

Factors related to lymphoedema coded with the International Classification of Functioning, Disability and Health

Viehoff PB, Hidding JT, Heerkens YF, van Ravensberg CD, Neumann HAM

Key words

Lymphoedema, International Classification of Functioning, Disability and Health, International Classification of Functioning, Disability and Health Core Set

PB Viehoff is Physical Therapy Scientist, Erasmus Medical Centre, Rotterdam, the Netherlands; JT Hidding, Physical Therapy Scientist, Radboud University Medical Centre, Nijmegen, the Netherlands; YF Heerkens, Human Movement Scientist, Dutch Institute of Allied Health Care, Amersfoort, the Netherlands; CD Van Ravensberg, Human Movement Scientist, Dutch Institute of Allied Health Care, Amersfoort, the Netherlands; HAM Neumann, Professor in Dermatology, Erasmus Medical Centre, Rotterdam, the Netherlands

Declaration of interest: None.

Abstract

Background: This article is the second arising from a literature review conducted as part of the development process for International Classification of Functioning, Disability and Health (ICF) Core Sets for lymphoedema. In the first article, meaningful concepts in lymphoedema-specific questionnaires were explored. **Aim:** To compare and analyse categories of the World Health Organization's ICF linked to meaningful concepts within studies focusing on lymphoedema with ICF categories linked to concepts derived from lymphoedema-specific questionnaires in the earlier study. **Methods:** Electronic searches of Medline, EMBASE, CINAHL, CENTral, and Pedro (2005–2010) were conducted. Meaningful concepts were retrieved from the included studies and linked to the ICF, and these were compared with the codes given to concepts derived from lymphoedema-specific questionnaires. **Results:** A total of 2381 studies were retrieved and, after reading the abstracts, 149 articles were included, of which 136 articles remained after reading the full text. Based on saturation in the detection of new concepts concerning upper-limb lymphoedema, 54 publications were excluded. Publications about combinations of regions were also excluded, resulting in the inclusion of 34 studies for upper-limb lymphoedema, 34 for lower-limb lymphoedema and five for midline lymphoedema ($n=73$). A total of 2488 concepts were extracted, of which 2269 (91.2%) could be linked to ICF categories. **Conclusion:** Studies of lymphoedema show a greater variety of meaningful concepts and ICF categories than do questionnaires, although the most frequently found ICF categories in studies and questionnaires are similar. The two methods complement each other and can be used together to produce more data than can be obtained through the use of either method alone. There is a need for lymphoedema-specific questionnaires concerning midline lymphoedema, and existing questionnaires should be adapted.

Lymphoedema is the abnormal accumulation of fluid and protein as a result of an imbalance between the flow and discharge of fluid (Stanton et al, 2009). Lymphoedema can occur in the upper and lower limbs as well as in the midline (head, neck, and thorax).

Lymphoedema can be subdivided into primary and secondary lymphoedema: primary lymphoedema, which is inherited, can occur in any part of the body but is most common in the legs; secondary lymphoedema is acquired following the removal or infection of lymph vessels and/or nodes (e.g. axillary lymph node dissection, inguinal lymph node dissection, or infection with lymphatic filariasis). Patients receiving

post-surgical radio- and/or chemotherapy have an increased chance of acquiring lymphoedema (International Lymphoedema Framework, 2006).

The epidemiology of lymphoedema is not precisely known because it is not a regular registered disease. According to a World Health Organization (WHO; 2010) report, approximately 120 million people in 72 countries have been infected with lymphatic filariasis, of whom 15 million have lymphoedema. Worldwide, the number of people with lymphoedema is estimated to be 140–250 million (Foldi and Foldi, 2006).

Symptoms of lymphoedema

Lymphoedema results in decreased mobility of the limb (Jacob et al, 2002),

slower wound healing, increased risk of infection, feelings of heaviness and tightness, discomfort and tight fitting of jewellery or clothing in the affected body parts (Fu et al, 2008; 2009; Norman et al, 2009). It leads to limitations in activities, such as a reduced ability to walk, and limitations in personal care, domestic life, occupation, and socialisation.

Lymphoedema is a chronic condition, and its influence on physical functioning can result in distress (Swenson et al, 2009) and loss of quality of life. Altered body shape and the necessity for daily use of specialised garments or bandaging as a standard treatment for stabilised lymphoedema make acceptance of the condition difficult for the patient, as well as for his/her family. In addition,

lymphoedema requires lifelong self-care: patients must follow prescribed regimens, such as daily bandaging or the use of special garments; utilise skin care regimens; and exercise caution with open wounds. Generally, the activity and participation levels of persons with lymphoedema are lower than those of their healthy peers.

ICF

The International Classification of Functioning, Disability and Health (ICF) is accepted by the World Health Assembly as the universal framework for classifying and describing functioning in people with specific health conditions (WHO, 2001). The International Society for Lymphology (ISL) emphasised the importance of the ICF in its 2013 revision of its lymphoedema consensus document.

The ICF organises information in two parts. Part one addresses functioning and is composed of the components “Body Functions and Structures” (divided into two classifications) and “Activities and Participation” (a single classification). Part 2 addresses contextual factors and consists of “Environmental Factors” (one classification) and “Personal Factors”. Personal Factors are not used as a classification in the ICF yet, because of the large social and cultural variance associated with these factors. The ICF makes it possible to link data across conditions or interventions with the goal of achieving efficient, transparent, and cost-effective health care (Ustün et al, 2004).

The present article is the second arising from a literature review conducted as part of the development process for ICF Core Sets (selections from the ICF for a specific group of clients) for lymphoedema. In the first article (Viehoff et al, 2013), meaningful concepts in lymphoedema-specific questionnaires were explored. One of the conclusions indicated a need to define what should be measured in lymphoedema care. In this second article, meaningful concepts in the studies themselves were coded with the ICF to define what researchers described as important items in lymphoedema. The ICF categories were then compared with the ICF categories linked to the

meaningful concepts found in the questionnaires to determine whether or not researchers explored these items.

Objectives

The objectives of the literature review were to: (i) identify literature describing problems in functioning of individuals with lymphoedema; (ii) identify meaningful concepts within the studies; (iii) link these meaningful concepts to the ICF; (iv) quantify these ICF-linked concepts (to count the frequency of occurrence of codes/classes); and (v) compare these codes with those that found in a previous study about lymphoedema-specific questionnaires.

Methods

Search strategy

Electronic searches of CINAHL, EMBASE, Medline, CENTRAL, and Pedro databases from 2005 to 2010 were carried out using the search terms “lymphoedema”, “elephantiasis”, “lymphatic edema”, “lymphostatic edema”, and “Milroy’s disease”. The detailed search strategy is available on request from the first author.

The target population of the included studies consisted of patients >18 years of age with a clinical diagnosis of lymphoedema. The following study types were included: randomised clinical trials; controlled clinical trials; observational studies (i.e. cohort studies and cross-sectional studies); guidelines; and qualitative studies. The publication language was English. If a study was described in a number of publications, only the publication in the journal with the highest impact factor was included.

Exclusion criteria were as follows: (i) studies with exclusively laboratory parameters; (ii) genetic studies; (iii) animal experiments; (iv) letters; (v) comments; (vi) editorials; (vii) systematic reviews; (viii) case reports; and (ix) meta-analyses.

Data collection procedure

Studies were selected based on title in the first instance. The abstracts of the retrieved studies were then checked for inclusion. For the selected abstracts, the entire publication was read and a decision was made as to whether it should be included in the review.

Next, meaningful concepts were extracted from the text of the selected publications. In this process, the following criteria were used: only the Methods and Results sections were assessed, and the concepts were focused on the patients themselves and their surroundings; examples and descriptions of the therapies and exclusion criteria used in the studies were not included. The measurement instruments used in the included studies are described elsewhere (Viehoff et al, 2013). After data extraction, two researchers (PV, JH) compared their results.

The retrieved meaningful concepts were then linked to ICF categories using standardised linkage rules (Cieza et al, 2002; 2005). Each relevant concept was linked by two researchers (PV, JH). Coding took place until the fourth level. Each class/category/code was used only once per article to avoid bias. After the linking process was complete, the two experts compared their results. Comparison of the meaningful concepts and the linking process took place at the same time. Initial disagreement was solved by discussion between the two experts. If disagreement remained, a third expert (YH or DvR) was consulted.

If a meaningful concept pertained to a personal factor (e.g. age, gender, or education), the code “pf” was attributed because personal factors are not coded within the ICF. If a concept described an aspect of functioning and health that is not covered by the ICF (e.g. taking up time or suffering), the code “nc” (not covered) was attributed. If a concept pertained to health conditions (e.g. lymphoedema or breast cancer), which are not included in the ICF, the code “hc” (health condition) was attributed. When the concept was related to quality of life, the code “nd-qol” (not definable-quality of life) was used. If the information provided by the meaningful concept was insufficient to allow the researchers to make a decision regarding which ICF category it should be linked to (e.g. side effects of therapy), it was assigned the code “nd” (not definable). Meaningful concepts referring to health in general were assigned the code “nd-gh” (not definable-general health).

Data analysis

The frequencies of the linked ICF categories were assessed. All of the

resulting ICF categories that referred to concepts measured in >5% of studies were reported (Post et al, 2010). ICF categories are presented at the second level. If a concept was linked to a third- or fourth-level ICF category, the corresponding second-level category was reported. This is appropriate because the lower-level categories share the attributes of the corresponding higher-level categories (ISL, 2013). Finally, the percentage frequencies of the meaningful concepts codes in the studies, and the codes of the meaningful concepts in the questionnaires, were compared.

Results

Of the 2381 titles retrieved, 149 studies were included. After reading the full-text articles, the investigators excluded an additional 13 studies because they did not meet the inclusion criteria (e.g. they represented comments or case reports or were not primarily focused on lymphoedema). Of the remaining 136 studies, 88 (65%) concerned individuals with upper-limb lymphoedema, 34 (25%) concerned individuals with lower limb lymphoedema, five (3%) reported on individuals with midline lymphoedema, and nine (7%) described patients with lymphoedema in multiple locations.

After 34 publications focusing on lymphoedema of the upper limb were assessed, saturation of the noticed meaningful concepts was reached. Because no further information was expected, it was decided that the remaining 54 publications on upper-limb lymphoedema would not be considered. Because no discrimination by location was possible in the studies that involved combined lymphoedema locations, these studies were excluded.

Ultimately, 73 publications were included (34 on upper-limb, 34 on lower-limb, and five on midline). Twenty of the included studies were intervention studies ($n=15$ for upper limb; $n=4$ for lower limb, and $n=1$ for midline), 37 were observational studies ($n=17$ for upper limb, $n=16$ for lower limb, and $n=4$ for midline), 13 were qualitative studies ($n=1$ for upper limb, and $n=12$ for lower-limb), and three were clinimetric studies ($n=1$ for upper limb and $n=2$ for lower limb). The studies of upper-

limb lymphoedema were all conducted in developed countries (i.e. Europe, USA, and Australia), whereas 22 of the 34 studies of lower-limb lymphoedema were conducted in Africa, Asia, and Central America.

Identified concepts and ICF codes

A total of 2488 concepts were extracted; 8.8% (219) of these concepts could not be linked to ICF categories. Of these 219 concepts, 76 concepts were considered “personal factors”, 75 represented “health conditions”, 37 were considered “not covered”, 21 were considered “not defined – quality of life”, and 10 were classified as “not defined-general health”. The remaining 2269 concepts could be linked to ICF categories. Three concepts (0.2%) were linked to an ICF component; 64 (2.8%) were linked to the first level categories, 935 (41.3%) to second-level, and 1265 (55.7%) to third-level. No concepts were linked to fourth-level categories. After the initial classification was made, the third-level categories were merged into the corresponding second-level categories.

Tables 1–4 list the 93 second-level ICF categories representing the meaningful concepts that were classified ≥ 4 studies involving upper-limb and/or lower-limb lymphoedema. This cutoff was chosen for readability. Because the number of midline studies was very small ($n=5$), all of those studies were taken into account. Thirty of the 93 categories (32.2%) belong to the component body functions, 12 (12.9%) to body structures, 27 (29.1%) to activities and participation, and 24 (25.8%) to environmental factors.

Tables 1–4 also show the division of the ICF categories linked to the meaningful concepts derived from the studies as well as the ICF categories linked to the meaningful concepts derived from the lymphoedema-specific questionnaires found in the identified studies (Viehoff et al, 2013). For comparison, the results of questionnaires for the upper extremity ($n=9$) and for the lower extremity ($n=2$) are presented separately. There were no questionnaires specific for midline lymphoedema.

Body functions

The most frequently found second-

level categories of the ‘body functions’ component for the upper limb were b435 (immunological system functions; 97% of studies; 89% of questionnaires), b280 (sensation of pain; 56% of studies; 78% of questionnaires), and b265 (touch functions; 32% of studies; 67% of questionnaires; Table 1). For the lower limb, the most frequently found categories were b280 (sensation of pain; 62% of studies; 100% of questionnaires), b435 (immunological system functions; 82% of studies; 50% of questionnaires), and b265 (touch functions; 32% of studies; 100% of questionnaires). In studies of midline lymphoedema, the categories b640 (sexual functions; 80% of studies) and b435 (immunological system functions; 60% of studies) were most often found. A similar pattern is apparent in the questionnaires, although class b455 (exercise tolerance functions) is not used to code meaningful concepts in the questionnaires for either the upper or lower limb. Several other categories also have no representation in the questionnaires (e.g. b160 – thought functions, b164 – higher-level cognitive functions, b270 – sensory functions related to temperature and other stimuli, b530 – weight maintenance functions, and b540 – general metabolic functions). Three ICF categories that are present in the questionnaires were not present in at least four studies: b134 (sleep functions), b550 (thermoregulatory functions), and b840 (sensation related to the skin).

Body structures

The most frequently found second-level categories of the ‘body structures’ component for the upper limb were s730 (structure of upper extremity; 91% of studies, 100% of questionnaires), s420 (structure of immune system; 68% of studies, and s810 (structure of areas of skin; 38% of studies; 33% of questionnaires; Table 2). For the lower limb, the most frequently found categories were s750 (structure of lower extremity; 94% of studies; 100% of questionnaires), s810 (structure of reproductive system; 26% of studies; 50% of questionnaires), and s420 (structure of immune system; 35% of studies).

In the midline studies, s630 (structure of reproductive system; 80% of studies),

s750 (structure of lower extremity; 60% of studies), and s810 (structure of areas of skin; 60% of studies) were the most often classified categories. No representation of s420 (structure of immune system) was found in the questionnaires, although this category had a high frequency in the studies. The questionnaires also

did not mention s410 (structure of cardiovascular system) or s430 (structure of respiratory system). It is remarkable that s730 (structure of upper extremity) has a representation in studies of the lower limb, and that, vice versa that s750 (structure of lower extremity) is represented in studies of the upper limb.

Activities and participation

In the component “activities and participation”, d570 (looking after one’s health; 71% of studies; 78% of questionnaires), d540 (dressing; 21% of studies; 78% of questionnaires), and d455 (hand and arm use; 21% of studies; 56% of questionnaires) were most often coded for the upper limb (Table 3).

Table 1. Frequency of second-level categories of the International Classification of Functioning, Disability and Health (ICF) linked to concepts contained in the 73 studies for the component Body Functions. For the studies of upper and lower limb lymphedema, only ICF codes that were present in at least four of studies are shown.

ICF code	Description	Upper limb n (%)		Lower limb n (%)		Midline Studies n (%)
		Studies	Questionnaires	Studies	Questionnaires	
Chapter 1: Mental functions						
b114	Orientation functions	–	–	10 (29)	–	–
b122	Global psychosocial functions	4 (12)	–	7 (21)	–	–
b126	Temperament and personality functions	5 (14)	3 (33)	10 (29)	–	1 (20)
b130	Energy and drive functions	–	–	10 (29)	–	1 (20)
b134	Sleep functions	–	4 (44)	–	1 (50)	–
b140	Attention functions	–	–	–	–	1 (20)
b152	Emotional functions	8 (24)	2 (22)	19 (56)	1 (50)	2 (40)
b160	Thought functions	–	–	8 (24)	–	–
b164	Higher-level cognitive functions	–	–	9 (26)	–	–
b180	Experience of self and time functions	6 (18)	3 (33)	8 (24)	1 (50)	1 (20)
Chapter 2: Sensory functions and pain						
b210	Seeing functions	–	–	5 (15)	–	–
b265	Touch functions	11 (32)	6 (67)	11 (32)	2 (100)	–
b270	Sensory functions related to temperature and other stimuli	5 (14)	–	7 (21)	–	–
b280	Sensation of pain	19 (56)	7 (78)	21 (62)	2 (100)	1 (20)
Chapter 4: Functions of the cardiovascular, hematological, immunological and respiratory systems						
b415	Blood vessel functions	–	–	5 (15)	–	–
b430	Hematological system functions	–	–	–	–	1 (20)
b435	Immunological system functions	33 (97)	8 (89)	28 (82)	1 (50)	3 (60)
b440	Respiration functions	–	–	–	–	1 (20)
b455	Exercise tolerance functions	14 (41)	–	13 (38)	–	–
Chapter 5: Functions of the digestive, metabolic and endocrine systems						
b530	Weight maintenance functions	4 (12)	–	4 (12)	–	–
b540	General metabolic functions	4 (12)	–	–	–	–
b550	Thermoregulatory functions	–	1 (11)	4 (12)	1 (50)	–
Chapter 6: Genitourinary and reproductive functions						
b610	Urinary excretory functions	–	–	–	–	1 (20)
b620	Urination functions	–	–	–	–	1 (20)
b640	Sexual functions	–	–	–	–	4 (80)
Chapter 7: Neuromusculoskeletal and movement-related functions						
b710	Mobility of joint functions	12 (35)	3 (33)	6 (18)	1 (50)	1 (20)
b730	Muscle power functions	9 (26)	1 (11)	6 (18)	1 (50)	–
b735	Muscle tone functions	–	–	–	–	1 (20)
Chapter 8: Functions of the skin and related structures						
b810	Protective functions of the skin	8 (24)	3 (33)	14 (41)	1 (50)	1 (20)
b820	Repair functions of the skin	8 (24)	1 (11)	18 (53)	1 (50)	2 (40)
b830	Other functions of the skin	–	–	4 (12)	–	–
b840	Sensation related to the skin	–	3 (33)	–	–	–

Yellow text, categories that have a mutual representation in both the studies and the questionnaires; *red text*, categories that are present in the questionnaires but have no representation in the studies; *blue text*, categories that are present in the studies but have no representation in the questionnaires.

Table 2. Frequency of second-level categories of the International Classification of Functioning, Disability and Health (ICF) linked to the concepts contained in the 73 studies for the component Body Structures. For the studies of upper and lower limb lymphedema, only ICF codes present in at least four of studies are shown.

ICF code	Description	Upper limb <i>n</i> (%)		Lower limb <i>n</i> (%)		Midline Studies <i>n</i> (%)
		Studies	Questionnaires	Studies	Questionnaires	
Chapter 4: structures of the cardiovascular, immunological and respiratory systems						
s410	Structure of cardiovascular system	-	-	6 (18)	-	1(20)
s420	Structure of immune system	23 (68)	-	12 (35)	-	1(20)
s430	Structure of respiratory system	4 (12)	-	-	-	-
Chapter 6: Structures related to the genitourinary and reproductive systems						
s610	Structure of urinary system	-	-	-	-	2 (40)
s620	Structure of pelvic floor	-	-	-	-	2 (40)
s630	Structure of reproductive system	6 (18)	5 (56)	9 (26)	1 (50)	4 (80)
Chapter 7: Structures related to movement						
s720	Structure of shoulder region	9 (26)	2 (22)	-	-	1 (20)
s730	Structure of upper extremity	31(91)	9 (100)	8 (24)	-	1 (20)
s740	Structure of pelvic region	-	-	6 (18)	-	-
s750	Structure of lower extremity	5 (15)	-	32 (94)	2 (100)	3 (60)
s760	Structure of trunk	6 (18)	3 (33)	-	-	2 (20)
Chapter 8: Skin and related structures						
s810	Structure of areas of skin	13 (38)	3 (33)	12 (35)	-	3 (60)

Yellow text, categories that have a mutual representation in both the studies and the questionnaires; red text, categories that are present in the questionnaires but have no representation in the studies; blue text, categories that are present in the studies but have no representation in the questionnaires.

For the lower limb, d570 (looking after one's health; 65% of studies; 50% of questionnaires), d850 (remunerative employment; 32% of studies; 50% of questionnaires), and d230 (carrying out daily routine; 29% of studies; 50% of questionnaires) were the most frequently found categories. In the midline studies, d570 (looking after one's health; 40% of studies) was the most frequently found category. In contrast to the upper extremity, the ICF codes for the lower limb – d415 (maintaining a body position) and d540 (dressing) – were not used in the questionnaires. This is also the case for many of the codes in Chapters 3, 4, 5, 7, 8, and 9. By contrast, d325 (communicating with – receiving – written messages), d330 (speaking), d440 (fine hand use), d445 (hand and arm use), d470 (using transportation), and d650 (caring for household objects) were not represented in the studies.

Environmental factors

For the component “Environmental factors”, the most frequently found categories for the upper limb were e580 (health services, systems and policies; 100% of studies; 67% of questionnaires), e355 (support of and relationships with health professionals; 85% of studies; 56% of questionnaires), and e115 (products and technology for personal

use in daily life; 68% of studies; 67% of questionnaires; Table 4). For the lower limb, these were e580 (health services, systems and policies; 100% of studies; 50% of questionnaires), e325 (support of and relationships with acquaintances, peers, colleagues, neighbours and community members; 91% of studies; 50% of questionnaires), and e355 (support of and relationships with health professionals; 79% of studies). Categories e580 (health services, systems and policies; 100% of studies), e325 (support of and relationships with acquaintances, peers, colleagues, neighbours and community members; 80% of studies), and e110 (products and substances for personal consumption; 60% of studies) were found in three or more studies of midline lymphoedema.

Comparison of the ICF codes in the studies and those in the questionnaires showed that in the case of e110 (products and substances for personal consumption), e115 (products and technology for personal use in daily living), and e355 (support of and relationships with health professionals) there was no representation of ICF codes in the questionnaires relating to the lower limb. This also applied to several other categories in the chapters concerning “environmental factors” (Table 4). The categories e125 (products

and technology for communication), e135 (products and technology for employment), e225 (climate), and e245 (time-related changes) are present in the questionnaires but not in the studies.

Discussion

Using the ICF as a reference, it was possible to identify and quantify meaningful concepts found in published studies that focused on individuals with lymphoedema. While a large number of these concepts could be linked to the ICF (91.2%), a small portion (8.8%) could not. Of these concepts, “health conditions” can be classified with the International Statistical Classification of Diseases and Related Health Problems 10 (WHO, 2010; 34%). Unfortunately, “personal factors” (3%) is a component of the ICF that remains under development, although some provisional lists for this component have been published (Kim do et al, 2010; Speck et al, 2010). The identification of ICF categories in the literature contributes to the development process of ICF Core Sets for lymphoedema predominantly from the researchers' point of view.

Whereas earlier systematic reviews for ICF Core Set development tended to focus on health-specific questionnaires (Post et al, 2010), in the present study meaningful concepts were extracted both

from the studies themselves and from the lymphoedema-specific questionnaires included in these studies. The results obtained using the questionnaires have been published elsewhere (Viehoff et al, 2013).

The most frequently found categories were e580 (health services, systems and

policies; 73 studies), b435 (immunological system functions; 64 studies), e325 (support of and relationships with acquaintances, peers, colleagues and community members; 59 studies), e355 (support of and relationships with health professionals; 58 studies), and d570 (looking after one's health; 48 studies).

Since the lymphatic system is part of the immunological system, this explains the frequency of the code b435 (immunological system functions), which implies the swelling of arms and legs. Swelling, especially in the genital region and breast, can also be a significant burden in midline lymphoedema.

Table 3. Frequency of second-level categories of the International Classification of Functioning, Disability and Health (ICF) linked to concepts contained in the 73 studies for the component Activities and Participation. For the studies of upper and lower limb lymphedema, only ICF codes present in at least four of studies are presented.

ICF code	Description	Upper limb n (%)		Lower limb n (%)		Midline Studies n (%)
		Studies	Questionnaires	Studies	Questionnaires	
d155	Acquiring skills	-	-	5 (15)	-	-
Chapter 2: general tasks and demands						
d230	Carrying out daily routine	6 (18)	3 (33)	10 (29)	1 (50)	-
Chapter 3: Communication						
d325	Communicating with – receiving – written messages	-	1 (11)	-	1 (50)	-
d330	Speaking	-	2 (22)	-	-	-
d335	Producing non-verbal messages	-	-	7 (21)	-	1 (20)
d340	Producing messages in formal sign language	-	-	6 (18)	-	-
Chapter 4: Mobility						
d410	Changing basic body position	-	-	7 (21)	-	-
d415	Maintaining a body position	6 (18)	3 (33)	16 (47)	-	1 (20)
d430	Lifting and carrying objects	6 (18)	2 (22)	-	-	-
d440	Fine hand use	-	2 (22)	-	1 (50)	-
d445	Hand and arm use	7 (21)	5 (56)	-	1 (50)	-
d450	Walking	-	2 (22)	11 (32)	-	1 (20)
d460	Moving around in different locations	-	-	5 (15)	-	-
d470	Using transportation	-	4 (44)	-	-	-
Chapter 5: Self-care						
d510	Washing oneself	-	-	8 (24)	-	-
d520	Caring for body parts	10 (29)	2 (22)	10 (29)	-	-
d530	Toileting	-	-	-	-	1 (20)
d540	Dressing	7 (21)	7 (78)	16 (47)	-	-
d570	Looking after one's health	24 (71)	7 (78)	22 (65)	1 (50)	2 (40)
Chapter 6: Domestic life						
d640	Doing housework	4 (12)	3 (33)	9 (26)	1 (50)	-
d650	Caring for household objects	-	2 (22)	-	-	-
d660	Assisting others	-	-	6 (18)	-	-
Chapter 7: Interpersonal interactions and relationships						
d710	Basic interpersonal interactions	-	-	4 (12)	-	-
d720	Complex interpersonal interactions	6 (18)	-	8 (24)	-	-
d750	Informal social relationships	-	-	7 (21)	-	-
d770	Intimate relationships	-	-	9 (26)	-	1 (20)
Chapter 8: Major life areas						
d845	Acquiring, keeping and terminating a job	-	-	7 (21)	-	-
d850	Remunerative employment	5 (15)	5 (56)	11 (32)	1 (50)	-
d870	Economic self-efficiency	-	-	10 (29)	-	-
Chapter 9: Community, social and civic life						
d910	Community life	7 (21)	-	10 (29)	-	1 (20)
d920	Recreation and leisure	5 (15)	5 (56)	11 (32)	-	1 (20)
d930	Religion and spirituality	-	-	8 (24)	-	-

Yellow text, categories that have a mutual representation in both the studies and the questionnaires; *red text*, categories that are present in the questionnaires but have no representation in the studies; *blue text*, categories that are present in the studies but have no representation in the questionnaires.

Inclusion of categories e580 (health services, systems and policies) and e355 (support of and relationships with health professionals) is due to the fact that in almost every study — regardless of design — the relevant therapy for lymphoedema was mentioned. The high frequency of e325 (support of and relationships with acquaintances, peers, colleagues and community members) derives from the fact that, in

the qualitative research, much emphasis is placed on focus groups and “fellow-sufferer” contacts. These were also coded with e325. For these underlying reasons, the classification e325 has been biased. Future research on the Core Sets development process should investigate whether this code is relevant.

The lack of literature concerning midline lymphoedema in this review was expected because midline

lymphoedema occurs less often than lymphoedema of the upper and lower limbs. The resulting codes for midline lymphoedema might be biased because four of the studies included focused on genital lymphoedema and one on breast lymphoedema. Because lymphoedema of the breast is seen more frequently than genital lymphoedema, there is a need for more studies of this condition. Furthermore, no studies concerning

Table 4. Frequency of second-level categories of the International Classification of Functioning, Disability and Health (ICF) linked to the concepts contained in the 73 studies for the component Environmental Factors. For the studies of upper and lower limb lymphedema, only the ICF codes present in at least four studies are presented.

ICF code	Description	Upper limb n (%)		Lower limb n (%)		Midline Studies n (%)
		Studies	Questionnaires	Studies	Questionnaires	
e110	Products and substances for personal consumption	13 (38)	4 (44)	26 (76)	-	3 (60)
e115	Products and technology for personal use in daily living	23 (68)	6 (67)	23 (68)	-	2 (40)
e125	Products and technology for communication	-	1 (11)	-	1 (50)	-
e135	Products and technology for employment	-	3 (33)	-	1 (50)	-
e140	Products and technology for culture, recreation and sport	4 (12)	-	-	-	-
e150	Design, construction and building products and technology of buildings for public use	-	-	-	5 (15)	-
e155	Design, construction and building products and technology of buildings for private use	-	-	9 (26)	-	1 (20)
e165	Assets	-	-	5 (15)	-	-
Chapter 2: Natural environment and human-made changes to environment						
e210	Physical geography	5 (15)	-	13 (38)	-	-
e215	Population	4 (12)	-	14 (41)	-	-
e225	Climate	-	2 (22)	4 (12)	-	-
e245	Time-related changes	-	2 (22)	-	-	-
e255	Vibration	-	-	-	-	1 (20)
Chapter 3: Support and relationships						
e310	Immediate family	8 (24)	2 (22)	16 (47)	-	1 (20)
e315	Extended family	-	-	8 (24)	-	-
e320	Friends	-	-	9 (26)	-	-
e325	Support of and relationships with acquaintances, peers colleagues, neighbours and community members	24 (71)	2 (22)	31 (91)	1 (50)	4 (80)
e330	People in positions of authority	-	-	6 (18)	-	-
e355	Health professionals	29 (85)	5 (56)	27 (79)	-	2 (40)
Chapter 4: Attitudes						
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	-	-	4 (12)	-	-
e460	Societal attitudes	-	-	4 (12)	-	-
e465	Social norms, practices and ideologies	-	-	4 (12)	-	-
Chapter 5: Services, systems and policies						
e535	Communication services, systems and policies	5 (15)	-	-	-	-
e555	Associations and organisational services, systems and policies	-	-	5 (15)	-	-
e570	Social security services, systems and policies	4 (12)	-	4 (12)	-	2 (20)
e580	Health services, systems and policies	34 (100)	6 (67)	34 (100)	1 (50)	5 (100)
e585	Education and training services, systems and policies	4 (12)	-	4 (12)	-	1 (20)

Yellow text, categories that have a mutual representation in both the studies and the questionnaires; *red text*, categories that are present in the questionnaires but have no representation in the studies; *blue text*, categories that are present in the studies but have no representation in the questionnaires.

head or neck lymphoedema were found, although these conditions frequently occur after surgery for cancer in this area. The present research also confirms the need for a lymphoedema-specific questionnaire for midline – especially genital – lymphoedema, and for head and neck lymphoedema because no such questionnaires were found in this literature research.

Body functions

Concerning the “body functions” categories, much lymphoedema research still focuses on the symptoms of pain and swelling, whereas other symptoms, such as skin problems (Chapter 8, Functions of the skin and related structures), are less well represented. If other research for the development of the ICF Core Sets for lymphoedema (i.e. the patient’s, clinician’s and expert’s points of view) indicates the importance of skin problems, the authors recommend that skin-related items be added to lymphoedema-specific questionnaires.

ICF “mental functions” categories is less well represented in studies of upper-limb lymphoedema (four categories) than in studies of lower-limb lymphoedema (eight categories). The included studies of lower-limb lymphoedema were mostly qualitative, whereas many studies involving upper-limb lymphoedema were controlled studies in which symptoms (e.g. pain and swelling) were measured.

Mental functions were more often described in qualitative research than in controlled studies. This difference can be explained by the fact that qualitative research produces more diverse data than controlled studies, which produce only predefined data. ICD “Mental functions”, as well as “Exercise tolerance functions”, were also better represented in the studies than in the questionnaires. Although researchers often describe problems related to mental function and exercise tolerance, they have not been used as outcome parameters in questionnaires; this gap could be filled by using generic questionnaires or performance tests. The under-representation could also be the result of the assumption that training is a contraindication for lymphoedema. More recent research devotes more attention to muscle power, coordination and endurance (Kim do et al, 2010; Speck et al, 2010; Tidhar and Katz-Leurer, 2010).

Body structures

Within the “body structures” component, codes s730 (structure of upper extremity), s750 (structure of lower extremity), and s420 (structure of immune system) were found most frequently. This is most likely because the swelling caused by the lymphoedema changed the appearance of the relevant body part. Code s420 (structure of immune system) is used often because of the description of lymphatic vessels and nodes. Concerning the codes s730 (structure of upper extremity) and s750 (structure of lower extremity), the same representations can be observed in questionnaires. On the other hand, more attention was paid to s630 (structures related to the genitourinary and reproductive systems) in questionnaires. The code s420 (structure of immune system) is lacking in the questionnaires. This is understandable given the difficulty of asking patients about their lymphatic vessels and nodes. Asking about the genitourinary region (genital lymphoedema) and the breast seems more appropriate.

That s730 (structure of upper extremity) was represented in studies of the lower limb and, conversely, that s750 (structure of lower extremity) was represented in one study of the upper limb can be explained by the fact that some disorders that cause lymphoedema (e.g. filariasis) have consequences for the entire body, although only one body part was examined in the study.

Activities and participation

In general, treatment of lymphoedema is divided into extensive and maintenance phases. In the second phase, self-management is of great importance. This explains the high frequency of the item d570 (looking after one’s health) in both the studies and questionnaires. Furthermore, researchers describe problems with maintaining a body position (d415; e.g. lying, sitting, and standing, especially for the lower limb; 47%), but do not ask about these problems in their questionnaires. This issue could also be relevant in upper limb lymphoedema, especially in patients with mastectomy and higher-grade lymphoedema. If maintaining a body position is a problem, patients should be questioned about it more often.

Most of the literature found concerned lymphoedema of the upper extremities ($n=88$), whereas lymphatic filariasis primarily damages the lymphatic system of the lower extremities (Kim do et al, 2010; Tidhar and Katz-Leurer, 2010; Devoogdt et al, 2011; Fayed et al, 2011; Tsauo et al, 2011; Pusic et al, 2012).

The category d450 (walking) had a low frequency (32%) in studies about lower-limb lymphoedema. Furthermore, walking is not mentioned in lymphoedema-specific questionnaires regarding the lower limb, although this may have been taken into account by using generic questionnaires or performance tests. There is an apparent need for research to provide more insight into the restrictions people with lymphoedema have with walking.

Environmental factors

Although the component “Environmental factors” is not frequently used to code meaningful concepts, the support of health professionals and the provision of therapy (e355) and health services, systems and policies (e580) seem to be important, given that they are mentioned in all included studies and half of all questionnaires. The categories e110 (products and substances for personal consumption; 42 studies) and e115 (products and technology for personal use in daily living; 48 studies) have a fairly high representation. The need for medication, the use of ointments for skin care and the use of bandages and hosiery are examples of the importance of these items to the lymphoedema patient. The results obtained from the questionnaires correspond to these findings, although, in general, there is relatively little representation of the ICF categories related to environmental factors in questionnaires related to lymphoedema of the lower limb (three categories). This may be because part of the research surveyed was conducted in communities in which access to health services is difficult.

Strengths and weaknesses of the study

The authors introduce a new approach to finding ICF categories in literature. Besides examining questionnaires, the studies themselves were assessed. The number of ICF categories is much greater

than the number of categories provided in the questionnaires. This has to be taken into account in the comparison of results. Nevertheless, by examining both the questionnaires and the studies themselves, a useful selection of ICF categories has been identified. The authors recommend the use of both questionnaires and studies because these approaches are complementary to each other and because the use of both methods yields more data than can be obtained by using either method alone.

All of the publications on upper-limb lymphoedema used in this review originated in developed countries. The literature on lower-limb lymphoedema came from both developed and developing countries. The fact that future Core Sets for lymphoedema will be designed primarily based on data produced in developed countries has to be taken into account.

The selection of studies might have been biased by the inclusion only of studies published in English. The literature search covered the period 2005–2010; it is possible that including more recent studies would have yielded different results (e.g. the Lymph-ICF cited by Devoogdt et al [2011]).

It was not always simple and straightforward to link concepts reported in clinical studies to the appropriate ICF categories, and this process required frequent discussion. More information regarding inter-coder agreement, by calculating kappa, might have been useful. Unfortunately, the required data were not recorded. However, linkage exercises have demonstrated that it is possible to examine and compare the content of measures based on the ICF framework and predefined linking rules with good reliability (Cieza et al, 2005; Cieza et al, 2002; Fayed et al, 2011).

The fact that the large majority of meaningful concepts found in studies concerning lymphoedema could be linked confirms the usefulness of the ICF classification. Recent publications (Devoogdt et al, 2011; Tsauo et al, 2011) stress these findings. Devoogdt et al (2011) constructed a questionnaire (Lymph-ICF) based on data from interviews with patients with breast cancer-related lymphoedema. It comprises 29 meaningful concepts

and their ICF codes. Although the classifications “body structures” and “environmental factors” are absent and the linking process remains unclear, their findings are in many ways similar to the results of the present review.

Conclusion

The ICF provides a valuable reference for identifying meaningful concepts among individuals with lymphoedema. In this and previous research on lymphoedema-specific questionnaires, a substantial number of meaningful concepts concerning lymphoedema were found, and most of them could be linked to the ICF. The most frequently found ICF categories in the studies and questionnaires are similar.

There is a need for lymphoedema-specific questionnaires concerning midline lymphoedema. Questionnaires for the lower limb should be adapted because many categories mentioned in the studies do not appear in the few questionnaires for the lower limb that were found.

The findings of this literature review indicate a need to define and to agree on what should be measured in lymphoedema care. Pusic et al (2012) reported a similar conclusion concerning data obtained from studies of health-related quality of life for patients with upper-limb lymphoedema.

References

- Cieza A, Brockow T, Ewert T et al (2002) Linking health-status measurements to the International Classification of Functioning, Disability and Health. *J Rehabil Med* 34(5): 205–10
- Cieza A, Geyh S, Chatterji S et al (2005) ICF linking rules: an update based on lessons learned. *J Rehabil Med* 37(4): 212–8
- Devoogdt N, Van Kampen M, Geraerts I et al (2011) Lymphoedema Functioning, Disability and Health Questionnaire (Lymph-ICF): reliability and validity. *Phys Ther* 91(6): 944–57
- Fayed N, Cieza A, Bickenbach J (2011) Linking health and health-related information to the ICF: a systematic review of the literature from 2001 to 2008. *Disabil Rehabil* 33(21-22): 1941–51
- Foldi M, Foldi E (eds) (2006) *Foldi's Textbook of Lymphology*. Elsevier, Munich
- Fu MR, Axelrod D, Haber J (2008) Breast-cancer-related lymphedema: information, symptoms, and risk-reduction behaviors. *J Nurs Scholarsh* 40(4): 341–8
- Fu MR1, Rosedale M (2009) Breast cancer survivors' experiences of lymphedema-related symptoms. *J Pain Symptom Manage* 38(6): 849–59
- International Lymphoedema Framework (2006) *Best Practice For The Management of Lymphoedema. International Lymphoedema Framework Consensus Document*. MEP Ltd, London
- International Society of Lymphology (2013) Consensus document of the international society of lymphology: the diagnosis and treatment of peripheral lymphoedema. *Lymphology* 46(1): 1–11
- Jacob SE, Sreevidya S, Chacko E et al (2002) Early and late morbidity associated with axillary levels I-III dissection in breast cancer. *J Surg Oncol* 79(3): 151–5
- Kim do S, Sim YJ, Jeong HJ, Kim GC (2010) Effect of active resistive exercise on breast cancer-related lymphedema: a randomized controlled trial. *Arch Phys Med Rehabil* 91(12): 1844–8
- Norman SA, Localio AR, Potashnik SL et al (2009) Lymphedema in breast cancer survivors: incidence, degree, time course, treatment, and symptoms. *J Clin Oncol* 27(3): 390–7
- Post MW, Kirchberger I, Scheuringer M et al (2010) Outcome parameters in spinal cord injury research: a systematic review using the International Classification of Functioning, Disability and Health (ICF) as a reference. *Spinal Cord* 48(7): 522–8
- Pusic AL, Cemal Y, Albornoz C et al (2012) Quality of life among breast cancer patients with lymphedema: a systematic review of patient-reported outcome instruments and outcomes. *J Cancer Surviv* 7(1): 83–92
- Speck RM, Gross CR, Hormes JM et al (2010) Changes in the body image and relationship scale following a one-year strength training trial for breast cancer survivors with or at risk for lymphedema. *Breast Cancer Res Treat* 121(2): 421–30
- Stanton AW, Modi S, Mellor RH et al (2009) Recent advances in breast cancer-related lymphedema of the arm: lymphatic pump failure and predisposing factors. *Lymphat Res Biol* 7(1): 29–45
- Swenson KK, Nissen MJ, Leach JW, Post-White J (2009) Case-control study to evaluate predictors of lymphedema after breast cancer surgery. *Oncol Nurs Forum* 36(2): 185–93
- Tidhar D, Katz-Leurer M (2010) Aqua lymphatic therapy in women who suffer from breast cancer-related lymphedema: a randomized controlled study. *Support Care Cancer* 18(3): 383–92
- Tsauo J, Hung H, Tsai H, Huang C (2011) Can ICF model for patients with breast-cancer-related lymphedema predict quality of life? *Support Care Cancer* 19(5): 599–604
- Ustün B, Chatterji S, Kostanjsek N (2004) Comments from WHO for the Journal of Rehabilitation Medicine Special Supplement on ICF Core Sets. *J Rehabil Med* (44 Suppl): 7–8
- Viehoff PB, Hidding JT, Heerkens YF et al (2013) Coding of meaningful concepts in lymphedema-specific questionnaires with the ICF. *Disabil Rehabil* 35(25): 2105–12
- World Health Organization (2001) *International Classification of Functioning, Disability and Health*. WHO, Geneva
- World Health Organization (2010) *International Classification of Diseases*. WHO, Geneva
- World Health Organization (2011) *Weekly Epidemiological Record* 86(35): 377–88