

The experience of children and families with lymphoedema – a journey within a journey

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ABSTRACT

This paper reports on a study in the UK that explored the experience of children suffering with Lymphoedema and that of their families. Qualitative data was collected from 20 children between the ages of 6 and 18 and their respective parents. Single, semi-structured interviews were used in which children and their parents were asked to share how lymphoedema impacted on their family life. Children were asked about their school experience, their dreams and their aspirations. Three categories emerged. Firstly, the negotiation of the health care system. Themes included correct diagnosis, finding robust information and reaching a knowledgeable expert. The second category explored the complex role of the parents as advocates. Themes within this category included the dilemmas of parenting and the increasing challenges as children reached adolescence. The final category involved the impact on the family unit. The first theme concerned the integration of lymphoedema into daily activities and the intrusion on family time. The second explored the impact on siblings and the final theme the changing dilemmas as children moved through the stages of childhood and faced adulthood.

Key words: Children with chronic illness • Expert knowledge • Impact on families • Lymphoedema • Parental advocacy

Key Points

- obtaining diagnosis is prolonged and difficult for parents causing stress
- inadequate information is available for parents and children
- information is inconsistent and confusing
- access to local treatment is poor
- access and choice of compression hosiery and footwear are problematic
- parents value expert help
- the impact of lymphoedema increases during adolescence
- families show resilience and courage
- lymphoedema impacts on many aspects of family life

INTRODUCTION

Lymphoedema is now recognized as a major health care problem affecting 1.3 per thousand in the general population (1). Causes include cancer treatment, parasitic infection and congenital problems in the development of the lymphatic system. In addition, lymphoedema can occur in patients with chronic diseases such as spina-bifida, rheumatoid arthritis and stroke. Children may develop lymphoedema as part of other conditions such as Turner's Syndrome. The epidemiology of lymphoedema

has received scant attention and the number of children suffering is unknown.

Children with lymphoedema

There is a paucity of evidence concerning the impact of lymphoedema on children with the condition and their families. The Lymphoedema Support Network, the patient support group in the UK, report that the condition significantly impacts on the lives of children and families. Todd in a study of six parents of children with lymphoedema reported that parents experienced particular difficulty before receiving a diagnosis and appropriate treatment (2). This situation was compounded by a general lack of professional awareness and knowledge of the condition. The day to day management of the condition was hampered by the psychological effects of the condition

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on their children (2). The research described in this paper was undertaken in the UK with 20 families. (20 children/20 parents) as part of a strategy to make international recommendations on the care of children with lymphoedema.

Study aim

The aim of the study was to explore the experience of children with lymphoedema and the impact on families. The study had a number of objectives:

- To describe and seek to understand the experience of children with lymphoedema and their families.
- To explore the impact lymphoedema has on the family and the process of adaptation
- To describe the experience of schooling in children with lymphoedema
- To capture the experience of the child and their family as they journey through the health care system.
- To determine how lymphoedema impacts on the child's social relationships with other children.
- To explore how the experience of having lymphoedema influences a child's dreams and aspirations for the future.

Overall research design

This was a qualitative study using in-depth interviews to form the basis of detailed case studies of the child and family. A purposive, stratified sampling approach was used to identify suitable patients and their families. Such a strategy sought to identify an information rich sample of 20 children of varying ages and with different presentations of lymphoedema (both the site of swelling and its severity). A mix of gender and ethnic background was ensured to capture the diversity of experience. For children under the age of 16 years, interviews were held with the child and at least one parent. For those over 16 years interviews were held separately unless the child requested the parent to be present. The study received approval by the local research ethics committee.

Sample details

Participants were recruited from children who attended a centre of lymphoedema excellence in London where the majority of children in

the UK are referred. The service register was used to ensure they met the requirements of the sampling strategy. Once selected, the consultant reviewed the suitability of the proposed children prior to contact in order to exclude any who may be considered particularly vulnerable or unsuitable for inclusion. The study had a number of criteria for inclusion. These included the following:

- Children under the age of 18 years of age with a diagnosis of lymphoedema of any cause.
- Those able to respond verbally to questions asked by the researcher.
- Those able to understand the reason for the research appropriate to their age and, where age permitted were able to provide informed written consent.

Additional inclusion criteria were set for parents or guardians of children. These included:

- Being a parent or guardian of a child with lymphoedema.
- The ability to give informed written consent.
- Parents had to be willing to allow their child to be included in the study.

Because of the diverse nature of why lymphoedema presents, a number of exclusion criteria were set. These included

- Children who were terminally ill.
- Children considered vulnerable or unsuitable by the medical consultant
- Children or parents who were unwilling to participate.

Obtaining consent from children and parents

Issues of consent were influenced by the age of the child. Informed consent was obtained from the parent or guardian of all children below the age of 16 years. Adolescents aged 16–18 years gave their consent and permission from the parents was also sought. For younger children the researcher explained the study with the help of the parents in ways most appropriate to the individual child. In all cases, it was essential that the child wished to take part in the study and this took precedence over the wishes of the parents.

The interviews

Interviews with young children can be challenging particularly when they are undertaken by a person with whom they are not familiar. All interviews were undertaken in the child's home. Interviews normally lasted about 1 hour for each child and a further hour for the parents. For younger children the interviews were sometimes shorter and combined the interview with the parents. Interview topic guides were used for both interviews.

Qualitative analysis

All interviews were tape recorded after having gained permission from the participants to do so and transcribed verbatim. To add additional rigour to this process, two independent researchers separately coded the initial transcriptions. Agreement was sort on any disparity of the initial coding.

The process of data analysis used the analysis method 'Framework' developed by the National Centre for Social Research (3). This is a matrix based analytic method that facilitates rigorous and transparent data management such that all stages involved in the analytical hierarchy can be systematically conducted. The framework classifies and organises data according to key themes, concepts and emergent categories. The study had a thematic framework comprising main themes, subdivided by a succession of related sub-topics. These were further refined through familiarisation with the raw data and cross sectional labelling. Once considered complete each main theme was charted on its own matrix. Every case was given a row and each column denoted a subtopic. Data was then synthesised within the thematic framework.

RESULTS

A diverse range of children with different causes of lymphoedema were recruited to this study. Table 1 outlines the demographic and clinical details.

Of the 20 children interviewed there were equal numbers of male and females. Two of the families came from non-European backgrounds. In 7 of the families there was a family history of lymphoedema but 17 of the children had been given no diagnosis of the type of lymphoedema they had. There

Table 1 Global and demographic details of study subjects

Number of children		20
Gender	Male	10
	Female	10
Age range	Male 6–10 years	6
	Male 11–17 years	4
	Female 6–10 years	6
	Female 11–17 years	4
Time of diagnosis	Before birth	4
	At birth	8
	Early childhood	8
Family history of lymphoedema		7
Types of lymphoedema diagnosed	Milroy's disease	2
	Lymphangectasia	1
	Distachsis	
Other medical conditions	Unnamed	17
	Autoimmune disease needing bone marrow transplant	1
	Lymphangectasia	2
	Lymphangioma	1
	Microcephaly with global developmental delay	1

were a number of children who had concurrent medical problems.

The results found three overarching categories that emerged with their related themes. The first category related to the parents' negotiation with the health service and their journey of exploration. Three themes emerged within this category. The first was the process of beginning the journey and the difficulties that were endured during this process. The second, the issues of finding robust information and the impact this process had on the family. The third, the parents and children's experiences of finally reaching the knowledgeable expert and the journey they endured before this occurred.

The second category identified the complex role that parents play as advocates for their child's well being and the changes and dilemmas that occur as the child moves to adolescence.

The final category explored the impact that lymphoedema has on the family unit. The first theme in this category explores the reality of living day to day with lymphoedema and the integration into everyday life of daily procedures and concern for complications occurring.

This theme also identified the complex issues for siblings of children with lymphoedema. The four final themes explore the uniquely different experience of younger children and that of adolescents where life becomes far more complex and lymphoedema more intrusive. The last theme explores the parents and children's perspective on the continuing journey of living with lymphoedema.

CATEGORY 1 NEGOTIATION WITH THE HEALTH CARE SYSTEM

Theme 1 – beginning the journey

For many of the parents in this study the realisation that their child had lymphoedema was an insidious experience. What appeared to be a small swelling of one limb was often dismissed by professionals who did not have the knowledge or skill to diagnose the problem and often attributed it to another cause such as an injury. One parent described their experience

She'd been at a disco, a school disco and her ankle was swollen. I noticed her ankle was swollen the next day. We just thought perhaps she'd twisted it and it just gradually got a bit worse, went to the doctors and they said, oh no, it's just this or that, keep an eye on it.

For others the condition was noted during an ultrasound scan during pregnancy. The shock of this finding was compounded on occasion by advice that maybe the pregnancy should be terminated causing enormous distress for the family. One mother's trauma is expressed below

It was diagnosed before birth. I think at the time it was that the other problem was she had this lymphangioma in her abdomen which was sort of causing a blockage, and it wasn't until after they discovered this blockage in the stomach that this leg was kind of a secondary thing, and at the 20 week scan we were told that her right leg was going to be bigger than her left leg and they thought at the time she was one of the worst cases. And we were, they actually recommended a termination because they said she was one of the worst cases and we wouldn't want to go ahead with a pregnancy like that would we?

In some cases the condition was diagnosed at birth or shortly afterwards. The child may have been acutely ill with another condition and parents were facing the possibility their child would not survive or that they would have to live with a significant health care problem.

In most cases there was considerable delay before a correct diagnosis and treatment was offered and parents were barraged with inaccurate information, further increasing their anguish. Most of the families had been referred to a number of different professionals before reaching an expert in lymphoedema able to correctly diagnose their condition. One family suffered a delay of 3 years before receiving a correct diagnosis. The experience of being sent to different professionals who had a limited or inaccurate knowledge of lymphoedema caused parents enormous distress and anxiety. One parent described their experience.

So the geneticist said something's not quite right but because they've ruled out Down's Syndrome then we'll leave it and wait and see what happens. . . . The paediatrician said ok well what we'll do is send you to an orthopaedic surgeon to have all his toes broken, to have his feet reshaped, which was an absolute madness wasn't it? And in the meantime we saw a plastic surgeon who said for goodness sake this baby has got lymphoedema.

A positive outcome of these difficulties was the strong parental advocacy roles that these parents developed in fighting for the best care for their children.

Parents reported that professionals often failed to listen to them or value the insight they had already gained about their child's condition. Parents with children born with lymphoedema reported that professionals made them feel that they had in some way contributed to the child developing lymphoedema. Mothers were repeatedly questioned on whether they had smoked or drunk during pregnancy adding to the guilt they already felt. One mother reported this experience

And every doctor went on about the pregnancy and did you smoke, and did you do this, and I began to feel it must have been something I'd done and nobody ever

considered us as people, I don't think, in the middle of all this. It was almost like well you've got this thing and it was awful, because he was this thing.

Families with a familial form of lymphoedema were often burdened with the reality that the condition had been passed down from parent to child.

Theme 2 – finding information 'I never find I really get the answers'

One of the most significant findings was the difficulty finding information about their child's condition. Parents spent a lot of time searching the internet, being shown relevant sections from medical textbooks, and other sources of literature. Many of these had pictures of extremely severe, complex patients with elephantiasis and this caused parents fear that this would be the outcome for their child. One mother recounts her experience

He read out the explanation from a big medical textbook. He knew nothing about it. So for us, I can remember my husband and I were quite devastated, we were absolutely crying our eyes out because it sounded so bad.

This was further aggravated by professionals being unable to predict the course the child's condition would take or the overall prognosis. The children support groups for lymphoedema were used frequently by parents who knew about them.

Despite this the level and quantity of information for parents and children in the UK is inadequate and parents have many unanswered questions. There was no literature for younger children with lymphoedema.

Parents wanted a single resource that told them of the different products, aids and range of hosiery that was available to them and mechanisms where they could buy extra products if they wished to add to those provided by the health service. Parents found that clinical information on aspects of care such as manual lymphatic drainage and compression hosiery was conflicting.

The parents reported that professional knowledge and information was poor and that they were frequently in a position of educating professionals such as GPs about the

needs of their child. This was not a comfortable position for many parents although they accepted this to ensure their child received the correct treatment. A particular difficulty was access to prophylactic antibiotics. Few GPs felt it was necessary despite letters from specialist therapists stating the child required them. Letters from hospital consultants held greater authority.

Parents found themselves acting as the sole source of information when their child went to school. They took responsibility for ensuring the information leaflets reached the school although this information was not always passed on when the child changed classes. Some teachers took an active interest and found extra information in order to enrich the child's experience at school. Parents continued to play a vital advocacy and protective role for their children while they were at school.

Theme 3 – reaching the expert

The importance of reaching the right expert cannot be overstated. The research indicated that this was often a long, painful and tortuous experience. The parents characterised this as a relationship of trust and a sense of knowing that the professionals really understood the condition. The relationship with key medical and therapists within these centres was the central reason why parents felt so supported.

... because until we saw this consultant, I've forgotten how long, 2 years probably 3 years ago now, we didn't really understand. We knew it was hereditary, and we knew there were things missing but he went into it in such a way that we learnt more in the hour that we saw him than we had for the last 15 years that she'd had it.

Parents experienced worry when specialist centres closed or when experts left a service causing problems with continuity of care.

However attending specialist centres required family commitment as many lived many miles away. Attending appointments required reorganisation of the whole family, not just the affected child. Siblings had to be left with friends or relatives, which could cause feelings of exclusion by brothers and sisters.

I think also we had so many appointments that she spent a lot of time with my mum

and dad because it wasn't really possible to keep taking her to the hospital all the time, and whilst, I mean she always thought it was good, or we always thought she thought it was good, you know, we're going to Nan and Grandad for a treat she now says she thought we were taking him off and doing something nice with him and I'm not going anywhere nice. So that has affected things I think.

Travel costs were high and families were often unaware that they might be able to claim expenses for travel. Appointments early or late in the day added to the expense as tickets during peak travel times cost much more. Long hospital appointments were found boring by many of the children and adolescents. Many children wished they could meet other children suffering with the condition during these visits.

While reaching a centre of excellence was very important, accessing local care often proved very difficult. Parents were frequently involved in appeals for funding of treatment and many resorted to paying for aspects of care such as manual lymphatic drainage as this was not provided for locally.

I think it helps him enormously actually, and it annoys me that his PCT won't fund it because as far as they're concerned it has no long-term benefits which makes me think they know nothing about lymphoedema at all. And I've sent them letters and my GP has tried to get funding but it's seen as quack medicine if you like. It's just written down on paper. I feel that I need to go somewhere to present my case but you're not given that opportunity. So that's the current situation but I'm not leaving it at that.

Children frequently suffered with veruccas and infections secondary to in-growing toe nails. Access to services such as podiatry was limited, with long delays for treatment. This was because lymphoedema was not recognised as a priority area unlike diabetes or the elderly.

CATEGORY 2 THE COMPLEX ROLE OF PARENTS

Theme 1 – parents as advocates for treatment

Parents spent much time and effort fighting to ensure their child received the best care

possible with the least interruption to family life. In many families one parent undertook this role more actively than the other did.

I always felt that I took on most of the responsibility and worry and everything like that although I know that X worries more than me about most things but I did always feel that most of the decisions came down to me.

This was often influenced by whether parents were working and the time available for providing treatment for the child. In some instances however, this seemed to be a coping mechanism. One parent found dealing with these issues very difficult and abdicated responsibility to the partner for finding out information and undertaking the treatment when she was available.

Parents struggled to ensure they had an adequate supply of hosiery. This was often difficult due to the delays in receiving orders of hosiery and the limits set on the numbers of garments that would be funded by hospitals.

We have 6 monthly appointments and they (hosiery) are absolutely worn out and holey by the time he goes to be measured, but it's all to do with funding again. And also they've stopped funding for the way stockings used to be finished off, The elastic finishes here and goes up the leg and round and because that's too expensive now it has to go straight across so they're missing out on a piece and that's all to do with money. His new stockings arrived last week and they were wrong so we've had to send them back. But what he's walking around in at the moment I'd be too embarrassed to show you. We have offered to buy them ourselves but you can't do that evidently.

The children and adolescents and their parents did not like the hosiery given to them. The styles were considered old fashioned and led some children to be bullied at school. A few children refused to wear the hosiery at all.

Provision of shoes was a major challenge for parents and children. While hospital orthotic departments made shoes for the children this frequently took weeks or months. This delay often meant that the shoes did not fit because

the child had grown during this period. The children complained about the style of shoes. Parents reported that several pairs of shoes for different purposes were required and these were not made available.

They've recently changed the way that I can order her shoes and it took seven and a half weeks just to get the order form. And then it's 8 weeks from when the form goes to the manufacturers so that's like 16 weeks for a pair of shoes.

There was one time when we were away and the appointment had got forgotten and we had to see a locum. At that time her feet had grown. She'd had three different types of footwear but never all at once. But as far as I could tell she needed a school shoe, something to wear at home and something for PE. And the locum said she couldn't have three and she asked me to choose. And I said I can't choose, you tell me which thing she can't do because as far as I'm concerned you can't wear your school shoes at home if you want to go out and play. They didn't get back in touch with me and I didn't know until I collected them what I was getting. And I thought that was a bit poor, and when I did collect them the lady said well you know the cost is hundreds of pounds

Theme 2 – parents as advocates at school

All parents were determined that their child should receive appropriate education and that the schools were prepared for the health needs of the child. Some children faced difficulties in integrating into school life because of the inflexibility of some schools in addressing the specific needs of the child if these differed from their standard practices. Issues such as pupils doing PE bare foot rather than in plimsolls were daily irritations that singled the child out as different and contributed to the need for constant parental vigilance.

Silly things about him not being allowed to wear plimsolls in PE and things, and the teachers saying if he wears his plimsolls it singles him out, which ok I can understand because his self-confidence is low so we don't want him singled out so that doesn't help. But then she says if he wears his plimsolls

he'll stand on somebody's fingers and we'll have this and we'll have that and it's because they don't understand.

Sporting activities were a constant challenge with staff not always available to take off or apply hosiery after activities. Some schools did not actively embrace children with disability and on a number of occasions suggested that these children should attend special schools.

The headteacher, she actually said to me one day – I've written it down, I've got a quote from her, 'people overestimate what mainstream schools are able to deal with'. And I said to her 'do you think X should be in a special school?' And she said 'yes'. So that's what we're dealing with, just dealing with this complete she's not supposed to be here attitude.

This situation was made more acute for children who were bullied at school because of their condition or the compression garments and shoes they wore causing them unhappiness and bewilderment

There were some examples of excellence where schools went out of their way to enhance the life of the child and promote independence.

Yes, I mean they've been very good, I have to say. When she was in infant school and junior school I had an association there because I worked in the infant school that was attached to it so I had a good relationship to the teachers there and everything. I was able to let them know what the problems were and issues. She went away on a residential, that was probably her last year, I would think so she'd be 11. The teachers were fantastic. The teachers came here to learn how to put her stockings on, to learn any signs, you know, what to pick up, so I couldn't fault them.

CATEGORY 3 LIVING WITH LYMPHOEDEMA – A FAMILY PERSPECTIVE

Theme 1 – living with 'our' lymphoedema

Lymphoedema impacted on many aspects of family life. One parent described this very well. When a family outing occurred they took the family and also the lymphoedema with them.

It's almost like an extra person in the family. We have X. We have Z. And we have lymphoedema because we have to consider it in everything that we do.

Despite this they made every attempt to 'normalise' life for the family and the affected child. Parents described an inherent tension between giving the child freedom to undertake normal childhood activities and the fear of harm from injury or infection.

If we go to the seaside I have a real problem there, nagging him to not muck about on the beach without his stocking on and worrying that he is going to get a cut or tread on something horrible. I've always found that a problem. I don't go for beach holidays consequently, we just don't do it. It's so stressful sand between the toes and all the rest of it. We do go to the seaside because after all children want to go and have fun, but I don't enjoy it

Parents showed different levels of parental control that ranged from giving in completely to the demands of the child to complete control of their treatment on a day to day basis. As children reached adolescence these issues became more difficult and parents often found difficulty relinquishing control because of their fear that the child would not adequately care for their condition leading to complications in adult life.

Theme 2 – the impact on siblings

The treatment of lymphoedema impacted on the time available for family life, although for the majority of children who developed the condition at an early age this was simply part of every day life. Parents reported there was less time available for siblings and that the sick child often took precedence over other family members. Parents were often distressed by these issues, recognising that siblings had to take a back seat and that this sometimes led to jealousy and frustration. However there were instances where very close sibling relationships were established. In one set of twins, the healthy twin gave treatment to the affected child. Parents appeared to expect that siblings took a degree of responsibility for the child and many showed maturity beyond their years.

Theme 3 – the experience of younger children

For many of the younger children with lymphoedema they had never known life without the condition and they accepted it or learned to ignore it. These sentiments are captured in the experience of one boy describing his feelings about his situation;

I was like oh my goodness gracious me, I'm not going to be able to get on with my life. But I learnt to ignore it. It was hard at first but I eventually ignored it. I learnt how to ignore it and now I'm just an average boy.

Most children had a basic understanding of their condition and that they would suffer with it for life. Some talked of hoping for a cure in the future.

Hopefully it won't get any worse but I don't think it will ever go away. I think you can have some relief but I don't think it will ever get 100% better.

Not at this precise moment but they're always making medical breakthroughs

Discussion with these children revealed that they had many dreams for the future and that lymphoedema did not appear to influence them. Many of the boys enjoyed sport and hoped to become famous footballers or sportsmen. However for some their condition reduced their effectiveness at sport and this caused them frustration.

I began to realise I was no faster at running than I used to be. It affects me cos when I'm doing sport I can't run. It slows me down. And also it like affects me because if before you had it you were quite athletic and you were really used to winning and you were like a great sport. Then all of a sudden you found out you'd got lymphoedema and like that really slowed you down a lot and then you'd start to get annoyed because you kept on losing to these other people that you'd kept on beating.

Many showed great adaptability when joining in games. Some children had developed effective strategies for releasing their frustration that they reported really helped them

When you do get days like that you just have to kick and punch the pillows and the duvets and if you do have a hill near you, going out to the hill and run down it screaming and waving your arms like a maniac and when you get to the bottom you're feeling much better. Then walk back up it or run up it again, do it as many times as you have to until you feel better. That's how I do it but people do it in different ways.

Many expressed a desire to meet others in a similar situation to their own so that they did not feel so alone.

It would be nice to have a place to go because as far as she's concerned she's the only person in the world isn't she?

But I'm quite excited because in the Christmas holidays there's this boy called G. and he's the same age as me and he's got lymphoedema in his leg, and what's going to happen is we both go and have our appointments on the same day, and I'll see the doctor and then we'll both have some time together so his mum can talk to my mum and then the one that's already had the appointment will go home and the other one has their appointment.

Theme 4 – the experience of adolescents

As children reached the stage of adolescence the lymphoedema had much greater impact on their lives. Issues such as body image and the ability to fit into a peer group were major problems. They demonstrated a range of coping mechanisms. For some they avoided telling others about their condition and girls dressed in trousers in order to hide the swelling from public view.

One teenager reported he had stopped talking about lymphoedema as this meant he no longer thought about it.

If I don't talk about it I don't think about it and I think I am a bit worse that way. Before I used to think about it but now I think if you don't think about it and forget about it then I don't think it's there kind of thing.

For some however they coped by hiding their emotions from others and withdrawing from family and friends.

Many found shopping particularly difficult, as they were unable to buy fashionable clothes or shoes that would accommodate the swelling. One teenager spent hours searching the internet for clothes and shoes and was hugely frustrated by the lack of choice.

Concordance with treatment was more difficult for teenagers and many refused to consistently wear their compression hosiery. One boy reported that his parents did not understand the impact the condition had on him and just thought he was being lazy.

I think it slows me down. Mum just thinks I'm lazy. I get very tired with it. This leg it wouldn't be tired. I can move that but this one it hurts more.

As with the younger children there was an even greater desire to meet with others who shared the condition.

Theme 5 – thinking about the future

On the whole the families showed acceptance of their situation. Many coped by comparing their situations to other sick children with more serious health problems.

Yes but like he said, I told him that at least he had legs to run with. You try and give him something positive like we see someone in a wheelchair and I say, and maybe I shouldn't, they can't run around and play like you.

They worried about the future, in particular how they would cope in adulthood.

Parents worried that their child would develop problems with their body image that would influence relationships. Two of the fathers expressed concern for sons with genital swelling and its impact on sexual relationships.

I know that his leg is a problem but I worry too much about his penis because it is getting bigger. After they said when he is five and we were just waiting, just waiting. I know that as he is older it is not too well. That's why I worry too much because I want for my boy.

As children moved into adolescence parents had to appraise how treatment was given. While a mother may have provided treatment for a boy during childhood this had to

alter as children reached adolescence where embarrassment became an issue.

And also he's going into puberty now and I'm still helping him with the stocking and he really couldn't do it himself. Luckily he's still not too embarrassed and I imagine my husband will have to take over fairly soon. He doesn't want his mum's head up his bottom you know.

DISCUSSION

This research identified the impact that Lymphoedema has on many aspects of family life and the emotional distress that occurred.

Research indicates that families caring for children who are chronically sick suffer a high degree of stress that is often poorly understood by professionals (4). A chronic illness impacts on many dimensions of family life. Financial and emotional difficulties are common and lead to greater strain than in families with healthy children (5).

In this study emotional distress occurred at different times. For those who discovered during pregnancy, they faced the dilemma of whether to undergo termination. For those with a diagnosis at birth or later there was the distress of not having a 'perfect' child.

We noted the stress that was caused to parents in trying to obtain a correct diagnosis. Some authors have reported that diagnosis and prognostic uncertainty are major psychological stressors (6). Parents feel overwhelmed at the point of diagnosis and need to know they are not alone in this situation. Starke and Moller (7) reported that parents coped with diagnosis by seeking information and gaining knowledge. It is proposed that this brings order into a chaotic situation. Parents in this study complained of lack of information that caused them continuing stress which was further exacerbated as they had to fight for appropriate treatment and resources from the health and education services. There was also evidence of stress within the family unit with parents showing different types of coping and methods of sharing the parental burden (8).

Parents of the children in this study sought to provide as normal a life as possible for their child and siblings. This was set against the continued concern of complications. The families exhibited varying levels of control

over the children's behaviour and inclusion in activities. Many struggled with relinquishing control as the child reached adolescence. This was exacerbated if the child was unwilling to care for their lymphoedema in the way the parents considered appropriate.

A number of authors have described the experience of developing a chronic illness. Bury (9) described this as a 'biographical disruption'. This requires the individual and family to 're-frame' their lives having acknowledged their altered situation. 'Biographical accommodation' is the sequel to this and requires a number of changes: keeping a sense of balance and control and adapting and allowing the illness to become part of the new biography of the child and family (10).

Children with chronic illness are known to be at risk of psychosocial adjustment problems (11). Much of the literature stresses the importance of families being able to live as normally as possible. This cognitive process has been described as 'normalisation' (12,13). There was evidence in this study that parent's also used normalisation and compared their child's situation with that of other children in worse situations. A number of studies have reported how parents interpret their lives by using a constant comparison to others in similar situations. These approaches include drawing on positive comparisons with others (14), emphasising growth in their child, however minimal (15) and gradual resignation when situations become too difficult or are deteriorating (16). Ray (15) stressed the importance of seeing one's child progress no matter how small the incremental gain. Even when a child was not improving a parent's ability to interpret their child as strong and in control was more sustaining than viewing their child as a helpless victim. The parents in this study were all very proud of what their child had achieved and the child thrived on this recognition.

Research suggests that children with chronic illness place great importance on attending school. Even in the absence of cognitive impairment, children with chronic illness perform less well academically than healthy children. A number of factors for this have been identified including absence from school and poor psychosocial well being due to the limitations their illness places on participation in school life. In the limited studies that have sought

the views of children, a number of problems have been identified about their experience of managing school. These include the effects that absence from school had on relationships with their peers and fear over keeping up with work. More direct communication and better information provided for schools was recommended in a number of studies (11). For the children in this study friends played a complex role in providing support that was both practical and emotional. Chronic illness research has shown that some children do have difficulty with relationships, although the pattern of evidence was inconsistent. Some found their condition had a positive effect, with friends watching out for them and offering practical help. Others suffered from being ignored, bullied or the focus of curiosity. Self-esteem, school functioning, and sports participation are among the most significant psychosocial issues that affect children and adolescents with chronic illness (17). Parents and teachers play an important role in normalising and providing gratifying opportunities for children who struggle to be seen as competent by their peers and will allow them to grow up happier and feeling better about themselves and enjoying more success (18).

Parents in this study were very aware of the impact on the child's siblings. However all showed a great understanding of the impact on their other children. They valued their willingness to support the sick child recognising that this caused tension at times within the family. Especially close relationships were seen in many families and parents recognised that siblings were often required to take a greater level of responsibility than they would have liked. Other research supports the findings from this study. The literature refers to siblings being described as 'lost in the shuffle' (19). Parents are acutely aware that the child who is ill becomes the central focus and that this is detrimental to the siblings (5,20). Siblings often take on a great deal of responsibility and adopt a monitoring role; this is particularly acute for girls (21).

Families in this study showed a high level of cohesion although some parents stated that the child's illness had placed stress on their relationship. This was because of the dedicated time that had to be spent on delivering treatment each day. For many this reduced the free time they could spend together and

reduced the ability to be spontaneous in planning activities that other families enjoy. Studies have shown that in families with a child with a chronic illness the relationship of parents with partners, friends and extended family are tested and changed in some way. Some relationships deepen while others drift away (22). The most important social support, particularly for mothers, is that of the partner. It is believed that the divorce rate is higher in those families with a sick child although the picture is not entirely clear. Some research has shown that couples stay together because of the needs of the child even within a poor relationship (15).

This study showed the increasing difficulties experienced by teenagers. Some faced significant problems of low self-esteem and poor body image. A number of authors have reported the difficulties of staring and teasing during social experiences (15). The literature reports a particular gap in understanding the views of disabled children with much of the literature relying on the views of parents as proxy measures for their children's views (23). However the quality of life literature suggests that children have a different opinion of the impact of their illness on their lives than those held by their parents (24,25).

The teenagers in this study were beginning to undertake their treatment with parental guidance. Parents were often fearful that their child did not understand their condition and were concerned that they would rebel. Research in other conditions shows a similar picture. Adolescents with cystic fibrosis reported their frustration when professionals or parents did not ask or consult them about their plan of care (26).

A literature review of the experience of parents with health professionals involved in caring for a child with a chronic illness found four common themes (27). The most vital factor was establishing trust. This was based on the assumption that health care professionals would always interact with families in a positive way. Information was very important to parents, reducing uncertainty and giving a greater sense of control. Parents became their child's advocate and wanted to participate in the routine and technical care for their child. Parents moved to collaborative decision making with health professionals as their knowledge and confidence increased over time (28).

One of the most fundamental assurances parents hoped for from health professionals was to know they were not totally alone and that they had somewhere to turn to if they needed help (15). In this study it was important for parents to find a professional who respected what they knew about their child and his illness and who was willing to find appropriate help and treatment if they themselves were not skilled to give this. The children gained from the parents' confidence in this professional that was passed down to them.

This study sought to understand the views of both parents and children. There were differences between their views in the condition. The dreams and aspirations of the younger children did not seem to be adversely affected. During adolescence there was a growing appreciation of the impact of their condition however the majority would not let this influence their lives.

Research has rarely focussed on the expressed views of children. Some parents choose not to focus on the future of their children and therefore do not encourage their children to talk about it either. It may be that their own fears for their children cause to silence them so that they do not have to think about any negative outcome. Societal attitudes to the passive voice of sick children may represent the wish to protect their welfare (29). Views are now changing with children considered to be active in the construction and determination of their own lives (30).

A key aspect of this research was the courage and resilience that the children and their families demonstrated in the face of many challenges, not least for the parents in accessing treatment for their children in a climate where lymphoedema is barely recognised by the professional community and services are sparse. We are very grateful for their willingness and openness in sharing their stories with us.

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We hope that this piece of research may lead to improved services for those children who suffer with lymphoedema.

REFERENCES

- Moffatt CJ, Franks PJ, Doherty DC, Williams AF, Badger C, Jeffs E, Bosanquet N, Mortimer PS. Lymphoedema: an underestimated health problem. *QJM* 2003;96:731–8.
- Todd M, Welsh J, Moriarty D. The experience of children with primary lymphoedema. *Int J Palliat Nurs* 2002;8:444–51.
- Ritchie J, Lewis J, editors. *Qualitative research practice*. London: Sage Publications, 2003.
- Pradeep R, Pradhan P, Shah H. Psychopathology and coping in parents of chronically ill children. *Indian J Pediatr* 2004;71:695–9.
- Williams PH, Williams AR, Graff J, Hanson S, Stanton A, Hafeman C, Liebergen A, Lenenberg K, Setter RK, Ridder L, Curry H, Barnard M, Sanders S. Interrelationships among variables affecting well siblings and mothers in families of children with a chronic illness or disability. *J Behav Med* 2002;25:411–24.
- Lenhard W, Breitenbach E, Ebert H, Schindelbauer-Deutscher HJ, Henn W. Psychological benefit of diagnostic certainty for mothers with disabilities: lessons from Downs syndrome. *Am J Med Genet* 2005;133(Part A, 2):170–5.
- Starke M, Moller A. Parents need for knowledge concerning the medical diagnosis of their children. *J Child Health Care* 2002;6:245–57.
- Cashin GH, Small SP, Solberg SM. The lived experience of fathers who have children with asthma: a phenomenological study. *J Pediatr Nurs* 2008;23:372–85.
- Bury. Chronic illness as a biographical disruption. *Sociol Health Illn* 1981;4:167–82.
- Sartain S, Clarke C, Heyman R. Hearing the voices of children with chronic illness. *J Adv Nurs* 2000;32:913–21.
- Lightfoot J, Wright S, Sloper P. Supporting pupils in mainstream school with an illness or disability: young people's views. *Child Care Health Dev* 1999;25:267–83.
- Knafel K, Deatrick J. How families manage chronic conditions: an analysis of the concept of normalisation. *Res Nurs Health* 1986;45:1433–47.
- Protudjer JL, Kozyrskyj AL, Becker AB, Marchessault G. Normalization strategies of children with asthma. *Qual Health Res* 2009;19:94–104.
- Cunningham SD, Warschau Sky S, Thomas PD. Parenting and social functioning of children with and without cerebral palsy. *Rehabil Psychol* 2009 Feb; 54(1):109–15.
- Ray L. Parenting and childhood in chronicity: making visible the invisible work. *J Pediatr Nurs* 2002;17:424–38.

- 16 Bruce EJ, Schultz CL, Smyrnois KX. A longitudinal study of the grief of mothers and fathers of children with intellectual disability. *Br J Med Psychol* 1996;69(Part 1):33–45.
- 17 Vitulano LA. Psychosocial issues for children and adolescents with chronic illness: self-esteem, school functioning, and sports participation. *Child Adolesc Clin N Am* 2003;12:585–92.
- 18 Meijer SA, Sinnema G, Bijstra JO, Mellenbergh GJ, Wolters WHG. Social functioning in children with a chronic illness. *J Child Psychol Psychiatry Allied Discip* 2000;41:309–17.
- 19 Breslau N, Prabucki K. Siblings of disabled children: effects of chronic stress in the family. *Arch Gen Psychiatry* 1987;44:1040–6.
- 20 Jackson C, Richer J, Edge JA. Sibling psychological adjustment to Type 1 diabetes mellitus. *Pediatr Diabetes* 2008;9(4 pt 1):308–11.
- 21 Klein SD, Schliefer MJ. *It isn't fair! Siblings of children with disabilities*. Westport: Bergin & Garvey, 1993.
- 22 Taanila A, Jarvelin J. The long term effects of children's early onset disability on marital relationships. *Dev Med Child Neurol* 1999;38:567–77.
- 23 Anthony KK, Gil KM, Schanberg LE. Parental perceptions of child vulnerability in children with chronic illness. *J Pediatr Psychol* 2003;28:185–90.
- 24 Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. *Arch Dis Child* 2001;84:205–11.
- 25 Bullinger M, von Mackensen S. Psychosocial determinants of quality of life in children and adolescents with haemophilia – a cross-cultural approach. *Clin Psychol Psychother* 2008;3:164–72.
- 26 Nuttal P, Nicholes P. Cystic fibrosis: adolescent and maternal concerns about hospital and home care. *Issues Compr Pediatr Nurs* 1992;15:199–213.
- 27 Dixon D. Unifying concepts in parents' experiences with health care providers. *J Fam Nurs* 1996;2:111–32.
- 28 Angst D, Deatrick J. Involvement in health care decisions. Parent and children with chronic illness. *J Fam Nurs* 1996;2:174–94.
- 29 Davie R, Varma V, Upton G, editors. *The voice of the child: a handbook for professionals*. London: Falmer, 1996.
- 30 James A, Prout A. *Constructing and reconstructing childhood; contemporary issues in the sociological study of childhood*. London: Falmer, 1990.

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