Audit to strategy: development of a national children and young people lymphoedema service

Abstract

Lymphoedema in children and young people (CYP) can cause significant impact affecting physical, psychological and social wellbeing. This audit of 286 CYP with Lymphoedema (2015–2018) is the first national cohort reported and provides new information on patient reported outcome (PROM) changes over time. Conservative therapy produced statistically significant change in outcome measures relating to swelling, infection, appearance and compression garments. Almost half of the children had primary lymphoedema of varying types. An overall prevalence of 31 per 100 000 CYP with lymphoedema was found among a population aged 0–25 over a 3-year period. This finding suggests a higher occurrence of lymphoedema in children and young people than previously reported and is important for service planning and health professionals' education.

Key words

Lymphoedema, paediatric, clinical audit, epidemiology, clinical protocols, prevalence

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Lymphoedema in children and young people (CYP) can affect physical, psychological and social wellbeing and cause significant impact on daily life (Moffatt and Murray, 2010). Lymphoedema results from the failure of the lymphatic system to drain lymph fluid from the interstitial spaces (International Lymphoedema Framework [ILF], 2010). The term encompasses a range of symptoms including swelling, pain, decreased mobility and skin conditions (Morgan et al, 2005). CYP with lymphoedema have experienced issues with bullying, difficulty finding fashionable clothes and shoes that fit, altered personal relationships with family and in school (Hanson et al, 2018). Complicating factors include the risk of cellulitis and the psychosocial issues from having a visible but rare condition (Quéré et al, 2021).

As with adults, lymphoedema can be secondary to trauma or other pathologies but the majority in childhood are due to primary malformation and/or dysfunction of the lymphatic system (Gordon et al, 2020). Despite advances in the possibility of molecular and genetic diagnosis of primary lymphoedema (e.g. Milroy disease), diagnosis for many, particularly late-onset (e.g. Meige) or syndromic and systemic types (e.g. generalised lymphatic dysplasia), diagnosis for many, particularly late-onset or syndromic and systemic types, has been delayed by years (Gordon et al, 2020). True prevalence in CYP is unknown but for almost four decades, it has been based on an estimated average annual incidence of 1.15 per 100 000 (Smeltzer et al, 1985). Local variance may depend on study methods and regional service provision. For example, a regional comparison of overall prevalence (adults and children) in the West Midlands and Southwest of England found regional differences in prevalence (3.58 per 1 000 and 2.29 per 1 000 respectively) but in both regions children represented only 1% of the overall caseload (Cooper and Bagnall, 2016). However, as children services develop and accrue data, figures suggest that true incidence may be much higher (Todd et al, 2014).

In 2011, National Health Service (NHS) Wales coordinated disparate local lymphoedema services into

an equitable national Adult Lymphoedema Service - Lymphoedema Network Wales (Thomas and Morgan, 2017). However, CYP struggled to gain access to specialist lymphoedema services locally in which staff members were confident and competent in treating children with lymphoedema, driving them to seek help outside of Wales. In November 2015, through fixed term funding from the Welsh Government, a CYP lymphoedema service commenced in Wales. This aimed to provide local assessment and support within each of the seven Health Boards in NHS Wales. A National Children and Young People Lymphoedema Specialist post was created to support local lymphoedema therapists in developing their skills to manage this distinct group of patients. Following guidelines from the Welsh Government on the transitioning of services between childhood and adulthood the upper age limit of the service was set at 25 years with discretion to extend where special needs existed, a policy which was the subject of a recent consultation (Welsh Government, 2020). The CYP service was formally evaluated in 2019 by auditing all existing patient data. This formalised the service from a pilot to 'business as usual' (i.e. being funded as a regular part of service provision) and prompted a step-change in improvements. The findings of the audit would also enable essential knowledge sharing with national and international colleagues.

Aims and objectives

The aim of this article is to review the results of the 2019 audit and reflect on how it strategically shaped future service delivery. Objectives identified for this audit included determining the prevalence of lymphoedema in CYP in Wales, understanding the complexity of lymphoedema in this population, and exploring their self-reported experience of lymphoedema through PROMs.

Method

In 2019, a local audit of all available records of care from each person referred to the national CYP lymphoedema service was undertaken. The audited period was November 2015 to December 2018. Records included active patient cases, discharged patients and any referred patients who were not yet seen. The data indicated below were transcribed from paper notes and the LymCalc database to Microsoft Excel on an NHS Wales Health Board computer. All identifiable patient information was removed.

Data collected included date of birth, sex, date of referral, referrer's profession, and date of first appointment. Information documented from the CYP or their parent/guardian/carer encompassed date of onset of lymphoedema, areas affected by the lymphoedema, family history, cellulitis history, and the amount of pain experienced with their lymphoedema using age-appropriate visual aids to capture a score from zero to ten and was reported at initial assessment and subsequent appointments where attended, corroborated by parent/guardian where relevant. All documented objective data were collected at each point of contact (initial assessment and follow up) which included skin and soft tissues conditions, pitting test and Stemmer's sign (ILF, 2010). Height and weight were measured, and body mass index (BMI) calculated for each CYP where possible.

There is a lack of international consensus for categorising lymphoedema in children. Lymphoedema Network Wales (LNW) utilises the British Lymphology Society's (BLS) Grouping from 0 to 3c (BLS, 2016), as well as the ISL staging from 0 to III (International Lymphology Society, 2013). The ISL classification considers the physical signs associated with and the appearance/extent of the swelling without taking into account psychological and psychosocial factors. The BLS grouping takes into consideration the impairment of function and mobility of the patient, as well as the physical appearance of the swollen areas. In LNW, the BLS classification of oedema in advanced disease (British Lymphology Society, 2016) is recorded as 'Group 4' for ease of capturing the number of patients that have advanced or palliative disease with secondary lymphoedema.

A treatment outcome tool used within the CYP lymphoedema service is Patient Reported Outcome Measures (PROM) (Gabe-Walters and Thomas, 2021). PROMs capture information with a scale of 0 to 4, where 0 = Not applicable; 1 = Not at all, 2 = A little bit, 3 = Quite a bit, and 4 = A lot. This allows statistical analysis of various descriptors over time including impact of lymphoedema on school/work, hobbies, clothing, shoes, etc.

Analysis

For statistical analysis, IBM SPSS Statistics version 26 was used to calculate the means, frequency counts, percentages, one sample and paired sample t-tests and Pearson's correlations of the data, all with a confidence interval (CI) of 95%.

This study design was reviewed by the Joint Study Review Committee at Swansea Bay University Health Board and deemed a service evaluation/data audit and did not require formal NHS ethics approval.

Results

Referrals and discharges

As of the 31st of December 2018, the CYP service had 200 active patient cases, 79 discharged and 7 were unseen (4 waiting to be seen and 3 did not attend). For a population in Wales of 909 346 from birth to 25 years in 2018 (Welsh Government, 2020) this gave a prevalence of 0.31 per 1 000; or limiting the age range to the typical school range of birth to 19 years the prevalence would be 190/704 040 or 0.27 per 1 000 as presented in *Table 1*.

There was a broad range of referral sources to the CYP service. The largest number of referrals were from the adult lymphoedema services (n=56; 19.6%) transferring care which was outside of their service remit. Next were physiotherapists (n=45; 15.7%), then burns and plastics,

Table 1. Number of CYP lymphoedema service users from birth to 25 years (2015–2018)

Wales data	Population* (mid-year 2018)	Population from birth to 25 years old	No. of patients referred	Prevalence per 1000 birth to 25 years old	Prevalence per 100 000 birth to 25 years old	Active cases	Referral not seen	Discharge
TOTAL	3 138 631	909 346	286	0.31	31	200	7	79

*StatsWales, 2020

and consultant paediatricians referring an equal number of patients (n=38; 13.3% each), finally family physicians/general practitioners (GPs) referring 12.6% (n=36) of the caseload. Smaller numbers came from a broad range of other health professionals/services returning patients back to Wales.

On analysing the discharge data (n=79), 19 were of a cancer-related lymphoedema category and 60 were non-cancer related. As shown in *Table 2*, the commonest reasons for discharge were patients self-managing their lymphoedema (n=22; 28%), followed by those referred to the adult service (n=20; 25%).

Table 2. Number of CYP Lymphoedema service users from birth to 25 years (2015–2018)

Numbers of patients discharged (79)	Number	Percentage
Discharged to adult service	20	25%
Declined Treatment	16	20%
Did Not Attend	7	9%
Self-managing lymphoedema as treatment provided	22	28%
Inappropriate referrals	8	10%
Deceased	3	4%
Out of area	3	4%

Waiting times

Once referred, the average number of days waiting to be seen was 16.9 days (SD 21.6 days). However, 126 (46.8%) waited 10 days or less. A further 43 (16%) were seen in 11–20 days and 51 (19%) within 21–30 days. Some of those who waited longer were young adults who chose to remain within the adult service.

Age at referral and age of onset

The age at referral to this new service ranged from birth into early adulthood (0–25), with a mean age of 15.3 years (SD 7.0 years). Only 34 (10.1%) of those referred were in the youngest age group, birth to 4 years. The proportion increased progressively with each older age band, with 112 (33.1%) referrals in the 20 years and older group.

At the initial lymphoedema assessment, the patient (and/or their parents/carer) was asked the age of lymphoedema onset. Response data were available for 257 patients. The largest group (n=105; 31.1%) described an onset in the first 4 years, of these 89 (84.8%), reported that their lymphoedema was present at birth. The next notable age group for onset was those transitioning into adolescence aged 10–14 (n=62; 18.3%).

Analysis of the data showed a considerable time lag between age of onset and the referral to the service (*Figure 1*). The lag between onset and being referred/seen in a CYP lymphoedema clinic had been over 5 years for 59.8% (147) of patients in this time period. The possible impact of such delays on children and families have been reported elsewhere (Quéré et al, 2021). However, this was partly due to the lack of a specific paediatric lymphoedema service within Wales until 2015. Prior to this date, health professionals identifying paediatric lymphoedema would have had to practise within the scope of their adult-based knowledge or refer the child to a specialist service in London.

Sex

The overall ratio of male to female at the date of review (31 December 2018) was 45:54, with 131 males and 155 females. However, combined analysis of the age of onset, age at referral and sex shows sex-based differences. In relation to age of onset, the ratio of male to female in children 9 years and younger is 50:50. However, more males were referred than females. This disparity remains until the older age group of 15–19 years when the ratio of referrals for females overtake the males. This is despite the onset of lymphoedema showing a higher proportion of females in the 10–14 age group.

Lymphoedema type and distribution of swelling

Analysing the data on lymphoedema type, 49% (n=140) were diagnosed with primary lymphoedema, 39.2% (n=112) with secondary to non-cancer, and 2.4% (n=7) as secondary lymphoedema to cancer.

Over half of CYP with lymphoedema had bilateral swelling in their limbs (i.e. both legs or arms) (n=142;

54.8%). Of those with unilateral lymphoedema, more were right-sided (n=65; 25.1%) than left-sided (n=52; 20.1%). During the initial lymphoedema assessment, affected segments (e.g. part of a limb) were documented. The most commonly identified category was the multi-segmental lower limb (n=113; 43.6%) (3 or 5 segments), followed by uni-segmental right lower limb (n=45; 17.4%) and uni-segmental left lower limb (n=32; 12.4%). Of those patients with upper limb involvement, 14/35 (40%) patients had symptoms in their right arm/hand, 13 (37.1%) patients had left arm/hand symptoms, and 8 (22.9%) had lymphoedema in both arms/hands. Five percent (n=13) of patients had involvement of their trunk, genitalia or face.

Family history

An underlying gene mutation may be detected in 25% of primary lymphoedema (Gordon et al, 2020) therefore the family history is important. However, it is highly dependent on family knowledge and recall. In this audit, 228 (79.2%) of patients (or their parents/carers) reported no known family history and 28 (9.7%) did not know. Only in the remaining 32 (11.1%) was there a reported family link.

Cellulitis

During the audit, 33 (12.4%) of CYP reported having had cellulitis previously, with over half experiencing 1 or more episodes in the last year, one person reported 5 episodes. Following individualised packages of lymphoedema treatment, which might include personalised skin care advice, bandaging and/or compression garment, lymphatic massage and exercise/activity advice (ILF, 2010), the number of episodes of cellulitis was found to be reduced (*Table 3*) with statistical significance ($p<0.05$). And the reduction of hospital admissions from a total of 42 to 3 is of economic importance.

Reported pain

There was a demonstrable improvement in levels of pain from initial assessment to first and second follow-up appointments. The mean pain score at the initial assessment was 2.97 (SD1.0), at the first follow-up 2.17 (SD 0.9) and at the second follow up the mean pain score was 2.29 (SD 0.8).

Physical characteristics and symptoms

There are physical anomalies that are typical of syndromic types of primary lymphoedema (Gordon et al, 2020).

Table 3. Cellulitis episodes recorded before and since lymphoedema treatment

Paired samples		Frequency		Correlations	
		Count	Percentage	Correlation (95%CI)	Sig.
Pair 1	Cellulitis initial assessment	33	12.4%	0.162	0.035
	Cellulitis review	9	5.3%		
Pair 2	Episodes in last year initial assessment	31		0.102	0.188
	Episodes in last year Review	16			
Pair 3	Hospitalised initial assessment	9	3.4%	-0.189	0.626
	Hospitalised review	2			
Pair 4	No. of admissions initial assessment	42		-	-
	No. of admissions review	3			
Pair 5	Prophylactic antibiotics initial assessment	5	1.9%	0.274	0.000
	Prophylactic antibiotics review	3	1.1		

Table 4. Crosstabulation of BMI by age and obesity class

BMI * Age at referral crosstabulation

		Age at referral					Total	Percentage
		Birth-4 years	5–9 years	10–14 years	15–19 years	20+ years		
BMI	Underweight <18.5	2	0	0	1	0	3	1.1%
	Normal weight 18.5–24.9	8	9	10	15	16	58	21.6%
	Pre-obese 25-29.9	0	1	8	12	12	33	12.3%
	Obese class 1 30–34.9	1	1	3	2	5	12	4.5%
	Obese class 2 35–39.9	0	0	1	5	5	11	4.1%
	Obese class 3a 40–49.9	1	2	0	0	6	9	3.3%
	Obese class 3b 50–89.9	0	2	0	2	4	8	3.0%
	Not reported	13	18	36	27	41	135	50.2%
Total		25	33	58	64	89	269	100.0%

Some known anomalies were documented including overgrowth of limbs (n=15), chest infections (n=7), distichiasis (n=1), warts (n=1), venous disease (n=3), neck webbing (n=1), ptosis (drooping of the eyelid) (n=5) and chronic diarrhoea (n=4). More recently recognised attributes which were not identified during this audit included wide-spaced nipples, ascites and hydrocele.

Examination of the skin data showed that 56% (n=160/285) patients had a normal, healthy skin, 17% (n=49) had dry skin, <7% (n=19) had skin folds, <6% (n=16) fibrosis, 4% (n=12) hyperkeratosis and <2% (n=5) papillomatosis. Other documented skin-related issues included fragile skin (n=1), taut skin (n=7), shiny skin (n=4), lymphorrhoea (n=1) and fungal infections (n=8). Ten patients had birthmarks and ten had fatty tissue as opposed to fluid. A positive pitting test was present in 40% (n=107/265) of those tested and a positive Stemmer's sign in 59% (n=147/248) of those tested.

Most of the reported CYP had a normal BMI of between 18.5 to 24.9kg/m2. However, those in the obese class 2 were identified in the teenage group (15–19 years), with obese class 3 clearly identified in young adulthood (20+ years) (*Table 4*).

Category and staging of lymphoedema

BLS grouping

There were 62 (23.2%) CYP identified as being at risk of lymphoedema (Group 0), 57 (21.3%) with early onset (less than 3 months) lymphoedema (Group 1) and 95 (35.6%) with uncomplicated lymphoedema (Group 2). Of the complex lymphoedema cases, 23 (8.6%) had one limb affected (Group 3a), 22 (8.2%) had multiple limbs with or without midline involvement (Group 3b) and seven (2.6%) had midline oedema (Group 3c). Midline oedema can be swelling of the head and neck, breast, trunk or genitalia. One person had lymphoedema due to advanced cancer.

ISL staging

From the data collected at first assessment, 65 (24.1%) patients had been classified as subclinical (Stage 0), 58 (21.5%) as early lymphoedema with pitting (Stage I), 122 (45.2%) as established, non-pitting lymphoedema with or without fibrosis (Stage II) and 22 (8.1%) as elephantatic type lymphoedema (Stage III).

Patient reported outcome measures (PROMs)

Table 5 demonstrates the Pearson's correlations (95% CI) for each descriptor at the two follow-ups. Each item of PROM was scored 0 to 4, where 4 indicates greatest impact of daily life. Therefore, a lower score is a perception of reduced impact or an improvement in condition. All descriptors were statistically significant at the first follow-up ($p<0.005$), even though correlations were positive but moderate at

Table 5: Pearson's correlations of PROMs (impact of lymphoedema)

PROMs descriptor	Pearson correlation initial assessment to follow-up 1	Sig. (2-tailed) follow-up 1	Pearson correlation follow-up 1 to 2	Sig. (2-tailed) follow-up 2
Swelling	0.631**	0.000	0.440	0.052
Garments	0.568**	0.000	0.290	0.229
Pain	0.639**	0.000	0.633**	0.004
Shoes	0.628**	0.000	0.756**	0.000
Skin tissue	0.570**	0.000	0.576**	0.010
Clothes	0.443**	0.000	0.803**	0.000
ROM	0.579**	0.000	0.797**	0.000
Exercises	0.541**	0.000	0.620**	0.005
Weight	0.607**	0.000	0.749**	0.000
Appearance	0.649**	0.000	0.413	0.079
Hobbies	0.486**	0.000	0.881**	0.000
Scars/discolouration	0.649**	0.000	0.535*	0.018
School/work	0.553**	0.000	0.631**	0.004
Relationships	0.587**	0.000	0.462*	0.046
Cellulitis/infection	0.576**	0.000	-0.056	0.821

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

*. Correlation is significant at the 0.05 level (2-tailed).

best. By the second follow-up, four descriptors were no longer statistically significant (p>0.05) (i.e. swelling, cellulitis/infection, appearance, and garments). In fact, cellulitis had a negative correlation (-0.056), indicating a change for the better in experience of cellulitis by those individuals.

Discussion

The results from this audit have directly shaped future service delivery by informing the CYP Strategy for Wales, 2020, as per the aim of the audit. Here we give examples of the audit results which shaped the strategy in the context of growing literature in the field.

Planning a health service would ideally be conducted with a known prevalence of a condition within a community. However, there was a distinct lack of international evidence on prevalence of lymphoedema in children. Our prevalence of 0.31/1 000 among 0 to 25-year-olds, or 0.27/1 000 in 0 to 19-year-olds were the first national prevalence figures for CYP lymphoedema. This fulfilled the first objective of this audit. It seems unlikely, however, that all CYP with lymphoedema in Wales had been identified by the end of 2018; thus, for planning purposes, this was considered a conservative figure for age-limited prevalence.

The upper age in paediatric services has been rising internationally (Sawyer et al, 2019). The age range of this service in the first few years had been deliberately broad to allow for young people who had paediatric forms of lymphoedema to gain an understanding of their condition before transitioning to the adult service. A fifth of our young people felt this was unnecessary and chose to attend (or remain with) adult services. Others, particularly the families of CYP with learning difficulties or special needs, found the opportunity useful. The transition of children with long-term conditions to adult services was a relatively new area of research at the time of the audit (Gabriel et al, 2017). A longitudinal study, based on the long-term conditions of type 1 diabetes, cerebral palsy and autism, found that UK-based services provided only some of nine proposed beneficial features of transition services (Colver et al, 2018). With this in mind, and based on our audit and clinic experience over the first years, we designed the transition of CYP to adult

services to include the following factors:

- Individuals were seen in age-appropriate clinics
- Promotion of self-efficacy with subtle changes from school to college to work in language used in assessment documentation
- Importantly, the same local therapist remained with the CYP into the adult service. This seamless transfer supported a coordinated and holistic approach in care transition.

Setting up a service for childhood conditions where previously there has been patchy, disparate or non-existent services will inevitably involve a mopping-up period, during which the average age will be high. However, noting the age of onset allowed us to see how the average age of the paediatric patient population would shift as the service matured. Based on age of onset, we could anticipate that with timely referral (through increased awareness), between 40% and 50% of future referrals would be aged birth to 4 years, and we could plan services and education accordingly. This information, along with the knowledge of who the referring agents had been up to 2019 would help us target 'early years' services and GP practices with awareness of our CYP lymphoedema services.

The ratio of male to female patients, defined as sex recorded in medical notes at time of referral (Clayton and Tannenbaum, 2016), reflects that of other reports; where in the younger age group (<12 yrs.) the boys outnumbered the girls, but in teens/young adults this is reversed (Gordon et al, 2020; Schook et al, 2011). In contrast, only 10% of our patients reported family history. This is likely to be low, based on recent reports (Gordon et al, 2020), and may reflect the lack of genotyping available to us at the time of the audit. Since completing the audit in 2019 we have added more physical anomalies onto assessment vigilance (e.g. ascites and hydrocele).

Education at an early age about healthy lifestyle needs to be integral in both the health and education sectors to help vulnerable and 'at risk' children from becoming obese in their early teens (Mikhailovich and Morrison, 2007; Freedman et al, 2020). Obesity is a factor, which in adults, contributes to the progression of lymphoedema and complicates its management (Cucchi et al, 2017), in addition to contributing to health risks such as diabetes, heart disease and cancer (Frank et al, 2019). Given the current teenage obesity in the caseload it is prudent to ensure this message is reinforced at the lymphoedema appointment (Aarthun et al, 2018), and that we ensure multi-professional links with dietitians as part of the strategy going forward. Obesity is a particular problem in Wales with 27.1% of reception school children (aged 4–5) in Wales found to be overweight or obese, compared with 22.6% in England, with greatest occurrence in areas of social deprivation (Public Health Wales NHS Trust, 2018).

Cellulitis has been attributed as both a cause and a complication of lymphoedema (Burian et al, 2021; Rodriguez et al, 2020) resulting in redness, pain and heat in the affected part and generalised feeling of being unwell. On rare occasions, this can progress to a serious infection requiring hospital admission, and differential diagnosis of sepsis in children can be particularly challenging (Brent, 2017). An episode of cellulitis can be distressing to a child and frightening for parents (Moffatt et al, 2019). In addition, recurrent episodes not only cause frustration but accumulative harm to the tissues (Rodriguez et al, 2020). The reduction in pain and cellulitis episodes seen in children once they attend the CYP service therefore had important health and economic implications which helped justify the strategy for service development going into 2020. The economic implications are currently being evaluated in detail.

The complexity level of the lymphoedema in this CYP group (objective 2) was mostly early onset, uncomplicated lymphoedema. This is in contrast to the adult lymphoedema population, where most cases are established and complex, often with skin and tissue changes, and in some cases, wounds. Treatment for the CYP group should in theory be easier; however, monitoring needs to be done more frequently (three- to four-monthly) due to the growth expected in the various age groups, a factor affecting the reliability of outcome measures (Phillips and Gordon, 2014; Moffatt et al, 2019). The frequency of review and the outcome tools that are used need to be considered when planning service delivery.

The third objective was exploring the experience of lymphoedema through the use of patient-reported outcome measures (PROMs) in this audit, which has fed into the development of a specific LYMPROM that is being validated in Wales (Gabe-Walters and Thomas, 2021). Our experience in feasibility testing LYMPROM during this audit period produced useful information about the specific items that should be included in the tool. LYMPROM is an important development as it will guide prioritisation and supports value-based health care.

Limitations

A limitation of this audit, as with any historical data, is that the science and understanding moves forward. In 2015, it was unknown what data would become significant. Retrospective medical record analysis can fill some gaps but not all. The data therefore had to be interpreted as it was. Data items for future audits have been added in line with the developing knowledge. Specifically in this field, the categorisation of paediatric and primary lymphoedema has developed as knowledge of genetic profiles have expanded (Gordon et al, 2020); although there remains some reliance on accurate family history. Knowledge sharing is particularly important with these rarer conditions and as different specialist services for CYP with lymphoedema establish themselves and share their data (e.g. Quéré et al, 2021; Vignes et al, 2021), the global knowledge level on paediatric lymphoedema will increase.

A further limitation was the vast difference in data from initial assessment to second follow-up where missing data accounted for up to 54%, due to patients not yet seen, which greatly affected the capture of patient experience of their lymphoedema through PROMs. This could be rectified with identified timescales for completion of datasets going forward. Furthermore, for young children aged under 10 years, it could be difficult for them to understand how to complete the PROMs, and further research is needed on completion of PROMs by parents/guardians/carers.

Recommendations for future research

As this clinical field grows, further research is required in clinical practice, outcome measures and the education needs of professionals. Despite the growth in lymphoedema services for CYP internationally, there remains a lack of empirical evidence for specific treatment modalities and appropriate outcome measures when applied to children (Phillips and Gordon, 2014). Further clinical research is needed. During the start of our service the importance of collecting PROMs was recognised, however, a validated QOL or PROM tool specifically for CYP remains to be developed. In parallel, as empirical evidence and experiential learning grows, the education need of health professionals proposing to treat CYP with lymphoedema, requires investigation. Lastly, this study has identified that the prevalence of lymphoedema in CYP is more common than previously reported; further studies are needed in different countries to examine the incidence and prevalence of lymphoedema in CYP (especially in specific groups such as children with cerebral palsy, spina bifida and other neurological conditions) to understand and support service development globally.

Implications for practice

This audit suggests that in countries where the prevalence is unknown there may be unmet needs in CYP with lymphoedema and their families. Education is therefore vital in improving awareness and recognition of lymphoedema among health professionals evaluating children, and that treatment can reduce pain and cellulitis, and improves PROMs. Furthermore, referral protocols need to be widely accessible to avoid significant delays in assessment and management of these children.

Service designs for CYP need to include minimum datasets to allow for comparison of services. These datasets need to include limb volume or tissue fluid content where possible to add more information about the complexity of the lymphoedema in the caseload. Set timescales for completion of data needs to be determined to allow for a more comprehensive audit of patient outcomes to inform future service delivery plans.

Sharing of data with peers across the UK and internationally will facilitate prudent development of CYP services and ensure they are designed to meet the needs of this specific population. In addition, since the correlation was poor to moderate in the PROMs scores for garments, indicating a displeasing impact of this essential treatment modality, discussion with manufacturers for ongoing development and design of compression garments is needed.

Conclusions

The audit of the CYP lymphoedema service has raised some interesting points from the three years since inception of the service. Firstly, lymphoedema in CYP appears to be more common than previously reported. Secondly, the lack of a coordinated national service meant that there had been significant delays in accessing CYP services for the older age groups, who may have had lymphoedema from birth or before the age of 4. It is anticipated that referrals will improve through education and raising awareness of lymphoedema at a young age, as well as streamlining of the CYP and adult services to ensure CYP are prioritised for assessment. Thirdly, there appears to be a good transfer rate to the adult services for continuity of the patient's lymphoedema management.

The audit demonstrated that specialist lymphoedema treatment for the CYP reduced the number of cellulitis episodes in a year. This has immediate and long-term health benefits. In addition, for the CYP, the most troublesome impacts of lymphoedema reported in PROMs were swelling, clothing and shoes. All three showed statistically significant improvement as treatment progressed. Such results were key to securing the future of the specialist service and developing the strategy for its future. Data collection is vital in providing robust information on the health of this population, as well as informing appropriate service development.

The results of this audit were used to shape the strategy for development of CYP lymphoedema service in Wales, a key feature being the employment of two permanent national lymphoedema CYP specialists. Further clinical research is needed in relation to the adaptation of treatment modalities for CYP with lymphoedema and into the education need of lymphoedema professionals treating them. **CHHE**

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