, MLD, multilayer short-stretch bandages for the lower limbs and genital area, and exercises.

The MLD in this particular case consisted of four basic Norton techniques (stationary circle, rotary, pump and scoop techniques) (Norton, 2018) and was performed in a proximal to distal direction with light skin strokes for 45 minutes. The pubic area, scrotum and penis were decongested with MLD (Figure 2) before the lower extremities were. The cervical area,

FIGURE 2. **Adolescent boy receiving MLD**

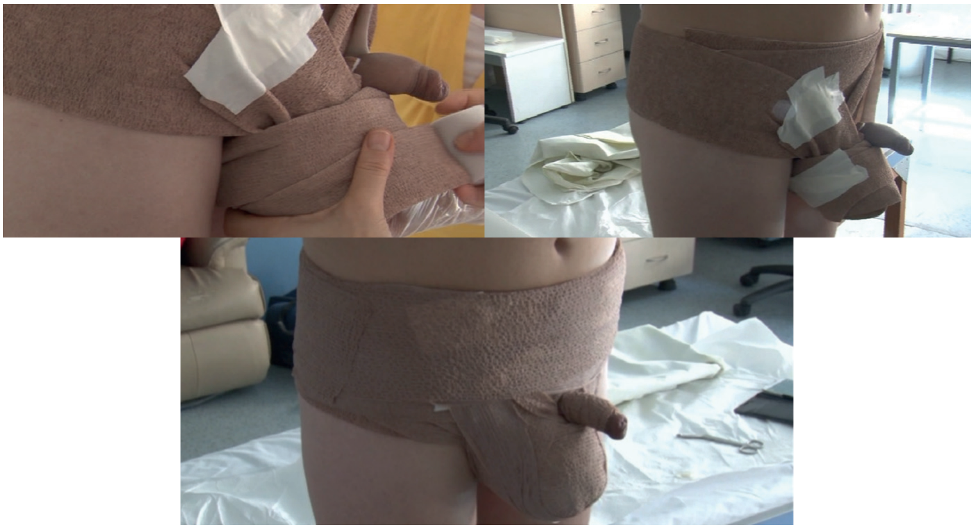

FIGURE 3. **Genital bandaging in case 1**

abdominal area and axillary lymph nodes were manipulated. Superficial and deep techniques were used for abdominal treatment. Anterior and posterior inguino-axillary anastomoses were activated on both sides by rotary techniques starting at the sagittal watershed to the side, followed by stationary circles toward the axilla. Bilateral lower abdominal quadrants were covered by rotary techniques toward the anastomosis.The pubic area was decongested toward to the lower truncal quadrants and anastomosis area by rotary and stationary circles. Then, stationary circle technique was performed on both sides of the scrotum to manipulate the lymph fluid toward the pubic area and, from here, toward the axillary lymph nodes using inguino-axillary anastomosis. The lower parts of the scrotum were decongested toward to the gluteal watershed with rotary and stationary circle techniques on both sides. After the scrotum, the penis was decongested. Using one hand as a firm support, the penis was rested and stationary circles were applied to the entire shaft of the penis focusing on a light circular motion directed toward the suprapubic area. Stationary circles were also performed to the foreskin using the fingertips (Figure 2). In other cases, MLD techniques from Casley-Smith, Foldi or Vodder might be used as appropriate.

Before the decongestion of the limbs, the scrotum and penis were wrapped with 3M Coban 2 layer bandages in a standard manner (Schuren et al, 2012; 3M, 2013) (Figure 3). Finally, the lower limbs were decongested from the proximal to distal parts of the extremity with great emphasis on avoiding the genitals and mid-sagittal watershed.

After MLD, short-stretch multilayer bandages were applied to the legs and changed daily, except at weekends. This allowed for washing of the short-stretch bandages between reapplications, if required. Multilayer compression bandages were applied such that greater compression was at the distal parts, and the compression decreased gradually in the proximal direction. This was staged, so that, in the first week, the legs were bandaged below the knee; in the second week, the left leg was bandaged up to the thigh; and, in the third week, the right leg was bandaged up to the thigh. The Coban bandages were changed every other day, while the conventional multilayer bandages were changed daily. Small adjustments were made as required, based on reassessment at each bandage change and the tolerance and feedback of the patient.

The patient had an individualised active exercise programme of 20 minutes per day under the supervision of the physiotherapist, including diaphragmatic breathing exercise, neck-and-shoulder stretching, foot pumping and non-isometric strengthening of the legs muscles, in order to help facilitate lymphatic flow and improve strength and range of motion. Pelvic floor and core exercsies were not necessary in this particular case. The exercises were performed with the bandages

FIGURE 4. **Case 1 after 3 weeks of therapy**

TABLE 1. **Lower limb volumes before and after treatment and volume difference**

Volume (cm3)	Before therapy	After therapy	Difference	Percentage change
Right leg	6881	5506	1375	-20%
Left leg	7808	5676	2132	-27%

kept on.

The baseline leg volumes calculated from circumferential measurements of the limb were found to have reduced significantly at the end of phase-1-CDT (Table 1). The patient was prescribed a pantyhose pressure garment (CCL3 pressure for the legs) with a genital padding area. He was also advised to wear cycle shorts and to continue self-massage and exercises in phase-2 CDT. Daily washing of the genitalia and application of moisturising agents to prevent skin breakdown were emphasised, and information about the signs and symptoms of cellulitis and education about healthy nutrition and weight control were included. At the follow-up visit 4 weeks after CDT, the improvement was stable and compliance was good.

The compliance to therapy was poor at the beginning, but with education and the help of his mother, the patient began to apply self-MLD to the genital area, and his compliance was better after he saw the improvement. He is still being followed up at 6 months, and his mother is motivating him. His quality of life has improved, along with his social interactions and self-confidence. At the last visit, the authors learned that he had a girlfriend. It would have been useful to add aquatic exercises, but the facilities were not available in this case.

CASE 2

A 49-year-old man with genital swelling was admitted to the lymphoedema unit. He had undergone surgery for penile cancer 18 months previously and was treated with chemotherapy and radiotherapy. No additional comorbidities were present. The swelling was noted after 12 months and gradually increased. He was a medical doctor and reported being uncomfortable during routine hospital work and, sometimes, had pain in the scrotal area. He had some problems with sexual interaction, and low self-esteem. With some understanding of lymphoedema, he was fearful about its progression.

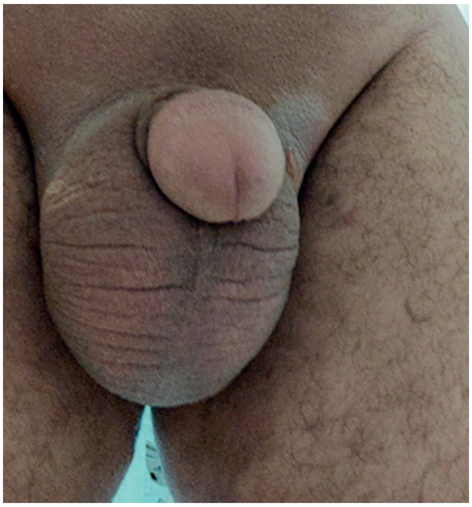

FIGURE 5. **Penile cancer treatment-related lymphoedema before CDT**

FIGURE 6. **Penile cancer treatment-related lymphoedema after CDT**

On physical examination, his vital signs were in the normal range, but his BMI was 32 kg/m^2. He had a slight, soft non-pitting oedema in the pubic area and mild scrotal oedema, which disfigured the genital contours (ISL stage 2). No oedema in the penis was observed. According to the genital lymphoedema severity scoring system (Yamamato et al, 2016), he had a score of 3 (see Box 1).

After his acceptance to the lymphoedema unit, he was educated about skin care, weight control and self-management techniques. Photographs of the patient were taken as routine work-up (Figure 5). The patient received four sessions of CDT (2 days/week), which comprised MLD and Coban-2 layer bandages for the genital area, with pubic padding. Diaphragmatic breathing exercises were demonstrated for him to practise.

The MLD covered the central areas, axillary region, inguino-axillary anastomosis

KEY WORDS

- Lymphoedema
- genital oedema
- case reports
- hydrocele
- treatment

FIGURE 7. **Genital lymphoedema in a patient treated for rectal lymphoma, before CDT**

and the genital area. After MLD, the scrotum was wrapped with Coban 2 layer bandages in a standard manner (Schuren et al, 2012; 3M, 2013), with padding to the pubic area. The bandages were changed once every three days, and MLD was performed four times over 2 weeks. The swelling reduced significantly within 2 weeks (Figure 6). After the active phase of CDT, he was prescribed special genital hosiery with pubic padding under cycle shorts. The patient was advised to continue to use the compression garment and perform self-MLD and skin care.

FIGURE 8. **Bandaging of genitals allowing for a stoma bag**

FIGURE 9. **After three sessions of CDT over 10 days**

CASE 3

A 60-year-old retired civil servant was admitted to the lymphoedema unit of a hospital with oedema of the genital area. He had a history of cancer surgery due to primary rectal lymphoma, which was diagnosed 13 months previously. He had received chemotherapy and radiotherapy, which ended 9 months previously. He initially presented with mild right lower extremity oedema that had been managed at another centre, but the oedema had progressed to the genital region. The swelling in the genital area had been noticed 10 months previously, before the end of chemotherapy and radiotherapy, and it had gradually increased. His past medical history included two attacks of cellulitis in the genital area during the previous year, for which he received antibiotics as an in-patient. As is often the case, the root cause of the cellulitis was unknown. He complained about difficulties with wearing trousers and lack of sexual intercourse since the cancer surgery. He had no comorbidities.

On physical examination, his vital signs were stable, and his BMI was 28.1. He had a colostomy bag on the left lower abdominal area (Figure 7). There was significant scrotal and pubic lymphoedema with fibrotic and keratotic skin changes and mild tenderness on the genital areas and bilateral mild hyperemia on the scrotal area (ISL stage 3). His penis was buried within the scrotum, and he had difficulty with urination. There was also mild lymphoedema on the upper right thigh. To appropriately evaluate the patient's genital oedema, the widest girth of the scrotum from the middle and the length of the scrotum (from the pubis to the inferior end) were measured before and after therapy.

The patient was educated about self-drainage and exercise, as well as skin care. His wife also learned all these same maintenance care and self-management techniques in order to assist him. The patient received three sessions of CDT (2 days/week), which comprised MLD and Coban 2 layer bandages with scrotal padding for the genital area (Figure 8). After application of urea-containing cream to the scrotal area to address the keratotic changes and reduce infection risk, the genital scrotal and pubic MLD sequences in addition to central decongestion (cervical, abdominal areas) and clearing anastomosis (anterior, posterior bilateral inguino-axillary) were conducted. The patient was also shown how to perform diaphragmatic breathing exercises.

After three sessions of CDT for just 10 days, the scrotal measurements had reduced (initial evaluation: 18 cm × 34 cm (length x girth) vs. discharge: 12 cm x 27 cm). A better scrotal contour was achieved, and the degree to which the penis was buried had decreased (Figure 9). The patient could extend the penis out for urination, and the urination was less painful. The authors believe that better results could have been achieved if he could have continued the CDT sessions, but, as he was living in another city and had accommodation problems, he could only attend treatment sessions for 10 days.

After the intensive phase of CDT, the patient was prescribed special genital hosiery with suspending scrotal padding under cycle shorts. His wife agreed to perform all maintenance procedures for him, which comprised skin care, individualized MLD and the compression garment. Placing the burden of care on a spouse is a careful consideration, but, in this case, it worked very well for the couple.

CASE 4

A 48-year-old man with bilateral lower limb lymphoedema and gross genital and abdominal lymphoedema was referred to the National Complex Lymphoedema Clinic by his local lymphoedema service in 2018. He had been regularly seen by his primary care general practitioner (UK) and had been reviewed by a vascular and urology surgeon, who diagnosed lymphoedema and made the initial referral to the local lymphoedema service in 2014. Having weighed 250 kg in 2016, the patient lost 70 kg by following a calorie-controlled diet. The lower limb oedema was controlled with compression garments, but the genital oedema remained hugely problematic. In the past year, he reported admission to hospital six times due to cellulitis in the abdominal panniculus and genital area. He also reported that, functionally, he found micturition extremely difficult due to the size and location of the oedematous area. Psychologically, the patient indicated that he felt his life was worthless; he was socially isolated due to this

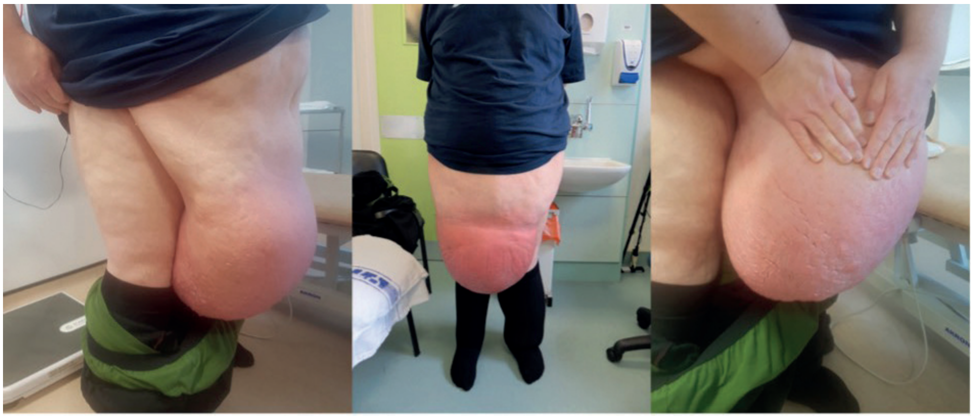

FIGURE 10. **Abdo-genital lymphoedema in a morbidly obese patient**

KEY POINTS

- **Genital lymphoedema is a challenging condition that causes long-lasting and debilitating physical, emotional, social and sexual problems, leading to impaired quality of life for affected patients and families**
- **On diagnosis of genital lymphatic disease, complex decongestive treatment (CDT), also known as decongestive lymphatic therapy (DLT), should be initiated unless there is clear indication for immediate surgery**
- **Conservative management of genital oedema requires imaginative use of compression bandages and support garments, adapted manual lymphatic drainage (MLD), enablement of exercise and activities required for school/ employment and diligent skin care to avoid cellulitis**
- **Patient and/or family education and infection prevention, as components of CDT, play a key role in the management, and lifestyle changes may be required**
- **Surgical management of genital oedema can be usefully integrated with conservative management.**

physical size and felt ridiculed when he went out (Figure 10).

The focus of the case report was the genital oedema assessment. On physical assessment, his BMI was found to be 51 kg/m^2. The extent of oedema was significant, extending from his umbilicus down to his shin, measuring 80 cm in length and 74 cm at the widest point. The distal area was hard, with a non-pitting solid lymphoedema with slight redness and areas of papillomatosis. There was no indication of active cellulitis. As the patient had expressed great difficulty in micturition, the mass was placed on a plinth and lifted, so that the condition of the structures underneath could be ascertained. Surprisingly, no genitalia were identifiable. The penis and scrotum had been encompassed within the abdominal/genital mass. A creased opening was visible, which was leaking urine, and the perineal area was macerated, with a fungal infection present. During the examination, the patient stated that he had limited sensation in the area. Based on the findings, his condition was categorised as complex lymphoedema, according to UK staging (BLS, 2016), or ISL stage 3 (Executive Committee of the ISL, 2020).

Initial treatment focused on medical management of the fungal infection and self-management, including skin care and massage of the oedematous mass while it was supported on a low table. Positively, even after a few minutes of gentle massage and rocking motion, the patient reported that he experienced sensation in that area of the skin for the first time in years. Healthy eating and weight loss were encouraged, along with general activity and raising exercise tolerance around the house and garden. As the patient had experienced numerous episodes of cellulitis within the past year, a discussion with his general practitioner initiated a prescription for prophylactic antibiotics. Compression was strongly indicated for the genital/abdominal area, and a short-stretch cohesive bandaging regime that lifted and compressed the area was commenced. However, this significantly altered the patient's line of gravity in relation to his base of support, so that he felt he was falling backwards. This also occurred when a made-to-measure flat-knit garment of jockstrap-type shorts with an abdominal pouch support was fitted. Further, this created difficulties as he had urinary incontinence and the garment was saturated. The team and the patient jointly decided that it was not safe or acceptable to reduce his mobility by introducing gait imbalance, so a more conservative approach was agreed. This meant reducing the compression to simpler cylindrical compression.


FIGURE 12. **Results after surgery integrated with conservative care**

Due to the seriousness of the ongoing cellulitis infections and the retraction of the whole of the genitalia into the abdominal mass, an urgent opinion from plastic surgery was sought. The patient is now on the surgical list for genital extraction and removal of the


FIGURE 11. **Genital oedema associated with recurrent cellulitis and obesity**

abdominal panniculus. Due to the COVID-19 pandemic, his surgery is likely to occur in late 2021. His weight has reduced by a further 15 kg, and the area is slightly softer with the support of cylindrical compression. Prophylactic antibiotics have reduced the frequency of cellulitis episodes, but not eliminated them.

CASE 5

A 39-year-old man with genital oedema was referred to the lymphoedema service from a urology consultant in 2015. Ultrasound, MRI scans and blood tests ruled out any malignancies. Based on the scans, a diagnosis of lymphoedema linked to obesity and recurrent cellulitis infections was made.

On examination, the patient presented with severe scrotal and penile swelling with papillomatosis and lymphorrhoea to the left and lower side of the scrotum (ISL stage 3) (Figure 11). The skin was clean, and the patient reported washing, drying and moisturising the area daily. His weight was 95 kg, and he had a BMI of 39 kg/m². He worked as a hotel porter, which involved 8 hours of standing 5 days a week; he felt this exacerbated the swelling.

Conservative compression therapy was applied, including self-bandaging and compression shorts, which, although supporting the genitals, did not reduce the oedema. He continued with healthy eating, skin care, exercises (core, pelvic floor and legs) and self-massage (simplified individualised MLD) to ensure that the area remained soft.

Due to ongoing issues with micturition, shape deformity and increasing swelling, an opinion from plastic surgery was sought. In April 2017, debulking of the genital area was completed, with good cosmetic outcomes (Figure 12). The patient was delighted with the surgery and was recommended to continue with skin care, exercise and compression shorts daily to maintain the improvements.

DISCUSSION

The aetiology of genital lymphoedema is commonly non-cancer (e.g. primary lymphoedema, inflammatory or infective causes), but cancer therapies, including lymph node dissection and radiation, may also cause lymphoedema in the genital area. This embarrassing chronic condition impairs voiding, social and sexual interactions and severely affects patients with regard to physical and psychosocial aspects and quality of life (Murillo and Fishman, 2015; Norton, 2018). Both patients and health professionals have barriers to conversations about genital lymphoedema (Noble-Jones et al, 2019). A lack of education among health professionals caring for these patients has been shown to be an international problem (articles 2 and 3) and, understandably, contributes to patients feeling frustrated by having to attend repeated visits to multiple health providers who are unable to provide proper care. The management of genital lymphoedema is challenging, as there is no standard algorithm or established treatment for this condition. Surgery is probably the most commonly suggested treatment available or a combination of conservative treatment (CDT) with surgery (Torio-Padron et al, 2015; Guiotto et al, 2019). In the context of endemic filariasis, surgery or no treatment at all may be the only pragmatic option (WHO, 2021). Further research is needed to establish whether greater awareness of conservative treatments would reduce this tendency towards surgery as the first choice. The application of compression bandages or garments on the genital area is also difficult due to the anatomical characteristics of the condition (Whitaker, 2007; Murillo and Fishman, 2015; Norton, 2018). Education on the treatment of genital lymphoedema is often neglected and can be lacking, even on certificated courses (article 2 of this supplement). A lack of confidence in health professionals' knowledge and skills causes further barriers to treatment (Moffatt et al, 2019b; articles 2 and 3). Lymphoedema therapists can be reluctant to perform MLD and bandaging for genital lymphoedema due to embarrassment and taboo (article 3 of this supplement). This can be a particular problem where cultural or religious requirements restrict treatment of male patients by female lymphoedema therapists. However, when performed competently by experienced therapists, CDT can lead to considerable reduction of genital volume, as demonstrated in the case reports in this supplement and other articles (Whitaker, 2007; Torio-Padron et al, 2015).

The five patients in these case reports had very different medical histories, but each responded well to conservative genital lymphoedema management, with the addition of surgery considered for two patients. The effects of CDT on genital lymphoedema have rarely been reported in the literature in comparison with surgery alone. Plastic surgery in combination with conservative therapy may yield dramatic differences in genital oedema shape and size. However, emphasis must remain on continued compression and skin care to maintain progress.

In conclusion, awareness of genital lymphoedema is important in order to prevent delayed diagnosis. Education and training for health professionals about the management genital lymphoedema and CDT are needed to prevent delayed treatment, which leads to physical and psychosocial disability and decreased quality of life in patients with this progressive condition. **JWC**

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An international investigation of the education needs of health professionals conservatively managing genital lymphoedema: survey findings

Rhian Noble-Jones, Melanie J. Thomas, Isabelle Quéré, Karen Morgan, Christine Moffatt

ABSTRACT

The education needs of health professionals supporting patients with genital lymphoedema (oedema) are unknown. An accurate prevalence of genital oedema, in men, women and children, has not been achieved. It may have many potential causes and multiple psychosocial influences. The vast variations in patient origins and access and delivery of service provision compound the complexity of the management of genital oedema.

The aim of this study was to understand the education needs of health professionals supporting patients with genital oedema and how best to meet these needs. This mixed-method project used an online survey, distributed through national and international professional lymphoedema groups, followed by focus groups. This article reports the results of the survey.

The views of 583 health professionals from 31 countries were collated. Only 42% respondents had completed specific education in treating genital lymphoedema, despite 92% feeling it was necessary. Only 7% reported that patient identification and referral was timely. Wound care skills had been used by 26% of respondents. Education subjects considered ‘essential’ and those of ‘least confidence’ were identified. E-learning packages, videos, printed materials and collaborative events with urology and pelvic health provide opportunities to address the identified needs.

This original project identified unmet needs among health professionals conservatively managing genital lymphoedema.

INTRODUCTION

There is very little guidance available for health professionals conservatively managing patients with genital lymphoedema. Practice is extrapolated from more commonly recognised presentations, such as breast cancer-related lymphoedema, and the specific evidence base is weak (Vignes, 2018, Quéré et al, 2019). Further, the true prevalence of the condition is not known, although data drawn from specific populations have been reported (Rockson et al, 2008; Schook et al, 2011). A low presentation rate at clinics also means that health professionals do not develop a sense of familiarity from regular contact with those with the condition. Neglect or poor management can lead to chronic, irreversible deterioration, with a reported risk of recurrent cellulitis (Hara and Mihara, 2020), meaning that there are implications for ongoing health costs, as well as physical and psychosocial impact on patients (Humphreys and Thomas, 2017; Eneanya et al, 2019).

Lymphoedema specialist health professionals are commonly therapists and nurses with supplementary education who predominantly treat lymphoedema of the limbs and breast. The frequency of contact with common presentations of lymphoedema leads to familiarity, experiential learning and, arguably, confidence in managing the conditions. In addition, education/training programmes use the swollen limb as the object for skill teaching, for example, when learning to apply compression bandaging. Anecdotally, the context in and frequency with which health professionals treat patients with genital oedema vary considerably.

Given this background, it might be expected that lymphoedema practitioners would describe lower confidence in their knowledge of genital oedema and its management than of the more common presentations of lymphoedema. Further, since research on which to base good education is lacking and

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practical training tends to reflect common practice, genital oedema can be anticipated to be an area of education need.

Little is known of how much use is made of tools for differential diagnosis of genital oedema, for example, Weinberger et al's diagnostic algorithm (2007) and National Institute for Health and Care Excellence (NICE) (2019) guidelines for male genital oedema, but fewer diagnostic guides are available for females (Pinto e Silva et al, 2015). Further, the proportion of patients with genital oedema accessing lymphoedema practitioners by self-referral (without medical examination) is also unknown. A knowledgeable assessment by the health professional in the lymphoedema clinic would be essential in such circumstances to identify red flags requiring medical investigation. The specific training/education received by therapists and nurses to assess and treat patients with genital oedema have not been previously investigated (Noble-Jones et al, 2019a).

AIM

The aim of this international project was to investigate the perceived education needs of health professionals who conservatively manage genital oedema/lymphoedema in adults and children and the preferred method of accessing that education.

METHOD

This mixed-method investigation used an online survey and three large focus groups. This report relates to the survey findings only, while the focus group findings are reported in a partner article (article 3 of this supplement). The survey questionnaire was developed through a systematic process of literature review, stakeholder consultation and piloting. The last included a workshop held during the International Lymphoedema Framework (ILF) conference, in Chicago, US, in June 2019. This was attended by 28 purposively selected lymphoedema experts from various countries (specialist clinicians, academics and researchers). The final versions of the questionnaire were accessed through two hyperlinks; the first was to an English language survey distributed by the ILF, Lymphoedema Network Wales (LNW), British Lymphology Society (BLS) and by cascading to connected professionals via Twitter. The second linked to a French version cascaded through the Société Française de Médecine Vasculaire (SFMV) in France and was made available to the ILF Framework in Canada.

Health professionals managing patients with lymphoedema in Wales were given access to the survey from 15 August 2019 to 15 September 2019. The survey was subsequently made available to a wider global audience from 20 September 2019 to 30 November 2019.

Data analysis used descriptive statistics and thematic analysis of open-text responses, primarily by the main author and subsequently checked by the research team.

APPROVALS AND FUNDING

The project was reviewed by the research and development department of Abertawe Bro Morgannwg University Health Board (now Swansea Bay University Health Board), which confirmed that, based on the information provided, the project was considered a service development. The project was funded by Tenovus Cancer Care (Wales) and sponsored by Swansea Bay University Health Board.

FINDINGS

Survey respondents

The survey received 688 respondents, of whom 583 were health professionals from 31 countries, and 91% of these professionals were women. The survey completion rate was 73%. It should be noted that the n value provided throughout this report will be the number of respondents to a particular question, for example,558 responded to the question regarding profession, while 571 responded to the one regarding years of experience.

There were respondents of many different registered professions and specialities. Many performed multiple roles. Although 49% (273/558) were physical/physiotherapists, 17% (96/558) were nurses and 11% (61/558) were medical doctors/surgeons, 55% of all the health professional respondents (308/558) described themselves as lymphoedema therapists, and 18% (103/558) described themselves as MLD/massage therapists. The majority were very experienced in working with lymphoedema patients; 67% (384/571) of the health professional respondents had over 5 years' experience. Further, most already managed genital oedema (77%; 442/572), and a further 14% (81/572) anticipated doing so soon.

Access to services, patient assessment and treatment

The most commonly described referral method of patients with genital oedema to lymphoedema services was specialist hospital physician to 73% (377/519) of respondents, followed by the family physician (51%; 265/519), patient self-referral (44%; 229/519) and clinical nurse specialist (42%; 217/519). Other sources of referrals reported by 12% (60/519) of respondents included allied health professionals, oncology team members, urology and vasectomy clinics, other types of medical or surgical physicians and community nursing teams. However, there was a common perception of late identification of the problem (79%; 409/517) and delayed referral (50%; 260/517). Only 7% (38/517) of respondents indicated experiencing early identification and prompt referral of patients with genital oedema.

Most respondents (83%; 410/494) used generic assessment documentation rather than a genital oedema-specific assessment or self-report tool. Only 16% (79/494) used previously published genital oedema-related tools (Carter et al, 2010; Yost et al, 2013; Noble-Jones et al, 2019a), and 7% (34/494) used other tools, such as ones they had devised themselves.

Common treatment approaches used were skin care advice, compression garments and manual lymphatic drainage (MLD), used by 85%, 84% and 83% of respondents, respectively (n=492). Respondents also reported using exercise (64%), bandaging (62%), kinesiotaping (48%) and external scar massage/work (33%). A range of other modalities was used by some respondents, including 26% (129/492) reporting those based on wound care skills and knowledge.

The most common onward referral was for reductive surgery (11% of respondents), followed by lymphatic surgery (8%) and a dietitian (8%); slightly fewer referred patients on to sexual health (7%) and mental health services (6%) (n=492).

Education received by lymphoedema health professionals

Only 5% (25/498) of the respondents indicated that existing lymphoedema specialist training (e.g. Certified Lymphoedema Therapist) was adequate to manage patients with genital oedema. Fewer than half (42%; 208/498) had completed supplemental genital oedema management training/education; of these, 90% (188/208) said this extra education was needed. This compares with over 93% (269/288) of

TABLE 1. **Difference in weighted average confidence in knowledge with/without supplemental GO education**

Topic	Years of experience	Weighted average confidence in knowledge score range 1-3		Difference in score	% difference confidence in knowledge
Clinical Reasoning in GO management		Without GO education	With GO education		
	0-1	1.21	1.70	0.49	24.5%*
	2-5	1.60	2.06	0.46	23.0%
	6-10	1.77	2.04	0.27	13.5%
	over 10	2.01	2.28	0.27	13.5%
Other physician/ medical treatments	0-1	1.15	1.11	-0.04	-2%*
	2-5	1.29	1.36	0.07	<4%
	6-10	1.26	1.48	0.22	11%
	over 10	1.48	1.64	0.16	8%

*not statistically reliable due to low number of respondents with 1 year or less experience (p=0.05). GO=genital oedema

TABLE 2. **Topics indicated as essential, useful and not required in GO education**

Topic	Essential		Useful but not essential	Not required	Don't know	Respondents n=
	responses	As % of n				
Skin care and reducing infection risk	429	97	11	1	1	442
Garments for compression and support of GO	426	96	16	1	1	444
Self-drainage/simple lymphatic drainage for GO	412	93	29	1	1	443
Common pathologies and medical intervention leading to a risk of GO	403	91	38	2	2	445
Overcoming difficult conversations in relation to GO	399	90	41	1	1	442
Patient factors that will impact on providing treatment for someone with GO (including psychosocial)	397	89	42	0	5	444
Manual lymphatic drainage (MLD) for GO	393	89	46	2	3	444
Assessment and evaluation techniques for GO	388	88	51	1	12	442
Common surgery leading to a risk of GO	383	86	59	3	2	447
Health professional factors that will impact on providing treatment for someone with GO (including psychosocial)	380	86	57	2	4	443
Cultural and ethical concerns in treating GO	376	85	55	4	7	442
Bandaging for GO	375	85	61	3	4	443
Clinical reasoning for GO-related treatment decisions	371	84	60	1	11	443
Exercise prescription for GO	369	83	61	3	10	443
Legal concerns in treating GO	361	82	58	7	17	443
Assessment documentation for GO	360	81	70	2	12	444
Anatomy of male pelvis and connected musculature	357	80	82	2	7	447
Anatomy of female pelvis and connected musculature	358	80	82	2	5	447

*not statistically reliable due to low number of respondents with 1 year or less experience (p=0.05). GO=genital oedema

those who had not yet completed supplemental education but felt it was needed.

CONFIDENCE IN KNOWLEDGE

A consensus process with lymphoedema, urology, gynaecology and pelvic floor specialists was used to develop a list of topics considered pertinent knowledge. Participants rated their confidence against each topic as 'not confident at all', 'somewhat confident', 'very confident' or 'not applicable'. For analysis, these responses were weighted 1, 2, 3 and 0, respectively. From the combined respondent scores, a weighted average was calculated (range, 1–3); for example, a score of 2.8 would indicate a high level of confidence across respondents, while 1.2 would indicate a very low level of confidence.

Across all respondents, the greatest confidence in knowledge was indicated for 'skin care and reducing infection risk', 'self-drainage/SLD' and 'manual lymphatic drainage (MLD)' (each scoring a weighted average of 2.4), followed by 'compression and support garments for genital oedema' (score=2.3).

Background knowledge, such as 'anatomy of the male and female pelvis,' and 'common pathologies', 'medical interventions' and 'surgery leading to risk of GO' each scored around the midpoint of 'somewhat confident' (1.9–2.2).

Respondents indicated least confidence in their knowledge regarding 'legal', 'cultural/ethical concerns' and 'genital oedema assessment/evaluation' (each scoring 1.7). The lowest scores were related to 'assessment documentation'

KEY WORDS
- Lymphoedema
- Education needs
- Health professionals
- Genital oedema
- Education resources

(1.6), 'self-report tools' (1.5) and 'other treatments offered by physicians' (1.4).

Impact of supplementary genital oedema education on confidence in knowledge

In all respondents who had worked at least 1 year in the field of lymphoedema, confidence in overall genital oedema knowledge was an average of 8%–12% higher in respondents with, rather than without, supplementary genital oedema education. The difference was even greater for some topics when analysed individually, for example, 'bandaging for genital oedema' showed a weighted average for 'confidence in knowledge' that was 21.5% higher in those with, than those without, supplementary genital oedema education. The topics for which supplementary education made the greatest perceived difference, regardless of years of experience, were bandaging (21.5%), clinical reasoning (19%), assessment documentation (18.5%), taping/other adjunct treatments (18.5%) and MLD (17%).

Health professionals' years of experience and impact of supplementary genital oedema education on confidence

There was a difference in the confidence score between those with and without supplementary genital oedema education across topics, and this difference was the most stark among respondents who were in the first 5 years of working with patients with lymphoedema. After >10 years of lymphoedema experience, the difference remained for all topics, although it was minimal for topics of least confidence, such as 'other physician/medical treatments for genital oedema' (Table 1). To understand whether the low-scoring topics were considered non-essential by respondents, the next key question addressed 'essentiality'.

Essential topics for education on managing genital oedema: respondents' views

Respondents were asked to indicate whether each topic presented was 'essential', 'useful but not essential', 'not required' or 'not applicable'.

Eighteen of the 24 given topics were identified as essential by 80% or more of respondents (Table 2).

The following topics were considered essential by 52–65% of the respondents: 'using self-report tool with genital oedema patients', 'awareness of surgical treatments for genital oedema', 'taping and adjunct treatments', 'other physician/medical treatments'. The two topics least frequently scored as essential and most frequently indicated as 'not required' were 'pneumatic pumps in genital oedema management' (31% (n=137), essential; 11% (n=49), not required; n=443) and 'electrotherapy techniques for genital oedema' (22% (n=98), essential; 10% (n=44), not required; n=443).

In the open-text responses, another 88 suggestions were made of essential topics, each by only one to three respondents, indicating the breadth of education need.

Respondents' personal top three education needs

A total of 1200 open-text response items were provided by 425 respondents in identifying their individual top three education needs for managing genital oedema. These were categorised by the research team into four themes: assessment, treatment, underpinning knowledge and education/service development. Table 3 shows the number of response items in each set within the four categories and as a percentage of respondents.

Despite respondents indicating a high level of confidence in knowledge regarding compression bandaging and garments (weighted average=2.3) earlier in the survey, the most common set of items in the top three needs were issues regarding compression treatment (70% of respondents). Further, compression was second only to skin care as an essential topic within education (96%). Other similar threads were drawn through the survey—for example, confidence of knowledge regarding legal concerns scored an average 1.7 across all responders (lower than 'somewhat confident'), 82% of respondents considered it essential content and 35% of respondents indicated that, for them, it was a top three individual need. It was a similar position in relation to clinical reasoning (weighted average confidence=1.9; 84%, essential; 47%, top three needs) and patient assessment (weighted

TABLE 3. **Individual respondents' top education needs by category with respondent number and percentage (n=425)**

Assessment-related items	**Treatment-related items**
194 (46%) Patient assessment	298 (70%) Compression
148 (35%) Psychological and legal	241 (57%) Self-management/other treatments
96 (23%) Outcome measures	180 (42%) MLD
45 (11%) Confidence and communication	155 (37%) Surgical and medical options
Education and service development	140 (33%) Skin care and cellulitis risk reduction
198 (47%) Learning and teaching other content e.g. clinical reasoning	131 (31%) Exercise and activity
94 (22%) Learning and teaching medium / method e.g. workshops, videos	108 (25%) Palliative/end-of-life care
23 (5%) Protocols and pathways of care	15 (3%) Continence and sexual health
14 (3%) Other general issues	10 (2%) Internal examination/treatment
13 (3%) Lobbying and raising awareness	**Supporting/underpinning knowledge**
	114 (27%) Anatomy and physiology
	59 (14%) Pathology and causes
	23 (5%) Gender-specific and paediatric

average confidence=1.7; 88%, essential; 46%, top three needs).

Format of education resources

Responses to the question of how best to deliver educational resources showed a range of views, with individuals often indicating a desire for multiple learning formats. The four formats considered essential by most respondents were:

- E-learning package (70%; 293/420)
- Film/video by clinical expert (68%; 285/424)
- Development of video prescriptions (education films for patients) (61%; 258/421)
- Leaflet/booklets/folder of printed materials (58%; 243/417).

Collaborative events with pelvic health specialists and urology specialists were considered essential by 55% (231/417) and 53% (221/417), respectively, and slightly fewer (50%; 210/419) considered an International Lymphoedema Framework-dedicated workshop and a collection of case studies as essential.

CONTRIBUTIONS OF NON-HEALTH PROFESSIONALS

The approvals for this project did not allow data collection from non-health professionals, but patient, carer and corporate respondents were appropriately signposted to contribute their expertise/experience or where appropriate, for advice. People with personal experience of genital oedema subsequently contributed to the creation of videos for patients, carers and health professionals through associated projects (e.g. www.medic.video/w-lymph-edu).

DISCUSSION

This unique enquiry to explore the education needs of lymphoedema practitioners in the management of genital oedema/lymphoedema was a pragmatic work to fill a distinct vacuum in existing knowledge. Spanning 31 countries and amassing the views of 583 health professionals, the survey had a higher number of respondents from more countries than expected. In addition, the survey completion rate of 73% was excellent for a questionnaire that was long and detailed. These are indicators of a high level of engagement with the subject (Cunningham et al, 2015; Corner and Lemonde, 2019). The findings of this survey, combined with those of the subsequent focus group, allow for a degree of confidence in the recommendations from this report (Table 4).

The finding that most respondents already manage genital oedema and could identify ongoing education needs may indicate that the complexity of managing this condition becomes more apparent in practice. Whether they had undertaken specific genital oedema education or not, most considered supplemental education specific to genital oedema necessary (90% and 93%, respectively). The impact of genital oedema education was demonstrated by the higher weighted average confidence in knowledge, regardless of years of experience but particularly the first 5 years of lymphoedema practice, among those who had received this education. In reviewing curriculum content and teaching methods, education providers should consider the topics where this education has made the greatest and least difference, particularly in relation to how essential health professionals indicated a topic to be.

With regard to specific topics, synthesis of findings across the survey questions produced interesting profiles. For example, with regard to compression (bandaging and garments) for genital oedema, the vast majority (96%) of respondents stated that education on this topic was essential, and respondents indicated a relatively high level of confidence in their knowledge overall. However, when asked for their top three education needs, 70% of respondents identified compression-related issues. This key element of conservative lymphoedema management is more difficult to achieve on the irregularly shaped genitals than on (broadly) cylindrical limbs. The prominence of compression as an education need likely reflects dissatisfaction with current compression methods, and the availability of garments and how the skill is taught must also be considered. This issue was discussed in the focus groups to generate a deeper understanding (see article 3).

Anecdotal reports of health professionals' concerns regarding legal, ethical and cultural issues in assessing and treating genital oedema prompted the inclusion of these topics in the survey. Synthesis across questions identified that confidence in relevant legal knowledge was low among respondents, despite 82% considering it essential content and, perhaps indicating that this is not covered satisfactorily in education. Over a third of respondents identified this as one of their top three education needs. There are no similar data available regarding treatment of arm, leg or even breast lymphoedema, but it seems likely that the concerns are heightened because of the intimate, private nature of the genitals. This was further explored in the subsequent focus groups.

'Clinical reasoning' and 'patient assessment' showed similar threads of low confidence, high sense of it being essential knowledge and a relatively high proportion of respondents identifying these in their top three education needs. Very few respondents (16%) reported using specific genital oedema assessment or self-report tools. Differential diagnosis of genital oedema can be complex, and, given the relatively infrequent contact with genital oedema, guidance by algorithm and/or assessment tools may be useful to identify important red flags (Weinberger et al, 2007; NICE, 2019). While not replacing the decision-making skills of a health professional, an algorithm and other decision-support tool can be useful (Semigran et al, 2016). There is a sparsity of genital oedema-specific assessment tools, and existing ones require validation for use in diverse contexts and languages before wider dissemination.

An unexpected finding was that over a quarter of the respondents had used wound care in their management of genital oedema. Consequently, this was not a separate question in relation to level of confidence, essentiality and specific education need. However, wound care skills were not reported as a top three need even in the open-text responses. It seems likely that those with existing knowledge of wound care were able to apply it when relevant, whereas others may be unaware that such skills and knowledge may help even in genital oedema management. This is a specific area that educationalists should consider; bridging knowledge between wound care specialists and tissue viability specialists would be useful, for example, in dealing with macerated skin and lymphorrhoea (weeping

TABLE 4. Recommendations from this education need project

Education
1. Supplemental genital-oedema-specific education should be available to lymphoedema therapists and those HCP expected to manage patients with genital oedema/lymphoedema.
2. Genital oedema education needs to be informed by contemporary patient experience, research evidence and multi-professional, inter-speciality collaboration.
3. Organisations, such as ILF, could play a role in drawing together international expertise in workshops (in person and virtual) and special interest groups to promote sharing and refine knowledge of genital lymphoedema. This should include, where appropriate, the experience of patients and carers either by direct involvement or from research of patient experience.
4. Genital oedema education needs to include resources that are accessible in multiple formats (e-learning packages and online information, videos for HCP and for patients, printed materials).
Service development: tools
5. The findings of this research suggest pragmatic development, validation and evaluation of: a) Algorithms for differential diagnosis for doctors and advanced practitioners/lymphoedema specialist nurses/therapists b) Specific assessment and evaluation tools for the genital oedema c) Treatment pathways in urology and gynaecology which identify people at risk.
Research
6. Research is needed to detect the incidence and prevalence of genital lymphoedema and to identify those patients who may be at greatest risk. This will require large population research in collaboration with pelvic health professionals (urology and gynaecology) and paediatric specialists.

skin) in the community (Lewis and Morgan, 2008).

Self-referral of patients with genital oedema was accepted by 44% of respondents. This may have been helped by earlier work to encourage reticent men to self-report genital oedema. However, over half of the respondents reported that they do not accept self-referral of patients. There are many possible explanations for this, such as contextual differences internationally, different accessibility to lymphoedema services, cultural attitudes and, perhaps, a lack of recognition among medical specialists leading to patients having to refer themselves. Further research is needed to better understand this finding.

Concurrently, understanding of genital oedema has benefited from advances in lymphatic vessel investigations (Hara and Mihara, 2017), developments in microsurgery (Aulia and Yessica, 2020) and continued development in the assessment of gynaecology-related lymphoedema (Carter et al, 2019). In addition, its management has featured in several recent national and international conferences and seminars (Australasian Lymphoedema Association (ALA), 2020; British Association of Urology Nurses (BAUN), 2020; British Lymphology Society (BLS), 2020; National Lymphedema Network (NLN), 2020). However, for the health professionals practising day-to-day in lymphoedema services, information remains dispersed, access to education varies widely and specific guidelines are lacking. An international position paper on the conservative management of genital lymphoedema is needed and is being discussed by the ILF as a priority.

The breadth of resource format types identified as useful by the surveyed population is a reminder to education providers that, although e-learning is widely used, there is still a need for more traditional resources. Over half of the respondents indicated a need for 'leaflets/booklets/folder of printed materials'. Despite availability of digital education materials, printed materials and their online format remain popular for health journals and position documents through the websites of organisations such as the International Lymphoedema Framework (ILF) and BLS and their international equivalents. Similarly, learning from case studies or patient vignettes is a very traditional form of learning, particularly where patient numbers may be too few for large randomised controlled trials. There are numerous dispersed case studies of genital oedema published, but no single collection where these could be contextualised for the learner. This would be a useful addition to an educational resource on genital lymphoedema.

Collaborative events with other professions/specialities, which can be in-person or online, provide an excellent opportunity for breaking down information silos, developing new understanding and facilitating joint projects that could benefit a wider population of patients, for example, lymphoedema and wounds (Lewis and Morgan, 2007) or lymphoedema and urology (Noble-Jones et al, 2019b). More multidisciplinary/cross-speciality collaboration and publication would help fill existing knowledge gaps.

The use of videos can enhance accessibility through mobile phones and other common technology to a global audience of professionals and patients. The inclusion of patients, some of whom had made contact through this project, in the creation of information videos on genital oedema for patients and carers has been very well received, and these resources have been accessed by users in many countries. They include ones specifically aimed at health professionals and are available at http://www.medic.video/ilf-lymph-edu

LIMITATIONS

The range of participating countries and number of respondents from each is likely to indicate the way the survey was distributed rather than the level of education need in each country. Pre-testing and other mitigating efforts sought to reduce the limitations common to online surveys, such as those of accessibility, self-selection bias and question interpretation, but these are acknowledged to exist nonetheless (Arafa et al, 2019). For example, with regard to question interpretation, respondents from Australia tended to include laser treatment as an electrotherapy technique, whereas this may not have been a consideration in counties where laser use is more limited and was instead identified as an issue in the open-text responses. In relation to the reach of the survey, dissemination of the questionnaire hyperlink could not be achieved equally in all countries. Genital lymphoedema from filariasis is an endemic problem in parts of India and Africa (Manokaran, 2005; National Health Portal India, 2016; Eneanya et al, 2019), but the survey did not reach health professionals managing this condition. Further research is needed into the particular education needs of those managing filariasis-related lymphoedema.

The need for education by health professionals does not occur in a vacuum; the wider population attitudes and healthcare practice, policy and procedure, as well as the co-existing lack of empirical evidence, are all relevant. This aim of understanding the impact of context underlay the rationale for holding focus groups with lymphoedema professionals in different countries and is reported separately.

CONCLUSION

An international survey identified education needs in relation to conservative management of genital oedema/lymphoedema. Health professional respondents identified specific individual educational needs as well as topics of knowledge they considered essential in education provision. Accessibility to resources, including online and printed resources, and inter-speciality knowledge sharing were identified as necessary. **JWC**

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KEY POINTS

- **There is very little guidance for the health care professionals (HCP) conservatively managing patients with genital lymphoedema.**
- **Algorithms and assessment-evaluation tools are needed to improve and expedite diagnosis.**
- **Health care professionals managing patients with genital oedema have education needs regarding skills such as bandaging and contextual knowledge such as legal and ethical issues.**
- **Supplemental education, specialist networking and online resources are needed by health professionals managing genital oedema**
- **Wound care in relation to genital oedema was unexpectedly common and requires further investigation.**

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Contextual factors affecting the education needs of lymphoedema health professionals regarding genital oedema

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ABSTRACT

The impact of the care context on education needs of health professionals who conservatively manage patients with genital oedema is unknown. This study aimed to explore the contextual factors surrounding the treatment of patients with genital oedema that affect the education needs of lymphoedema practitioners, and identify interventions that might support both practitioners and patients. Following scoping groups in Australia and the US, formal focus groups were held with lymphoedema practitioners in England, Wales and the US, as part of an international mixed-method project in 2019. Ethics review from Swansea Bay University Health Board Research and Development Department found ethics approval unnecessary. This project was funded by Tenovus Cancer Care. A total of 69 lymphoedema practitioners (21 from England, 20 from Wales and 28 from the US) self-identified a personal education need and a need in other health workers who should be identifying and referring patients to their services. Genital oedema was considered to be under recognised, of complex aetiology and psychosocially challenging. A need for specific assessment tools, outcome measures and a diagnostic algorithm was identified. In addition, improved availability of acceptable compression products was suggested to possibly help normalise discourse around genital oedema with patients and its management. The payer model in the US means an early intervention/risk reduction approach is more difficult. Importantly, learning support resources need to be readily accessible. The focus groups provided contextual understanding regarding the genital oedema education needs of lymphoedema practitioners and the factors affecting patient treatment.

INTRODUCTION

The specific education received by health professionals (nurses, physiotherapists, occupational therapists, radiographers and doctors) to assess and treat patients with genital oedema has not been previously investigated (Noble-Jones et al, 2019). This article reports the findings of focus group discussions conducted in the US, England and Wales to explore the factors influencing the education needs of health professionals in these countries. These occurred in conjunction with an international online survey of education needs (reported separately in this supplement) and followed initial scoping discussion groups with lymphoedema practitioners in Brisbane, Australia, and Chicago, US.

Most health professionals managing patients with lymphoedema in the NHS are registered health professionals with post-registration certification in lymphoedema management (British Lymphology Society (BLS), 2016). Similarly, in the US, practitioners are licensed health professionals who are encouraged to complete lymphoedema certification training to be a certified lymphoedema therapist (CLT). Some may choose to complete an advanced national certification through the Lymphology Association of North America (LANA) (2020). Anecdotally, in the UK, US and Australia, and in contrast with learning lymphoedema management of the arm or lower limb, the treatment of genital oedema has predominantly been learnt experientially (on the job) rather than within formal education. It was unknown whether lymphoedema practitioners perceived this, or other factors, as affecting education needs regarding assessing and treating genital oedema.

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AIM

This study aimed to explore the contextual factors surrounding the treatment of patients with genital oedema that affect the education needs of lymphoedema practitioners, and identify developments that might support the health professional and patient.

METHOD

This report relates to findings from focus group discussions in a mixed-method investigation (online survey and three large focus groups). Lymphoedema practitioners from Wales attended a focus group on 16 September 2019, and those practising in England attended on 8 October 2019. Similarly, a focus group was held in Atlanta, US, on 18 October 2019. The questioning route of the discussion (Krueger and Casey, 2009) was based on the themes of the associated online survey. The groups each lasted approximately an hour, were audio-recorded and immediately transcribed by the lead author. Data underwent thematic analysis, initially by the lead author and, subsequently, by the research team.

APPROVALS AND FUNDING

The project was reviewed and confirmed as a service development by the Joint Study Review Committee within the Research and Development Department of Swansea Bay University Health Board (formerly known as Abertawe Bro Morgannwg University Health Board). The project was funded by Tenovus Cancer Care (Wales) and sponsored by Swansea Bay University Health Board.

FINDINGS

Focus group participants

For the purposes of reporting, the generic term 'lymphoedema practitioner' is used for a registered health professionals who has completed full certified training in lymphoedema management, regardless of whether they are a nurse, physiotherapist, doctor or other allied health professional (AHP). Quotations are identified by participant number (e.g. P4), and then W for Wales, E for England and A for the US (e.g. P4A).

Attendees were predominantly nurses, physiotherapists and occupational therapists, but included one other AHP and one doctor. In total, there were 69 participants (66 women and three men). Each focus group was large (20 participants in Wales, 21 in England and 28 in USA) and facilitated by the lead author with a local co-researcher as moderator/scribe (Krueger and Casey, 2009).

Discussion themes

The key themes identified in the discussions were factors that created a perception of education needs in genital oedema, how these are managed and suggestions for developments.

- Creating education needs
  - Infrequent contact with patients with genital oedema
  - Impact of context and wider organisation structure
  - The uniqueness of genital oedema–patient awareness, behaviour and emotions
  - Perception of genital oedema as complex
- Existing workarounds in the context of genital oedema education needs
- Education and product developments suggested by focus group participants

Infrequent contact with patients with genital oedema

While the focus group participants acknowledged that the true prevalence of genital oedema in men and women requires further research, they said that referral rates of patients with genital oedema to lymphoedema services were below their expectations.

'When you read the publications, the prevalence for gynae[cology] cancers is really high, and yet we don't see anything like that.' (P4W)

A common experience for most participants was assessing a new patient with lower limb lymphoedema, only to identify co-existing genital oedema on examination or discussion with the patient. Participants attributed this to ambivalence or a lack of awareness among the health professionals who were referring patients to lymphoedema services, such as those from community healthcare, specialist urology, gynaecology and cardiac services.

'In the doctor's notes, the patient kept going and complaining about leg and genital

swelling. He had removed the pelvic lymph nodes, they'd done radiation, and, in the notes, it said something like "once his lymphatic system has calmed down, the swelling will resolve." And by the time it didn't calm down enough, and the patient was beside himself ... now we've got this horrible situation.' (P11A)

'**The patient went to their GP, the GP didn't even look at the genitals.**'(P6W)

'**District nurses in that situation might not realise that there's genital oedema there, even though they're going in dressing a wound [on the leg]**.' (P2W)

'**Making heart failure nurses aware that it [genital oedema] could be an issue for their patients, so that they can refer on and they can recognise that it's an issue for patients ...**' (P3E)

Participants in the US identified issues related to the insurance company payer model followed in the country. The insurance companies typically only reimburse for treatments for identified medical problems, rather than preventative or risk-reduction treatments, as is the case in the UK. The health professionals believed that this practice reduced their exposure to patients with genital oedema at an early stage.

'**We have a reactive payer model here, [unlike] in Europe where it's all about prevention; you're trying not to spend money ... they know oedema is going to lead to wounds, pain, skin tissue changes ... it's hard to change a mindset here [in the US]**.' (P10A)

For participants from the US, there was also a cost benefit consideration to accessing training, especially advanced training on topics like genital lymphoedema. Their organisation rarely paid for them to attend courses or provided study leave, so they would have to attend on their own time and pay out of their own pocket for this training. For several, the return in terms of reimbursement would not justify the loss of income. However, others considered going to great lengths to seek genital oedema education.

'**I had two or three patients with genital oedema, and the certification course is [only] kinda touching on it. So, I thought, I've got to find a course. The only course I could find was in Germany, and I figured the hospital was not going to pay for me to get to Germany; so now, I'm excited to find out that there's a more advanced course [here in US]**.' (P4A)

In both the UK and US, participants reported that surgeons tended not to advise patients of a timeframe for acute post-operative genital swelling to be resolved, an issue which was recognised as being challenging.

'**After gynae[cology] surgery, they're told to expect swelling, [but] when do they expect not to have swelling? They're not advised about that.**' (P9E)

The US focus group noted that formal recognition of oedema or lymphoedema was widely lacking and remained missing from hospital registration documents or doctors' office forms (edema/lymphedema is the US spelling).

'**About 3 years ago, I did a project looking to who assessed lymphoedema, and I gathered up [data from] the major university hospitals across the United States. I only found one hospital ... that any place on those papers ask anything at all about edema or swelling or lymphedema**.' (P3A)

The lack of administrative recognition of lymphoedema led to discussion of other concerns around wider service organisation influencing the education needs of lymphoedema practitioners regarding management of genital oedema.

In the UK and US, the infrequent contact with patients with genital oedema not only reduced potential experiential learning opportunities but also meant that, when decisions were made regarding spending resources (time and money) on professional development, the more common presentation of lymphoedema (breast cancer-related or chronic bilateral leg oedema) took priority over genital oedema. For a few participants, the lack of regular exposure to patients with genital oedema led them to question the relevance of additional education until the referral rate was improved. In contrast, others argued that an awareness of what was possible was still useful even if referrals remained low.

'**[But] if you've done it as an essential [training], you know it exists, even though** you mightn't have used it, you think "oh, I know I should be doing something about this".' (P1W)

For those who could not justify attending courses, online and other resources that could be accessed on demand were popular suggestions (discussed below).

The lack of opportunities for experiential learning contributed to a lack of confidence for some, for example, in accurately predicting the outcomes of treatment, particularly in contrast with the treatment of the more familiar swollen leg or arm.

'When you see something less frequently, it's harder to be able to predict for the patient and for yourself ... you think, "what am I expecting to happen to this now. Should it be much better than this? Is this what it should be doing?" Because you don't have ... the normal path.' (P4W)

Context and wider service organisation

Another contextual issue highlighted was ownership, in that no single professional group sees it as its role to deal with genital oedema in the acute or community healthcare setting.

'No one else is going to do it. No one is going to bandage your leg and notice there's a big lump in the middle [genital oedema], male or female ... who's going to go there, no one is, it's got to be us.' (P8E)

One participant suggested that practising in geographical isolation sometimes made accessing support or advice more difficult and that large regional oncology services (UK), such as gynaecology/urology, were not always aware of small local lymphoedema services.

'I think being in a more rural area, we don't have immediate access like regional centres would have or the links with the consultants. So, you're expected, you, that one person, to look after that individual without any support from anywhere else.' (P5W)

Compounding uncertainty in clinical decision making for lymphoedema practitioners was a lack of sufficient pertinent patient information from referrers.

'There's hardly anything on our referral form ever. Trying to find out exactly what they've had done ... it could be made a lot more streamlined if they actually put more information.'(P10W)

In addition, the US group pointed out that treatment of complex lymphoedema, such as genital oedema, was seen as time consuming and not popular in a payer model favouring quick or group treatment.

'The trouble is everything is time consuming. I mean, just traditional OT (occupational therapy) clinic can have four people at a time, running the different exercise programme, you know they have one therapist. But, with lymphoedema, it's one on one. So they [the employer] don't really like that.' (P9A)

'Because were not profitable, that way.' (P5A)

Therefore, a combination of organisational processes and low referral rate had the two-pronged effect of reducing experiential learning and deprioritising education in genital oedema management.

Uniqueness of dealing with genital oedema–patient awareness, behaviour and emotions

Heightened communication skills were recognised as needed to sensitively navigate patient behaviour and for open discourse due to the lack of awareness of risk or the lack of relevance of genital oedema symptoms among patients.

'Lots of our patients wouldn't realise that it [genital oedema] was even relevant ... written it off as something else.' (P4W)

'They [patients] don't know where to go ... because they [patients and health professionals] don't even know what we do or what we are capable of doing.' (P9A)

Even if a patient was aware of the potential management of their genital oedema symptoms, they were not always ready to accept treatment. This would be true of any self-care advice, but the participants felt it was particularly relevant to this subject. Moreover, while the care of each patient needed to be individualised, some standardisation and guidance would be welcomed, including protocols for other health professionals.

'[It] can be very specific to each person. Their main concern may be not necessarily to do with the oedema. [We need to know] the services that we can signpost (direct patients) to'.' (P12W)

Others' experiences demonstrated the range of different ways that patients might behave in disclosing this potentially embarrassing problem and the skills needed by the health professional.

'He [the patient] was just loitering for a bit, trying not to end the conversation, and [suddenly] he was like, "Well, what are you going to do about this?" And he dropped his trousers in front of me; I tried not to look too shocked. But he said he'd spoken to his GP about [it] and had had treatment for cellulitis, because he said he was sore. He had quite a large scrotum, but he had no penis at all. So, yes, I think it's about having more information so that you're not so shocked about things, and you have more of an idea of what you would expect.' (P4W)

In discussing emotional and psychosocial barriers to conversations between patients and health professionals about the genitals, the adjectives used by the focus group participants included 'emotive,' 'tricky,' 'private,' 'intimate' and 'a guarded area.'

'It's complicated by the patient's perception. It's a private place that some people can't talk about, or even to admit they've got it.' (P8E)

The emotional impact of genital oedema on the patient could be profound, and this can have a knock-on impact on the attending professional, as this same participant described.

'So, I was trying to show him it [how to bandage], and he got it. So, I said, "Let's you do it," and he said, "It hurts, it hurts." So, I said, "Where does it hurt?," and he said, "Here, [pointing to his heart] I can't do this." That was a massive wake up for me … it's emotive. It's a body part that's not just a breast, or an arm or a leg.' (P8E)

A participant from the US group described the inability of patients to adhere to treatment recommendations when they felt overwhelmed and when compression bandaging/garments affected their appearance and work.

'I feel the frustration with my men, in particular, with the bandaging aspect, and that idea of having to re-bandage, maybe twice a day or three times a day. After a while, it just becomes too overwhelming. It gets really bulky for them, especially if they are in sales, where image is everything. It's hard for them to buy into, and, if they're not buying into it, it's hard to treat them.' (P2A)

The focus group participants suggested that more specific genital oedema education would enable them to deal with such unpredictable psychosocial demands.

Participants from all three focus groups discussed the emotions they experienced in managing this patient group. These included shock at the appearance or the manner in which the genital oedema presented, feelings of helplessness in dealing with patient misunderstanding or health beliefs, their own uncertainty, fear of being out of their depth, feeling overwhelmed and frustrated. In addition, they felt concern about their own and their patient's vulnerability, which included feeling incompetent, embarrassing the patient and concerns around accusations of misconduct in treating an intimate body part.

'You don't want to be fumbling in front of your patient. They're embarrassed already. They want you to look confident.' (P3E)

'That kind of thing can make you feel quite vulnerable, because you don't know where you can refer on to, or there doesn't feel like there's anybody out there to refer on to. So, again, it's exposing the vulnerability of the patient, but also you as an individual clinician; it can be difficult. It's about consent and protection of us as staff and, you know, particularly opposite sexes.' (P5W)

Concerns were expressed in all three focus groups that practice was not standardised, for example, in checking for genital oedema as a co-existing problem when patients are referred with bilateral leg swelling.

'Making sure that we were being consistent in our approach: are we really addressing those people when they are quite likely to have genital swelling but not likely to divulge it?' (P4W)

'There's not much written about it at all, and we tend to rely on experience of what we've talked about it at conferences and things, but it's not actually a document. How do I know that the way my team and I are treating patients is that what we should be doing?' (P1E)

Perception of genital oedema as complex

As described above, the participants cited several reasons for this patient group having complex needs, but there was underlying recognition that the aetiology of genital oedema could also be complex.

'I think it's not knowing the underlying cause; with genital oedema, it's not so straightforward. It's having that support medically as well and confidence to refer patients on that actually need further investigation.' (P8W)

'There's not much understood, there's not much written about genital oedema and the underlying pathology behind it; so, why is that person getting genital oedema, rather than just one leg or two legs. It's about being able to assess genital oedema as part of a holistic assessment. But having been taught to do that, then it certainly helps to know why it's done in the first place.' (P3E)

'Then, there's the complexities of genital oedema within a very complex patient, that has the heart failure and he's end of life.' (P7E)

Obesity is a common factor affecting outcomes for patients lymphoedema, and it is particularly salient in relation to genital oedema management, both in terms of physical access to the oedematous part and as a contributing factor to the oedema. The willingness and capability of patients to address their obesity was identified as a factor affecting management of genital oedema.

'I feel that I am quite knowledgeable and trained to handle genital oedema, but I know if I don't handle the obesity, I still won't achieve the outcome that I want.' (P6W)

Differential diagnosis can be particularly important in paediatrics and in relation to repeated infections.

'There are other diagnoses that have to be considered. You know they might not think to ask about diarrhoea, history of Crohn's disease of the gut. Then, there's the recurrent cellulitis issue, that's a problem I find: do you treat them with the same antibiotic as you would a recurrent cellulitis in the limb? Sometimes not. There are so many different medical reasons, I think, that need to be considered, and that's why I think it's quite a daunting topic.' (P1E)

The likelihood of comorbidities and the complexities of presentation of genital oedema could lead to decisions regarding the priorities of care that may omit/delay genital oedema care.

'In terms of the tension at the time of the appointment, those patients also have bilateral legs [swelling] and, invariably, lymphorrhoea with it, and if it's that hour appointment, clearly, you need to prioritise. So, I make that decision ... I cannot do it all.' (P12E)

Sometimes, the inherent complexity of genital oedema was practical in nature. The shape variations in genital oedema often means that made-to-measure compression garments are required. Even with training, these can be difficult to get right, and the shape can be changeable.

'[Made-to-measure garments] It's quite a difficult thing to get right [even] when you know what you're doing. So, if it's then wrong, I think both therapist and patient think, "Well, I'm not going there again," "made-to-measure hasn't worked." So, it can be quite exasperating.' (P9E)

Many people in the focus group acknowledged a fear of finding themselves out of their depth in a conversation with the patient. This included lacking the time to deal with issues of intimacy and the importance of being able to signpost appropriately for the safety and wellbeing of the patient and to ensure the health professional remains within their scope of practice.

'We can deal with certain emotional issues, but I think you've got to recognise a point whereby you're not able to help that patient any further, and you've got to know where to signpost them.' (P13W)

'I'm always anxious hearing (HCPs) talking about that, you open a can of worms. If they open it, I don't want to leave the patient high and dry. I've had a situation where I've had a gentleman with genital oedema who actually opened up to me and said that he was abused when he was young.' (P6W)

The importance of the lymphoedema practitioners recognising the boundaries of their competence and responsibility, particularly with this group of patients, was recognised.

In contrast to the complexity perceived

by lymphoedema practitioners, a frustration for one of the US participants was that the surgeons and physicians oversimplified the management of genital oedema. Participant P2A suggested that this was part of the reason for a lack of referrals to therapists.

'I think it has a lot to do with just the fact that, if you look at the history of oedemas, well people live with them. Standard teaching in medical schools [is] the oedema is there, throw Lasix at them or some compression hos(iery), and that's what they did for years. And then, here comes therapists along, who can now treat it. Well, they got along fine without us before, right? Because no-one died immediately at the first sign of swelling.' (P2A)

In summary, a wide range of emotions and practical reasons specific to managing this group of patients were described, along with associated factors that affected the confidence of the health professional to manage genital oedema as a complex condition. It was recognised that some difficulties might be mitigated and greater confidence might be gained through genital oedema-specific education, but it would not resolve all difficult situations.

EXISTING MITIGATING ACTIONS TO ADDRESS GENITAL OEDEMA EDUCATION NEEDS

The participants in these three large focus groups described actions, such as seeking support on an individual basis, to mitigate any impact on the patient from an unmet education need.

'I purchased, before I could find a course, a video. So, what I would do is, I would watch it every time I would get a patient. Even though I don't do it that often, when I do it, I want to know how to do it right.' (P4A)

Participants described the levels of support as local, regional, national and remote, and the support depended on the context in terms of type of patient and location of service. One-to-one support occurred between those of different professional backgrounds within and outside of the speciality, for example, a lymphoedema-specialist physiotherapist drawing on support from a lymphoedema nurse colleague. In some places, larger lymphoedema team discussions occurred, particularly when the problems presented by patients seemed increasingly complex. One team had worked to establish useful phrases for dealing with difficult conversations with patients.

'I think it's having a few phrases that are key ways of just starting that conversation. [Within teams, asking each other] "What do you use? How do you say it? When do you do it? At which point do you do it?"' (P4W)

In summary, mitigating the impact of unmet education needs on patients included seeking within-team and inter-professional support, resources such as videos and courses, and in the UK, national expertise where this was accessible.

DEVELOPMENTS SUGGESTED BY FOCUS GROUP PARTICIPANTS

Three main approaches were suggested by the participants to bridge the gap between genital oedema education needs and patient care: ways of addressing the knowledge requirements of health professionals managing genital oedema, improving health service processes to support identification and treatment decisions and making changes in the wider context to raise awareness.

Addressing the education need of lymphoedema practitioners

Means of gaining greater clarity on the risk of genital oedema from different primary pathology and interventions were indicated, including physical, social and psychological red flags and what outcomes to expect. Some participants suggested that the lack of direct experience could be addressed by sharing example case reports. Others proposed large projects such as the creation of diagnostic algorithms or protocols, alongside small changes to existing assessment documentation to include prompts to check for the presence of genital oedema. Separate documentation for assessing genital oedema in detail was also considered useful and educational.

'Some kind of protocol to follow ... you know, like an aide memoire, because you might not have seen those patients for a long time.' (P3W)

'It could also include when to refer for

KEY POINTS

- **Genital oedema is perceived by health professionals as a complex and daunting condition to manage**
- **Genital oedema must be normalised in conversations and not classed as an uncomfortable or sensitive topic**
- **Health professionals support the need for genital oedema assessment tools, outcome measures and a diagnostic algorithm to improve care**
- **There is a need to review the types of patient compression products available internationally**
- **The topic of genital oedema must be included in learning resources readily available for health professionals.**

surgery.' (P6E)

'I think it's about having a pathway, you know, of treatment, which you can initiate quite early on, even if you [later] go in a different direction because there's no response to that initial treatment, but starting with the basics and guiding the way, as it were.' (P9E)

Mentor support was seen as ideal but difficult to achieve. Videos, which could be easily accessed to remind the lymphoedema practitioner of practical skills, were commonly suggested.

'This might not be workable, but what about some kind of mentor? "I got a so-and-so coming in today, can I have a quick chat?" It would have helped me, it did help me.' (P8E)

'I think a series of videos ... because there's lots on MLD [lymphatic massage], on fluoroscopy, exercise, arm bandaging, leg bandaging, but there's no videos of managing genital oedema. So, to have an equivalent in that would be really useful.' (P3E)

Other online educational resources and ways of working were suggested, including webinars, but whether these should be live (synchronous) or asynchronous, given the previously mentioned time constraints, was equivocal. A key point overall was access to supporting resources on an as-needed basis that could supplement some core knowledge in genital oedema management.

Multidisciplinary working and collaboration

Each group identified that early intervention required multidisciplinary collaboration. Patient education initiatives for those at risk of genital oedema were described in some areas.

'We've had fantastic relationship with the gynae specialist nurses for over 10 years, so we do have prevention clinics for gynae as well as for breast, and it's the same format. We get them in, and we do prevention education, and we measure their legs, not genitals.' (P6W)

However, elsewhere, attempts at upstream collaboration through multidisciplinary team (MDT) meetings proved less successful in increasing timely referrals, suggesting that the health beliefs of some health professionals can be difficult to change.

'We used to have a member of staff who used to go to the MDT meeting every week, and the gynae specialist nurse was involved, and we learned from them. They learned from us. (But) After 5 years of input, and probably more than that, she's still not referring.' (P9W)

Normalising conversations about genital oedema

Enabling patients, the public and health professionals to speak more comfortably about problems of genital oedema was unanimously voiced as a positive aim and a step to normalising conversations.

'If we were able to raise awareness among the general public much more about genital lymphoedema, it might be less of a taboo subject and more like breast oedema.' (P2W)

'I just think it needs to be normalised a little bit. I think it needs to, to stop being this area that we're so uncomfortable with, and we just need to sort of completely try and normalise it more, you know, if we can.' (P11E)

'We need to have the confidence to actually ask the question. We need to actually open up that conversation and say "it's absolutely fine to talk to me about that, and I might not be able to do everything, but I can signpost you or guide you to someone who [you] can access."' (P13E)

Having a range of suitable compression products available, in the same way as compression stockings in a large range, would also help normalise the situation for the patient. Products needed to be suitable, easily accessible and affordable. Alluding to the previous example of men working in sales, where image is crucial, the bulk and inconvenience of currently available bandages and supports was considered unacceptable.

'Fast and easy for the patient to put on. Even though a lot of them don't become independent anyway, they're living this chronic lifestyle for a very long time, but getting them that [means] they can manage their dressings.' (P9A)

In summary, developments suggested by the focus groups in the UK and US included decision tools, access to mentors, videos of practical skills and short courses; Multi-Disciplinary Teams (MDTs) or sharing inter-professional knowledge; raising awareness among the public on the issue of genital oedema and normalising (and improving) those conversations; and better availability of a range of compression products.

DISCUSSION

No previous studies have explored the factors creating a perception of education needs in genital oedema management among health professionals and lymphoedema practitioners' views of the impact of these unmet education needs on patients and themselves. Issues were found to be contextual, substantive (of genital oedema itself) and psychosocial.

Neither the profession of registration (e.g. nurse, occupational or physical therapist, doctor) nor the particular 'school' of lymphoedema training made a significant difference to the views expressed by the participants in the focus groups. All participants felt that the content of lymphoedema certification programmes needed to have more content on genital oedema or that it needed to be offered as a follow-up 'masterclass' type course.

Notwithstanding the lack of large-scale studies regarding prevalence, infrequent contact with patients with genital oedema was largely attributed to a lack of vigilance and referrals from health professionals dealing with patients at risk of genital oedema. A likely reason for this is the heterogeneity of causes of genital oedema and the consequent lack of focal health speciality. Genital oedema can be a developmental anomaly (Gordon et al, 2020), symptom of disease, such as filariasis (WHO, 2020), or cancer and its treatment (Vural et al, 2020), obesity (Lister and Noble-Jones, 2017) or trauma (NICE, 2020). The findings of this report indicate that this is a chicken-and-egg situation: the limited referral rates of patients with genital oedema may be a result of the identified unmet education needs of patients, the public and health professionals, and vice versa.

In relation to the need to spend valuable resources on education about genital oedema, the rarity of the condition relative to lymphoedema of the limbs was counterpointed by its perceived complexity and the inherent need of the health professional to feel competent. There is an association between workplace motivation, learning and competence (Ryan and Deci, 2017; Noble-Jones et al, 2019). Therefore, it is even more important in rarer presentations of illness that education needs are analysed for quality, content and pragmatic means of responding. The concurrent international survey presented in article 2 of this supplement investigated the content and means of delivering education regarding genital oedema. The focus groups added insight to the difficulties that can result from ignoring such education needs for health professionals and patients and how the provision of education/training in multiple formats may pragmatically address the need.

A move in recent years to centralise services for rare cancers, for example, penile cancer, has arguably increased the gulf between multidisciplinary oncology specialists and the local services providing follow-up or long-term care (Noble-Jones and Thomas, 2021). Means of addressing isolation of lymphoedema practitioners through support networks and/or mentors are particularly relevant in these rarer presentations of lymphoedema. In England, national lymphoedema centres of excellence are available. In Wales, Lymphoedema Network Wales runs regular national clinics for patients with complex lymphoedema issues. Known colloquially as 'complex clinics', smaller local lymphoedema services can send their patients to these clinics for a care review. Health professionals are provided the learning opportunity of attending the national clinic with the patient and asking questions. No such formal support system was described in the US focus group, where practitioners instead rely on informal networks. Local peer learning and webinars were reported as the major source of lymphoedema education by the US participants, but these had not, at the date of the meeting, covered genital oedema management; however, short, weekend specialist courses were mentioned as a recent development.

Difficult conversations and taboo subjects are encountered in many areas of health and

KEY WORDS

- Lymphoedema
- Education needs
- Health professionals
- Genital oedema
- Focus groups

social care, death and dying, sexuality and sex, dementia and mental health, all of which can be more difficult with the young (Perez et al, 2020) or where language and culture add complexity (Watts et al, 2017). A limitation of the present study was that the focus groups were limited to the UK and US, which means that, although there are some differences in the care context (e.g. funding), they are not as stark as they might be among other countries, for example, Ethiopia (Wharton-Smith et al, 2019) or Zambia (Maritim et al, 2021). The focus of the Wharton et al (2019) and Maritim et al's (2021) studies was people with lymphatic filariasis (which can cause genital oedema), and they highlighted that gender, taboo and stigma can be significant barriers to accessing care, as well as highlighting the important role of education of both patients and carers in a community. Despite the differences, there is also striking resonance between the nuances of the 'hidden' parts of the body described by Wharton-Smith et al (2019) and the adjectives used by the participants in the present study ('emotive', 'tricky', 'private', 'intimate' and 'a guarded area'). Similarly, within the focus groups, the health beliefs of both health professionals and patients were found to be barriers to appropriate care, even if expressed quite differently from those in Zambia (Martim et al, 2021).

Undoubtedly, there are useful transferable skills from specialities such as oncology and palliative care for health professionals who have worked in those contexts. However, specific additional resources may be needed for management of genital oedema. Psychosocial red flags, in addition to physical ones, could be a useful addition to an assessment algorithm to identify when outward referral for psychological or cultural support may be appropriate. In addition, as with health professionals who commence working in specialist areas such as sexual health, specific training in dealing with embarrassing conditions may be helpful. A systematic review by Fennell and Grant (2019) found that published research on how nurses learn to be comfortable talking about sexual health and related matters is sparse, and guidelines for education on these potentially embarrassing topics are lacking. They concluded that knowledge, attitudes and beliefs, nurses' comfort and perceived barriers needed to be addressed at three levels—education, workplace and professional society. The findings of the present study with health professionals managing patients with genital oedema echo their recommendations for increased multilevel knowledge on matters relating to the genitalia and living with genital oedema. Increasingly, patients will access their own sources of knowledge. Trust between health professionals and patients is a complex balance of scientific knowledge and care (Baron and Berinsky, 2019). Studies such as this one, which identify specific education needs and the most practicable means of reaching the health professional, can focus scarce health resources to where they can be most impactful.

LIMITATIONS

All three focus groups were convenience samples, in that the participants were meeting for another purpose, but two (England and US) were self-selecting, in that the participants had an interest in genital oedema and/or lymphoedema education. In contrast, the participants of the Welsh focus group were meeting for a different purpose and represented a sample of lymphoedema clinical staff from NHS bands 5–8 (registered health professionals usually with over 2 years' or more of experience). All three groups were fairly large, which ensured heterogeneity of experiences but may have restricted the depth of conversation from each participant (Krueger and Casey, 2009). This was mitigated to some extent by the experienced facilitator, use of counterpoint questions and the use of a local co-researcher as moderator, who ensured that each participant was asked if they had more they wanted to contribute. Plans to have a research focus group in Australia had to be changed due to a delay in getting ethics approval, but the inclusion of Australian participants in the pre-study scoping exercise was useful to the research team for subsequent data interpretation. Similarly, although the accompanying survey had a wide global reach, time and access limitations meant that developing countries could not be included. However, as highlighted, there are a number of studies on lymphatic filariasis in some of these countries that provide useful contextual understanding.

CONCLUSION

Lymphoedema practitioners described the care context that created perceptions of education needs in relation to managing genital oedema. This included a lack of opportunities for experiential learning and a lack of recognition of the condition as treatable by referring agencies and patients themselves. Once referred, the condition was considered complex in both practical and psychological terms. Flexible and multiple means of ad-hoc support and education were considered more pragmatic than traditional face-to-face education, although a baseline of lymphoedema knowledge was considered necessary and could be included in the curriculum of lymphoedema management certification. **IFL**

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