Development of ICF Core Sets for Lymphedema

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Development of International Classification of Functioning, Disability, and Health (ICF) Core Sets for Lymphedema

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Ontwikkeling van International Classification of Functioning, Disability and Health (ICF) Core Sets voor lymfoedeem

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“All ends with beginning”

Get lucky - Daftpunk

voor mijn ouders

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Chapter 1

Introduction

Lymphedema

Lymphedema is a multi-causal chronic condition that can lead to symptoms and complaints that can be described in terms of impairments of functions and structures, limitations in activities, and participation restrictions and that often result in distress and loss of quality of life (1). Lymphedema presents clinically as swelling that is due to an imbalance between interstitial filtration, fluid and protein discharge caused by compromised lymphatic system transport capacity (2). This impairment can be relative or absolute. In the former, hyper filtration causes fluid overflow in the tissue despite maximal or normal lymphatic function (high output lymphedema) (2). With regard to absolute impairment, which is more common, lymphatic vessel transport capacity is decreased due to insufficient physical activity, a genetic predisposition, or lymphatic vessel obstruction or absence (low output lymphedema) (2). Lymphedema can also be subdivided into primary and secondary lymphedema (figure 1).

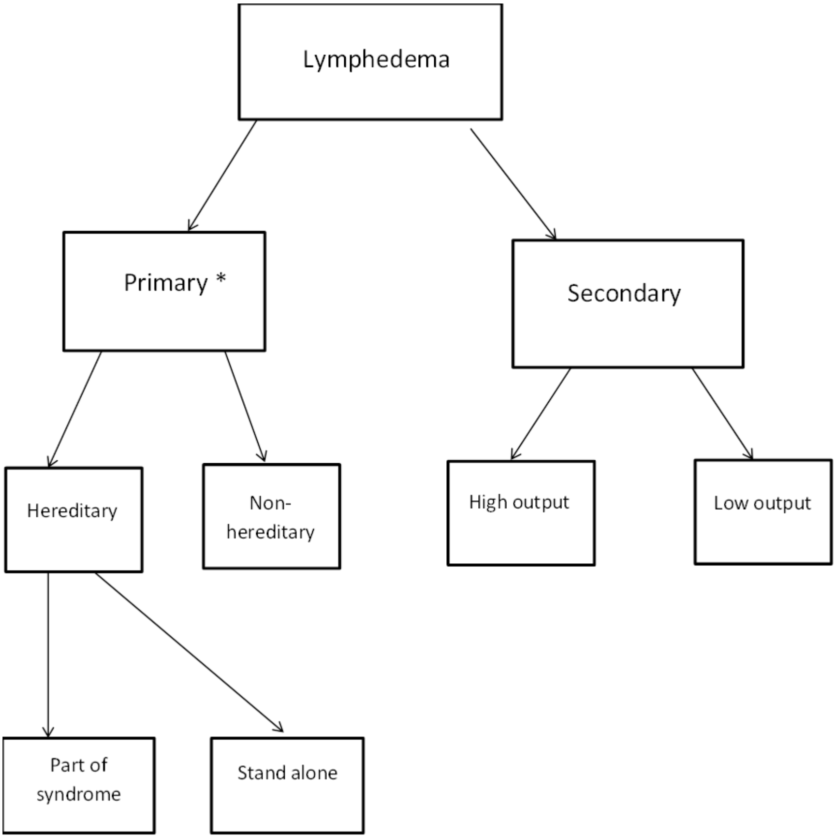

Figure 1. Subdivision of lymphedema. * primary lymphedema can vary among praecox and tarda forms.

Primary lymphedema

Primary lymphedema, a congenital and sometimes hereditary disease, may be present at birth; however, most cases of primary lymphedema manifest themselves during puberty and even later (lymphedema tarda). Different types of gene mutations lead to lymph vessels and/or nodes with an abnormal structure and/or function. The final result of an abnormal or even absent system is a serious disturbance in the clearance of interstitial fluid. This condition is most commonly observed in the legs but can occur throughout the entire body. Genital region involvement needs special attention when intestinal lymphatic impairment is present. Connell et al. (3) proposed a flowchart presenting a subdivision of primary lymphedema based on the clinical diagnosis (phenotype). This subdivision results from the following decision points (yes or no) (3):

1. syndromic

2. systematic/visceral involvement pre- or postnatal onset

3. disturbed growth / cutaneous manifestations / vascular anomalies with lymphedema

4. distichiasis present (a genetic multisystem disorder characterized by the development of extra eyelashes and swelling of the legs because of fluid accumulation)

5. congenital (onset less than a year after birth) .

In the last decade, genetic mapping has changed the primary diagnostic tool from a phenotypic diagnosis into a diagnosis that is based on the underlying genetic disturbance (genotype). All genetic diseases are classified according the OMIM (Online Mendelian Inheritance in Man) database (3). This database counts 79 different genotypes of lymphedema. For some examples, see table 1.

Table 1. Well-known examples of genetic classification of primary lymphedema (from: OMIM database www.OMIM.org (3)).

Name	Gene	OMIM code
Lymphedema-distichiasis syndrome	FOXC2	602402
Milroy disease	FLT4	136352
MLCRD syndrome	KIF11	148760
Lymphedema late-onset	FLT4	136352
Hennekam lymphangiectasia syndrome	CCBE1	612753
Hereditary primary lymphedema	GJC2	608803
Emberger syndrome	GATA2	137295
Vascular endothelial growth factor-related protein lymphedema	VEGFC	615907

Secondary lymphedema

Secondary lymphedema is much more common and is the result of an overload of interstitial fluid due to an increase of capillary filtration (as in chronic venous insufficiency) or a blockage/interruption of one or more major lymph vessels (e.g., after trauma, surgery, infection or radiotherapy). In the case of overload, dermal backflow (re-entry of more proximal lymph fluid into the skin) leads to lymphedema. In the case of a lymph blockage, stasis due to diminished or blocked lymph transport is the result. In cases of overload, secondary lymphedema is classified as high output lymphedema (4). Infections can be present due to a completely different etiology, which can be bacterial, viral, fungal, or parasitic. Erysipelas, an obligate streptococcal skin infection, is often thought to be an end result of lymphedema. However, this appears to be incorrect: subclinical (congenital) lymphedema is more prone to streptococcal infections than normal skin. For this reason, erysipelas is usually the first clinical sign of lymphedema (5). In the tropical, underdeveloped world, lymphedema mainly occurs as the result of an infection. As for parasitic infections, according to a WHO report, approximately 120 million people in 72 countries were infected with a parasite (lymphatic filariasis) in 2010 (6). Of these infected individuals, 15 million people suffered from consequent manifestations of lymphedema (filarial elephantiasis), which were a serious burden (6). Podoconiosis is an endemic non-parasitic and non-infectious form of elephantiasis due to long-term barefoot exposure to irritant volcanic soils in tropical Africa, leading to serious problems in that part of the world (7).

Localization of lymphedema

Lymphedema may occur anywhere in the body, but it mostly occurs in the upper and lower limbs and the midline (head, neck, thorax and genital region). Abdominal lymphedema is very rare.

Lymphedema in an upper limb occurs most often as a result of breast cancer treatment because of the resection of axillary lymph nodes and/or radiation therapy. In the lower limbs, lymphedema originates both secondary to cancer treatment and primarily as a genetic predisposition.

Midline lymphedema located in the head/neck-region is also almost exclusively due to cancer treatment in that region, although some non-oncological types exist: solid facial edema, Ascher’s syndrome, M. Morbihan, and Melkersson-Rosenthal syndrome are well-known examples (8). Lymphedema in the genital region often results from the oncological treatment of the internal or external genital organs, from lymphatic injury in the pelvic region, or as part of M. Milroy (Table 1).

Genital lymph drainage is bidirectional: one route flows directly to the superficial regions of the groin and the other flows through the pelvic region. If one of the routes is spared, it can serve as an alternative pathway. Abdominal lymphedema occurs most frequently because of an obstruction of the lymph vessels caused by cancer processes.

Incidence and prevalence of lymphedema

Unfortunately, the incidence and prevalence of lymphedema are not precisely known because it is not a well-registered disease worldwide and because epidemiological studies have been scarce. Its prevalence worldwide is estimated to be approximately 140-250 million people (4). In the Netherlands, information about the incidence of lymphedema is hard to find because of the great variance in studies, symptoms, methods of lymphedema diagnosis, and lack of sufficient registration of lymphedema patients because patients are predominantly classified in hospital registration systems according to the 'primary' disease, such as cancer. Moreover, in most patients lymphedema develops after hospital discharge. Sound epidemiological data are always mandatory when organizing health care programs. For a serious problem, such as lymphedema, a registration system is needed. The consensus document of the International Society for Lymphedema (ISL) (9) and the best practice document of the International Lymphedema Framework (ILF) (10) give guidelines to solve this problem. However, in many countries there have been no governmental programs or supportive actions by health insurance companies to develop a registration system according to these guidelines.

Symptoms of lymphedema

Lymphedema is a slowly progressive disease, often suddenly inflicted by a (minor) trauma. Early in the disease process, the lymphedema is pitting, although it is a protein rich edema, and the affected skin feels soft (see figure 2).

Figure 2. Example of pitting lymphedema in the foot.

Figure 3. Example of lymphedema in the lower extremity.

Figure 4. Example of lymphedema in the upper extremity. (all photos published with permission of the patients).

In later stages, the skin may feel hard and thickened as a result of chronic inflammation, leading to fibrosis of the dermis and subcutis as well as of the adipose tissue in the swollen area (see figures 2 and 3). In this stage the edema is non-pitting and irreversible. In severe long-lasting lymphedema, secondary skin changes are visible, such as papillomatosis and verrucosis (see figure 4). If untreated, lymphedema leads to further deterioration, a high risk of infection (especially erysipelas), swelling, and feelings of heaviness and tightness. In addition, patients report that jewelry and clothing feel uncomfortable or tight on the affected body parts (11-13). Moreover, other impairments occur, such as decreased limb mobility, and, as a consequence, limitations in walking and personal care and restrictions in domestic life, occupation, and socialization (7). Altered body shape (an impairment in structure), often leading to stigmatization as well as the need to use garments or bandaging daily, makes acceptance difficult for the patient as well as his/her family (14). Untreated lymphedema can lead to significant loss of health-related quality of life and even social isolation (14). Thus, lymphedema is a serious disease that needs the full attention of the medical world. The prognosis for the patient is best when appropriate care is provided starting from the first signs (15). However, the author has commonly found that the vast majority of lymphedema patients receive the care they need too late. As lymphedema is not curable, lifetime treatment is mandatory. For optimal lifetime care, good motivation of the patient, and the commitment of specialized health care professionals are the keys to success.

Treatment

The treatment of lymphedema is very challenging. Therapy is roughly divided into two major steps: 1. the initial phase and 2. the maintenance phase. The therapeutic options for lymphedema include conservative and surgical modalities and should be fine-tuned to the patient's personal circumstances, including work and home environments as well as personal factors, such as age, co-morbidities, (malignant) disease prognosis, psychosocial aspects, and physical potential (10). The goal of conservative treatment in the initial treatment phase is to eliminate edema by reducing interstitial fluid production and stimulating lymphatic propulsion by compression therapy (16). In addition, lymph flow is stimulated by manual lymph drainage (MLD) (16). Exercises aimed at improving mobility and physical strength and reconditioning are important to stimulate natural lymph drainage from the tissues (16). Pursuit of a normal body mass index (BMI) and weight control are essential components of lymphedema treatment (17). To minimize the risk of infection, the maintenance of skin integrity and special skin care are mandatory (18). Decongestive lymphatic therapy and a combination of various evidence-based conservative interventions are used to reduce the edema as much as possible and to obtain a shape as normal as possible. In conclusion, in the initial phase, the treatment consists of edema reduction, prevention of infection and maintaining a lifestyle to cope with the results of the lymphedema. Once the maximal initial therapeutic result is achieved, the maintenance phase begins. This phase - starting when there is no more pitting edema and maximal edema reduction is reached - depends mainly on patient self-management, including strict compliance in wearing compression garments (bandages, wraps and/or hosiery) and an adapted lifestyle (16). These aspects are essential to preserve the treatment results of the initial phase and to accommodate long-term management. In severe lymphedema with concurrent lipedema, circumferential suction-assisted lipectomy can be applied to alleviate irreversible alterations, such as adipose tissue formation and fibrosis (19).

In 2012, the ILF published a 'best practice document' for the management of lymphedema (10). It proposes a comprehensive lymphedema service based on the chronic care model and use of the International Classification of Functioning, Disability and Health (ICF) as a vehicle for determining and delivering care fine-tuned to the patient’s needs (10).

For an in-depth understanding of optimal lymphedema management and an objective follow-up, the use of reliable and valid assessment instruments and systematic consideration of the ILF recommendations are needed. To gather data for monitoring individual progress, the ICF can be of great value.

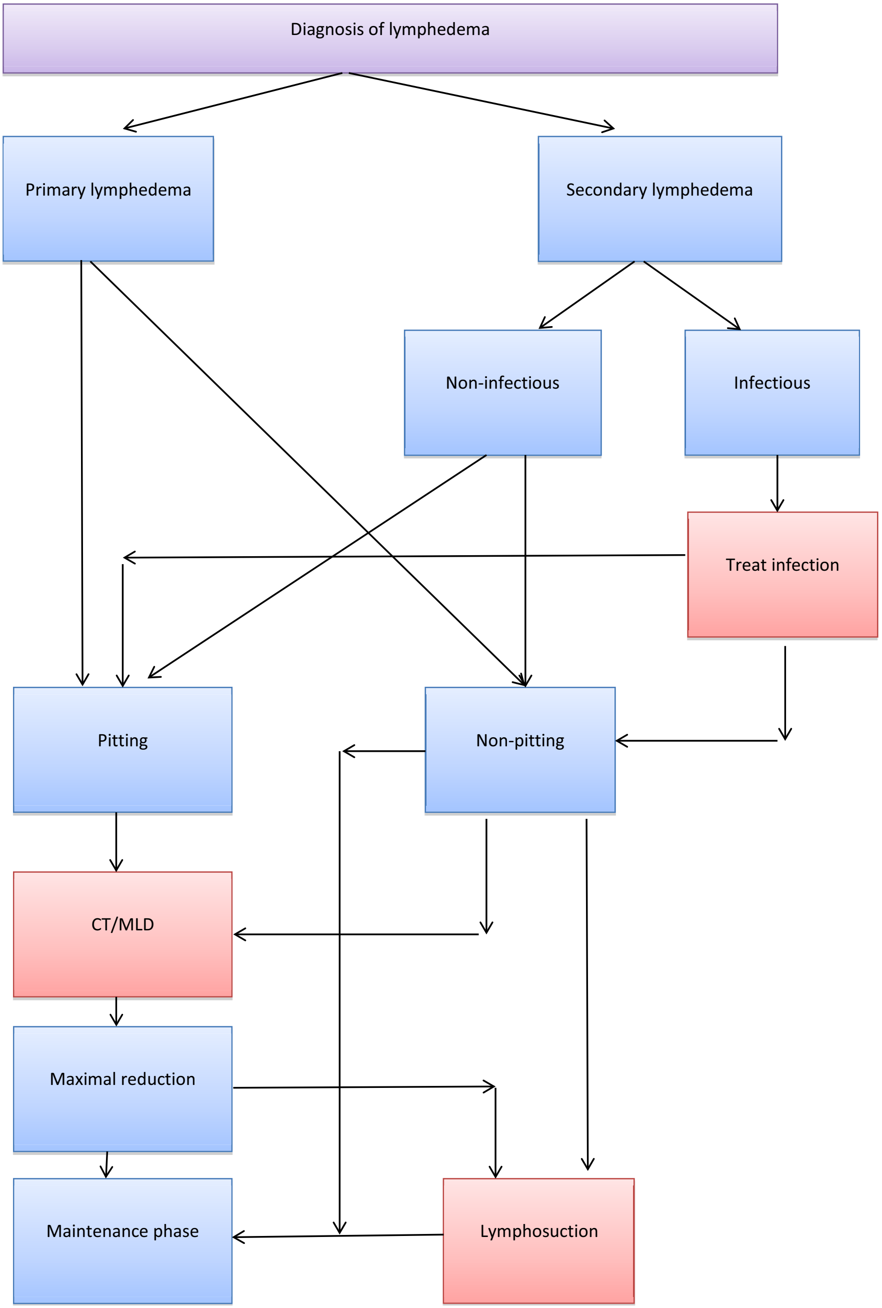

Figure 5. Schedule of lymphedema treatment. Blue sections represent classifications; red sections represent treatments.

International Classification of Functioning, Disability and Health (ICF)

The ICF provides a comprehensive framework of human functioning (see figure 6) as well as a classification system (20). The ICF offers a universal language that is understood by health professionals, researchers, policy makers, patients, and patient organizations.

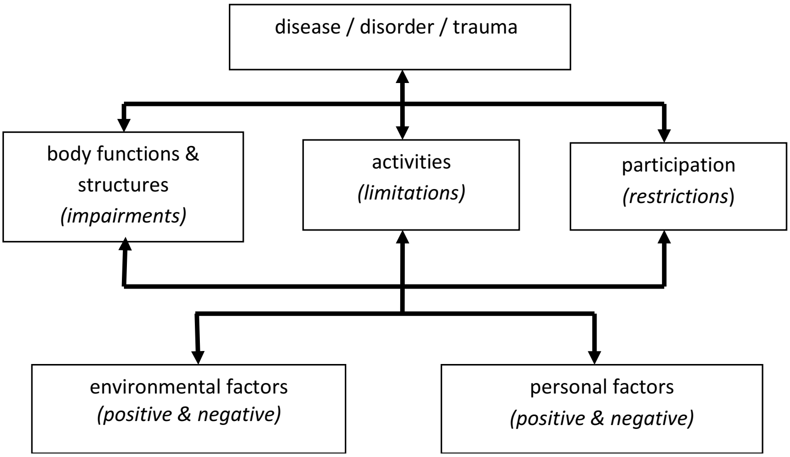

Figure 6. ICF-scheme.

The ICF consists of two separate parts. Part 1 addresses functioning and is composed of three components: ‘Body Functions’, ‘Body Structures’, and ‘Activities and Participation’. Part 2 addresses contextual factors and is composed of two components: ‘Environmental Factors’ and ‘Personal Factors’. However, ‘Personal Factors’ are not elaborated as a classification in the current ICF because of significant social and cultural variations (21).

Each ICF category is assigned an alphanumerical code consisting of a letter and one or more numbers. The letter refers to the classification component (b: Body Functions, s: Body Structures, d: (domains of) Activities and Participation, and e: Environmental Factors). Each letter is followed by one or more numbers, starting with the first level (chapter) (one digit), followed by second-level specifications (three digits) and the third- and fourth-level specifications (four and five digits), where applicable. In registration systems, ICF Qualifiers are to be added to indicate the severity of the indicated features in the individual patient.

Although its classifications with more than 1400 categories can serve as a reference, the ICF is not easily applicable in daily clinical practice. For this reason, tools, such as ICF Core Sets (20) (ICF selections applied to a specific diagnostic group), have been developed to make the ICF more useful for health care providers (22). The selected items are investigator-independent (established for universal application) and generic for all patients with a certain health condition. The ICF Core Sets facilitate the description of the functioning of patients and the formulation of treatment goals. Description uniformity is needed to compare data for evaluations of treatment efficacy and to collect data for research. The ICF Core Sets may be used as a guide for clinical reasoning and to record the results of clinical reasoning in electronic patient files. They can help the health care provider in the objective follow-up of their patients if valid and reliable measurement instruments are being used.

Two types of Core Sets exist, brief Core Sets and comprehensive ones. The brief ICF Core Set is used as a quick scan of the patient’s functioning in a clinical study or clinical situation (mono-disciplinary); the comprehensive ICF Core Set guides multidisciplinary assessments (20). Both sets include a selection of ICF categories; as few categories as possible are included for practical purposes, but as many categories as needed to describe the typical spectrum of functional problems in clinical studies, clinical encounters, multidisciplinary assessments, and treatment evaluations. Core Sets can be used by health care providers, researchers, health insurance companies, and policy-makers. Today, ICF Core Sets have been developed for various burdensome chronic conditions, such as cerebral stroke, multiple sclerosis and breast cancer (23-25). Core Sets have also been developed for acute hospital and early post-acute rehabilitation facilities (26-28). They have been proven to be of great value in daily practice (26-28). Because lymphedema is a serious, chronic and worldwide problem in health care, specific Core Sets for lymphedema could help to describe and record the total scope of the problems in functioning of these patients worldwide. To date, these Core Sets have not yet been developed.

The value of ICF Core Sets for lymphedema

The value of ICF Core Sets for lymphedema expresses itself in various ways:

1. *ICF Core Sets can act as a guideline for the choice of relevant outcomes from different perspectives.*
   The most dramatic symptom of lymphedema is swelling of the affected body part. In terms of the ICF, this is an impairment of the lymphatic function, belonging to the component Body Functions, and results in an anatomical change of the body part (second qualifier, code 7: 'qualitative change in structure, including accumulation of fluid'), belonging to the component Body Structures (21). For the greater part, studies in lymphedema have used volume as an outcome measure. Using the ICF, which is a bio-psycho-social conceptual framework, it is apparent that there are also other components that are important, such as activities and participation and that functioning of the patient is influenced by contextual factors (environmental and personal factors). Tsauo et al. (29) used the ICF in a study about breast cancer-related lymphedema, and their findings indicate that according to the patients themselves, their major problems were limitations in activities and participation restrictions. In their studies about health related quality of life in lymphedema patients, DeVoogdt et al. also distinguished a number of additional items as being important for these patients other than swelling alone, such as mental functions, household activities and social activities (30, 31).
2. *ICF Core Sets can facilitate the choice of measurement instruments.*
   It must be clear which items, in ICF terms, measurement instruments contain. To analyze this, measurement instruments can be linked to the ICF with a standard procedure and linking rules (32, 33). The results of the linking process can subsequently be compared with the ICF Core Sets, creating an image of the extent to which a measurement instrument covers one or more items from the ICF Core Sets. Given this information, the researcher or clinician can decide whether he will use the instrument or not. In conclusion, the patient information registered using the ICF Core Sets can help to decide what to measure. This concerns not only their use for scientific research but also the choice of outcomes and corresponding measurement instruments in clinical practice.

3. *The ICF Core Sets can be used as a framework for guidelines.*
   In the latest version of the Dutch multidisciplinary guideline for lymphedema, the Core Sets are frequently mentioned (15), but only as being ‘under construction’. When ICF Core Sets for lymphedema are developed and adopted by the field they can be implemented in the guideline.
4. *ICF Core Sets for lymphedema can bring structure to the data to be recorded and facilitate their incorporation into the files.*
   As a result of advancing informatics in health care, electronic patient files are used more often and data are recorded in a comprehensive way. However, the description of the functioning of lymphedema patients is not possible with the existing files.
5. *ICF Core Sets can help to describe the severity of the disability for lymphedema patients.*
   For each ICF-category of the Core Sets, so called qualifiers can be assigned. One of these qualifiers denotes the severity of the problems. A study by Uhlig et al. (34) examining the responsiveness of the ICF Core Set for rheumatoid arthritis using this qualifier demonstrated moderate responsiveness in the real-life setting of patients where minor changes occurred during treatment with a specific medication.
6. *ICF Core Sets can be used to generate lymphedema-specific data.*
   As already indicated, data on the incidence and prevalence of lymphedema are scarce (4, 15). ICF Core Sets for lymphedema could help to generate more information about this health condition, not only about the occurrence of the specific components of the ICF but also about the impact of the disease on daily life.
7. *Both ICF Core Sets and the ICF scheme can be helpful for patient education and for enhancing self-management.*
   With the ICF scheme and the ICF Core Sets, it is possible to give the patient more insight into his/her functioning and into the factors influencing this functioning. Therefore, the ICF can be used in patient education and in teaching self-management strategies. The formulation of treatment goals by the patients themselves can be facilitated with the ICF Core Sets, as they provide an overview of the relevant items. In a study about geriatric rehabilitation, the association between goal attainment and improved overall functioning underlined the necessity of considering the patient’s perspective in the rehabilitation process (35).

8. *ICF Core Sets for lymphedema can facilitate the multidisciplinary cooperation.*
   As it is a universal language, the ICF and the ICF Core Sets can not only be applied in the conversation with the patient but also in consultation settings of different health professionals. The Comprehensive Core Sets lend themselves to these multidisciplinary settings, whereas the Brief Core Sets can be applied in one on one situations (20, 22). A study by Huber et al. showed that the ICF Core Sets can be used to record precise information on patients' functioning in a multidisciplinary setting. Typical impairments, limitations and restrictions, intervention goals and methods of functioning could be documented (36). Finally, broad information about lymphedema can be collected worldwide and compared by using the universal framework of the ICF and ICF Core Sets.

Aim of the thesis

For the above mentioned reasons, the introduction of specific ICF Core Sets could significantly and positively impact care for lymphedema patients. As seen previously, lymphedema presents roughly in a consistent manner, but may have different causes. ICF Core Sets will cover the whole spectrum of this health condition. The purpose of this thesis is to develop ICF Core Sets for lymphedema. Because lymphedema can present in different regions of the human body, we decided to construct three separate ICF Core Sets: for the upper limb, lower limb, and midline lymphedema. This process will result in six Core Sets, as both a Brief and a Comprehensive Core Set will be developed for each region. To achieve this goal, we followed the method of Stucki and Cieza, who constructed a standard method for this development process (20) (figure 7). This method consists of a preparatory phase with several studies, phase 1, in which the first versions of the Core Sets are concluded, and phase 2, in which the versions are validated and implemented.

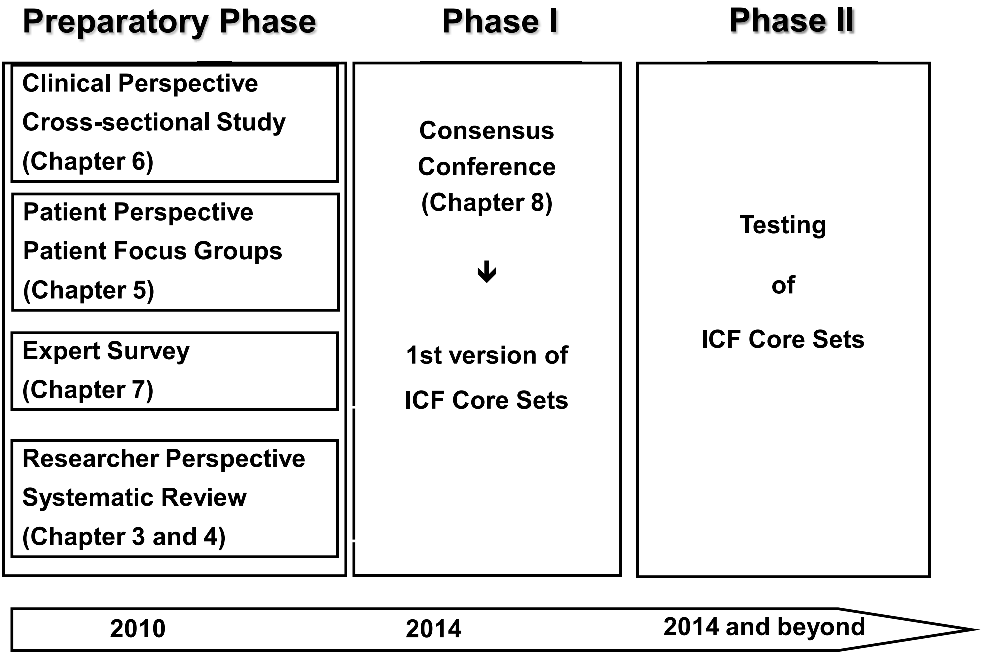

Figure 7. Development of the ICF Core Sets for lymphedema: phases of the project (from: Stucki et al., 2002 (20)).

Contents

In Chapter 2, a position paper is presented about the project in which the procedure is discussed.
The following research questions were asked:

1. What are the areas of functioning, disability and health reported in peer reviewed studies on patients with lymphedema?
2. Which concepts of the ICF can be linked to the ULL27, a French health-related quality of life questionnaire for upper limb lymphedema?
3. What are the most common problems of patients with lymphedema from the clinician's perspective using the ICF?
4. What is the perspective of patients on their functioning and health and which concepts of functioning and health are important to these patients using the ICF?

5. What is the perspective of experts in the field of lymphedema on the functioning and health of patients with lymphedema and which concepts of functioning and health are important using the ICF?
6. What is the content of ICF Core Sets for lymphedema, taking into account the researcher's, patient's and professional's perspective?

To answer these questions the procedure starts with four preparatory studies: a literature review (Chapter 3 and 4), a qualitative study with lymphedema patients (Chapter 5), a cross-sectional clinical study (Chapter 6), and an international e-mail-based survey of lymphedema experts (Chapter 7). The findings of these preparatory studies were presented at an international consensus conference (phase I), which led to the first versions of the ICF Core Sets for lymphedema (Chapter 8).

In Chapter 9, the results of a translation and validation study of the ULL27 are presented. Following the linking rules of Cieza et al. (32, 33), this questionnaire - included in the literature study (Chapter 3) - is linked to the ICF and is used as an example of the use of the ICF Core Sets in the General Discussion.

In the General Discussion (Chapter 10), we critically review our research methods and our results, and we have given recommendations for the use of the ICF Core Sets for lymphedema in practice and for further research, including validation (phase II).

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Chapter 2

Human functioning in lymphoedema

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Abstract

Background: The description and testing of the different levels of human functioning is essential prior to beginning the treatment of patients with lymphedema.

Aims: The International Classification of Functioning, Disability and Health (ICF) offers an international framework for the classification of health and health-related domains. ICF Core Sets are lists of selected ICF categories concerning the important aspects of functioning that are most likely to be affected by a specific disease. These Core Sets make it easier and faster to describe the patient’s problems and to define treatment goals. Furthermore, they are available to healthcare providers of all professions, researchers, health insurance companies and policymakers. The objective of this document is to outline the development process for ICF Core Sets for lymphedema.

Methods: The ICF Core Sets for lymphedema will be defined at a consensus conference, on the basis of data derived from the following preparatory studies: a) a systematic review b) a qualitative study c) an expert survey and d) a cross-sectional study.

Results: Not available at the time of writing.

Conclusions: The ICF Core Sets for lymphedema are being designed with the aim of providing a useful standard for both research and clinical practice.

Declaration of interest: None.

Introduction

The course of lymphoedema is unpredictable and varies among patients. Various types of impairments, activity limitations and restrictions in physical and psychosocial participation can have important consequences for each individual patient with lymphoedema, influencing the treatment programme. Describing and testing the different levels of human functioning is essential prior to embarking on the treatment of patients with lymphoedema, enabling treatment goals to be defined and adapted to the functioning of each individual patient. To achieve this goal, a framework and classification system is needed, which can serve as an objective, universal yet standardised language understood by health professionals, researchers, policymakers, patients, and patient organisations. The International Classification of Functioning, Disability and Health (ICF), approved by the World Health Assembly, offers standard terms and schemes for classifying and describing functioning, disability and health in people with lymphoedema. The International Society for Lymphology (ISL) mentions The International Classification of Functioning, Disability and Health (ICF) in its latest revision of the lymphoedema consensus document (2009).

The ICF framework consists of the following components:

- Body Structures and Functions
- Activities and Participation
- Personal and Environmental factors.

Personal factors are not yet classified, but the other components contain more than 1,400 ICF categories. This is simply not workable in practice and necessitates the creation of a practical tool to incorporate such a large number of categories. In order to reduce the amount of categories, only those terms/concepts that are the most relevant for individuals with a defined health condition are selected by the experts attending the consensus conference.

Core sets

These summaries are called ICF 'Core Sets', which are practical tools designed for a range of purposes. They allow health professionals to classify and describe an individual's functioning using a universal and standardised language. Therefore, the Core Sets offer a chance for international studies to be conducted and open up the possibility of comparing the impact of different conditions. Moreover, the ICF Core Sets can be used in electronic healthcare records.

To date, ICF Core Sets have been developed for several chronic conditions — see Cieza et al (2004), Stucki and Grimby (2004), Üstün et al (2004). For every health condition, both a brief ICF Core Set (for clinical or epidemiological studies), as well as a comprehensive ICF Core Set (for multidisciplinary assessments) has been established. The development of ICF Core Sets for lymphoedema started as a cooperative effort between the Erasmus Medical Centre (Department of Dermatology) in Rotterdam and the Dutch Institute of Allied Health Care in Amersfoort, under the auspices of the WHO Collaborating Centre for the Family of International Classifications (WHO-FIC) in Bilthoven (all in The Netherlands).

Methods

There are three phases in the development of the ICF Core Sets for lymphoedema: a preparatory phase, phase I and phase II (Figure 1).

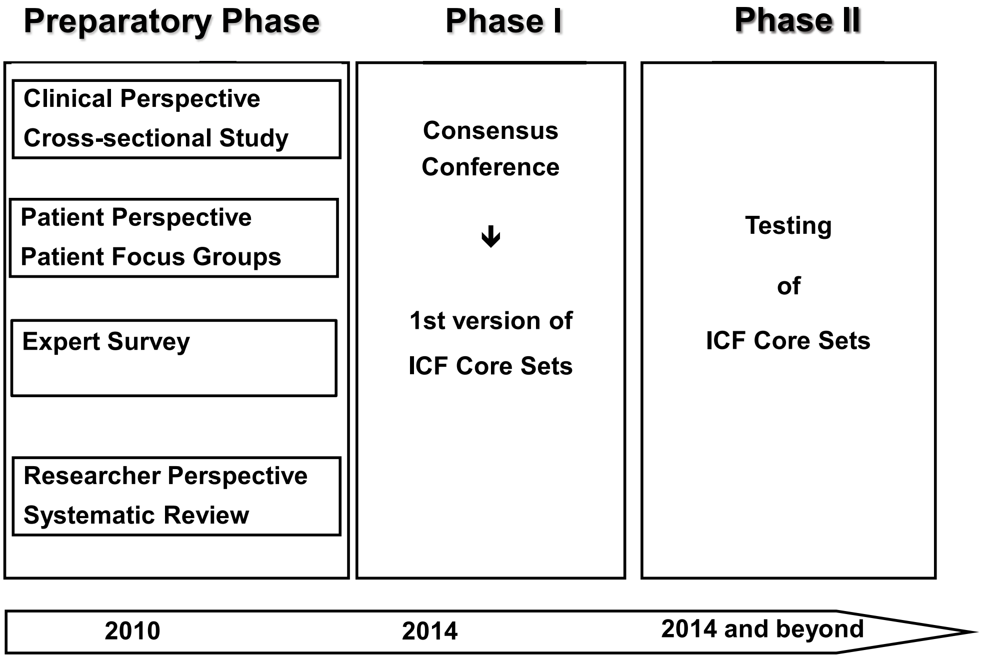

Figure 1. The three phases in the development of the ICF Core Sets for lymphoedema.

Preparatory phase

Within the preparatory phase, studies will be conducted from four different perspectives.

Systematic review: researcher perspective.

A systematic review of English language literature will be performed: (1) to identify meaningful concepts reported in studies published between 2005 and 2010 that involve patients with lymphoedema; and (2) to link these concepts to ICF codes and quantify them to indicate their importance in the literature. The literature will be retrieved from the Medline, Central, Cinahl, Embase and Pedro databases.

Qualitative study: patient perspective.

Focus group interviews will be performed with individuals with lymphoedema to identify the aspects of functioning, disability and health that are important to the individual in question. These will then be listed using the ICF as a reference. Different focus groups will be interviewed, such as those with upper extremity, lower extremity, midline lymphoedema and those with lymphoedema that may or may not be related to cancer.

Expert survey: expert perspective.

An internet-based survey will be performed to gather the opinions of an international pool of experts across various health professions (for example, physicians, nurses, physical therapists and skin therapists) regarding the most relevant aspects to be considered in persons with lymphoedema.

Empirical cross-sectional study: clinical perspective.

A cross-sectional study with 400 patients will be performed: (1) to describe functioning, disability, health and the relevant personal and environmental factors of individuals with lymphoedema and (2) to identify whether problems in functioning can be unequivocally classified using the ICF. This study will be executed partly in Australia in cooperation with Flinders University Medical Centre Lymphoedema Assessment Clinic Adelaide (Professor Neil Piller).

Phase I

The data collected during the preparatory phase will be presented at a international ICF Core Set consensus conference and experts in the field of lymphoedema will work together to reach a consensus on the most adequate ICF categories to be included in both the comprehensive and the brief ICF Core Sets for lymphoedema.

Phase II

In Phase II of the project — the testing and implementation of the ICF Core Sets for lymphoedema — an international, cross-sectional, multi-centre validation study involving patients with lymphoedema will be performed, to assess the content validity and feasibility of the comprehensive and brief ICF Core Sets for lymphoedema.

The development of ICF Core Sets for lymphoedema is an inclusive and open process and the project will be conducted in accordance with the ethical principles of the Declaration of Helsinki. Anyone who wishes to participate actively in this process is invited to contact the project co-ordinator (p.viehoff@erasmusmc.nl). The project will be guided by a steering committee comprising the authors of this article.

Discussion

ICF Core Sets for lymphoedema offer a dedicated tool to guide research, with the ultimate aim of improving understanding of functioning, disability and health in people with lymphoedema.
Tsauo et al (2010) already proved the usefulness of the ICF in patients suffering from lymphoedema due to breast cancer. They found that data classified with the ICF could well predict the health-related quality of life for these patients.
The ICF Core Sets for lymphoedema will also form the basis for the development of assessment instruments to quantify the severity of the impact of lymphoedema on functioning; to measure change over time; and to measure the effectiveness of interventions. They can also serve as the basis for setting clinical significance thresholds for diagnostic purposes. Finally, it is hoped that this will lead to effective intervention methods that improve functioning and minimise disability among people with lymphoedema throughout the world.

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Chapter 3

Coding of meaningful concepts in lymphedema-specific questionnaires with the ICF

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Abstract

Purpose: To identify and quantify the meaningful concepts within questionnaires focusing on lymphedema using the International Classification of Functioning, Disability and Health (ICF).

Methods: Electronic searches of Medline, EMBASE, CINAHL, CENtral and Pedro (2005–2010) were conducted. The concepts in the questionnaires were retrieved from the included studies and linked to the ICF.

Results: Of the 2381 abstracts retrieved, 136 studies were included. The study population suffered from lymphedema in the upper limb (65%), in the lower limb (25%), in the midline (3%) and in combinations of these areas (7%). In total, 12 lymphedema-specific questionnaires were found (nine for the upper limb, two for the lower limb and one for lymphedema in general). A total of 301 concepts were extracted from the questionnaires, of which 271 (90%) could be linked to the ICF. There were 45 two-level ICF categories linked to concepts in 2 questionnaires; 13 in Body Functions, 6 in Body Structures, 16 in Activities and Participation and 10 in Environmental Factors. The most frequently measured categories were ‘‘Structure of upper extremity’’, ‘‘Immunological system functions’’, ‘‘Looking after one’s health’’, ‘‘Sensation of pain’’, ‘‘Touch functions’’, ‘‘Dressing’’ and ‘‘Health services, systems and policies’’.

Conclusion: The ICF provides a valuable reference to identify concepts in questionnaires focusing on individuals with lymphedema

Introduction

Lymphedema is a chronic condition resulting in an abnormal accumulation of extracellular fluids as a result of disturbed lymphatic flow, primarily in the subcutaneous tissues and subfascial compartments. In lymphedema, the transport capacity of the lymphatic system is compromised because of less activity or less capacity of the lymphatic vessels or obstruction of the lymphatic system [1]. Lymphedema can occur in both upper and lower limbs and in the midline (head, neck, abdomen and thorax).

The precise epidemiology of lymphedema is unknown because it is not a regularly registered disease. The number of lymphedema patients worldwide is estimated to be approximately 140–250 million [2].

Lymphedema is divided into primary lymphedema and secondary lymphedema. Primary lymphedema is inherited as it is caused by gene mutations and starts at adolescence, or sometimes earlier, and affects mainly the foot and calf. Some congenital types are already visible at birth (Milroy's disease). Secondary lymphedema is acquired as a result of an injury or infection of the lymph system. In the Western world, 80% of lymphedema cases are acquired after cancer treatment as a result of the removal of lymph nodes (mostly in the axilla or groin) and/ or radiotherapy [3]. In the developing world, parasites (filariasis) are responsible for millions of patients with lymphedema [4].

Lymphedema after cancer treatment

Williams et al. [3] stated that the incidence of lymphedema is 30% after axillary lymph node dissection. Norman et al. [5] reported a cumulative incidence of subject-reported lymphedema 5 years after axillary lymph node dissection in 50% of patients younger than 50 years, in 40% of patients between 50 and 70 years and in 26% of patients older than 80 years. Eighty percent of lymphedema patients developed the condition in the first 2 years after the operation. Lee et al. [6] described factors, such as higher age, post-operative wound infections (or later infections on the operated side) and obesity as cumulative risk factors for lymphedema after axillary node dissection. Obesity as a risk factor for developing lymphedema was described by several authors [7–14].

Cormier et al. [15] described an overall incidence of lymphedema of 15.5% after cancer treatment beyond breast cancer, 16% after cancer treatment for melanoma, 20% after cancer treatment for gynaecological cancer, 10% after cancer treatment for genitourinary cancer, 4% after cancer treatment for head/neck cancer and 30% after cancer treatment for sarcoma. Increased lymphedema risk was described for pelvic dissections (22%) and radiation therapy (31%). Incidence reporting has increased using objective measurement methods and longer follow-up.

Lymphatic filariasis

According to a WHO report, 120 million people in 72 countries were infected with lymphatic filariasis in 2010, of which 15 million people suffered from the stigmatizing and disabling manifestations of lymphedema (elephantiasis) [4].

Symptoms of lymphedema

Initially lymphedema is non-pitting and the affected skin feels soft. In later stages, the skin may feel hard and thickened as a result of fat deposits and fibrosis in the swollen area. In severe, long-lasting lymphedema, skin changes are visible as keratosis. Lymphedema results in many symptoms such as decreased mobility of the limb [16], slower wound healing, increased risk for infection, feelings of heaviness and tightness, discomfort and tight fitting of jewelry or clothes on the affected body parts [5,17,18]. It leads to impairments such as limited walking distance in leg lymphedema and to limitations in personal care, domestic life, occupation and socialization. Lymphedema is a chronic condition and its limitation on physical functioning can result in distress [7] and loss of quality of life. Generally, the activity level of lymphedema patients is lower compared with healthy persons of the same age. Altered body shape and the necessity of daily use of special garments or bandaging as a standard treatment for stabilized lymphedema make acceptance of lymphedema difficult for the patient and for his family.

Treatment and health classifications

Lymphedema requires lifelong skin care and precautions to minimize (recurrent) swelling. Patients are advised to practice certain regimens, such as daily bandaging or the use of special garments, skin care and caution with open wounds.

Today medical treatment of patients with lymphedema is primarily aimed at reducing volume, but treatment should most likely be aimed at other problems as well. An important basis for the optimal treatment and management of lymphedema is an in-depth understanding, systematic consideration and sound measurement of its consequences at different levels of a person’s functioning. To achieve this goal, it is necessary to develop a comprehensive framework and classification which can serve as a universal language, understood by health professionals, researchers, policy makers, patients and patient organizations. The International Classification of Functioning, Disability and Health (ICF) [19] is a widely used classification system in which terms and classes are designed to describe functioning, disability and health in people with health disorders. The terms and classes in the ICF consist of both descriptions and codes. Descriptions are culturally tuned and provided in many languages. The pertinent codes (letters and numbers) are identical world- wide. The ICF terms can be understood by health professionals, researchers, policy makers, patients and patient organizations. The International Society for Lymphology (ISL) mentioned the importance of the use of the ICF in their latest revision of lymphedema consensus document (2009) [20].

The ICF contains two parts. Part 1 describes Functioning and Disability and includes three components: Body Functions, Body Structures and Activities and Participation. Part 2 addresses Contextual Factors and includes two components: Environmental Factors and Personal Factors. However, ‘‘Personal Factors’’ is not elaborated as a classification in the ICF because of the large social and cultural variety associated with these factors [19].

With the ICF codes, linkage among data across conditions or interventions is possible as a necessary step in the development of efficient, transparent and cost-effective health care [21]. Although the ICF classification system with more than 1400 categories can serve as a reference, the ICF is not easily applicable in clinical practice. Therefore, tools such as ICF Core Sets (health-specific selections of ICF categories) [22] are helpful to make the ICF more appealing for the implementation in medical/allied health care [23]. Health-specific Core Sets choose the appropriate terms and classes to describe the functioning of the patient and make the formulation of treatment goals easier, faster and more transparent. Therefore, these data are also appropriate for use in electronic health records. Core Sets are available to all types of health care providers, researchers, health insurance companies and policy makers. Once a Core Set is developed, one can select measurement instruments for each code (or combination of codes), thereby creating a basic set of measurement instruments as well. To date, ICF Core Sets have been developed for various burdensome chronic conditions [21,24,25], such as stroke, multiple sclerosis and breast cancer. Core Sets have also been developed for an acute hospital and early post-acute rehabilitation facilities [26–28]. ICF Core Sets for lymphedema are not yet developed.

Stucki et al. [22] have composed a standard method to develop ICF Core Sets. The development of ICF Core Sets for lymphedema will follow this method. The method consists of the following component studies: (a) a literature review: a search for meaningful concepts (i.e. a unit of text that conveys a single theme [29]) in the literature regarding the outcomes used in clinical trials, qualitative research and selected observational studies, representing the researchers' perspective; (b) focus groups of patients with lymphedema (patients' perspective); (c) an expert survey on the outcomes gathered in studies (a) and (b) using the Delphi technique (perspectives of various relevant groups); and (d) an empirical, cross-sectional study to gather concepts that are named in clinical situations (health care providers' perspectives). The final composition of the ICF Core Sets for lymphedema will take place at an ICF Core Set Consensus Conference (study e), which will integrate evidence from the previous studies (a–d). The development of ICF Core Sets on lymphedema (a–e) will be carried out in 3 years. This article represents the researchers' view (study a). The literature will be systematically searched for meaningful concepts, especially meaningful concepts within lymphedema-specific questionnaires. An analysis of meaningful concepts within the text of published studies (also part of study a) will be published separately.

Objectives

The objectives of the literature review are as follows: (1) to identify questionnaires in studies focusing on patients with lymphedema and (2) to link meaningful concepts within these questionnaires to the ICF.

Methods

Search strategy

Electronic searches were carried out in CINAHL, EMBASE, Medline, CENtral and Pedro databases from 2005 to 2010 using the search terms lymphedema, elephantiasis, lymphatic edema, lymphostatic edema and Milroy's disease (the detailed search strategy is available upon request). The target population in the included studies was patients who were 18 years or older and had a clinical diagnosis of lymphedema. Studies with one of the following designs were included: (1) randomized clinical trials, (2) controlled clinical trials, (3) observational studies, i.e. cohort studies and cross-sectional studies, (4) guidelines, and (5) 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. The exclusion criteria for the studies were as follows: (1) studies with exclusively laboratory parameters, (2) genetic studies, (3) animal experiments, (4) letters, (5) comments, (6) editorials, (7) systematic reviews, (8) case reports, and (9) meta- analyses.

Data collection procedure

In the first step, the abstracts of the retrieved studies were checked for the inclusion criteria. For the selected abstracts, the whole publication was studied and a decision was made as to which of the publications should be included in the study. In the second step, measurement methods, including questionnaires, were identified. In the third step, meaningful concepts were extracted from the selected questionnaires. The identification of meaningful concepts was performed separately by two researchers (PV and JH) who are trained in this type of identification before the research, using the first specific rule for health-status measures of Cieza et al. [30] (''Before starting the process of linking health-status measures to the ICF categories, identify all meaningful concepts within each item of the health status measure under consideration.''). The concepts should be focused only on patients themselves and their surroundings. Examples of therapies mentioned were not included. In the fourth step, the meaningful concepts were linked to ICF categories using rule 2 (''Each meaningful concept is linked to the most precise ICF category'') of Cieza et al. [30,31].

Coding principles

Coding was performed until the fourth level of the ICF. Each defined class/category/code was used only once for each questionnaire (e.g. pain has been marked as a meaningful concept; when pain in the foot and pain in the leg were described in the same questionnaire, only one single ICF code (b28015: pain in lower limb) was registered). If a meaningful concept pertained to a personal factor, such as ''age'', ''sex'' and ''education'', the concept was coded as ''pf'' because various personal factors are only named as examples but are not yet properly classified within the ICF. If a concept described an aspect of functioning and health that is not covered by the ICF, such as ''taking up time'' and ''suffered'', the code ''nc'' (not covered) was attributed. If a concept pertained to health conditions, such as ''lymphedema'' and ''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 given.

Inter-coder agreement

The following steps of the study were performed by two researchers (PV and JH) independently: (1) eligibility check of the abstracts, (2) extraction of the meaningful concepts of all selected questionnaires, (3) specification of the concepts and (4) linking of the concepts to the ICF. After steps 1 and 2, both researchers compared their results. Initial disagreement was resolved after the discussion. If any disagreement remained, a third person (YH or DVR) was consulted. After steps 2, 3 and 4 (taken together), the inter-coder agreement was quantified by calculating the kappa with its 95% confidence interval (CI). Calculations were performed with the statistical software package SPSS, version 19.0 (IBM SPSS, Armonk, NY). This was performed for all of the questionnaires. After the kappa calculations, disagreement (first on extraction/specification of concepts, second on proper ICF code) was also resolved together or by a third person. These steps together provided the final codes of the meaningful concepts that are presented in this study.

Data analysis

Frequencies of the linked ICF categories are reported when measured in more than two questionnaires. If a concept was linked to a third or fourth level ICF category, the corresponding second- level category is reported in this article to avoid long lists of ICF categories. This is appropriate because the lower level categories share the attributes of the higher level categories [19].

Results

From the 2381 abstracts retrieved, 149 studies were selected based on the inclusion and exclusion criteria. Reading of the publications occurred in alphabetical order based on the name of the first author. After the full-text articles were read, an additional 13 studies were excluded because they did not fit into the inclusion criteria (e.g. being comments, case reports or not primarily focusing on lymphedema), which was not evident from the abstracts. In the remaining 136 studies, 88 (65%) of the study population consisted of patients with lymphedema in the upper limb, 34 (25%) of the study population consisted of patients with lymphedema in the lower limb, 5 (3%) of the study population consisted of patients with midline lymphedema and 9 (7%) of the study population consisted of patients who had lymphedema in combinations of locations. The included studies consisted of 46 intervention studies (40 for upper limb; 4 for lower limb; 1 for midline; 1 for combinations), 67 observational studies (43 for upper limb; 16 for lower limb; 4 for midline; 4 for combinations), 19 qualitative studies (4 for upper limb; 12 for lower limb; 3 for combinations) and there were 4 clinimetric studies (1 for upper limb; 2 for lower limb; 1 for combinations). The measurement methods described and used in the 136

studies are listed in Table 1. The next step was to filter out the questionnaires that were used (Table 2). Finally, only the lymphedema-specific questionnaires were selected [5,7,17,32–40] (Table 3).
In total, 12 lymphedema-specific questionnaires were found (10 for the upper limb, 1 for the lower limb and 1 for lymphedema in general). From these questionnaires, all meaningful concepts were determined and linked to the ICF.

Table 1. Measurement methods used in the 136 studies.

Questionnaire	114
Circumferential measurement	49
BMI	28
Water displacement device	23
VAS	17
Bio impedance	12
Perometer	9
Goniometry	9
Lymphoscintigraphy	7
Palpation	4
Sonography	4
Tonometry	3
Blood samples	3
Grip strength	3
Diary	2
Water composition	2
Duplex Ultrasound	2
3D Measuring	1
MRC	1
Optoelectronic system	1
Lymphangiography	1
Pletysmography	1
Functional Tests	1
Electrical Impedance Spectrography	1
Magnetic Resonance Imaging	1
Computer Tomography	1
1 RM test	1

Table 2. Questionnaires used in the 136 studies.

Name of questionnaire	**Number of studies**
Short Form-36	14
Functional Assessment of Cancer Therapy Breast plus 4 subscale	7
Lymphedema and Breast Cancer Questionnaire	7
European Organization for Research and Treatment of Cancer –Quality of Life Questionnaire –Cancer 30	5
Dermatology Life Quality Index	4
Disabilities of Arm Shoulder and Hand	3
European Organization for Research and Treatment of Cancer –Quality of Life Questionnaire –Breast 23	3
Nottingham Health Profile	3
Upper Limb Lymphedema 27	3
Borg Rate of Perceived Exertion Scale	2
Center for Epidemiologic Studies of Depression Scale	2
General Health Questionnaire -30	2
McGill Pain Questionnaire	2
Short Form -12	2
World Health Organization Disability Assessment Schedule 2	2
Selfreport (Norman)	2
Various	51

Table 3. Lymphedema-specific questionnaires used in the 136 studies (5, 7, 17, 32-40).

Name	**Author**	**Location (A = arm, B = leg, C = combinations)**	**Number of studies**
Lymphedema and Breast Cancer Questionnaire	Armer et al (32)	A	7
Upper Limb Lymphedema 27	Launois et al (37)	A	3
Arm Symptom Distress Scale	Mak et al (36)	A	1
Edema Scale	Belcaro et al (39)	C	1
Gynaecologic Cancer Lymphedema Questionnaire	Carter et al (33)	A	1
Lymphedema and Pain Questionnaire	Oliveri et al (35)	A	1
Lymphedema Risk-Reduction Behaviors	Fu et al (17)	A	1
Lymphedema Signs and Symptoms Questionnaire	Dawes et al (34)	A	1
Measure of Arm Symptom Survey-Lymphedema	Swenson et al (7)	A	1
Memorial Symptom Assessment Scale (adapted)	Norman et al (5)	A	1
Questionnaire for Lymphedema-Related Subjective Symptoms	Szolnoky et al (38)	A	1
Supportive Care Needs Survey Short Form (Lymphedema supplement)	Beesley et al (40)	B	1

Inter-coder reliability

The inter-coder agreement for the determination of meaningful concepts was 80% (kappa 0.55; 95% CI 0.48–0.63). For the linking with the ICF, the inter-coder agreement was 63% (kappa 0.28; 95% CI 0.18–0.37). Disagreement originated most often in the same direction by the same differences in coding (e.g. when both researchers consistently coded the same meaningful concept in different ways. For instance ‘‘Do you have limited movement of your foot’’ was coded ‘‘b7100 Mobility of a single joint’’ by one researcher and ‘‘b7101 Mobility of several joints’’ by the other continuously). In only three cases, no consensus was reached by the two researchers about linking a meaningful concept to the ICF. These could be resolved by the third researcher.

Identified concepts and ICF codes

A total of 301 concepts were extracted from the 12 questionnaires. Out of these, 4 concepts in a total of 3 questionnaires were considered as ‘‘not covered’’ (‘‘nc’’) by the ICF, 13 concepts in a total of 11 questionnaires as ‘‘health condition’’ (‘‘hc’’), 1 as ‘‘quality of life’’ (‘‘nd-qol’’) and 12 concepts in a total of 4 questionnaires as ‘‘personal factor’’ (‘‘pf’’). Examples of Personal Factors found were ‘‘Date of birth’’, ‘‘Age’’, ‘‘Lifestyle’’ and ‘‘Race’’. The other 271 concepts (representing 90%) could be linked to a total of 117 different ICF categories. In all, 1 concept was linked at the level of an ICF chapter only, 6 concepts were linked to the first level of the ICF categories, 116 concepts were linked to the second level and 148 concepts to the third level. Not a single concept was linked to a fourth-level category. Later, the third- level categories were merged into the corresponding second-level categories for purposes of this article. Tables 4–7 list the 45 second-level ICF categories representing the concepts contained in two or more questionnaires. Of these 45 categories, 13 categories belong to the component ‘‘Body Functions’’, 6 to the component ‘‘Body Structures’’, 16 to the component ‘‘Activities and Participation’’ and 10 to the component ‘‘Environmental Factors’’. The second-level categories of the ‘‘Body Functions’’ component that were found most frequently were ‘‘b435, Immunological system functions’’ (10 questionnaires), ‘‘b280, Sensation of pain’’ (9 questionnaires) and ‘‘b265, Touch Functions’’ (8 questionnaires) (Table 4). Of the ‘‘Body Structures’’ component, the most frequently found categories were ‘‘s730, Structure of upper extremity’’ (10 questionnaires) and ‘‘s630, Structure of reproductive system’’ (6 questionnaires) (Table 5). The most frequently found categories in ‘‘Activities and Participation’’ were ‘‘d570, Looking after one’s health’’ (9 questionnaires), ‘‘d540, Dressing’’ (7 questionnaires), ‘‘d445, Hand and arm use’’ (6 questionnaires) and ‘‘d850, Remunerative employment’’ (6 questionnaires as well) (Table 6). The most frequently found ‘‘Environmental Factors’’ were ‘‘e580, Health services, systems and policies’’ (7 questionnaires) and ‘‘e115, Products and technology for personal use in daily living’’ (6 questionnaires) (Table 7).

Table 4. Frequency of second level categories of the ICF linked to concepts contained in the 12 questionnaires: component Body Functions (A = code found in upper limb questionnaire, B = code found in lower limb questionnaire, C = code found in lymphedema general questionnaire).

ICF code	**Description**	**Number of questionnaires**
Chapter 1: mental functions		
b126 (A)	Temperament and personality functions	3
b134 (A,B)	Sleep functions	5
b152 (A,B)	Emotional functions	3
b180 (A,C)	Experience of self and time functions	4
Chapter 2: sensory functions and pain		
b265 (A,B)	Touch functions	8
b280 (A,B)	Sensation of pain	9
Chapter 4: functions of the cardiovascular, haematological, immunological and respiratory systems		
b435 (A,B,C)	Immunological system functions	10
Chapter 5: Functions of the digestive, metabolic and endocrine systems		
b550 (A,B)	Thermoregulatory functions	2
Chapter 7: neuromusculoskeletal and movement-related functions		
b710 (A,B)	Mobility of joint functions	4
b730 ((A,B)	Muscle power functions	2
Chapter 8: functions of the skin and related structures		
b810 (A,B,C)	Protective functions of the skin	5
b820 (A,B)	Repair functions of the skin	2
b840 (A)	Sensation related to the skin	3

Table 5. Frequency of second-level categories of the ICF linked to concepts contained in the 12 questionnaires: component Body Structures (A = code found in upper limb questionnaire, B = code found in lower limb questionnaire, C = code found in lymphedema general questionnaire).

ICF code	**Description**	**Number of questionnaires**
Chapter 6: Structures related to the genitourinary and reproductive systems		
s630 (A,B)	Structure of reproductive system	6
Chapter 7: Structures related to movement		
s720 (A)	Structure of shoulder region	2
s730 (A,C)	Structure of upper extremity	10
s750 (B,C)	Structure of lower extremity	3
s760 (A)	Structure of trunk	3
Chapter 8: skin and related structures		
s810 (A)	Structure of areas of skin	3

Table 6. Frequency of second-level categories of the ICF linked to concepts contained in the 12 questionnaires: component Activities and Participation (A = code found in upper limb questionnaire, B = code found in lower limb questionnaire, C = code found in lymphedema general questionnaire).

ICF code	**Description**	**Number of questionnaires**
Chapter 2: General tasks and demands		
d230 (A,B)	Carrying out daily routine	4
Chapter 3: Communication		
d325 (A,B)	Communicating with –receiving- written messages	2
d330 (A)	Speaking	2
Chapter 4: Mobility		
d415 (A)	Maintaining a body position	3
d430 (A)	Lifting and carrying objects	2
d440 (A,B)	Fine hand use	3
d445 (A)	Hand and arm use	6
d450 (A)	Walking	2
d470 (A)	Using transportation	4
Chapter 5: Self-care		
d520 (A)	Caring for body parts	2
d540 (A)	Dressing	7
d570 (A,B,C)	Looking after one’s health	9
Chapter 6: Domestic life		
d640 (A,B)	Doing housework	4
d650 (A)	Caring for household objects	2
Chapter 8: Major life areas		
d850 (A,B)	Remunerative employment	6
Chapter 9: Community, social and civic life		
d920 (A)	Recreation and leisure	5

Table 7. Frequency of second-level categories of the ICF linked to concepts contained in the 12 questionnaires: component Environmental Factors(A = code found in upper limb questionnaire, B = code found in lower limb questionnaire, C = code found in lymphedema general questionnaire).

ICF code	**Description**	**Number of questionnaires**
Chapter 1:products and technology		
e110 (A)	Products and substances for personal consumption	4
e115 (A)	Products and technology for personal use in daily living	6
e125 (A,B)	Products and technology for communication	2
e135 (A,B)	Products and technology for employment	4
Chapter 2: Natural environment and human-made changes to environment		
e225 (A)	Climate	2
e245 (A,C)	Time-related changes	3
Chapter 3: Support and relationships		
e310 (A,B)	Immediate family	2
e325 (A,B)	Acquaintances, peers colleagues, neighbours and community members	3
e355 (A)	Health professionals	5
Chapter 5: services, systems and policies		
e580 (A,B)	Health services, systems and policies	7

Discussion

Using the ICF as a reference, it was possible to identify meaningful concepts in questionnaires focusing on patients with lymphedema. Many of these concepts could be linked to the ICF. The identification of the ICF categories contributed, from the researcher's point of view, to the development of ICF Core Sets for lymphedema. This research covered a period of 5 years. Most literature reviews concern the evidence for certain treatments or diagnostic methods, often using a period of more than 5 years. In this particular case, the review aims at classifying meaningful concepts used in scientific literature concerning lymphedema. A 5-year-period is considered to be sufficient for this particular goal [41,42]. Coincidentally, between 2005 and 2010 literature about lymphedema focused more on other items than only swelling and there was paid more attention to lymphedema other than caused by breast cancer than in the years before. This enlarged the variety of meaningful concepts in this research. Compared to previously published systematic reviews for ICF Core Set development [43], this study not only included RCTs but also studies with other designs, such as observational studies and qualitative studies. The reason for including observational and qualitative studies was that these type of studies might be more appropriate to disclose the everyday problems related to lymphedema. Expanding the scope of studies included offering a chance to reveal more meaningful concepts.

The ICF codes were summarized into two-level codes to ensure readability and practical use. This way of presenting was performed previously in ICF Core Set development [41], although this procedure seems to be contrary to Coding Rule 2, as described by Cieza et al. [30], which states that the most precise ICF category should be identified. More detailed data will be used for the next studies on developing the Core Sets (studies b–e).

The somewhat low kappa of 63% of the inter-coder reliability in comparison to other studies (e.g. Geyh et al. [44]; 82%) can be partly explained by the fact that in our study kappa values were calculated after a fully independent formulation of meaningful concepts by two persons and the linking of the concepts to ICF codes. Inter-individual differences can result from differences in the identification of meaningful concepts as well as in differences in the selection of ICF codes. In the study of Geyh et al. [44] the kappa values were calculated after consensus was reached for the meaningful concepts. Furthermore, it was not simple or straight-forward to link meaningful concepts derived from the questionnaires to the appropriate ICF category because concept wordings are often not very precise as to their exact meaning and are not equal to the ICF terms. Thus, the coding frequently requires discussion. Recent linkage exercises, however, have demonstrated that it is possible to examine and compare the content of measures based on the ICF framework and predefined linking rules [30,31,45]. A third explanation might be that both reviewers had good theoretical knowledge of the ICF, but less experience in the linking process.

In the lymphedema-specific questionnaires, the most frequently measured categories were ‘‘s730, Structure of upper extremity’’ (10 questionnaires), ‘‘b435, Immunological system functions’’ (10), ‘‘d570, Looking after one’s health’’ (9), ‘‘b280, Sensation of pain’’ (9), ‘‘b265, Touch functions’’ (8), ‘‘d540, Dressing’’ (7) and ‘‘e580, Health services, systems and policies’’ (7). Because most of the measurement instruments concerned about upper extremity lymphedema, it is clear that ‘‘s730, Structure of upper extremity’’ was found most frequently. Lymphedema is a chronic health condition that presents with swollen extremities. This explains the high frequency of ‘‘b435, Immunological system functions’’, implying the swelling of arms and legs. Self-management of lymphedema seems to be important, considering the high frequencies of ‘‘d570, Looking after one’s health’’ and ‘‘d540, Dressing’’. Many questionnaires contain items related to therapy and caregivers, which can be deduced from the high frequency of the category ‘‘e580, Health services, systems and policies’’, indicating their importance for patients with lymphedema.

Concerning the Body Functions category, much lymphedema research still focuses on the symptoms, pain and swelling, and little attention is paid to other problems, such as skin problems (Chapter 8 – Functions of the skin and related structures). More research from the patient’s, clinician’s and expert’s point of view will show whether skin problems really are of less importance.

Although lymphatic filariasis (a mosquito-transmitted chronic, incurable disease often found in Third World countries) damages mostly the lymphatic system of the lower extremity [46–51], most of the questionnaires were found to be concerned about lymphedema of the upper extremity. This is the reason why, for example, ‘‘d450, Walking’’ has a relative low frequency. There is a need in this field of research to provide more insight into these limitations.

The other measurement methods (not the questionnaires) that were identified in the studies were almost always related to the classification ‘‘Body Functions’’ of the ICF. These methods are concentrated on clinical symptoms, such as swelling and pain. However, from a more holistic point of view (e.g. using the perspective of the ICF), there are other fields important to describe the patient’s functioning as well.

Considering the questionnaires, it is striking that there are 51 different questionnaires in the included studies, so it seems that every researcher used his/her own questionnaire. To achieve more uniformity in this perspective, the ICF, and more specifically the Core Sets for lymphedema, can work as a framework in the future.

Most of the included meaningful concepts could be linked to the ICF (90%), but a small proportion could not. Of these, ‘‘Health Conditions’’ can be classified with the ICD-10. Unfortunately, the ‘‘Personal Factors’’ component, as part of the ICF, is still under development.

The selection of studies might have been biased by including only studies and questionnaires published in English. The search concerned publication years 2005–2010, so it is possible that more recent studies would have revealed different results.

Because the large majority of meaningful concepts found in the questionnaires concerning lymphedema could be linked to the ICF, this type of classification appears workable. Recent publications [52,53] stress the importance of implementing the ICF.

The ICF provides a valuable reference to identify and quantify the meaningful concepts focusing on individuals with lymphedema. The findings of this literature review indicate a need to define and to agree on ‘‘what should be measured’’ in lymphedema care. This is one of the goals of the ICF Core Set development for lymphedema. ICF Core Sets should allow for a comparable and comprehensive description of patient populations, their functioning and health across studies and interventions.

Declaration of interest

The authors report no conflict of interest or financial ties in this research.

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Chapter 4

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

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Abstract

Background: This article is the second arising from a literature research study that was conducted as part of the development process of ICF Core Sets (selections from the ICF for a specific group of clients) for lymphoedema. In the first article, meaningful concepts in lymphoedema-specific questionnaires were explored.

Objectives: To compare and analyze categories of the WHO's International Classification of Functioning, Disability and Health (ICF) linked to meaningful concepts within studies focusing on lymphoedema with ICF categories linked to meaningful 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 meaningful concepts derived from lymphoedema-specific questionnaires.

Results: 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 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.

Introduction

In lymphoedema, there is an abnormal accumulation of fluid and protein as a result of an imbalance between the flow and discharge of fluid (1). Lymphoedema can occur in the upper and lower limbs as well as in the midline (head, neck and thorax). It 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; in this condition, the lymph vessels and/or nodes have been removed or infected. Examples of secondary lymphoedema are upper limb lymphoedema after axillary lymph node dissection, lymphoedema of the lower limb after a gynecological operation with inguinal lymph node dissection, and infection with lymphatic filariasis, a mosquito-transmitted, chronic, incurable disease often found in third-world countries. Patients receiving post-surgical radiotherapy and/or chemotherapy have an increased chance of acquiring lymphoedema (2). The epidemiology of lymphoedema is not precisely known because it is not a regular registered disease. According to a WHO report,(2010) approximately 120 million people in 72 countries were infected with lymphatic filariasis, of whom 15 million suffered from lymphoedema (3). Worldwide, the number of persons with lymphoedema is estimated to be 140-250 million (4).

Symptoms of lymphedema

Lymphoedema results in decreased mobility of the limb (5), slower wound healing, increased risk of infection, feelings of heaviness and tightness, discomfort and tight fitting of jewelry or clothing in the affected body parts (6-8). It leads to limitations in activities, such as a reduced ability to walk, and limitations in personal care, domestic life, occupation and socialization. Lymphoedema is a chronic condition, and its influence on physical functioning can result in distress (9) and loss of quality of life. Altered body shape and the necessity for daily use of specialized garments or bandaging as a standard treatment for stabilized lymphoedema make acceptance of the condition difficult for the patient as well as for his or her family and environment. In addition, lymphoedema requires lifelong self-care: patients must follow prescribed regimens, such as daily bandaging or the use of special garments; utilize 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) (10) is accepted by the World Health Assembly as a universal framework for classifying and describing functioning in people with specific health conditions. The International Society for Lymphology (ISL) emphasized the importance of the ICF in the latest revision of its lymphoedema consensus document in 2013 (11). The ICF organizes information in two parts. Part 1 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 the components '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 (10). The ICF makes it possible to link data across conditions or interventions with the goal of achieving efficient, transparent, and cost-effective health care (12). This article is the second arising from a literature research study that was conducted as part of the development process of ICF Core Sets (selections from the ICF for a specific group of clients) for lymphoedema. In the first article, meaningful concepts in lymphoedema-specific questionnaires were explored (13). One of the conclusions indicated a need to define and agree upon '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. Afterwards, these ICF categories were compared with the ICF categories linked to the meaningful concepts found in the questionnaires (13) to determine whether researchers indeed explored these important items.

Objectives

The objectives of the literature review were 1) to identify literature describing (problems in) functioning of individuals with lymphoedema; 2) to identify meaningful concepts within the studies; 3) to link these meaningful concepts to the ICF; 4) to quantify these ICF-linked concepts (to count the frequency of occurrence of codes/classes); and 5) to compare these codes with those that were found in a previous study about lymphoedema-specific questionnaires.

Methods

Search strategy

Electronic searches were carried out in the CINAHL, EMBASE, Medline, CENtral and Pedro databases from 2005 to 2010 using the search terms ‘lymphoedema’, ‘elephantiasis’, ‘lymphatic edema’, ‘lymphostatic edema’ and ‘Milroy’s disease’. The detailed search strategy can be obtained from the first author upon request. The target population of the included studies consisted of patients over 18 years old with a clinical diagnosis of lymphoedema. Studies with the following designs were included: 1) randomized clinical trials; 2) controlled clinical trials; 3) observational studies, i.e., cohort studies and cross-sectional studies; 4) guidelines; and 5) 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. The exclusion criteria for the studies were as follows: 1) studies with exclusively laboratory parameters; 2) genetic studies; 3) animal experiments; 4) letters; 5) comments; 6) editorials; 7) systematic reviews; 8) case reports; and 9) meta-analyses.

Data collection procedure

In a *first step*, the studies were selected based on title. The abstracts of the retrieved studies were checked for inclusion. For the selected abstracts, the entire publication was studied and a decision was made as to whether the publication should be included in the study.

In a *second step*, 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 studies are described in the previous article (13). After data extraction, two researchers (PV, JH) compared their results.

In a *third step*, the retrieved meaningful concepts were linked to ICF categories using standardized linkage rules (14, 15). Each relevant concept was linked by two researchers (PV, JH). Coding took place until the 4th 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 person (YH or DvR) was consulted.

If a meaningful concept pertained to a personal factor such as ‘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, such as ‘taking up time’ or ‘suffering’, the code ‘nc’ (not covered) was attributed. If a concept pertained to health conditions such as ‘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, the meaningful concept was assigned ‘nd’ (not definable) (e.g., ‘side effects’ of therapy). Meaningful concepts referring to health in general were assigned ‘nd-gh’ (not definable-general health).

Inter-coder agreement

The following steps of the study were performed by two researchers (PV and JH) independently of each other: 1) eligibility check of the abstracts; 2) data extraction of the included full-text articles and specification of the concepts; and 3) linking of the concepts to the ICF. After each step, the researchers compared their results. Initial disagreement was solved after discussion. If disagreement remained, a third person (YH or DVR) was consulted.

Data analysis

The frequencies of the linked ICF categories were reported. All of the resulting ICF categories that referred to concepts measured in more than 5% of the studies were reported (16). 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 (10). Finally, the percentage frequencies of the codes of the meaningful concepts in the studies and the codes of the meaningful concepts in the questionnaires were compared to each other.

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 concerning patients with 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 of the location was possible in the studies that involved combined lymphoedema locations, these studies were excluded. In the end, the included research studies included 73 publications (34 on upper limb lymphoedema, 34 on lower limb lymphoedema, and five on midline lymphoedema). Twenty of the included studies were intervention studies (n = 15 for upper limb, n = 4 for lower limb, and n = 1 for midline lymphoedema), 37 were observational studies (n = 17 for upper limb, n = 16 for lower limb, and n = 4 for midline lymphoedema), 13 were qualitative studies (n = 1 for upper limb and n = 12 for lower limb lymphoedema), and three were clinimetric studies (n = 1 for upper limb and n = 2 for lower limb lymphoedema). The studies of upper limb lymphoedema were all conducted in Western countries (i.e., Europe, the United States of America and Australia), whereas 22 of the 34 studies of lower limb lymphoedema were conducted in Africa, Asia or 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. In all, three concepts (0.2%) were linked to an ICF component; 64 (2.8%) were linked to the first level of the ICF, 935 (41.3%) were linked to second-level ICF categories, and 1265 (55.7%) were linked to third-level ICF categories. 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 through 4 list the 93 second-level ICF categories representing the meaningful concepts that were classified in four or more 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%) belong to the component Body Structures, 27 (29,1%) belong to the component Activities and Participation and 24 (25.8%) belong to the component Environmental Factors. Tables 1 through 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 previous study (13). For comparison, the results of the questionnaires for the upper extremity (nine questionnaires) and for the lower extremity (two questionnaires) 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 the studies, 89% of the questionnaires), ‘b280, Sensation of pain’ (56% of the studies, 78% of the questionnaires), and ‘b265, Touch functions’ (32% of the studies, 67% of the questionnaires) (see Table 1). For the lower limb, the most frequently found categories were ‘b280, Sensation of pain’ (62% of the studies, 100% of the questionnaires), ‘b435, Immunological system functions’ (82% of the studies, 50% of the questionnaires) , and ‘b265, Touch functions’ (32% of the studies, 100% of the questionnaires). In studies of midline lymphoedema, the categories ‘b640, Sexual functions’(80% of the studies) and ‘b435, Immunological system functions’ (60% of the studies) were most often found. A similar pattern is apparent in the questionnaires, although the class ‘b455, Exercise tolerance functions’ is not used to code MCs in the questionnaires for either the upper or the 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; these are ‘b134, Sleep functions’, ‘b550, Thermoregulatory functions’, and ‘b840, Sensation related to the skin’.

Table 1 Frequency of second-level categories of the 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 the studies are shown.
( ) = the corresponding percentage of studies or questionnaires.
Yellow = categories that have a mutual representation in both the studies and the questionnaires.
Red = categories that are present in the questionnaires but have no representation in the studies.
Blue = categories that are present in the studies but have no representation in the questionnaires.

ICF code	Description	Upper Limb		Lower Limb		
		Number (%) of studies	Number (%) of question-naires	Number (%) of studies	Number (%) of question-naires	Number (%) of studies Midline
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)	-	-	-

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 the studies, 100% of the questionnaires), ‘s420, Structure of immune system’ (68% of the studies), and ‘s810, Structure of areas of skin’ (38% of the studies, 33% of the questionnaires) (see Table 2). For the lower limb, the most frequently found categories were ‘s750, Structure of lower extremity’(94% of the studies, 100% of the questionnaires), ‘s810, Structure of reproductive system’ (26% of the studies, 50% of the questionnaires), and ‘s420, Structure of immune system’ (35% of the studies). In the midline studies, ‘s630, Structure of reproductive system’ (80% of the studies), ‘s750, Structure of lower extremity’ (60% of the studies), and ‘s810, Structure of areas of skin’ (60% of the 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 vice versa that ‘750, Structure of lower extremity’ is represented in studies of the upper limb.

Table 2. Frequency of second-level categories of the 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 4 of the studies are shown.
( ) = the corresponding percentage of studies or questionnaires.
Yellow = categories that have a mutual representation in both the studies and the questionnaires.
Blue = categories that are present in the studies but have no representation in the questionnaires.

ICF code	Description	Upper limb		Lower limb		
		Number (%) of studies	Number (%) of question-naires	Number (%) of studies	Number (%) of question-naires	Number (%) of studies Midline
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)

Activities and participation

In the component Activities and participation, ‘d570, Looking after one’s health’(71% of the studies, 78% of the questionnaires), ‘d540, Dressing’ (21% of the studies, 78% of the questionnaires), and ‘d455, Hand and arm use’ (21% of the studies, 56% of the questionnaires) were most often coded for the upper limb (see Table 3). For the lower limb, ‘d570, Looking after one’s health’ (65% of the studies, 50% of the questionnaires), ‘d850, Remunerative employment’ (32% of the studies, 50% of the questionnaires), and ‘d230, Carrying out daily routine’ (29% of the studies, 50% of the questionnaires) were the most frequently found categories. In the midline studies, ‘d570, Looking after one’s health’ (40% of the studies) was the most frequently found category. In contrary to the upper extremity, the ICF codes of the lower limb ‘d415, Maintaining a body position’ and ‘d540, Dressing’ are not used in the questionnaires. This is also the case for many of the codes in chapters 3, 4, 5, 7, 8, and 9. On the other hand, ‘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’ are not represented in the studies.

Table 3.Frequency of second-level categories of the 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 4 of the studies are presented.
( ) = the corresponding percentage of studies or questionnaires.
Yellow = categories that have a mutual representation in both the studies and the questionnaires.
Red = categories that are present in the questionnaires but have no representation in the studies.
Blue = categories that are present in the studies but have no representation in the questionnaires.

ICF code	Description	Upper limb		Lower limb		
		Number (%) of studies	Number (%) of question-naires	Number (%) of studies	Number (%) of question-naires	Number (%) of studies Midline
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)	-	-

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 the studies, 67% of the questionnaires), ‘e355, Health professionals’ (85% of the studies, 56% of the questionnaires), and ‘e115, Products and technology for personal use in daily life’ (68% of the studies, 67% of the questionnaires) (see Table 4). For the lower limb, these were ‘e580, Health services, systems and policies’ (100% of the studies, 50% of the questionnaires), ‘e325, Acquaintances, peers, colleagues, neighbors and community members’ (91% of the studies, 50% of the questionnaires), and ‘e355, Health professionals’ (79% of the studies). The categories ‘e580, Health services, systems and policies’ (100%), ‘e325, Acquaintances, peers, colleagues, neighbors and community members’ (80%), and ’e110, Products and substances for personal consumption’ (60%) 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, Health professionals’, there is no representation of ICF codes in the questionnaires relating to the lower limb. This also applies to several other categories in the chapters concerning Environmental Factors (see 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.

Table 4. Frequency of second-level categories of the 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 4 studies are presented.
( ) = the corresponding percentage of studies or questionnaires.
Yellow = categories that have a mutual representation in both the studies and the questionnaires.
Red = categories that are present in the questionnaires but have no representation in the studies.
Blue = categories that are present in the studies but have no representation in the questionnaires.

ICF code	Description	Upper limb		Lower limb		
		Number (%) of studies	Number (%) of question-naires	Number (%) of studies	Number (%) of question-naires	Number (%) of Studies Midline
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	Acquaintances, peers colleagues, neighbors 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, neighbors 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 organizational 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)

Discussion

Using the ICF as a reference, it was possible to identify and quantify meaningful concepts found in published studies that focus on individuals with lymphoedema. While a large number of these concepts (2269 out of 2488) could be linked to the ICF, a small portion (8,8%) could not. Of these concepts, Health Conditions can be classified with the ICD-10 (34%). Unfortunately, Personal Factors (3%), which is a component of the ICF, is still under development, although some provisional lists for this component have been published (17, 18). 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 (16), 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 separately (13).

The most frequently found categories were 'e580, Health Services, systems and policies' (73 studies), 'b435, Immunological system functions' (64 studies), 'e325, Acquaintances, peers, colleagues and community members' (59 studies), 'e355, Health professionals' (58 studies), and 'd570, Looking after one's health' (48 studies).

Lymphoedema is a chronic health condition that presents most often with swollen extremities. 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 area and breast, can also be a significant burden in midline lymphoedema. The categories 'e580, Health services, systems and policies' and 'e355, (support of and relationships with) Health professionals' originate from 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 researches 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 indicate whether this code is relevant.

The minor representation 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 have been biased because the study includes four studies about genital lymphoedema and one about breast lymphoedema. Because lymphoedema of the breast is seen more frequently than genital lymphoedema, there is a need for more studies involving this condition. Furthermore, no studies concerning 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 chapter 1: Mental Functions is less represented in studies of upper limb lymphoedema (4 categories) than in studies of lower limb lymphoedema (8 categories). The surveyed studies involving lower limb lymphoedema were mostly qualitative, whereas many of the studies involving upper limb lymphoedema were controlled studies in which symptoms such as 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. Mental Functions, as well as Exercise Tolerance Functions, are also much better represented in the studies than in the questionnaires. Although researchers often describe problems related to mental function and exercise tolerance, there seems to be no need to use these areas as outcome parameters in questionnaires; the gap concerning these areas can most likely be filled by using generic questionnaires or performance tests. The underrepresentation 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 (17-19).

Body structures

Within the Body Structures component, the 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 is 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 quite understandable because it is difficult to ask 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’ has a representation in studies of the lower limb and, conversely, ‘750, Structure of lower extremity’ has one in studies 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 an extensive and a maintenance phase. 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 the studies as well as in the 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 seems to be 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 (18-23). The category ‘d450, Walking’ has a rather low frequency (32% of the 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, Health professionals’ and ‘e580, Health services, systems and policies’) seem to be important because they are mentioned in all of the studies and half of the 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 (3 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 of finding ICF categories in literature. Besides examining questionnaires, the studies themselves were observed as well. 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 study originated in Western countries. The literature on lower limb lymphoedema came from both Western and developing countries. The fact that future Core Sets for lymphoedema will be designed primarily based on data produced in the Western countries has to be taken in 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 mentioned by Devoogdt (20)). Furthermore, 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 the inter-coder agreement, by calculating kappa, might have been useful. Unfortunately, the required data were not recorded. Linkage exercises have demonstrated, however, that it is possible to examine and compare the content of measures based on the ICF framework and predefined linking rules with good reliability (14, 15, 21).

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 (20, 22) stress these findings. Devoogdt et al (20) constructed a questionnaire (Lymph-ICF) based on data from interviews with patients with breast cancer-related lymphoedema. It consists of 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 this study.

Conclusion

The ICF provides a valuable reference for identifying meaningful concepts related to 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 (23) reported a similar conclusion concerning data obtained from studies of health-related quality of life for patients with upper limb lymphoedema.

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Conflict of interest

The authors report no conflict of interest or financial ties in this research.

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Chapter 5

Upper limb lymphedema 27 (ULL27): Dutch translation and validation of an illness-specific health-related quality of life questionnaire for patients with lymphedema

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Lymphology. 2008 (41): 131-138.

Abstract

The health-related quality of life questionnaire for lymphedema of the upper limb (ULL27) was translated into Dutch according to international guidelines and validated. Eighty-four patients with lymphedema that occurred after axillary surgery for breast cancer and subsequent radiotherapy, chemotherapy, or hormonal therapy, completed the translated version of the ULL27 and the RAND36. Severity of upper limb edema was measured by specialized physiotherapists. The internal consistency of the domains of the questionnaire was good as were the convergent validity and discriminant ability. Upper limb volume and the domains of the ULL27 were not correlated. The Dutch translation of the ULL27 questionnaire has good internal consistency and validity but further research is needed to determine its responsiveness.

Keywords: validation, lymphedema, health-related quality of life, ULL27, RAND36, breast cancer

Introduction

Breast cancer is diagnosed in 9,000 women a year in the Netherlands. The prevalence of upper limb lymphedema is 25% after modified radical mastectomy and 40% after adjuvant radiotherapy in the axillary region (1). Although upper limb lymphedema is sometimes treated surgically, it is usually treated conservatively, with manual lymphatic drainage (a mild form of massage), ambulant compression, exercise, and advice (2). The aim of such treatment is to reduce the edema and to improve the function of the upper limb (especially kinesiophobia, pain and limited shoulder function and usage). Functional limitations of the affected upper limb have a much greater impact on well-being than the increase in the size of the upper limb (3-5). Moreover, impairments such as swelling, numbness, and pain are not correlated with upper limb function (6). Both Bosompra et al (6) and Johansson et al (7) stressed the importance of assessing patients not only at the level of impairment but also for practical and psycho- social problems experienced by patients in daily life. Indeed, most investigators stress the importance of using health-related quality of life (HRQOL) questionnaires (8-14) to determine patient well-being. Several HRQOL questionnaires are being used to assess patients with cancer, and breast cancer in particular, such as the European Organization for Research and Treatment of Cancer Quality of Life Questionnaire (EORTC QLQ-C30) (15), the Functional Assessment of Cancer Therapy-Breast with 4 additional questions about the upper limb (FACT-B+4) (8), the Short Form 36 (SF36) (16) and the Nottingham Health Profile (17). However, none of these, mostly generic, questionnaires specifically focus on lymphedema and its consequences on HRQOL. The only lymphedema-specific HRQOL questionnaire currently available is the French language Upper Limb Lymphedema 27 (ULL27) (9). Cross-cultural validation of an existing specific HRQOL scale has the advantage of avoiding the initial stage of development of a new questionnaire. The aim of this study was to translate the ULL27 into Dutch and to determine its internal consistency and validity for assessing upper limb lymphedema in the Dutch population of patients with lymphedema.

Patients and methods

ULL27

The ULL27 is a self-report questionnaire encompassing 27 questions with answers given on a 5-point Likert scale, ranging from “never” to “always” and is considered to measure the effects of lymphedema on HRQOL. The questionnaire measures three domains: physical (15 items), psychological (7 items) and social (5 items), with scores ranging from 0 to 100 (100 being the best score possible). Internal consistency (Cronbach’s alpha) was reported to be > 0.82. Positive correlations between the ULL27 subscales and homologous SF-36 domains were significant. Sensitivity analysis in patients with active lymphedema demonstrated a significant effect size (9).

Translation

The questionnaire was translated according to established international guidelines (18-24). Two translators independently translated the questionnaire from French into Dutch, and a first version was established at a consensus meeting. The translation took into account the cultural and lifestyle context, making use of appropriate idioms [i.e., cross- cultural adaptation (20)]. Subsequently, a third translator translated the Dutch questionnaire back into French (19). This process of forward and back translation was repeated until a satisfactory translation was achieved [model of Brislin (19)].

As stipulated by Guillemin (20), the final translation was tested on patients, to clarify any ambiguities, in one-to-one interviews. Five patients commented on the difficulty, clarity and language of the questionnaire using a Dutch translation of the EORTC debriefing questionnaire for this purpose (15). Based on this information, a new version of the Dutch questionnaire was generated, which was translated into French by a fourth translator.

The final version of the questionnaire and its back translation, were then sent to the originator of the ULL27, Professor Launois (21), for comments. These were incorporated in the definitive version of the Dutch translation of the ULL27.

RAND36

The RAND 36-item Health Survey (RAND36) is a generic HRQOL questionnaire. It contains 8 domains: Physical Functioning (PF), Social Functioning (SF), Role Physical (RP), Role Emotional (RE), Mental Health (MH), Vitality (VT), Pain (P) and General Health (GH). Internal consistency was > 0.71 and construct validity, compared to similar scales, was significant (25).

Validation

The questionnaire was validated in twenty-three physiotherapy practices by physiotherapists specialized in edema therapy. Patients who matched the inclusion criteria were asked to participate in this study. These criteria were (1) women only, (2) unilateral edema of the upper limb, (3) no distinction between primary and secondary lymphedema, and (4) women should have fluency in Dutch, sufficient to understand the questionnaire. Patients with: (1) progressive cancer or (2) who had lymphangitis or infections of the upper limb in the past 2 months were excluded from this study.

All participants were asked to complete a general questionnaire concerning demographic data, the ULL27 and the RAND36. The physiotherapists were asked to determine the severity of edema, following a specific protocol in which the size difference of both upper limbs was measured in a standardized way. According to the protocol of Kühnke (26) with the volume of the hand added to the total of the upper limb. Completed questionnaires were returned and data were anonymized. Informed consent was not needed, since all measurements were part of good clinical practice.

The discriminant validity was determined in an age-matched group of 61 women without symptoms of the neck, shoulder or upper limb and had been recruited by the practice, family, and friends.

Statistical Analysis

The data were checked for normal distribution. Thereafter the internal consistency of the three domains was determined with Cronbach's Alpha, with a value of 0.7 or higher being considered adequate. The item internal consistency was also determined, with an item-domain correlation of 0.4 or higher being considered sufficient. The item-domain discriminant validity was also determined. The concurrent validity was established by comparing the scores of the three ULL27 domains with the scores of the eight domains of the RAND36, and the discriminant ability was tested by comparing the total score and the scores of the three domains of the patients with those of the age-matched group of women without upper limb lymphedema. A Spearman correlation coefficient of 0.40 or higher was considered to indicate a significant correlation (19). Finally, the correlation between the mean scores of the ULL27 and the severity of edema was determined.

This research was conducted in accordance with the Declaration of Helsinki.

Results

The Translation

Two translation rounds were needed to obtain the definitive version of the questionnaire that was tested on patients in the pilot study. During this study, one patient commented that questions on psychological topics might raise expectations that physiotherapists would also treat these aspects. This can be avoided by giving patients adequate information about treatment goals before therapy starts. Another patient suggested that the word “restricted” for questions 24 to 27 should be replaced by “hindered.” She considered that “restricted” meant that a person could not perform an action whereas “hindered” implied that a person could still perform the action, albeit to a diminished extent. The questionnaire was then discussed with Professor Launois, in English. The use of the word “hindered” or “restricted” in questions 24 to 27 was replaced by “are you feeling any difficulty,” and the questionnaire was approved.

The Validation Research

Twenty-nine physiotherapists specialized in edema were recruited between February and August 2006. They recruited 84 patients, the characteristics of whom are given in Table 1.

Most patients (mean age 59) were housewives (42.4 %), and 94% had undergone axillary surgery with or without surgical dissection of the breast. Three patients had not had surgery, and one patient had undergone extirpation of a melanoma on the shoulder.

Table 1. Characteristics of the Research Population (N=84)

Characteristic	Mean	Sd	Range
Age	59	11.79	34-80
Size difference in ml.	360.4	338.3	-170-1628
Start of oedema after the operation in months	26	56.51	0-360
Duration of oedema in months	35.51	45.14	0.5-276
Number of physiotherapy sessions	115	184.74	0-1100

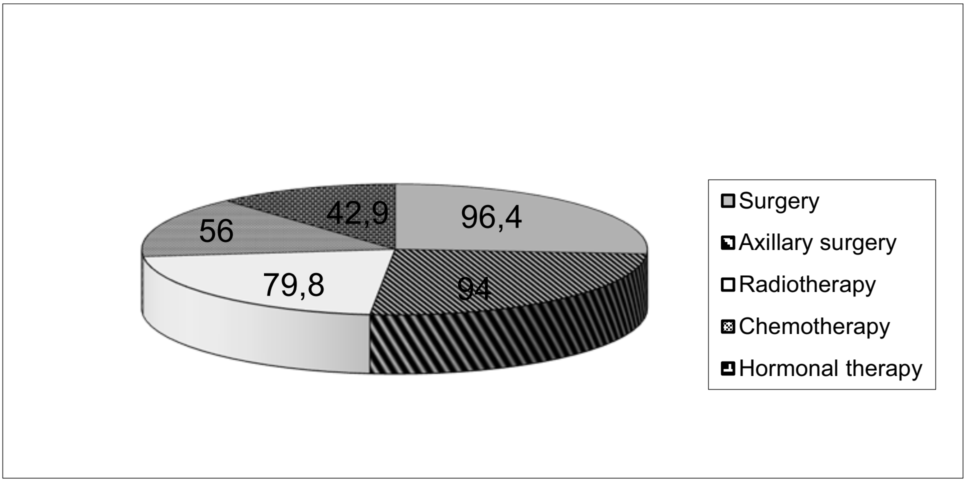

Fig. 1. Various treatments for breast cancer (%).

Surgery involved breast amputation (n=49) and breast-saving techniques (n=31). Patients received various postoperative treatments (see Fig. 1); 47 patients wore therapeutic hosiery for treatment of their edema.

Patients were classified by the severity of edema (Table 2), and then the mean ULL27 total score and subdomain scores were calculated for each edema severity (Fig. 2).

Kolmogorov-Smirnov analysis showed the data not to be distributed normally. An ANOVA with Bonferroni correction showed that there was no significant difference between the total and subdomain scores of the ULL27 in each of the four grades of edema severity.

The internal consistency of the Dutch ULL27 was good (all Cronbach's alpha values >0.7; Table 3) and the item-domain internal consistency was considered sufficient except in two cases (> 0.4). Question 20 ("Did you feel confident about the future during the last 4 weeks?") was scored 0.29 and question 22 ("Did you feel well and at ease during the last 4 weeks?") was scored 0.25. The item-domain discriminant validity was determined by examining the correlations of the items with all three domains. In two cases (also questions 20 and 22) the items showed a better correlation with a domain other than its corresponding domain.

Table 2. Classification of the Severity of Edema

Grade	Description	Volume difference	Number of patients
1	Not measurable oedema	150-299 ml	45
2	Small-volume oedema	300-499 ml	17
3	Medium-volume oedema	500-800 ml	15
4	Large-volume oedema	> 800 ml	7

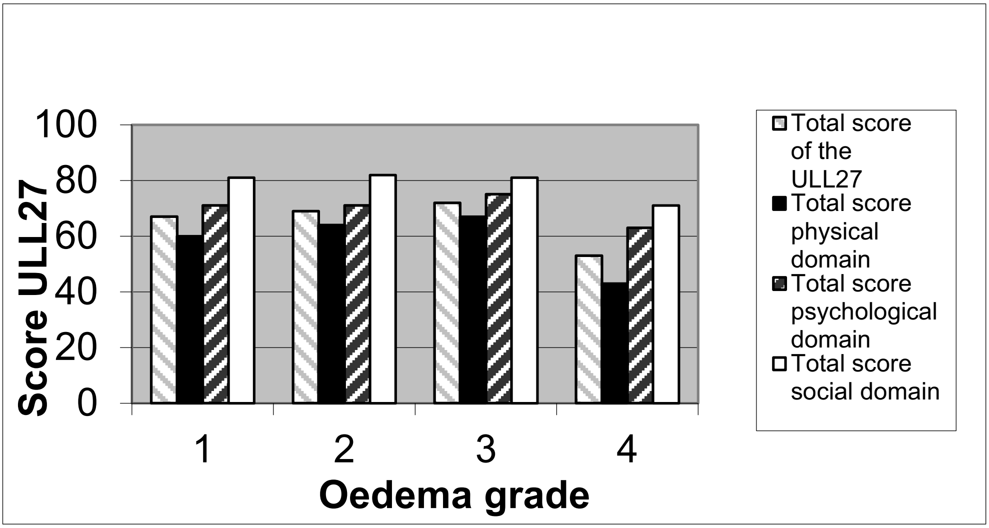

Fig. 2. Mean ULL27 score by severity of lymphedema.

Table 3. The Internal Consistency of the ULL27

Domain	Dutch version	French version
Physical domain	0.92	0.93
Psychological domain	0.78	0.86
Social domain	0.79	0.82

The concurrent validity was examined by associating the outcomes of the ULL27 with the RAND36. The domains of the Dutch ULL27 were significantly correlated with most of the corresponding domains of the RAND36 (Table 4).

Lastly, the total score and subdomain scores of an age- matched group of women without symptoms of the neck, shoulder or upper limb (N=61, mean age (SD) 54 (9.4)) were compared with those of 61 patients, using an unpaired T-test. The total score and subdomain scores of the two groups were significantly different ($p<0.001$), with the control group scoring higher on the ULL27.

Table 4. Correlations Between RAND36 and ULL27

RAND36	Physical domain ULL27	Psychological domain ULL27	Social domain ULL27
Physical Functioning (PF)	0,38	0,21	0,64
Role Physical (RP)	0,48	0,35	0,32
Bodily Pain (BP)	0,69	0,38	0,40
General Health (GH)	0,60	0,54	0,56
Vitality (VT)	0,47	0,55	0,44
Social Functioning (SF)	0,55	0,51	0,45
Role Emotional (RE)	0,39	0,42	0,36
Mental Health (MH)	0,52	0,66	0,53

Discussion

The purpose of this study was to translate the French ULL27 questionnaire into Dutch and to validate it in a Dutch population of patients with lymphedema in the upper limb. The availability of such a questionnaire makes it possible to compare the data collected in other international studies. The questionnaire was validated in 84 patients but the distribution of edema severity in this population (grade 1 53.6%, grade 2 20.2%, grade 3 17.9% and grade 4 8.3%) was not similar to that of the French population (n=304) used to validate the original questionnaire (grade 1 13%, grade 2 20%, grade 3 27%, grade 4 40%), which means that it was not possible to compare the data directly.

Edema severity and ULL27 scores were not correlated, as has been found in other studies showing a lack of correlation between the difference in size between the affected and non-affected upper limbs and HRQOL (3-7). The internal consistency of the three domains was good and similar to that of the French study. The item-domain internal consistency and item-domain validity differed in two cases: questions 20 and 22 showed a better correlation with the social domain than with their own psychological domain. These two questions have a different direction in their presentation than the other 25 questions and serve as control questions. It may be appropriate to ask patients to pay special attention to these questions. Otherwise, these items may need to be deleted in a future version. The physical domain of the ULL27 was poorly correlated with the Physical Functioning domain of the RAND36 (r =0.4). This can be explained by the fact that the questions of the

RAND36 are focused more on the lower limb and those of the ULL27 more on the upper limb. The strongest correlations between the scores of the psychological and social domains of the ULL27 were found for the General Health (GH), Vitality (VT), Social Functioning (SF), Bodily Pain (BP) and Mental Health (MH) domains of the RAND36 and ranged from rs 0.55-0.69, supporting concurrent validity. The correlations were less strong for the other domains, as was also found with the original French questionnaire (manuscript in preparation).

Lastly, the Dutch ULL27 showed strong discriminant validity between groups of women with and without symptoms of lymphedema of the upper limb.

However, a distinction in severity of the edema could not be made using the ULL27. A potential limitation of this study is that the severity of edema was measured according to a written protocol, and the measurements had not been practiced in advance. However, since specialized edema physiotherapists routinely measure the upper limb volume, it is expected that the measurements were accurate, with the only difference from normal practice being the measurement of the hand volume as part of the total volume. Moreover, it was the difference in hand/arm volume between the two upper limbs that was important.

The finding that HRQOL is not correlated with the difference in volume between the two upper limbs is consistent with earlier findings and supports the use of the ULL27 to provide additional information about lymphedema of the upper limb, because it offers better understanding of the patient in total. A minor limitation of the questionnaire is that it does not include information about upper limb complaints based on pathology other than edema. Therefore some open questions in the French, as well as in the Dutch version, are added to the 27 questions.

The design of this study was aimed at the validity of the ULL27. Its sensitivity to change in clinical use, however, is not yet determined. Further research is needed to establish the responsiveness of the Dutch ULL27.

Conflict of interest

The ULL27 was developed by Professor Robert Launois with an educational grant from REES France. Any person who wishes to use the questionnaire should contact Professor Robert Launois (reesfrance@wanadoo.fr). The Dutch translation and validation were prepared without funding.

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Chapter 6

Functioning in lymphedema from the patients' perspective using the International Classification of Functioning, Disability and health (ICF) as a reference

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Abstract

Purpose: To identify and quantify meaningful concepts in lymphedema from the patients' perspectives using the International Classification of Functioning, Disability and Health (ICF).
Methods: Six focus group interviews in five different centers were organized, audiotaped, transcribed verbatim and analyzed.
Results: A total of 2681 relevant ICF linkings were performed with the focus group data, resulting in 130 different second-level categories. Of these 130 second-level categories, 41 (31.5%) categories were categorized as Body Functions, 20 (15.5%) as Body Structures, 41 (31.5%) as Activities and Participation, and 28 (21.5%) as Environmental Factors. Overall, the most important issues according to the patients were the use of hosiery and bandages, support and relationships, and the shape of structures related to movement.
Conclusion: Based on their experiences with lymphedema, patients reported activity limitations and participation restrictions combined with impaired body functioning. Anatomical changes (Body Structures) were also often mentioned as a problem in daily life. Environmental factors may act as a barrier or facilitator for patient functioning. The ICF provides a valuable reference to identify concepts in statements from lymphedema patients. The results of this research will be used in the development of ICF Core Sets for lymphedema.

Introduction

Lymphedema is a chronic condition that can lead to physical functioning problems, often resulting in distress (1) and loss of quality of life (2, 3). Lymphedema presents clinically as swelling given the imbalance between interstitial filtration and fluid and protein discharge caused by compromised lymphatic system transport capacity. This condition may occur in upper and lower limbs as well as the midline (head, neck and thorax) and can be subdivided into primary and secondary lymphedema (4). Primary lymphedema, a congenital and sometimes hereditary disease, may exist at birth; however, most cases of primary lymphedema are manifested during puberty (4). The lymph vessels and/or nodes are not well constructed or do not function properly because of gene mutations (4). This condition is most commonly observed in the legs but can occur throughout the entire body, including the genital region. Secondary lymphedema results from lymph vessel and/or node damage, which occurs as a complication of oncologic surgery and/or radiotherapy, infections or trauma (5). According to a WHO report, approximately 120 million people in 72 countries were infected with a parasite (lymphatic filariasis) in 2010 (6). Of these infected individuals, 15 million people suffered from the consequent lymphedema manifestations (filarial elephantiasis). Podoconiosis is endemic non-filarial elephantiasis due to long-term barefoot exposure to irritant volcanic soils in tropical Africa (7). Lymphedema epidemiology is not exactly known because it is not a well-registered disease. The number of patients worldwide is estimated to be approximately 140-250 million (5).

Symptoms of lymphedema

If untreated, lymphedema leads to irreversible tissue changes, a high risk of infection and feelings of heaviness and tightness; in addition, patients report that jewelry or clothes feel uncomfortable or tight on the affected body parts (8-10). These features lead to impairments, such as a decreased range of movement of the limb involved, as well as to limitations in walking, personal care, domestic life, occupation and socialization (7). Altered body shape, often leading to stigmatization, as well as the need to use garments or bandaging daily, make acceptance difficult for the patient as well as his/her family (11).

Treatment

Lymphedema therapeutic options include conservative and operative modalities and should be fine-tuned to the patient's living circumstances, including work and home environments, as well as to personal factors, such as age, co-morbidities, (malignant) disease prognosis, psychosocial aspects, and physical potential. The general goal of conservative treatment is to eliminate edema; by compression therapy in combination with manual lymph drainage (MLD) (12), exercises, proper weight reduction and weight control (13), and special skin care to prevent infection (14). When maximal initial therapeutic result is achieved, the emphasis of the treatment will be on enhancing patient self-management, including strict compliance in wearing compression garments (bandages, wraps or hosiery) (12). In severe lymphedema stages, circumferential suction-assisted lipectomy can be applied to alleviate irreversible alterations, such as adipose tissue formation and fibrosis (15). In 2012, the International Lymphedema Framework (ILF) published the 'best practice document' for the management of lymphedema (16). It proposes a comprehensive lymphedema service based on the chronic care model and use of the International Classification of Functioning, Disability and Health (ICF) as a vehicle for determining and delivering a patient's needs (16).

ICF

The ICF provides a comprehensive framework of human functioning as well as a classification system, based on a bio-psychosocial model (17, 18). In addition, it offers a universal language understood by health professionals, researchers, policy makers, patients, and patient organizations.

The ICF consists of two separate parts. Part 1 addresses functioning and is composed of three components: 'Body Functions', 'Body Structures', and 'Activities and Participation'. Part 2 addresses contextual factors and is composed of two components: 'Environmental Factors' and 'Personal Factors'. However, 'Personal Factors' are not described as a classification in the ICF yet, because of the significant social and cultural variation (11).

Each ICF category is assigned an alphanumerical code, a letter that refers to the classification components (b: Body Functions, s: Body Structures, d: Activities and Participation, and e: Environmental Factors). Each letter is followed by a number or numbers starting with the chapter number (one digit) and followed by second-level specifications (three digits) and further third- and fourth-level specifications (four and five digits), where applicable.

Although its classifications with more than 1400 categories can serve as a reference, the ICF is not easily applicable in clinical practice. For this reason, tools, such as ICF Core Sets (17, 18) (a core set is an ICF selection developed for a

specific diagnostic group), make the ICF useful for health care providers (19). The Core Sets facilitate the use of ICF-terminology to describe the functioning profile of a patient (a ‘functional’ diagnosis) and the formulation of treatment goals. Description uniformity is needed to compare data for the evaluation of treatment efficacy and to collect data for research. The ICF Core Sets may be used during the process of clinical reasoning and to record the results of clinical reasoning and treatment in electronic health care records.
Today, ICF Core Sets have been developed for various burdensome chronic conditions (18, 20-22) and since lymphedema is a serious, chronic and worldwide problem in health care, there is a great need for specific Core Sets for this disease (3, 23, 24) . The development of Core Sets for lymphedema consists of several preparatory studies leading to an international consensus conference. To obtain a complete spectrum of the effects of lymphedema, these studies have different points of view, i.e. the researcher, the clinician, the patient and the international expert.

Aim of the study
The aim of this study is to determine relevant aspects of functioning as well as relevant environmental and personal factors from the lymphedema patients’ perspective and to quantify these, using the ICF.

Methods

Design
A Dutch multicenter qualitative study using focus group interviews was conducted as part of a larger project on the development of ICF Core Sets for lymphedema (25). Patients were recruited from five different centers during 2012-2013. Stucki and Cieza (17) developed a method to establish ICF Core Sets for specific conditions. The development of ICF Core Sets for lymphedema follows this procedure. The study was approved by the Ethics Committee of the Erasmus Medical Centre in the Netherlands and performed in accordance with the Declaration of Helsinki.

Participants

To obtain a comprehensive understanding of the continuum of clinical care, patients from various treatment stages (control, after first contact, during initial treatment phase and maintenance phase) were included. Because lymphedema occurs in various locations and results from various causes, six different focus groups were used, each with 3-8 participants. These groups consisted of patients with: (A) lymphedema with non-oncology background (mixed locations), (B) lymphedema with oncology background (mixed locations), (C) lymphedema of the upper limb (mixed causes), (D) lymphedema of the lower limb (mixed causes), (E) lymphedema in the genital region (mixed causes), and (F) lymphedema in the head and neck region caused by cancer-treatment.

All participants had to meet the following inclusion criteria: the individual (1) had a diagnosis of lymphedema determined by a physician, (2) was older than 18 years, (3) was informed of and understood the purpose and rationale of the study, and (4) signed the patient consent form. Before they participated, patients had to fill in a form with questions about socio-demographic items and comorbidities and they had to range their perceived lymphedema severity on a scale from zero to ten.

Questions

The following open-ended questions, based on ICF components, were used in the focus groups (26-28):

1. If you think about your body and mind, what does not work the way it is supposed to? (Body Functions)
2. If you think about your body, in which parts are your problems located? (Body Structures)
3. If you think about your daily life, which problems do you experience performing the activities or things you want or are necessary? (Activities and Participation)
4. If you think about your environment and living conditions, what do you find helpful or supportive? (Environmental Factors-facilitators)
5. If you think about your environment and living conditions, what barriers do you experience? (Environmental Factors-barriers)
6. If you think about yourself, what is important about you and the way you handle your disease or the problems associated with your disease? (Personal Factors)

Data collection

The same moderator (a member of the Dutch Society for Physical Therapists within Lymphology (NVFL)) led the focus groups. When collecting information about the nature and course of the study, the moderator emphasized that the individual retained the right to refuse participation at any time without any treatment consequences. Each focus group discussion was digitally recorded and transcribed verbatim. When the transcription was completed, the members of each focus group were asked – by email - if they agreed with the contents of the transcription (member check). If not, alterations were made until there was agreement.

Data analysis

The data analysis was conducted in four steps and followed the method of 'meaning condensation' (29). In the first step, the transcribed focus group data were read to obtain an overview of the collected data. In the second step, the data were divided into 'meaning units', and the theme that dominated a meaning unit was determined. A 'meaning unit' was defined as a specific unit of text, either a few words or a few sentences, with a common theme (30). The text was divided as soon as the researcher discerned a shift in the meaning (29). In the third step, 'meaningful concepts' contained in the meaning units were identified. A 'meaningful concept'(MC) was defined as a unit of text that conveys a single theme (31). A meaning unit could contain more than one MC. This procedure was conducted using the Kwalitan software program, version 6.02.

In step four, each MC was linked to one or more ICF categories according to published linking rules (32, 33). Linked ICF categories are defined as relevant concepts of functioning for individuals with lymphedema. The following concepts cannot be classified using the ICF: personal factors (indicated with 'pf'), health conditions ('hc'), concepts related to the ICF but cannot be assigned a particular component or code (not definable 'nd'), and concepts beyond the ICF framework (not covered 'nc'). The MCs were linked independently of the six questions (e.g., codes referring to the Body Functions component could also arise when the group answered one of the five other questions). After each focus group, the linked ICF categories were added to a list of ICF categories selected so far from earlier studies (34)

Quality of data

To assure reliability, two health professionals (PV and PG) separately performed step three and four in the first focus group to achieve agreement concerning the implementation of the linking rules for this specific health condition. The procedure was performed according to the method proposed by Stucki and Cieza for the development of ICF Core Sets (32, 33). Afterwards, the MCs and linking processes identified by the two health professionals were compared. The degree of agreement between the two health professionals (PV and PG) regarding the identified MCs as well as the linked ICF categories in the first focus group was calculated by kappa statistics. The kappa coefficient values generally range from 0 to 1, where 1 indicates perfect agreement and 0 indicates no agreement beyond what is expected by chance alone. A kappa value of 0.6 or greater is considered sufficient (35). Disagreement was resolved by structured discussion.

The data analysis for all focus groups

After analyzing data from the first focus group and ICF linking, three individuals independently linked all MCs from all focus groups to the ICF (PV, YH, DvR). Disagreement was resolved after structured discussion to create consensus for a final version of the linked ICF categories. This extra ICF coding (agreement) was performed to obtain the best possible ICF linking from the patients' perspectives. The degree of agreement concerning the linking of all MCs between the main researcher (PV) and the other researchers (YH, DR) was subsequently assessed by kappa statistics.

Results

Description of the focus groups patients

A total of 31 patients with a mean age of 55 years participated in six focus groups; the majority of the participants were female (67.7%). Of the 31 patients, the lymphedema location included seven exclusive upper limb cases, 11 exclusive lower limb cases, five exclusive midline cases and eight cases displaying a combination of locations (6 lower limb-genital region cases and two upper limb-breast cases). In total, 18 patients reported a history of cancer, and 13 reported no history of cancer (i.e. primary lymphedema). Lymphedema severity, as judged by the participants themselves, was the highest in the lymphedema group with an oncologic cause (6.5 on a scale of 0 to 10) and the genital lymphedema group (6.0). Further details of the patient characteristics are shown in Table 1.

Table 1. Patient characteristics. [( ) = standard deviation.]
(A) Lymphedema with non-oncology background (mixed locations), (B) lymphedema with oncology background (mixed locations), (C) lymphedema of the upper limb (mixed backgrounds), (D) lymphedema of the lower limb (mixed backgrounds), (E) lymphedema in the genital region (mixed backgrounds) and (F) lymphedema in the head and neck region.

Variables	Total (n = 31)	A (n = 5)	B (n = 6)	C (n = 5)	D (n = 6)	E (n = 6)	F (n = 3)
Age in years	55 (13.6)	37.8 (8.5)	61.1 (13)	61.6 (12.9)	51.5 (12.2)	56.2 (11.9)	65.3 (3)
Women : men	21 : 10	5 : 0	6 : 0	5 : 0	5 : 1	0 : 6	0 : 3
Duration of lymphedema in months	124 (160)	186 (132.9)	39 (20.1)	49.4 (50.2)	218.5 (231.8)	184.3 (205.2)	5.3 (1.5)
Upper limb : lower limb : midline : combination	7 : 11 : 5 : 8	0 : 5 : 0 : 0	2 : 2 : 0 : 2	5 : 0 : 0 : 0	0 : 4 : 0 : 2	0 : 0 : 2 : 4	0 : 0 : 3 : 0
Seriousness of lymphedema on a 11-point scale	5.7 (2.3)	5 (1.2)	6.5 (0.5)	5.6 (3)	5.5 (2.3)	6 (4)	5.3 (2)
Employment	15	4	2	3	3	2	1

Data analysis and meaning units

The following meaning units were observed in most of the groups:

- Lymphedema is accepted as a burden that one has to learn to live with.
- Hosiery and bandages are viewed as barriers to daily life that also alleviate the disability.
- Persons with lymphedema experience problems with clothing choices.
- Swimming is an activity that provides relief during the activity itself. However, patients do not choose to go to the beach or swimming pool, due to either feelings of shame or problems with their stockings.
- Most lymphedema (swelling) problems are experienced during summer. However, some patients report fewer problems in summer compared with winter.
- Contact with health professionals is rather ambivalent. The patients report good and bad experiences. In addition, the patients feel as if health professionals are ignorant and downplay lymphedema, because they do not really know what it means to have lymphedema.

- Numbness and hardened skin are the most frequently mentioned problems concerning the skin.
- Patients report that distress has a negative effect on lymphedema; stress worsens lymphedema and is a risk factor for erysipelas.
- Contact with fellow sufferers is worthwhile.
- Patients with lymphedema localized in the genital area as well as in the head and neck region are most likely to discuss the impact and consequences of operations compared with the other groups.
- Patients with lower limb lymphedema experience swelling that always begins in the feet and expands up the leg, whereas patients with upper limb lymphedema describe swelling at various sites of the arm.
- Individuals with lymphedema in the lower limbs mentioned pain more often than those with lymphedema in other locations.
- The group with genital lymphedema uses specific devices, such as general lymphedema devices or devices that support urination (e.g., the Whittaker pouch and self-made constructions to regulate urination), most often compared with other groups.

Linking MCs to the ICF

Based on the focus group data, a total of 2681 relevant ICF linkings were performed. In total, 12 first-level categories, 78 second-level categories, 146 third-level categories, and 7 fourth-level categories for ICF linkings were identified. For clarity and readability, the third- and fourth-level categories were merged into the second-level categories, resulting in 130 different second-level categories. Of the 130 second-level categories, 41 (31.5%) categories were identified as Body Functions, 20 (15.5%) as Body Structures, 41 (31.5%) as Activities and Participation, and 28 (21.5%) as Environmental Factors. Of the 282 MCs that could not be given an ICF code (9.5% of all MCs), 135 (48%; 4.6% of all MCs) were coded as Personal Factors, 9 (3%; 0.3% of all MCs) were coded as 'nc', 12 (4%; 0.4% of all MCs) were coded as 'nd', 7 (2%; 0.2% of all MCs) were coded as 'nd-gh', and 119 (43%; 4% of all MCs) were coded as 'hc'.

Body Functions

Table 2 (tables 2-5 published on the Acta Oncologica website) shows the first- and second-level categories identified for the Body Functions component. The top five frequently mentioned categories were 'b435, Immunological system functions' (impairments in the lymphatic system); 'b152, Emotional functions' (emotions such as fear, anger, joy); 'b280, Sensation of pain'; 'b126, Temperament and personality functions' (including psychic stability, confidence and optimism); and ‘b840, Sensations related to the skin’ (including itching and tingling). This pattern is observed in most of the groups, with a few differences. In the group with lymphedema in the head and neck region (F), the most mentioned category is ‘b510, Ingestion functions’. In the group with upper limb lymphedema (C), the most frequent category is ‘b455, Exercise tolerance functions’. Various quotes from patients describing some of these categories are found below.

“ … heavy feeling and tiredness. Especially in the summer when it is hot, you get the feeling that your leg is swelling and you can’t move forward. Previously I did not have that problem but ever since the edema it bothers me” (patient from group B lymphedema with oncology background (mixed locations)).

“…it looks as if mental things have more influence on the edema than physical. When, for instance, I witness a funeral the edema gets worse” (patient from group B lymphedema with oncology background (mixed locations)).

Body Structures

Table 3 displays the first- and second-level categories identified for the Body Structures component. In this component, the top five mentioned categories were ‘s750, Structure of lower extremity’; ‘s730, Structure of upper extremity’; ‘s630, Structure of reproductive system’; ‘s710, Structure of head and neck region’; and ‘s760, Structure of trunk’. In the genital lymphedema (E) group, the most mentioned category was ‘s630, Structure of reproductive system’. In group F (head and neck lymphedema), the most mentioned category was ‘s710, Structure of head and neck region’. The following quotes illustrate the Body Structures component:

“...but in fact I already have thick feet and swollen legs since 1978” (patient from group D lymphedema of the lower limb (mixed causes)).

“…but it is especially the deformation of my face. That is very annoying. It does not bother me much, except that my lips get swollen. They do not occlude enough and you make a mess when drinking” (patient from group F lymphedema in the head and neck region).

“…when I was being operated and my lymph nodes were removed, they told me that there was a chance I could get swollen legs. But about genital lymphedema they did not say anything. You had to find that out afterwards by yourself” (patient from group E lymphedema in the genital region).

Activities and Participation

Table 4 indicates the first- and second-level categories identified for the Activities and Participation component. The top five categories identified were ‘d920, Recreation and leisure’; ‘d415, Maintaining a body position’ (lying, sitting, standing, etc.), ‘d570, Looking after one’s health’ (ensuring comfort, maintaining health and managing diet and fitness); ‘d450, Walking’; and ‘d475, Driving’. In focus group C (upper limb lymphedema with mixed backgrounds), ‘d640, Doing housework’ occurred most frequently, whereas ‘d415, Maintaining a body position’ occurred most frequently in the genital lymphedema group (E).

Typical quotes from the Activities and Participation component are found below:

“…you are always busy considering your legs. Because you can’t work and practice a sport and doing things at home as well” (patient from group A lymphedema with non-oncology background (mixed locations)).

“…the problem with driving is the pain in your legs. But since I have a car with cruise control it doesn’t bother me anymore” (patient from group D lymphedema of the lower limb (mixed causes)).

Some statements regarding the impact of genital lymphedema (group E patients) are:

“…I can’t walk very far, I can’t sit for a long time, I can’t lay down and I can’t bend over because I lose my balance and then I fall down on the ground. This is hindering my daily activities, although I try to make the best out of it”.

“…I also can’t urinate in a normal way. At home I use an urinal and when I leave home I always take little cups with me because it sprinkles everywhere. I use a diaper and every morning I bandage my scrotum”.

Environmental Factors

Table 5 displays the first- and second-level categories identified for the Environmental Factors component. The categories that occurred most often were ‘e115, Products and technology for personal use in daily living’ (non-adapted items include clothes, shoes, textiles, furniture, and tools; adapted items include stockings and prostheses); ‘e580, Health services, systems and policies’ (all services, systems and policies for the prevention and treatment of health problems that provide medical rehabilitation and promote a healthy life style); ’e355, Health professionals’; ‘e310, Immediate family’; and ‘e110, Products and substances for personal consumption’. The examples below illustrate these findings:

“…what really works is a hula-hoop. It helps draining the lymphedema in my legs when I use it daily” (patient from group D lymphedema of the lower limb (mixed causes)).

“…the insurance company does not always co-operate. They compensate two pairs of stockings within 12 months, which is not sufficient at all. I just have to pay them myself” (patient from group E lymphedema in the genital region (mixed causes)).

Personal Factors

Personal factors can be broadly divided into socio-demographic factors (including gender and race), personal living situations and coping strategies.

Various quotes applicable to these factors are found below:

"…I don't know how to explain it. Everybody would like to assist you but in some sort of way you live in a cocoon. You go your own way and you seclude yourself from them. There will be a moment in time that you will realize that you can't do it alone, but I am not at that point yet" (patient from group A lymphedema with non-oncology background (mixed locations)).

"...always being busy. With a group of friends practicing sports and afterwards drinking a pint. Not sitting at home and thinking about the problems" (patient from group E lymphedema in the genital region (mixed causes)).

Not covered or definable items

Statements that were not covered or definable by the ICF were for instance: "daily life", "handicap", "dead", "tissue" and "being sickly".

Quality of data

Inter-coder reliability in the first focus group (PV and PG).

The inter-coder agreement for the determination of MCs was 63.8% (kappa 0.31). Regarding the linking of MCs to the ICF, the inter-coder agreement was 73% (kappa 0.46). As previously mentioned, the MCs determination and the ICF linking process were performed simultaneously. Disagreements most often originated from the fact that both researchers possess different points of view (e.g., both researchers consistently coded the same MC in different ways).

Inter-coder reliability for all focus groups (PV, YH and DvR).

The inter-coder agreement for ICF linking of the meaningful concepts between PV and YH was 75% (kappa 0.72) and 55% (kappa 0.49) between PV and DR. In this case, YH and DR linked MCs with unknown contexts (i.e., the coders did not know which focus groups supplied the MCs). These MCs could be interpreted in various ways. Examples are MCs like: "I am not a man anymore", "Visit" and "Stress". In addition, these statements are also examples of the previously cited problem of coding the same MC in different ways.

Discussion

As part of the development process for ICF Core sets for lymphedema, the purpose of this study was to determine relevant aspects of functioning as well as environmental and personal factors from the perspective of lymphedema patients. Based on their experiences with lymphedema, patients predominantly faced activity limitations and participation restrictions (31.5% of MCs) combined with impaired body functions (also 31.5% of MCs). Environmental factors were indicated in 21.5% of MCs, and these factors were considered to act as barriers (e.g., hosiery) or facilitators (e.g., spouses).

Anatomical changes (body structure impairments) were given less attention and identified in 15.5% of MCs.

From the focus groups it can be derived that lymphedema is a chronic health condition, which needs constant medical care and attention. In many parts of the world lymphedema is treated in rehabilitation settings. A common complaint of patients is that they have a chronic medical problem and need ongoing medical care and not only one period of treatment in a rehabilitation setting. The findings in this study support this patients' point of view. Although recent guidelines(12, 36) put more emphasis on self-management and education of the patient to become less dependent on health care, regular monitoring and support is advised. Depending on the severity of the condition and the level of self-management, patients will need more or less frequent treatment and lifelong monitoring.

Meaning units

Problems with accepting lymphedema were mentioned in every group; however, these problems were cited less frequently in primary lymphedema patients. This finding is likely attributable to the fact that these patients have had more time to accept their disease as they have experienced lymphedema longer than the other groups. Many patients stated that movement, particularly swimming, relieves lymphedema symptoms during the activity itself. Consistent with this finding, Tidhar et al. (37, 38) designed a water exercise program to improve mobility and lymphatic drainage. Most swelling problems occur during summer; however, some patients experience fewer problems in summer than winter. Higher summer temperatures, resulting in greater subcutaneous fluid in the suprafascial compartments by enhanced capillary filtration, leads to more swelling. The available literature does not explain why warm weather provides relief to some patients.

The most frequently mentioned skin complaints consist of a hardened and numb feeling. This complaint could be a negative side effect of radiotherapy; however, various patients with primary lymphedema who have not received radiotherapy cite similar problems.

Patients with lower limb lymphedema complain more about pain than people with lymphedema in other locations. This observation can be explained by the fact that, due to gravitation, more pressure exists in the lower vs. upper limbs.

Body Functions

The focus group participants reported a variety of impairments in Body Functions. All of the chapters in this component were covered, especially the chapters regarding mental functions as well as functions of cardiovascular, hematological, immunological and respiratory systems. The mental effects of lymphedema pertain to topics that were also reported in previous studies, such as impairments in body image (39, 40), distress (41, 42), and more fatigue (42). Person et al. (43) recommended that physiological distress management should serve as a significant component of lymphedema management programs in developing countries. Although impairments in sexual functions can play an important role (44), these problems were only mentioned in the genital lymphedema group. It is possible that these sexual problems are underreported and will be more easily expressed in one-to-one interviews.

Body Structures

Altered body shape is a problem discussed in the groups and found in the Body Structures component. The shape of the lower limb was particularly cited as problematic. This finding was also mentioned in earlier publications. (39, 40, 42).

Activities and Participation

All categories of the Activities and Participation component were mentioned. Mobility items were mentioned most often, followed by self-care, recreation and leisure, and domestic life. Other studies partially support these findings. Self-management to prevent lymphedema worsening is emphasized in the focus groups (11, 45); however, the impact on recreation and leisure was not found in earlier literature. The patients also stated that they had problems with creating and maintaining interpersonal relationships, which made them isolated in society. This isolation was described in various studies(41, 46, 47). In the focus groups, remunerative employment was only partially discussed; this observation is likely because half of the patient population was not working at the time of the interview. It appears that lymphedema has a minor impact on work in Western countries compared with developing countries (47, 48).

Environmental Factors

The top three mentioned environmental factors are Products and technology, Support and relationships, and Health services, systems and policies. The literature supports these findings.

The impact of bandages and hosiery is reported in the literature. Although studies on compression garments are scarce or only described using small patient groups, Janda et al. (49) highlighted the financial burden of compression garments (only 15 patients). Frid et al. (39) mentioned the loss of autonomy as patients are dependent on nursing staff for dressing. Sawan et al. (50) concluded that the use of hosiery had no impact on health-related quality of life (14 patients). The findings of Sawan et al. are not supported by the patients of these focus groups. In every focus group bandaging and hosiery were discussed in detail and it was a recurring topic. Overall the conclusion is that it is a burden, but one with which they cannot live without. Presumably it does affect their health-related quality of life. Thus, the influence of this environmental factor should be more broadly examined in the future.

In the search for a correct diagnosis and effective treatment (45), interactions with other persons are perceived as both positive and negative, often leading to stigmatization (39, 40, 47, 48, 51). Although stigmatization was mentioned in some Western studies, the impact of its consequences likely plays a greater role in developing countries (48).

Health professionals are perceived as lacking education in this respect and give too less post-intervention support (40, 46, 48, 51). From these patients' observations it can be derived that lymphedema is not well-known and sometimes underestimated by health professionals. So there is a need for more education for those who could encounter lymphedema (e.g. general practitioners and surgeons), especially when working with oncology patients. Financial support from government and insurance companies is however lacking (51).

Personal Factors

In the qualitative research literature, personal factors play an important role in describing the coping strategies of lymphedema patients (11, 39, 46, 48). The outcomes of this study support these findings. Unfortunately, personal factors cannot be coded with the ICF; however, various suggestions on the subdivision and classification of personal factors have been made (52-54).

Limitations of the research

Although several meaning units were identified and most of the ICF domains were addressed, various limitations to this study exist. By merging the third and fourth ICF levels into the second level, some specific codes (corresponding with MCs) are not expressed in the analysis. These codes were presented at the concluding consensus conference, which is the last step in the Core Sets development. Based on the findings of this research, it cannot be concluded that all of the ICF categories that were identified are equally relevant and represent all forms of lymphedema because the focus groups were comprised of lymphedema patients with various etiologies and clinical locations. Although the researchers attempted to obtain data on a variety of health conditions, the predominance of cancer-related lymphedema (18 of the 31 patients) can serve as a bias. Cross-cultural variation was not considered because the study was conducted in one (Western) country, the Netherlands. Only one person with a different cultural background was part of the focus group. Lymphedema can be inborn; by excluding individuals younger than 18 years, some age-related issues were potentially missed.

This research is part of the preparatory studies for the development of ICF Core Sets for lymphedema and provides quantitative data for the consensus conference, where the final version of the Core Sets was formulated (25). This study uses a qualitative approach, seeming the best method to determine the patients' point of view. For the presentation at the international consensus conference, being the final part of the development of the ICF Core Sets, the qualitative data had to be made quantitative. This study followed the method of Stucki and Cieza (14) in which this is stated. Many researches followed this procedure ever since (21, 28-30).

Furthermore, the research bias could result from the fact that the six open questions are leading, referring to specific ICF components. However, all participants could freely speak about issues important for them during the focus group discussions. Above all, it is important to take into account that the qualitative approach in this study was performed to identify the broadest possible range of problems, also to provide a decision base for the consensus process in the development of ICF Core Sets for lymphedema.

The low kappa value of the inter-coder reliability between PV and PG (0.46) and PV and DvR (0.49) compared with other studies (e.g., Geyh et al. (55); range: 0.46-0.84) can be partly explained by the fact that the Kappa values were calculated after a fully independent formulation of MCs by two persons and concept linking to ICF-codes. Inter-individual differences can result from variation in the identification of MCs as well as the selection of ICF-codes. In the study of Geyh et al. (55), the kappa values were calculated after consensus was reached for the MCs.

Compared to the literature review (34), which was conducted as part of the lymphedema ICF Core set development, some points of interest are worth mentioning. Although Randomized Clinical Trials (RCTs) and qualitative research are commonly considered the most valuable type of research , qualitative lymphedema research is limited. Furthermore, in the existing lymphedema RCTs, little attention is given to the following ICF chapters: Functions of the skin and related structures, Recreation and leisure, and Support and relationships. However, patients indicated that they consider these topics important. These findings should be considered when composing the first version of the lymphedema Core Sets. They also should be respected by health care providers and should be considered as variables in future RCTs.

Conclusion

Systematic research on the effects of lymphedema from a patient's point of view using the ICF has not been previously performed. This study provides sufficient data for the development of Core Sets for lymphedema using the bio-psychosocial model. The data help to describe the functioning of lymphedema patients using international standards. Various lymphedema locations were examined, indicating outcome differences and similarities between the groups. Patients with lower limb lymphedema tend to have more pain than other patients. Overall, the most important issues to the patient were the use of hosiery and bandages, support and relationships, and the shape of structures related with movement. The impact of hosiery and bandages on daily life in particular needs more attention in clinical practice and therefore further investigation is needed to obtain objective measurements on this topic.

Finally, from these focus groups it appeared that lymphedema is a chronic health condition which needs ongoing medical care and attention.

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Table 2. Frequency of the 44 first- and second-level categories of the Body Function component.
(A) Lymphedema with non-oncology background (mixed locations), (B) lymphedema with oncology background (mixed locations), (C) lymphedema of the upper limb (mixed backgrounds), (D) lymphedema of the lower limb (mixed backgrounds), (E) lymphedema in the genital region (mixed backgrounds) and (F) lymphedema in the head and neck region.

ICF code	Description	Total	(A)	(B)	(C)	(D)	(E)	(F)
b		4						4
Chapter 1: Mental functions								
b114	Orientation functions	1					1	
b126	Temperament and personality functions	58	3	21	9	8	13	4
b130	Energy and drive functions	17	4	2	3		7	1
b134	Sleep functions	12	2	5	2			3
b140	Attention functions	2				1	1	
b144	Memory functions	1			1			
b147	Psychomotor functions	1			1			
b152	Emotional functions	83	6	17	19	13	7	21
b160	Thought functions	1			1			
b164	Higher-level cognitive functions	14	3	6	2		3	
b180	Experience of self and time functions	7	2		2			3
Chapter 2: Sensory functions and pain								
b230	Hearing functions	1						1
b235	Vestibular functions	4		3			1	
b240	Sensations associated with hearing and vestibular function	2			1			1
b250	Taste functions	1						1
b255	Smell function	1						1
b260	Proprioceptive function	6	5	1				
b265	Touch functions	18	2	4	6	2	3	5
b270	Sensory functions related to temperature and other stimuli	3	1		2			
b280	Sensation of pain	65	16	11	6	22	4	6
Chapter 4: Functions of the cardiovascular, hematological, immunological and respiratory systems								
b4		1						1
b415	Blood vessel functions	8	4	4				
b435	Immunological system functions	159	34	36	17	38	23	11
b440	Respiration functions	5	2	2				1
b450	Additional respiratory functions	4						4

b455	Exercise tolerance functions	42	6	6	20	4	4	2
b460	Sensations associated with cardiovascular and respiratory functions	4						4
Chapter 5: Functions of the digestive, metabolic and endocrine systems								
b510	Ingestion functions	26						26
b530	Weight maintenance functions	11	4		2	3	1	1
b535	Sensations associated with the digestive system	3						3
b555	Endocrine gland functions	1			1			
Chapter 6: Genitourinary and reproductive functions								
b620	Urination functions	5					5	
b640	Sexual functions	5					2	3
b660	Procreation functions	5	4			1		
Chapter 7: Neuromusculoskeletal and movement-related functions								
b710	Mobility of joint functions	11	6	5				
b730	Muscle power functions	6		1		3	1	1
b735	Muscle tone functions	2						2
b760	Control of voluntary movement functions	23	4	3	9	2	4	1
b780	Sensations related to muscles and movement functions	26	6	6	5	6		3
Chapter 8: Functions of the skin and related structures								
b8		1				1		
b810	Protective functions of the skin	18	2	3		3	4	6
b820	Repair functions of the skin	2				1	1	
b840	Sensations related to the skin	47	5	8	3	8	3	20

Table 3. Frequency of the 23 first- and second-level categories of the Body Structures component. (A) Lymphedema with non-oncology background (mixed locations), (B) lymphedema with oncology background (mixed locations), (C) lymphedema of the upper limb (mixed backgrounds), (D) lymphedema of the lower limb (mixed backgrounds), (E) lymphedema in the genital region (mixed backgrounds) and (F) lymphedema in the head and neck region.

ICF code	Description	Total	(A)	(B)	(C)	(D)	(E)	(F)
s		2						2
Chapter 1: Structures of the nervous system								
s1		1		1				
Chapter 2: The eye, ear and related structures								
s2		1						1
s220	Structures of eyeball	2				2		
Chapter 3: Structures involved in voice and speech								
s320	Structure of mouth	11						11
s330	Structure of pharynx	3						3
s340	Structure of larynx	2						2
Chapter 4: Structures of the cardiovascular, immunological and respiratory systems								
s410	Structure of cardiovascular system	3					1	2
s420	Structure of immune system	9	1		3	2	3	
s430	Structure of respiratory system	2		1				1
Chapter 5: structures related to the digestive, metabolic and endocrine system								
s510	Structure of salivary glands	1						1
s540	Structure of intestine	1					1	
Chapter 6: Structures related to the genitourinary and reproductive systems								
s610	Structure of urinary system	2					2	
s630	Structure of reproductive system	35	2	4	6	1	22	

Chapter 7: Structures related to movement								
s710	Structure of head and neck region	20			5	3		12
s720	Structure of shoulder region	12		6	3			3
s730	Structure of upper extremity	58		25	29	1	2	1
s740	Structure of pelvic region	10				1	9	
s750	Structure of lower extremity	179	54	25	7	82	11	
s760	Structure of trunk	19	2	2	3	8	4	
s770	Additional musculoskeletal structures related to movement	3			1	1		1
Chapter 8: Skin and related structures								
s810	Structure of areas of skin	11			1	1	7	2
s840	Structure of hair	1			1			

Table 4. Frequency of the 43 first- and second-level categories of the Activities and Participation component.
(A) Lymphedema with non-oncology background (mixed locations), (B) lymphedema with oncology background (mixed locations), (C) lymphedema of the upper limb (mixed backgrounds), (D) lymphedema of the lower limb (mixed backgrounds), (E) lymphedema in the genital region (mixed backgrounds) and (F) lymphedema in the head and neck region.

ICF code	Description	Total	(A)	(B)	(C)	(D)	(E)	(F)
d		3		1	1		1	
Chapter 1: Learning and applying knowledge								
d115	Listening	2					2	
d166	Reading	2	1					1
d170	Writing	2			1			1
Chapter 2: General tasks and demands								
d230	Carrying out daily routine	10		2	2	1	4	1
d240	Handling stress and other psychological demands	1						1
Chapter 3: Communication								
d325	Communicating with – receiving- written messages	1	1					
d330	Speaking	8		1			1	6
d335	Producing non-verbal messages	2		2				
d350	Conversation	1					1	
d355	Discussion	1						1
d360	Using communication devices and techniques	11	1	4	2	2		2
Chapter 4: Mobility								
d410	Changing basic body position	26	8	4	3	4	4	3
d415	Maintaining a body position	59	11	15	9	13	11	
d430	Lifting and carrying objects	22		5	13	1	1	2
d440	Fine hand use	1		1				
d445	Hand and arm use	6	1		3		1	1
d450	Walking	45	8	8	5	18	6	
d455	Moving around	32	12	4	6	10		
d470	Using transportation	4		1		2		1
d475	Driving	38	7	4	8	12	4	3
Chapter 5: Self-care								
d510	Washing oneself	6	1	3			2	

d520	Caring for body parts	3				1		2
d530	Toileting	4	1	1			2	
d540	Dressing	2		1	1			
d550	Eating	11						11
d560	Drinking	5				1		4
d570	Looking after one's health	57	5	12	3	16	5	16
Chapter 6: Domestic life								
d610	Acquiring a place to live	2		2				
d620	Acquisition of goods and services	15	3	4	2	6		
d630	Preparing meals	9		1	1	3		4
d640	Doing housework	33	1	5	24	3		
d650	Caring for household objects	2	1		1			
d660	Assisting others	2			2			
Chapter 7: Interpersonal interactions and relationships								
d7		5			1	1	2	1
d710	Basic interpersonal interactions	2		1			1	
d750	Informal social relationships	8	1		5	2		
d760	Family relationships	1					1	
d770	Intimate relationships	1						1
Chapter 8: Major life areas								
d850	Remunerative employment	31	2	6	12	4	4	3
d860	Basic economic transactions	1					1	
Chapter 9: Community, social and civic life								
d910	Community life	2					1	1
d920	Recreation and leisure	100	22	22	12	21	8	15

Table 5. Frequency of the 32 first- and second-level categories of the component Environmental Factors.

(A) Lymphedema with non-oncology background (mixed locations), (B) Lymphedema with oncology background (mixed locations), (C) Lymphedema of the upper limb (mixed backgrounds), (D) Lymphedema of the lower limb (mixed backgrounds), (E) Lymphedema in the genital region (mixed backgrounds) and (F) Lymphedema in the head and neck region.

ICF code	Description	Total	(A)	(B)	(C)	(D)	(E)	(F)
e		2						2
Chapter 1: products and technology								
e110	Products and substances for personal consumption	64	10	3	9	5	9	28
e115	Products and technology for personal use in daily living	288	64	60	17	97	36	14
e120	Products and technology for personal indoor and outdoor mobility and transportation	6			1	2	1	2
e130	Products and technology for education	5			1			4
e135	Products and technology for employment	3	1	1		1		
e140	Products and technology for culture, recreation and sport	3		3				
e155	Design, construction and building products and technology of buildings for private use	1		1				
Chapter 2: Natural environment and human-made changes to environment								
e2		3		3				
e210	Physical geography	1		1				
e220	Flora and fauna	1				1		
e225	Climate	44	13	20	1	1	2	7

Chapter 3: Support and relationships								
e3		6					4	2
e310	Immediate family	88	10	4	35	19	14	6
e315	Extended family	1	1					
e320	Friends	17	2	3	6	0	4	2
e325	Acquaintances, peers colleagues, neighbours and community members	23	2	3	12	4	1	1
e330	People in positions of authority	6			3		2	1
e335	People in subordinate positions	6		1	2	2	1	
e345	Strangers	3		2	1			
e350	Domesticated animals	3		1	2			
e355	Health professionals	156	15	26	27	34	14	40
Chapter 4: Attitudes								
e4		1					1	
e410	Individual attitudes of immediate family members	22		1		5		16
e425	Individual attitudes of acquintances, peers, colleagues, neighbours and community members	1	1					
e450	Individual attitudes of health professionals	10	3	7				
e460	Societal attitudes	19	1	6	6	4		2
Chapter 5: services, systems and policies								

e540	Transportation services, systems and policies	1					1	
e555	Associations and organizational services, systems and policies	9	1	1	5	1		1
e565	Economic services, systems and policies	4	2	2				
e580	Health services, systems and policies	199	29	24	32	22	30	62
e585	Education and training services, systems and policies	1		1				

Chapter 7

Identification of relevant ICF (International Classification of Functioning, Disability and Health) categories in lymphedema patients: A cross-sectional study

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Abstract

Background: To describe functioning and health of lymphedema patients and to identify their most common problems using the International Classification of Functioning, Disability and Health (ICF) as part of the preparatory studies for the development of ICF Core Sets for lymphedema.

Methods: Cross-sectional study in a population of lymphedema patients (n = 200), undergoing treatment in a Dutch lymphedema-specialised hospital. The second-level categories of the ICF were used to collect information on patients' problems in daily functioning.

Results: The mean age of the study group was 56 years (22-84). 78.5% of the patients were female. The most frequent mentioned items were: In the Body Functions component: muscle power and mobility of joints , in the Activities and Participation component: doing housework, and changing and maintaining a body position , in the Environmental Factors: Health professionals, who can act as both facilitators and barriers. Interestingly, patients assessed their health more positively than health professionals do.

Conclusion: By using the ICF, a considerable part of the broad spectrum of problems in functioning of lymphedema patients was reported.

Introduction

Congenital (primary) or acquired (secondary) lymphedema can affect patient's quality of life both physically and psychosocially (1). It is a chronic condition which is often progressive in time (2). Factors influencing the quality of life are pain, swelling and disturbed feelings in the affected body parts (2). Patients can experience a change in body image or psychological stability, limitations in activities and restrictions in participation and all this can depend on the localization of the lymphedema as well as on personal and environmental factors (3).

Describing the functioning, disability and health of a lymphedema patient, including facilitators and barriers, is an important part of understanding the life of a lymphedema patient and will help to define treatment goals.

The International Classification of Functioning, Disability and Health (ICF), is the World Health Organization (WHO) framework for measuring functioning and disability at both individual and population levels and is approved by the World Health Assembly (4).

The ICF can be used for many different purposes such as:

- a scientific basis for understanding and studying health and health-related states, outcomes and determinants
- a common language for describing health and health-related states in order to improve communication between different users, such as health care workers, researchers, policy-makers and the public, including people with disabilities
- a tool for comparison of data across countries, health care disciplines, services and time
- a systematic coding scheme for health information systems (4).

More than 1400 categories are described in the ICF, which is not very applicable in daily practice. Working with ICF Core Sets for a specific health condition (Comprehensive Core Set of 75-150 categories; Brief Core Set of 10-15 categories) allows the ICF to be a practical tool for health professionals (5).

Since there are no ICF Core Sets for lymphedema, a project was started, following the procedures of Cieza et al (6). According to these procedures, four preparatory studies were performed from: 1) the researchers' perspective (systematic review); 2) the patients' perspective (qualitative study); 3) the expert perspective (expert survey); and 4) the clinical perspective (cross-sectional study). These preparatory studies were followed by an international consensus conference, meant for experts in the field of lymphedema to decide on the ICF categories to be included in the ICF Core Sets for lymphedema. This article reports on the results of the cross-sectional study. The objective was to describe the functioning and disability of lymphedema patients and to identify the most common problems in lymphedema using the ICF.

Methods

Study design

The study design was a cross-sectional study among lymphedema patients. Patients were recruited from the department of the Dutch expert centre of lymphovascular medicine Nij Smellinghe Hospital, Drachten, the Netherlands between March 2013 and March 2014. This hospital is specialized in the treatment of lymphedema and the patients recruited were either inpatients or outpatients. In accordance with the declaration of Helsinki, a positive vote of the ethics committee was obtained prior to start and informed consent was obtained from all the patients.

Assessment

The ICF has two parts, each containing two separate components. Part 1 covers functioning and disability and includes the components Body Functions (b), Body Structures (s), and Activities and Participation (d). Part 2 covers contextual factors and includes the components Environmental Factors (e) and (non-classified) Personal Factors. In the ICF classification, the letters b, s, d, and e, that refer to the components of the classification, are followed by a numeric code starting with the chapter number (one digit) followed by the second level categories (two digits), and the more detailed third and fourth level categories (one digit each). For this study, the second level categories of the ICF were used. The ICF provides qualifiers which range from 0 – 4 for each category. Each category of the components Body Functions, Body Structures and Activities and Participation was graded with the qualifiers:
0 for ‘no impairment/limitation/restriction’,
1 for ‘mild impairment/limitation/restriction’,
2 for ‘ moderate impairment/limitation/restriction’,
3 for ‘severe impairment/limitation/restriction’ and
4 for ‘complete impairment/limitation/restriction’.
The categories of the component Environmental Factors were graded with
0 for ‘No facilitator/no barrier’,
+ 1 for ‘ mild facilitator’ and – 1 for ‘mild barrier’,
+ 2 or – 2 for ‘moderate facilitator or barrier’,

+3 or – 3 for ‘severe facilitator or barrier’ and
+ 4 or – 4 for ‘complete facilitator or barrier’.
Additionally the qualifier ‘8’ had to be used if the available information was not sufficient and ‘9’ if the category was not applicable (7).
The interviewers were instructed to report only limitations and impairment due to the lymphedema. If a patient had a limitation due to comorbidity that was not associated with lymphedema, this did not contribute to the prevalence of the corresponding ICF category and was given the qualifier ‘C’. Because it was not patient-friendly to question all categories of the ICF only those second-level ICF categories, that came forward from a previous literature study of lymphedema-specific questionnaires (8) were used for this study (i.e. 102 categories). A standardized form collected socio-demographic variables and comorbidities.
Additionally, health professionals had to rank the patients’ health and the magnitude of the problems of the functioning of the patient in everyday life on a scale ranging from 0 (no problem) to 10 (complete problem). Patients rated their health status in the same way as well.

Data collection procedures
Patients were recruited and interviewed by health professionals from several specialities trained in the principles of the ICF. Each health professional was given a personal training how to obtain the best information by performing a patient interview. Before an interview started, each patient’s medical record sheet was checked and relevant information on socio-demographic variables and diagnoses was extracted. During the interview the information was recorded using the online database Parantion (Parantion Web Survey 6.8), patient data being anonymous.

Data processing
For the ICF components Body Functions, Body Structures and Activities and Participation absolute frequencies of impairments/limitations mentioned by the study population were reported. For Environmental Factors absolute frequencies of specific categories that were named to be a barrier or facilitator were reported. ICF categories that were not mentioned by at least 10% of the study population were considered not relevant, except for the midline lymphedema items, because this condition is relatively rare. Responses of patients with combinations of lymphedema locations were excluded from the analysis.

Results

Two hundred patients were included in the study. Patients' ages ranged from 22 to 84 years, with a mean age of 56. Most patients were female (87,5%). The mean duration of lymphedema was 11 years (range of three months to 77 years). There were 88 patients with upper limb lymphedema, 89 patients with lower limb lymphedema and 8 patients with midline lymphedema (i.e. 2 patients with lymphedema of the thorax, 1 with lymphedema in the head and neck region and 5 patients with genital lymphedema). In addition, there were 15 patients with combinations of locations with lymphedema, who were excluded from the study.

Table 1. Characteristics of the patients (ULL = Upper Limb Lymphedema, LLL = Lower Limb Lymphedema)

Characteristic	Total (n=185)	ULL (n=88)	LLL (n=89)	Midline (n=8)
Mean age (range)	55 (22-83)	60 (23-83)	50 (22-78)	57 (42-72)
Female (%)	81%	98.9%	69.7%	12.5%
Duration lymphedema (years)	11.6	7.5	15.8	8.9
Employment (%)	44.8%	26.1%	60.7%	75%

There were 142 outpatients and 58 inpatients (treated for lymphedema). 27% had primary lymphedema. 20 health professionals interviewed the 200 patients: 56 by physicians (28%), 71 by nurses (35%), 57 by physical therapists (29%), and 16 by skin therapists (8%). These professionals rated the patients' health with a mean of 4.2 and the magnitude of the problems in functioning of the patient in everyday life with 4.4 on a scale ranging from 0 (no problem) to 10 (complete problem). Patients themselves rated their health at a mean of 2.5 and 38% (76 patients) reported no problems in daily life (mean 4.6).

Tables 2-5 show the most mentioned ICF categories with impairments, limitations, barriers or facilitators for the four ICF components.

Table 2 Most mentioned impairments in the component Body Functions.
N = 88 patients with lymphedema of the upper extremity (U; white coloured columns), N= 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). N= 8 patients with midline lymphedema (M; blue coloured columns). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no impairment			1 = mild impairment			2 = moderate impairment			3 = severe impairment			4 = complete impairment			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
b126	Temperament and personality functions	82	76	62	14	13	13	3	7	25	0	1	0	0	1	0	1	2	0
b152	Emotional functions	65	62	50	22	24	37.5	8	7	0	3	3	12.5	0	1	0	2	3	0
b530	Weight maintenance functions	51	64	75	35.5	26	25	10.5	9	0	2	1	0	0	0	0	1	0	0
b640	Sexual functions	72	79	50	8	10	12.5	0	4	0	1	0	12.5	2	0	12.5	17	7	12.5
b710	Mobility of joint functions	41	49.5	75	26.5	21.5	0	18.5	26	12.5	5	0	0	1	0	0	7	3	12.5
b730	Muscle power functions	43	58	75	24	25	12.5	20	14	0	5	2	0	0	0	0	7	1	12.5

Table 3 Most mentioned impairments in the component Body Structures.

N = 88 patients with lymphedema of the upper extremity (U; white coloured columns), N= 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). N= 8 patients with midline lymphedema (M; blue coloured columns). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no impairment			1 = mild impairment			2 = moderate impairment			3 = severe impairment			4 = complete impairment			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
s420	Structure of immune system	88		87.5	6		12.5	5			1						0		0
s720	Structure of shoulder region	48		75	22		12.5	16		12.5	7			2			5		0
s730	Structure of upper extremity	56		87.5	18		12.5	15			8						3		0
s740	Structure of pelvic region		68			18			3									11	
s750	Structure of lower extremity		30.3			30.3			30.3			6.1			1			2	
s810	Structure of areas of skin	84	74	75	7	18	12.5	8	6		1	2	12.5				0	0	0

Table 4 Most mentioned limitations and restrictions in the component Activities and Participation. N = 88 patients with lymphedema of the upper extremity (U; white coloured columns), N= 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). N= 8 patients with midline lymphedema (M; blue coloured columns). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no limitation/ restriction			1 = mild limitation/ restriction			2 = moderate limitation/ restriction			3 = severe limitation/ restriction			4 = complete limitation/ restriction			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
d410	Changing basic body position	68	49.5	4	16	30.5	12.5	11	16	12.5		1	12.5				5	3	12.5
d415	Maintaining a body position	50	66.3	62.5	28	21.7		14	10	12.5	5					12.5	3	2	12.5
d430	Lifting and carrying objects	27	59	62.5	32	19		30	9	12.5	5		12.5	3			3	2	12.5
d445	Hand and arm use	26		87.5	32			24		12.5	14			1			3		0
d450	Walking	67	36	62.5	10	36		8	18	12.5		6	12.5	1			14	4	12.5
d640	Doing housework	24	64.3	50	35	27.7	12.5	30	4	37.5	10	1					1	3	0
d920	Recreation and leisure	61.3	57.3	5	17	22.4	2	12.7	13.4	1	5	4.5		2			2	2.4	0

Table 5 Most mentioned barriers and facilitators in the component Environmental factors.
N = 88 patients with lymphedema of the upper extremity (U; white coloured columns), N= 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). N= 8 patients with midline lymphedema (M; blue coloured columns). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no barrier| facilitator			1 = mild barrier| facilitator			2 = moderate barrier| facilitator			3 = severe barrier| facilitator			4 = complete barrier|facilitator			Total 8 + 9 + C		
	Location	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
e125	Products and technology for communication	65	80	50	2|10	2.2|3.3		5|6	0|3.5		0|2	1|2	0|12.5	0|1			9	8	37.5
e225	Climate	41	36	50	20.4|6.6	20|6	25|0	20|2	26|3		2|1	1|1			0|1	12.5|0	7	6	12.5
e310	Immediate family	32	39.3	25	5|19	2.2|20	0|25	2|23	2.2|23	0|25	2|14	0|10	12.5|12.5	0|2	0|2.2		1	1.1	0
e320	Friends	45	49.7	50	3|24	2.2|29.2	0|25	0|13	1.1|7.8		0|2	0|7.8	0|25	|2			0	2.2	0
e355	Health professionals	30	26	25	5|19.3	8|16	0|37.5	1.1|20.4	7|20	12.5|0	2.3|19	0|20	12.5|12.5	0|2	0|2		1	1	0
e460	Societal attitudes	76	74.2	62.5	7|9	12.4|3		2|5	6|1.1	0|12.5	1|0		25|0				0	3.4	0
e580	Health services, systems and policies	49.2	45	37.5	9.1|16	7|19	0|37.5	2.3|10	6|10	12.5|0	0|9	0|5		0|1	1|1	0|12.5	3.4	6	0

The complete tables with all categories that were reported by at least 10% of the study sample are provided as Supplementary material online (Tables 6-9).

Body Functions

Most mentioned categories in Body Functions were for both the upper and lower limb affected patients: ‘b710, Mobility of joint functions’, ‘b730, Muscle power functions’, and ‘b530, Weight maintenance functions’. For the midline location the most mentioned categories were: ‘b152, Emotional functions’, ‘b126, Temperament and personality functions’, and ‘b640, Sexual functions’.

Body structures

For the upper limb the most mentioned categories were: ‘s720, Structure of shoulder region’, ‘s730, Structure of upper extremity’, and ‘s810, Structure of areas of skin’. The most mentioned categories in the lower limb were: ‘s750, Structure of lower extremity’, ‘s810, Structure of areas of skin’, and ‘s740, Structure of pelvic region’. For midline lymphedema patients the following categories were the most important: ‘s810, Structure of areas of skin’, ‘s720, Structure of shoulder region’, and ‘s420, Structure of immune system’.

Activities and Participation

In this component the most mentioned items for the upper extremity were: ‘d640, Doing housework’, ‘d445, Hand and arm use’, and ‘d430, Lifting and carrying objects’. For the lower limb these were: ‘d415, Maintaining a body position’, ‘d450, Walking’, and ‘d410, Changing basic body position’. For midline patients the most important categories were: ‘d640, Doing housework’, in‘d410, Changing basic body position’, and ‘d920, Recreation and leisure’.

Environmental factors

In this component there was a division in facilitators and barriers. The following categories for the upper extremity were most mentioned as facilitators: ‘e355, Health professionals’, ‘e310, Immediate family’, and ‘e320, Friends’. Whereas ‘e125, Products and technology for communication’, ‘e355, Health professionals’, and ‘e225, Climate’ were seen as barriers. Facilitators for the lower limb were the same as for the upper limb. For the barriers there was a slight difference: ‘e225, Climate’, ‘e460, Societal attitudes’, and ‘e355, Health professionals’ were most mentioned. Most mentioned facilitators for the midline region were: ‘e310, Immediate family’, ‘e355, Health professionals’, and ‘e580, Health services, systems and policies’. Barriers were: ‘e225, Climate’, ‘e355, Health professionals’, and ‘e460, Societal attitudes’.

Discussion

In this cross-sectional study the most common problems of lymphedema patients were explored using the ICF. The findings of this research make clear that all four components of the ICF (Body Functions, Body Structures, Activities and Participation, and Environmental factors) are relevant for patients with lymphedema.

In the component Body Functions, functions of joint mobility, muscle power, and weight maintenance were impaired most frequently for upper and lower extremity. A literature study about lymphedema-specific questionnaires presented impairments of the immunological system (swelling), pain and touch functions as most measured categories (8). Furthermore in another study using focus groups the most frequently mentioned categories were: immunological system functions, emotional functions (emotions such as fear, anger, joy); pain, temperament and personality functions (including psychic stability, confidence and optimism), and sensations related to the skin (including itching and tingling) (9). The results of these two studies show a rather broad spectrum of impairments, but the findings of this study are slightly different with more attention to mobility and muscle strength. Weight maintenance is of great importance, because overweight is one of the risk factors for developing lymphedema. This is in line with previous studies (10, 11). In this study 74.5% of the patients considered pain as no problem and 78.9% had no problems concerning swelling. In the focus groups all patients mentioned pain and swelling as problems, whereas in the lymphedema-specific questionnaires 75% contained questions about pain and 83% about swelling. This could be related to the specific treatment the patients received with more focus on mobility and muscle strength and the fact that most of the patients were in the maintenance phase (when the lymphedema has stabilized), in which swelling was no limitation any more. In the maintenance phase the lymphedema already exists for a longer period. Since the mean duration of the lymphedema was 11.6 years (see Table 1), it may be concluded that most of the patients were in this phase. In addition, midline patients have more problems mentally and with sexual functions. Although the patients with upper limb and lower limb lymphedema also mentioned problems with sexual functions (11.3% and 14.6% respectively), it is quite understandable that patients with lymphedema in the genital region had more problems (37.5%), considering the impact of having lymphedema in that region.

In the component Activities and Participation, most relevant categories pertained to recreation and leisure, doing housework and maintaining a body position. These are also the results of the focus group research (9), whereas in the literature review these items are less prominent (8). In the lymphedema-specific questionnaires the most occurring item is 'looking after one's health'. In the focus group research, all patients mentioned problems with self-management, which is no or only a small problem (6.5%) for the patients of this study. Probably this is a result of the self-management training as part of the treatment policy of the participating hospital.

Family, health professionals and friends were perceived as facilitators for the upper and lower limb in the component Environmental factors. For midline patients health services, systems and policies were also important facilitators. This emphasizes the importance of patients' social network and the influence of the health professionals. Although no discrimination was made between facilitators and barriers in the literature review as well as the qualitative review with focus groups, the same categories are important with one exception: 'Products and technology for personal use in daily living' was the most mentioned item in these studies (8, 9). This category contains items like bandaging and hosiery; which are important items for lymphedema patients. It remains unclear why these items play a less important role in this present study. Perhaps the patients in this study are more adapted to the use of these assistive products. Health professionals are also mentioned as important barrier. Patients describe the ignorance and play down of the health professionals when speaking of lymphedema during cancer treatment. In one of the interviews a patient gave an example of how the surgeon reacted, when she mentioned her problems with her swollen limb: "Why are you complaining about swelling in your arm? You are lucky to be alive." This is in line with the previous qualitative research. Consequently, there is a need for more education and a change in attitude concerning lymphedema among health professionals.

The participating health professionals rated the patients' health with a mean of 4.2 and the magnitude of the problems of the functioning of the patient with 4.4. Whereas the patients themselves rated their health with 2.5 and the problems of their daily living with 2.4. It seems that the health professionals overrate the level of problems that patients experience. The data of the patients are not in line with the previous qualitative research. In the latter, patients experience more problems than in this present research. This could be a result of the fact that in

the focus groups the topics were discussed more profound and the rather small sample (n = 31).
This study has some potential weaknesses. All patients were recruited from one single hospital. This could serve as a bias, because of the policy of treatment of lymphedema (focus on self-management) brings forward special attitudes of health professionals and patients, which cannot be generalised for the whole population. On the other hand this can be of advantage when patient groups from several institutes will be compared in future research; either from a therapeutic approach or demographic point of view. Furthermore, the recruitment was not performed at random or blinded. Although both limitations are obvious, the data of this study represent a considerable part of the distribution of problems in functioning of patients with lymphedema.

Conclusion

The purpose of this cross-sectional study was to provide data of patients with lymphedema as one of the preparatory studies for the development of ICF Core Sets for lymphedema. This resulted in a representation of the assessment of the patients' perspective by health professionals, using the ICF. The in-depth knowledge of the typical spectrum of problems encountered in lymphedema patients can contribute to the optimal management of lymphedema patients, the teaching of professionals, the planning of studies and the development of assessment instruments.

Conflict of interest
The authors report no conflicts of interest or financial ties in this research. Also there was no funding in any kind of way.

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Table 6. Percentages of the severity of the impairments in the component Body Functions.

Only those functions are included in the table in which there is an impairment (severity of 1 (mild impairment) or higher) in at least 10% of the 88 patients with lymphedema of the upper extremity (U; white coloured columns) or at least 10% of the 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). The blue coloured columns include the data of the eight patients with midline lymphedema (M). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no impairment			1 = mild impairment			2 = moderate impairment			3 = severe impairment			4 = complete impairment			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
Chapter 1: Mental functions																			
b122	Global psychosocial functions	90		75	7		12.5	2		12.5	1		0	0		0	0		0
b126	Temperament and personality functions	82	76	62	14	13	13	3	7	25	0	1	0	0	1	0	1	2	0
b130	Energy and drive functions	75	78	74	14	10	13	7	10	0	1	1	13	0		0	3	1	0
b152	Emotional functions	65	62	50	22	24	37.5	8	7	0	3	3	12.5	0	1	0	2	3	0
b160	Thought functions	88		87.5	8		12.5	1		0	2		0	0		0	1		0
b164	Higher-level cognitive functions			87.5			12.5			0			0			0			0
b180	Experience of self and time functions	89	89	87.5	9	4.5	12.5	1	5.5	0	1	0	0	0	0	0	0	1	0

Chapter 2: Sensory functions and pain																			
b265	Touch functions	67	85		19	10		7	3		2	1		1	0		4	1	
b270	Sensory functions related to temperature and other stimuli	76	71		16	16		5	6		1	4		1	0		1	3	
b280	Sensation of pain	72	76	88	15	16	13	6	7	0	1	0	0	0	0	0	6	1	0
Chapter 4: Functions of the cardiovascular, haematological, immunological and respiratory systems																			
b435	Immunolo-gical system functions	74	83	87.5	17	12.5	12.5	6	4.5	0	2	0	0	0	0	0	1	0	0
b455	Exercise tolerance functions	61	74		20	16		7	6		5	2		0	0		7	2	
Chapter 5: Functions of the digestive, metabolic and endocrine systems																			
b530	Weight maintenance functions	51	64	75	35.5	26	25	10.5	9	0	2	1	0	0	0	0	1	0	0
b540	General metabolic functions			87.5			12.5			0			0			0			0
b550	Thermoregula-tory functions	85.5		87.5	10.5		12.5	0		0	1		0	0		0	3		0
Chapter 6: Genitourinary and reproductive functions																			
b640	Sexual functions	72	79	50	8	10	12.5	0	4	0	1	0	12.5	2	0	12.5	17	7	12.5

Chapter 7: Neuromusculoskeletal and movement-related functions																			
b710	Mobility of joint functions	41	49.5	75	26.5	21.5	0	18.5	26	12.5	5	0	0	1	0	0	7	3	12.5
b730	Muscle power functions	43	58	75	24	25	12.5	20	14	0	5	2	0	0	0	0	7	1	12.5
Chapter 8: Functions of the skin and related structures																			
b810	Protective functions of the skin	67	64	87.5	19	22.5	12.5	8	8	0	3.5	1	0	0	0	0	2.5	4.5	0
b820	Repair functions of the skin	72	67	75	19	20	25	7	9	0	1	2	0	1	0	0	0	2	0
b830	Other functions of the skin	81	84	100	13	8	0	4	3.5	0	0	0	0	0	0	0	2	4.5	0

Table 7. Percentages of the severity of the impairments in the component Body Structures.
Only those functions are included in the table in which there is an impairment (severity of 1 (mild impairment) or higher) in at least 10% of the 88 patients with lymphedema of the upper extremity (U; white coloured columns) or at least 10% of the 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). The blue coloured columns include the data of the eight patients with midline lymphedema (M). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no impairment			1 = mild impairment			2 = moderate impairment			3 = severe impairment			4 = complete impairment			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
Chapter 4: Structures of the cardiovascular, immunological and respiratory systems																			
s420	Structure of immune system	88		87.5	6		12.5	5			1						0		0
Chapter 6: Structures related to the genitourinary and reproductive systems																			
s630	Structure of reproductive system			87.5			12.5												0
Chapter 7: Structures related to movement																			
s720	Structure of shoulder region	48		75	22		12.5	16		12.5	7			2			5		0
s730	Structure of upper extremity	56		87.5	18		12.5	15			8						3		0
s740	Structure of pelvic region		68			18			3									11	
s750	Structure of lower extremity		30.3			30.3			30.3			6.1			1			2	
s760	Structure of trunk	67	79		10	12		2			1						20	9	
Chapter 8: Skin and related structures																			
s810	Structure of areas of skin	84	74	75	7	18	12.5	8	6		1	2	12.5				0	0	0

Table 8. Percentages of the severity of the limitations and restrictions of the component Activities and Participation.
Only those limitations or restrictions are included in the table in which there is a limitation or restriction (severity of 1 or higher) in at least 10% of the 88 patients with lymphedema of the upper extremity (U; white coloured columns) or at least 10% of the 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). The blue coloured columns include the data of the eight patients with midline lymphedema (M). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

ICF code	Description	0 = no limitation/ restriction			1 = mild limitation/ restriction			2 = moderate limitation/ restriction			3 = severe limitation/ restriction			4 = complete limitation/ restriction			Total 8 + 9 + C		
Location		U	L	M	U	L	M	U	L	M	U	L	M	U	L	M	U	L	M
Chapter 1: Learning and applying knowledge																			
d155	Acquiring skills	83		87.5	5			8			2					12.5	2		0
d170	Writing	65			14			4			3						2		
d172	Calculating			87.5									12.5						0
Chapter 2: General tasks and demands																			
d230	Carrying out daily routine	61	75	75	16	15	25	16	8		3	1		1			2	1	0
Chapter 3: Communication																			
d330	Speaking			87.5						12.5									0
d335	Producing non-verbal messages			87.5			12.5												0
d340	Producing messages in formal sign language			87.5			12.5												0
d350	Conversation			87.5			12.5												0
d360	Using communication devices and techniques	90			8						2						0		

Chapter 4: Mobility																			
d410	Changing basic body position	68	49.5	4	16	30.5	12.5	11	16	12.5		1	12.5				5	3	12.5
d415	Maintaining a body position	50	66.3	62.5	28	21.7		14	10	12.5	5					12.5	3	2	12.5
d430	Lifting and carrying objects	27	59	62.5	32	19		30	9	12.5	5		12.5	3			3	2	12.5
d445	Hand and arm use	26		87.5	32			24		12.5	14			1			3		0
d450	Walking	67	36	62.5	10	36		8	18	12.5		6	12.5	1			14	4	12.5
d460	Moving around in different locations	81	59	87.5	7	19		3	7		2	2	12.5				7	2	0
Chapter 5: Self-care																			
d510	Washing oneself	80			13			5						1			1		
d520	Caring for body parts	82		87.5	11		12.5	5						1			1		0
d540	Dressing	76			15			5			1			2			1		
d550	Eating			87.5			12.5												0
d570	Looking after one’s health	85.2			10.3			3.5						1			0		
Chapter 6: Domestic life																			
d640	Doing housework	24	64.3	50	35	27.7	12.5	30	4	37.5	10	1					1	3	0
d660	Assisting others	69			15			11									5		
Chapter 7: Interpersonal interactions and relationships																			
d750	Informal social relationships	90	87	87.5	8	10	12.5	2	2								0	1	0
d770	Intimate relationships	84	74	75	3.4	16	12.5	5.6	5	12.5	1	2			1		6	2	0
Chapter 8: Major life areas																			
d845	Acquiring, keeping and terminating a job	33	57	62.5	2	7	12.5	9	7		8	4		3	1	12.5	45	24	12.5
d850	Remunerative employment	30	57	62.5	5	9	25	6	3		7	6		2			50	25	12.5
d870	Economic self-efficiency	54	71	87.5	7	7		6	4		5	6				12.5	28	12	0
Chapter 9: Community, social and civic life																			
d910	Community life	80	72	87.5	10	19		6	6	12.5	2						2	3	0
d920	Recreation and leisure	61.3	57.3	5	17	22.4	2	12.7	13.4	1	5	4.5		2			2	2.4	0

Table 9. Percentages of the severity of the barriers and facilitators in the component Environmental factors.
Only those environmental factors are included in the table that are barriers or facilitators in at least 10% of the 88 patients with lymphedema of the upper extremity (U; white coloured columns) or at least 10% of the 89 patients with lymphedema of the lower extremity (L; yellow coloured columns). The blue coloured columns include the data of the eight patients with midline lymphedema (M). 8 = not specified; 9 = not applicable; C = Comorbidity. The numbers for 8, 9 and C are added (Total 8,9,C).

<table>
<tr><th>ICF code</th><th>Description</th><th colspan="3">0 = no barrier| facilitator</th><th colspan="3">1 = mild barrier|facilitator</th><th colspan="3">2 = moderate barrier|facilitator</th><th colspan="3">3 = severe barrier|facilitator</th><th colspan="3">4 = complete barrier|facilitator</th><th colspan="3">Total 8 + 9 + C</th></tr>
<tr><td colspan="2">Location</td><td>U</td><td>L</td><td>M</td><td>U</td><td>L</td><td>M</td><td>U</td><td>L</td><td>M</td><td>U</td><td>L</td><td>M</td><td>U</td><td>L</td><td>M</td><td>U</td><td>L</td><td>M</td></tr>
<tr><td colspan="20">Chapter 1: Products and technology</td></tr>
<tr><td>e110</td><td>Products and substances for personal consumption</td><td>53.4</td><td>58.4</td><td>50</td><td>6.8|15</td><td>6.6|12.5</td><td>12.5|25</td><td>3.4|11.3</td><td>3|5</td><td></td><td>1.1|2</td><td>1|7.5</td><td>0|12.5</td><td>1|1</td><td>0|2</td><td></td><td>5</td><td>4</td><td>0</td></tr>
<tr><td>e115</td><td>Products and technology for personal use in daily living</td><td>61.3</td><td>63.5</td><td>62.5</td><td>2.7|15</td><td>1.5|15</td><td>0|12.5</td><td>3|3</td><td>0|6</td><td></td><td>0|0</td><td>2|1</td><td></td><td>0|1</td><td>0|1</td><td></td><td>14</td><td>10</td><td>25</td></tr>
<tr><td>e125</td><td>Products and technology for communication</td><td>65</td><td>80</td><td>50</td><td>2|10</td><td>2.2|3.3</td><td></td><td>5|6</td><td>0|3.5</td><td></td><td>0|2</td><td>1|2</td><td>0|12.5</td><td>0|1</td><td></td><td></td><td>9</td><td>8</td><td>37.5</td></tr>
<tr><td>e130</td><td>Products and technology for education</td><td>45.4</td><td></td><td>62.5</td><td>3.4|3.4</td><td></td><td></td><td>1|3.4</td><td></td><td></td><td>1|1</td><td></td><td></td><td></td><td></td><td>12.5|</td><td>41.4</td><td></td><td>25</td></tr>
<tr><td>e135</td><td>Products and technology for employment</td><td>53.4</td><td>70</td><td></td><td>2|5.6</td><td>0|8</td><td></td><td>0|1</td><td>1|0</td><td></td><td>1|1</td><td>0|1</td><td></td><td>1|0</td><td></td><td></td><td>35</td><td>20</td><td></td></tr>
<tr><td>e140</td><td>Products and technology for culture, recreation and sport</td><td>69.3</td><td>69.6</td><td>75</td><td>6|6</td><td>5.3|6.1</td><td></td><td>2|2</td><td>4|3</td><td></td><td>1|2</td><td>0|3</td><td>0|12.5</td><td></td><td></td><td></td><td>11.7</td><td>9</td><td>12.5</td></tr>
<tr><td>e165</td><td>Assets</td><td>72</td><td></td><td>37.5</td><td>1|1</td><td></td><td>0|25</td><td>2|5</td><td></td><td></td><td>1|0</td><td></td><td></td><td></td><td></td><td></td><td>18</td><td></td><td>37.5</td></tr>
</table>

Chapter 2: Natural environment and human-made changes to environment																			
e210	Physical geography			37.5												12.5|0			50
e225	Climate	41	36	50	20.4|6.6	20|6	25|0	20|2	26|3		2|1	1|1			0|1	12.5|0	7	6	12.5
Chapter 3: Support and relationships																			
e310	Immediate family	32	39.3	25	5|19	2.2|20	0|25	2|23	2.2|23	0|25	2|14	0|10	12.5|12.5	0|2	0|2.2		1	1.1	0
e315	Extended family	67.4	74.2	50	4.3|9.3	2.2|11.6	0|25	0|6	2|1		0|5	0|1					7	8	25
e320	Friends	45	49.7	50	3|24	2.2|29.2	0|25	0|13	1.1|7.8		0|2	0|7.8	0|25	|2			0	2.2	0
e325	Acquaintan-ces, peers colleagues, neighbours and community members	52.2	58.4	62.5	2.2|19	3.4|27	0|25	5|10.2	3.4|5.6	0|12.5	0|8	0|1.1		0|2.2			1.2	1.1	0
e330	People in positions of authority	47	45	62.5	5|5	3|11	0|12.5	2|3	|11		1|3	1|1	12.5|0	1|0			33	17	12.5
e355	Health professionals	30	26	25	5|19.3	8|16	0|37.5	1.1|20.4	7|20	12.5|0	2.3|19	0|20	12.5|12.5	0|2	0|2		1	1	0

Chapter 4: Attitudes																			
e410	Individual attitudes of immediate family members	44	55	37.5	5|18	3.3|12	0|25	6|16	0|21.3		1|7	0|7.3	12.5|37.5	0|1			2	1.1	0
e415	Individual attitudes of extended family members	70	82	50	7|5	0|7	0|12.5	2|6	1|2		0|2	1|1	0|12.5				8	6	25
e420	Individual attitudes of friends	57	65	50	5|20	2.2|16.8	0|25	0|8	2.2|7.8		1|8	1|3	0|25	0|1			0	2	0
e425	Individual attitudes of acquintan-ces, peers, colleagues, neighbours and community members	66	73	62.5	5|10	3.4|12.4	0|12.5	3|10	4.5|4.5	0|12.5	0|5	0|1.1	0|12.5	0|1			0	1.1	0
e460	Societal attitudes	76	74.2	62.5	7|9	12.4|3		2|5	6|1.1	0|12.5	1|0		25|0				0	3.4	0
e465	Social norms, practices and ideologies	81	82	75	5.4|3.4	9|1.1		3.4|3.4	3.4|1.1	0|12.5							3.4	3.4	12.5

Chapter 5: Services, systems and policies																			
e535	Communica-tion services, systems and policies	76	81		2|5	0|6		1|6	0|2		1|0	0|2					9	9	
e570	Social security services, systems and policies	61.4	70	62.5	7|12.6	4.5|3.4	2.5|12.	1|5	4.5|2		1|1			1|0	1|		10	14.6	12.5
e580	Health services, systems and policies	49.2	45	37.5	9.1|16	7|19	0|37.5	2.3|10	6|10	12.5|0	0|9	0|5		0|1	1|1	0|12.5	3.4	6	0
e585	Education and training services, systems and policies	46.6		50	1.1|2.3			4.5|3.4		12.5|0	1.1|2.3						38.7		37.5

Chapter 8

Functioning in lymphoedema from the health professionals' perspective using the International Classification of Functioning, Disability and Health (ICF) as a reference

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Abstract

Purpose: To capture the views of different health care providers involved in the treatment of patients with lymphoedema from various countries around the world on the functioning of lymphoedema patients and the factors influencing functioning of these patients using the International Classification of Functioning, Disability and Health (ICF).

Method: A world-wide e-mail survey with questions based on components of the ICF.

Results: In total, 142 health professionals from seven different health professions and 20 different countries answered the questions. The aspects of functioning that were named by the health professionals could be linked to 359 different ICF categories. Of these categories, 109 belonged to body functions (30.4%), 55 to body structures (15.3%), 121 to activities and participation (33.7%), and 74 to environmental factors (20.6%). Overall, the most mentioned items were: Health services, systems and policies, Immunological system functions, Looking after one's health, Products and technology for personal use in daily life, and Dressing .

Conclusions: The ICF provided a valuable reference for identifying concepts in statements from international health care professionals experienced in the treatment of lymphoedema patients. The results of this research will be used in the development of ICF Core Sets for lymphoedema.

Introduction

Lymphoedema is a chronic condition that is characterised by abnormal accumulation of lymph fluid in the body. It occurs when the lymph vessels are unable to drain this fluid. It is caused by damage to or the absence of lymph vessels and/or excessive production of extracellular fluid. Lymphoedema can occur in both the upper and lower limbs as well as in the midline (head, neck, abdomen, and thorax). There are two types of lymphoedema: primary and secondary (1).

Primary lymphoedema
Sometimes lymphoedema is genetic, which is called primary lymphoedema. Either there are fewer lymphatic vessels or lymph nodes or the lymphatic vessels/nodes work less effectively. Some genetic types are visible at birth (Milroy's disease), others can occur later in life (Lymphedema Tarda). Primary lymphoedema can occur in any part of the body, but it is most common in the legs, and it starts during adolescence or sometimes earlier (2-7).

Secondary lymphoedema
Secondary lymphoedema is more common than primary lymphoedema. Secondary lymphoedema is caused by damage to the lymph vessels or lymph nodes; it can occur after an injury such as surgery or due to infection. In the Western world, lymphoedema is well-known after cancer treatment as a result of the removal of lymph nodes (mostly in the axilla or groin) and/or radiotherapy (8). Besides the result of cancer treatment in the pelvic organs, lymphoedema in the legs can also be caused by past infections (erysipelas). In the developing world, parasites (filariasis) are responsible for millions of cases of patients with lymphoedema (9). Patients with lymphoedema experience slower wound healing and heaviness and tightness of the limbs, and they have a higher risk of infection (10-12), all of which can lead to loss of quality of life because of the limitations in activities related to personal care, occupations, domestic life and socialisation (13). Lymphoedema requires lifelong skin care and precautions to minimise swelling or recurrent swelling. Patients are advised to practice certain regimes, such as daily bandaging or the use of special garments, caution with open wounds, and skin care.

ICF

An important basis for the optimal treatment and management of lymphoedema consists of an in-depth understanding, systematic consideration, and sound measurement of the consequences of lymphoedema at different levels of a person's functioning. To obtain this basis, it is necessary to develop a comprehensive framework and classification that can serve as a universal language, understood by health professionals, researchers, policy makers, patients, and patient organisations. The International Classification of Functioning, Disability and Health (ICF) (14, 15) is an international classification system in which the terms and classes have been designed to describe the functioning, disability and health of people. The classes/categories of the ICF consist of a title, a description and a code. The ICF organises information into two parts. Part 1 describes functioning and consists of the components 'Body Functions and Structures' and 'Activities and Participation'. Part 2 addresses contextual factors and consists of 'Environmental Factors' and 'Personal Factors'. There is not yet a list of Personal Factors in the ICF because of the large social and cultural variations associated with these factors. The ICF has a hierarchical structure of different levels (1 to 4). Each component contains four levels that can be used to describe items more detailed. So how higher the level , the more detailed the description of the item. The ICF has made it possible to link data across conditions or interventions with the goal of achieving efficient and cost-effective health care (16). The ICF classification system contains all items to describe functioning of patients, regardless their health condition. For every health condition health-specific selections of ICF categories (ICF Core Sets) (17) can be determined, making the ICF easier to implement in medical/allied health care (18). ICF Core Sets include the correct terms and classes to describe the functioning of persons with specific health problems, making the formulation of treatment goals easier, more transparent, and faster. ICF Core Sets for lymphoedema have not yet been developed. Stucki et al. introduced a standard method (see Figure 1) to develop ICF Core Sets (17). The expert research described in this article is one of the 'preparatory studies', which results will be incorporated into the international consensus conference to construct the first versions of ICF Core Sets for lymphoedema. The objective of this expert survey was to explore expert perspectives regarding the relevant problems of individuals with lymphoedema. The specific aims were as follows: (1) to identify problems in functioning that are important to patients with lymphoedema; and (2) to quantify these problems using the ICF.

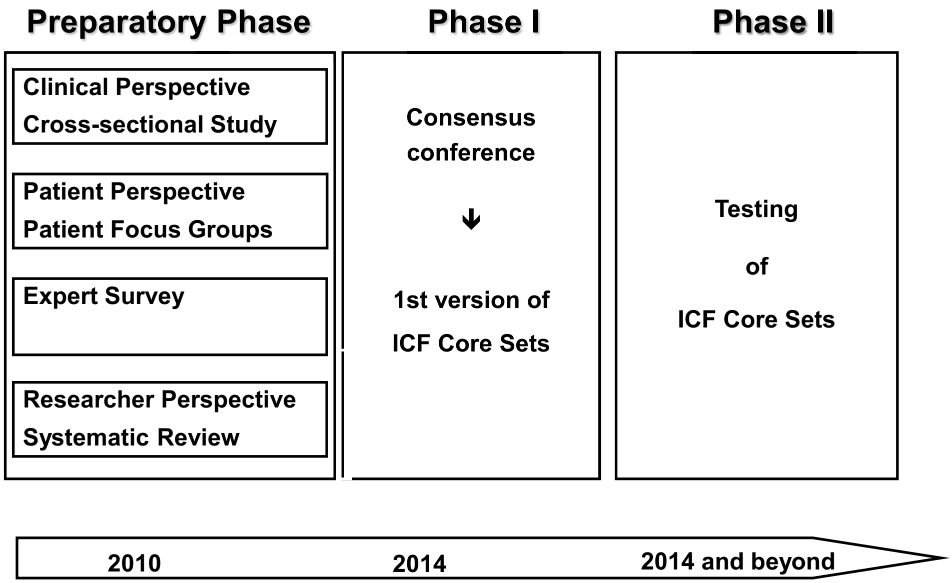

Figure 1. Development of ICF Core Sets for lymphedema: phases of the project.

Methods

Design

In an international Internet-based survey, health professionals from different health professions, who were experts in the treatment of lymphoedema, were asked five open-ended questions about factors relevant to describe the functioning of patients suffering from lymphoedema. In addition, basic socio-demographic characteristics of the experts were recorded. The questions and answers were provided in plain English and were subsequently linked to ICF categories by two independent researchers (PV, LP) according to the linking rules of Cieza et al. (19, 20).

Participants

Health professionals from all six WHO world regions were included: Africa, North and South America, the Eastern Mediterranean region, Europe, Southeast Asia, and Western Pacific. The following health professions were included in the study: physical therapists, nurses, occupational therapists, skin therapists, psychologists, phlebologists, dermatologists, and lymphologists. These health care providers are considered relevant for the treatment of lymphoedema. The inclusion criteria required experience with lymphoedema patients for at least one year and mastery of the English language.

During the preparatory phase of the study, professional associations of health professions engaged in the treatment of lymphoedema worldwide were identified and contacted by means of an Internet search. Furthermore, another Internet search and personal recommendations were used to identify health professionals experienced in the treatment of patients with lymphoedema. Finally, an informative recruitment presentation was given at the international congress of the International Society for Lymphoedema (ISL) in Rome in September 2013. All associations were asked to distribute a letter among their members with information about the study and how to participate. Each association that did not respond within two weeks received a reminder, as well as a second reminder after four weeks.

Data collection

All health professionals who were willing to participate were invited to enter an Internet-based survey platform with a password. They were asked to answer five open-ended questions that asked them to name relevant changes due to lymphoedema and its treatment. The questions covered the ICF chapters on Body Functions, Body Structures, Activities and Participation, and contextual Environmental and Personal Factors that affect life (Table 1). For each question, a health professional could write as many items as he or she considered relevant. The participants could not see each other's answers. Open-ended questions were applied to avoid any bias included in predetermined closed-ended questions and to obtain as much information as possible. Furthermore, the participants were asked to provide some (socio-demographic) information about their age, gender, health profession, country of practice, and years of professional expertise.

Table 1 Questions presented in the survey.

ICF component	Survey question
Body functions	If you think about the body and mind of individuals with lymphedema, what does not work the way it is supposed to?
Body structures	If you think about the body of individuals with lymphedema, in which parts are their problems ?
Activities and participation	If you think about the daily life of individuals with lymphedema, what are their problems?
Environmental factors	If you think about the environment and the living conditions of individuals with lymphedema, what is supportive and/or what is hindering for them?
Personal factors	If you think about individuals with lymphedema, what is important about them and the way they handle their condition?

Linking process

After receiving answers in plain English from all of the participating health professionals, the answers were transmitted (linked) to the ICF, based on the linking rules of Cieza et al (19, 20). This survey followed rule 1 ("Before starting the process of linking to the ICF categories, identify all meaningful concepts within the answers under consideration.") and 2 ("Each meaningful concept is linked to the most precise ICF category". An answer could contain several meaningful concepts, depending on the number of themes contained in the answer, and each meaningful concept could be linked to one or more ICF categories. Linking was performed using all levels (1 to 4). To overcome the possible bias of any health profession, the linking was performed by two different health professionals: a physical therapist (PV) and a skin therapy student (LP). The linking was independently performed, and when the professionals could not agree, an expert in ICF (YH or DvR) was consulted.

Analyses of answers not linked to detailed ICF categories

Some indicated concepts could not be classified within the ICF, namely personal factors ('pf'), health condition ('hc'), concepts that related to the ICF but could not be assigned a particular component or code ('nd'), concepts referring to health-related quality of life ('qol'), concepts related to general health ('gh'), concepts related to physical health ('ph'), and concepts outside of the ICF framework ('nc').

Accuracy of the linking process

The degree of agreement between the two health professionals regarding the linked ICF categories was calculated using kappa statistics, with 95% bootstrap confidence intervals. The kappa coefficient values generally range from 0 to 1, where 1 indicates perfect agreement and 0 indicates no agreement beyond what is expected by chance alone. A kappa value of 0.6 or greater is considered sufficient (21).

Results

Participants

All of the data were obtained between November 2013 and March 2014. One hundred four associations were approached, of which nine responded to the first letter and one after the first reminder (9.6%). The other associations did not respond, although it is possible that they sent the information letter to their members without responding to the invitation e-mail. The reasons for not answering are unknown but might include faulty e-mail addresses, spam filters, and a lack of interest in the survey.

In total, 142 health professionals answered the questions. The participating health professionals had a median age of 49 years (age range, 26-74 years) and had been working in the field of lymphoedema for a median of 13 years (range, 1-40 years). The male to female ratio was 19:123. The 142 health professionals belonged to seven different health professions: 86 (60.6%) were physical therapists, 19 (13.4%) were nurses, 17 (12.0%) were lymphologists, 10 (7.0%) were occupational therapists, six (4.2%) were dermatologists, three (2.1%) were skin therapists, and one (0.7%) was a phlebologist. In terms of the WHO world regions in which the respondents practiced, four of the six regions were represented: there were 86 participants (60.6%) from the European region (Austria, Denmark, France, Germany, Ireland, Israel, Italy, the Netherlands, Portugal, Romania, Spain, Sweden, Switzerland, Turkey, and the United Kingdom), 32 (22.5%) from the Western Pacific region (Australia and New Zealand), 21 (14.8%) from North America (Canada and the United States), and three (2.1%) from Southeast Asia (India). There were no participants from Africa, the Eastern Mediterranean region or South America. Of the participants, 73 (51.4%) were working in hospitals, 69 (48.6%) in private practices, 16 (11.3%) in rehabilitation centres, 11 (7.7%) in university settings, eight (5.6%) in inpatient practices, six (4.2%) in homes for the elderly, and five (3.5%) in nursing homes. Some of the participants worked in more than one location.

Linking process

In total, the answers provided by the health professionals included 7373 aspects of functioning, of which 6296 (85.4%) could be linked to 359 different ICF categories. Of the 359 identified ICF categories, 109 were body functions (30.4%), 55 were body structures (15.3%), 121 were activities and participation (33.7%), and 74 were environmental factors (20.6%).

One thousand seventy-seven answers (14.6%) could not be linked to ICF categories, of which 472 answers (6.4%) referred to the Personal Factor components, and 267 answers (3.6%) covered aspects of the underlying health condition and, consequently, were labelled ‘health condition’. All of the answers that were too general to be linked to an ICF category but that belonged to domains of the ICF (n=158) were labelled ‘not definable’ (2%). Nineteen were labelled ‘not definable-general health’ (0.25%), sixteen were labelled ‘not definable-quality of life’ (0.2%), and fifty-six were labelled ‘not definable-physical health’ (0.76%). Eighty-nine answers (1.2%) were labelled ‘not covered’ (see Table 2).

Table 2. Categories not linked to the ICF.

Code	Meaning	N
nc	Not covered	89
nd	Not definable	158
nd-gh	Not definable- general health	19
nd-hc	Not definable-health condition	267
nd-ph	Not definable –physical health	56
nd-qol	Not definable-quality of Life	16
pf	Personal factor	472
Total		1077

The final inter-linker concordance was 0.619 (95% confidence interval, 0.606-0.632). After the linking process of every 20th expert, there was consultation between the two health professionals.

Frequency analysis of ICF categories

There were a total of 359 different ICF categories. Of these, 26 (7.2%) pertained to first-level categories (that is on the level of ICF chapters, not further detailed), 120 (33.5%) to second-level categories, 201 (56%) to third-level categories, and 12 (3.3%) to fourth-level categories. For reasons of readability, all of third and fourth categories (the more detailed descriptions) were merged into the second-level categories, resulting in 181 first- and second-level categories. Tables 3 through 6 list the first- and second-level ICF categories, linked to ten or more meaningful concepts. This cut-off was also chosen for readability.

Body functions

After merging the third and fourth levels into the second-level categories, there were 54 first- and second-level Body Functions categories, of which 24 categories were used more than 10 times. The five most frequently used second-level categories of the Body Functions component were:

1) Immunological system functions (b435), e.g., impairments of the lymphatic function leading to swelling
2) Temperament and personality functions (b126), e.g., openness to trying different or new treatments, optimism and psychological and emotional stability
3) Emotional functions (b152), e.g., fear, anger, sadness and happiness
4) Experience of self and time functions (b180), e.g., body image, feeling too heavy, and
5) Sensation of pain (b280).

The most used first-level category was Chapter 7 ‘Neuromusculoskeletal and movement-related functions’.

Table 3. First and second-level ICF categories Body Functions (n ≥ 10)

ICF Code	Description	Total
b1	CHAPTER 1 MENTAL FUNCTIONS	24
b126	Temperament and personality functions	197
b130	Energy and drive functions	59
b152	Emotional functions	127
b160	Thought functions	10
b164	Higher-level cognitive functions	62
b180	Experience of self and time functions	122
b280	Sensation of pain	99
b415	Blood vessel functions	26
b435	Immunological system functions	563
b440	Respiration functions	11
b455	Exercise tolerance functions	28
b530	Weight maintenance functions	35
b540	General metabolic functions	13
b640	Sexual functions	19
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS	54
b710	Mobility of joint functions	44
b730	Muscle power functions	15
b755	Involuntary movement reaction functions	15
b780	Sensations related to muscles and movement functions	14
b8	CHAPTER 8 FUNCTIONS OF THE SKIN AND RELATED STRUCTURES	39
b810	Protective functions of the skin	35
b820	Repair functions of the skin	22
b840	Sensation related to the skin	57
Total		1690

Body structures

There were 23 first- and second-level Body Structures categories, of which 10 were used more than 10 times. In this component, the five most frequently used second-level categories were:

1) Structure of lower extremity (s750)
2) Structure of upper extremity (s730), both because of altered representation of the limb
3) Structure of reproductive system (s630), e.g., breasts and genital regions
4) Structure of trunk (s760), and
5) Structure of immune system (s420), lymphatic vessels and nodes.

Of the first-level categories Chapter 7 ‘Structures related to movement’ was the most frequently used.

Table 4. First and second-level ICF categories Body Structures (n ≥ 10)

ICF code	**Description**	**Total**
s	BODY STRUCTURES	39
s420	Structure of immune system	72
s630	Structure of reproductive system	85
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT	62
s710	Structure of head and neck region	63
s730	Structure of upper extremity	135
s750	Structure of lower extremity	150
s760	Structure of trunk	77
s8	CHAPTER 8 SKIN AND RELATED STRUCTURES	13
s810	Structure of areas of skin	48
Total		744

Activities and participation

In the component Activities and Participation, there were 53 first- and second-level categories, of which 26 were used more than 10 times. The five most frequently used second-level categories in this component were:

1) Looking after one’s health (d570), e.g., maintaining one’s health and managing diet and fitness
2) Dressing (d540), e.g., finding appropriate clothing and shoes
3) Recreation and leisure (d920)
4) Remunerative employment (d850), and
5) Doing housework (d640).

The most frequently used first-level category was Chapter 4 ‘Mobility’.

Table 5. First and second-level ICF categories Activities and Participation (n ≥ 10).

ICF code	**Description**	**Total**
d230	Carrying out daily routine	63
d360	Using communication devices and techniques	13
d4	CHAPTER 4 MOBILITY	72
d410	Changing basic body position	23
d415	Maintaining a body position	50
d430	Lifting and carrying objects	22
d440	Fine hand use	11
d445	Hand and arm use	25
d450	Walking	48
d455	Moving around	36
d470	Using transportation	19
d475	Driving	29
d5	CHAPTER 5 SELF-CARE	44
d510	Washing oneself	18
d520	Caring for body parts	54
d540	Dressing	285
d570	Looking after one’s health	385
d620	Acquisition of goods and services	12
d640	Doing housework	66
d650	Caring for household objects	13
d7	CHAPTER 7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS	35
d770	Intimate relationships	14
d845	Acquiring, keeping and terminating a job	13
d850	Remunerative employment	118
d870	Economic self-sufficiency	35
d920	Recreation and leisure	176
Total		1679

Environmental factors

Of the 51 first- and second-level categories belonging to the component ‘Environmental factors’, 24 were used more than 10 times. The five most frequently used categories were:

1) Health services, systems and policies (e580), e.g., all medical treatments and institutions
2) Products and technology for personal use in daily life (e115), e.g., bandages and hosiery
3) Health professionals (e355)

4) Acquaintances, peers, colleagues, neighbours and community members (e325), and
5) Climate (e225).

The most frequently used first-level category was Chapter 3 ‘Support and relationships’. Environmental factors can be both inhibiting and facilitating.

Table 6. First and second-level ICF categories Environmental factors (n ≥ 10).

ICF code	**Description**	**Total**
e	ENVIRONMENTAL FACTORS	10
e110	Products or substances for personal consumption	14
e115	Products and technology for personal use in daily living	299
e120	Products and technology for personal indoor and outdoor mobility and transportation	15
e135	Products and technology for employment	12
e155	Design, construction and building products and technology of buildings for private use	42
e165	Assets	12
e210	Physical geography	19
e225	Climate	64
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS	109
e310	Immediate family	32
e320	Friends	24
e325	Acquaintances, peers, colleagues, neighbours and community members	109
e340	Personal care providers and personal assistants	22
e355	Health professionals	135
e4	CHAPTER 4 ATTITUDES	12
e450	Individual attitudes of health professionals	58
e460	Societal attitudes	53
e535	Communication services, systems and policies	11
e540	Transportation services, systems and policies	10
e555	Associations and organizational services, systems and policies	11
e570	Social security services, systems and policies	27
e580	Health services, systems and policies	686
e585	Education and training services, systems and policies	35
Total		1789

Overall, the most frequently used second-level items were:

1) Health services, systems and policies (e580): 686 times
2) Immunological system functions (b435): 563 times
3) Looking after one's health(d570): 385 times
4) Products and technology for personal use in daily life (e115): 299 times, and
5) Dressing (d540): 285 times.

Discussion

In this research, experiences with the treatment of lymphoedema from the perspective of health professionals were collected. In this procedure, the ICF was used to translate multiple answers into one standard (WHO) language. Most of the aspects of functioning of the lymphoedema patient, valued by the health professionals, were covered by the ICF. This is in line with previous studies in the field of lymphoedema (22-24). By linking to the ICF it is possible to compare study results obtained from different regions of the world, regardless of the study designs and outcome measures (18, 20, 25).

A very small portion of the answers (1.2%) referred to aspects that are labelled 'not covered' by the ICF, because they are outside of the ICF framework. This finding emphasises the relevance of the ICF for lymphoedema. Examples of these aspects included life, morbidity, cosmetics and pathophysiology.

Personal factors are not yet covered by the ICF, although they belong to the contextual factors of the WHO bio-psychosocial model of functioning (15, 26). In this survey, 6.4% of the mentioned aspects referred to personal factors. Frequently mentioned personal factors were comorbidities, lifestyle, and coping style. Including a list of personal factors within the ICF framework seems appropriate and will increase the relevance of the ICF classifications for describing functioning of clients with different health conditions, including clients with lymphoedema (27). Some attempts to make such a list have already been undertaken (28-30).

Most of the health professionals were physical therapists (n=86; 60.6%), whereas 24 physicians (16.9%) participated. The very high response rate of physical therapists compared with physicians can be partly explained by the high involvement of physical therapists in the treatment of clients with lymphoedema. Another reason for the high response rate of the physical therapists could be that they are more used to the ICF and perhaps had more time to participate. The overrepresentation of physical therapists can be considered a limitation of the present study. On the other hand, the answers of the physicians were not very different compared to those of the physical therapists and the distribution of the population reflects the real-life distribution of disciplines working in the field of lymphoedema. A limitation was that psychologists are not included in the survey, which might have underestimated the value of psychological categories. Comparable problems were also seen in similar other studies (31, 32).

This study was also designed to cover perspectives from different cultural backgrounds. However, equal distribution in terms of WHO regions could not be attained. Although health professionals in various countries were initially contacted, in the final sample there was a predominance of respondents from the Western world (Europe, Australia and North America). There were no experts involved from the Eastern Mediterranean region, South America and Africa. The distribution across the WHO world regions however is consistent with that of previous ICF Core Set projects (33, 34). A reason for this might be the inclusion criterion of 'mastery of the English language' in countries where English is not the first language. Other reasons could be related to the health care infrastructure for lymphoedema and the accessibility of the Internet. For these reasons, the conclusions drawn from this study are only applicable to the Western world. The value of this study for the other WHO regions could therefore be low, although many aspects could be of importance for those regions also.

The response to the mail directed at the professional organisations was low. As the reasons for not answering are unknown, this could serve as a bias. However, it could well be that the associations did indeed disseminate the invitations among their members, but the members did not cooperate. The most plausible reason, besides language, in this study for the low response seems to be the time required to read all of the information sent and to answer the questions.

Frequently, the health professionals described the concepts in more detail than the corresponding categories in the ICF; e.g., hyperaesthesia (b2702, Sensitivity to pressure) and driving a bicycle (d4750, Driving human-powered transportation). Regarding this difference, one should bear in mind that the ICF is a reference classification to be used worldwide in all health care situations. It forms the basis for the description of the functioning of the patient, but additional information - information specific to the health condition, the setting and/or personal situation of the patient - must be added. Earlier, Weigl et al (25) described these findings as well. With regard to the concepts that were not able to be linked to an ICF category or domain, it is important to note that the ICF is a classification of functioning not of disease (25% of the non-ICF-linkable concepts are health conditions). For health conditions or diseases the International Classification of Diseases (ICD) can be used. The fact that the experts respond to the question about functioning by bringing up the health condition indicates perhaps that there is still great potential to convince experts in the field that functioning is complementary to the health condition and not solely as a consequence of a health condition. Moreover, ICF and ICD can be used complementary (35, 36).

The authors have presented the five most frequently mentioned categories for the second-level categories of the ICF. Also presented in Tables 3-6 are the categories of the first level of the ICF that were used more than 10 times. These categories have been used to code meaningful concepts that were too broad to classify in detail but that represented a significant proportion of the results. The five most frequently used first levels were Support and relationships (d7), Mobility (d4), Structures related to movement (s7), Neuromusculoskeletal and movement-related functions (b7), and Self-care (d5). Topics related to first levels ‘Support and relationships’ and ‘Self-care’ were also prominently represented in the top five most frequently found second-level categories. In contrast, the remaining first levels -- ‘Mobility’, ‘Structures related to movement’ and ‘Neuromusculoskeletal and movement-related functions’ (all related to movement and not represented in the top five of the second-level categories) --- emphasised the importance of these items to the contributing health professionals, although they provided rather vague descriptions.

The results of the present study resemble the findings of a literature review of ICF categories on lymphoedema-specific questionnaires, which was also a part of the project to develop ICF Core Sets for lymphoedema (22). In this review, the second-level categories of the ‘Body Functions’ component that were found most frequently were ‘(Immunological system functions (b435)’ (10 questionnaires), ‘ Sensation of pain (b280)’ (9 questionnaires) and ‘ Touch functions (b265)’ (8 questionnaires). In the ‘Body Structures’ component, the most frequently found categories were ‘ Structure of upper extremity (s730)’ (10 questionnaires) and ‘ Structure of reproductive system (s630)’ (6 questionnaires). The most frequently found categories in ‘Activities and Participation’ were: ‘ Looking after one’s health (d570)’ (9 questionnaires), ‘ Dressing (d540)’ (7 questionnaires), ‘ Hand and arm use (d445)’ (6 questionnaires) and ‘ Remunerative employment (d850)’ (6 questionnaires as well). The most frequently found ‘Environmental Factors’ were: ‘ Health services, systems and policies (e580)’ (7 questionnaires) and ‘ Products and technology for personal use in daily living (e115)’ (6 questionnaires). Except for the items related to movement, one can see that all of the categories important to the experts were represented in the questionnaires. Thus, it is advisable to insert some questions about mobility into future questionnaires as well.

In a qualitative research study, also part of the project on ICF Core Sets for lymphoedema (publication in preparation), important categories from the patient perspective were studied. The results correspond to the findings of this expert research. However, the following categories were more valued by patients than by health professionals: Sensations related to the skin (b840) (including itching and tingling), Maintaining a body position (d415) (lying, sitting, standing, etc.), Walking (d450), Driving (d475), Immediate family (e310), and Products and substances for personal consumption (e110). The category Experience of self and time functions (b180) (body image) was not mentioned often by the patients but was frequently noted by the health professionals. Most likely, the patients in this research did not have many difficulties with their body image.

This survey was one of the preparatory studies in the development of ICF core sets for lymphoedema and it tried to capture the views of different health care providers involved in the treatment of patients with lymphoedema from various countries around the world on the functioning of lymphoedema patients and the factors influencing functioning of these patients. The ICF has proven to be a practical tool for the translation of problems encountered in lymphoedema, and it is sufficiently comprehensive, although not always detailed enough, to address the points of view of different health professionals involved in the treatment of patients with lymphoedema.

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Conflicts of interest

The authors report no conflicts of interest or financial ties in this research.

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Chapter 9

Development of consensus international classification of functioning, disability, and health (ICF) Core Sets for lymphedema

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ABSTRACT

To understand the challenges of patients with lymphedema it is important to describe functioning and to measure the effectiveness of treatment in changing functioning. The International Classification of Functioning, Disability and Health (ICF) offers an international framework to classify functioning of persons in their personal environment. ICF Core Sets are lists of selected ICF categories concerning those important aspects of functioning that are most likely to be affected by a specific health problem or disease. These Core Sets make it easier and faster to describe and communicate the patient's problems and to define treatment goals. Furthermore, they are available to health care providers of all professions, researchers, health insurance companies and policy-makers. The objective of this document is to present the outcomes of a consensus conference held to determine the first versions of the ICF Core Sets for lymphedema. Frequency rankings were made of the ICF categories derived from four preparatory studies, being: a) a systematic review; b) a qualitative study; c) an expert survey; and d) a cross-sectional study. By means of working group discussions and plenary sessions, a final consensus on ICF categories was achieved and Comprehensive and Brief Core Sets for lymphedema for the upper limb, lower limb, and midline lymphedema were defined. These ICF Core Sets contain different items in each region. Future validation of these Core Sets for health professions and for countries is needed.

Keywords: International Classification of Functioning, Disability and Health (ICF), ICF Core Set, lymphedema, function, disability, consensus

Introduction

Professionals working in lymphology (e.g., health professionals, researchers, health insurance companies and policy makers) often have different terminologies, frameworks, and approaches, which can make the communication regarding health problems challenging. A common generally accepted framework and terminology (term sets) could facilitate comparability of research findings and clinical outcomes (1) and improve the communication process among health professionals, researchers, health insurance companies, the government, and patient organizations (2). Such a framework can be provided by the International Classification of Functioning, Disability and Health (ICF) developed by the World Health Organization (WHO) (3). The approval of the ICF in 2001, provides a universal conceptual framework of domains and classifications to describe the functioning of individuals suffering from lymphedema. The ICF describes functioning and disability of a patient as a result of the interaction between different components: body functions (b), body structures (s), activities and participation (d), environmental factors (e), and personal factors (4). The system contains a hierarchical structure, using first, second, third, and fourth levels. However, using the entire ICF with over 1,400 categories is very time-consuming, and its use in daily practice is not very practical. Therefore, focused lists of items (categories) that are relevant and important for patients with lymphedema (Core Sets) are needed (5,6). There are two versions of the ICF Core Sets: a Comprehensive and a Brief Set of categories and codes. The Comprehensive Core Set is a list of ICF categories that describes the problems in functioning of patients with lymphedema in a multidisciplinary assessment. The Brief ICF Core Set is a list of ICF categories that are essential to describe the impairments, limitations, and restrictions in functioning and the environment of patients with lymphedema, and at the same time short enough to be practical in clinical studies or trials (4,7). These sets of ICF categories allow health professionals to classify and describe an individual's functioning using a universal and standardized language. In this way the Core Sets offer a chance for international studies and the possibility to compare the impact of different conditions, especially if used in electronic health care records.

To date, ICF Core Sets have been developed for several chronic conditions (4,7-9). The objectives of this paper are: (1) to describe the consensus process, and (2) to present the lists of ICF categories and codes within the Comprehensive and Brief ICF Core Sets for lymphedema.

METHODS

The development of ICF Core Sets for lymphedema started as a cooperative effort in a Steering group of the Erasmus Medical Centre (Department of Dermatology) in Rotterdam and the Dutch Institute of Allied Health Care in Amersfoort, under guidance of the Dutch WHO Collaborating Centre for the Family of International Classifications (WHO-FIC) in Bilthoven (all in the Netherlands) (*Fig. 1*).

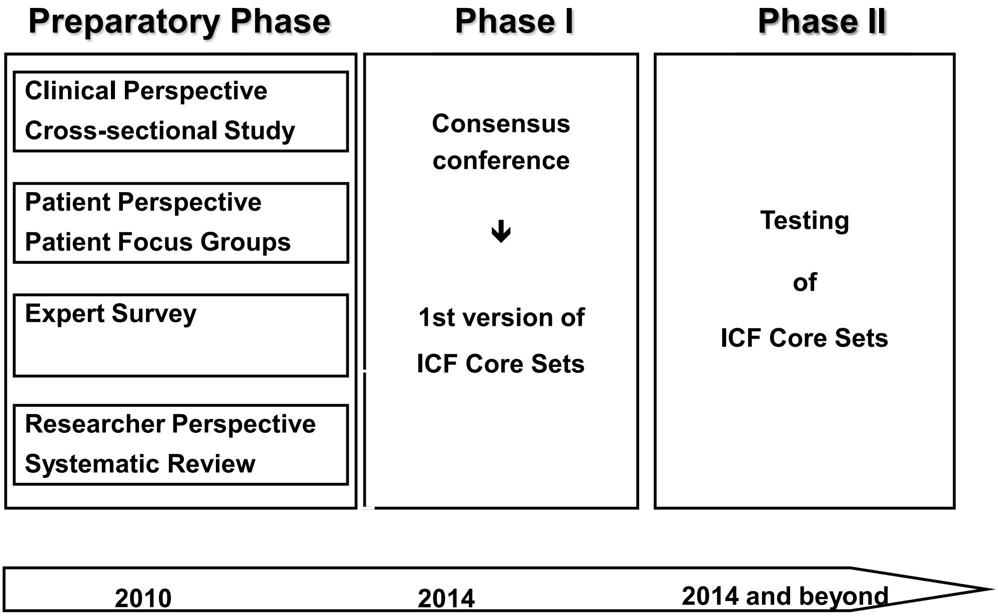

Figure 1. Development of ICF Core Sets for lymphedema: phases of the project.

Overview of the Process

The ICF Consensus Conference took place on the 4th of June, 2014 preceding the 5th congress of the International Lymphedema Framework in Glasgow, United Kingdom. Data from four preparatory studies were sent to experts before the conference, and outcomes of the preparatory studies were briefly reported at the beginning of the process. Preparatory studies included a systematic review of questionnaires used in lymphedema studies (10), a survey of international experts involved in lymphedema (n = 142) (publication in preparation), a study of focus groups of patients with lymphedema (qualitative study) (11), and a cross sectional study which took place in both The Netherlands (n= 200) (12) and in Australia (n = 93) (publication in preparation). A division in three regions was made: upper limb, lower limb, and midline lymphedema, because each of these lymphedema regions might cause specific functioning problems. Afterwards, the frequencies of every occurring ICF category were ranked for each study region, with the most occurring frequency given the lowest rank (number 1) and so on. The frequencies of registered ICF categories from the Netherlands and Australian cross sectional studies were combined to obtain a total data set. Next, the total ranking numbers between the separate studies were equalized due to different numbers of ICF categories per study. Then, frequency rankings of the defined ICF category of all studies were summed up resulting in a final list of ranked ICF categories for each region separately. These ranking lists were divided into three parts: the first 100 and second 100 categories (divided by a red line) and the remaining categories (divided by a blue line). Finally, these rankings of ICF categories were presented to the consensus participants (experts) as an Excel file. In column 1, the ICF code was provided and the title of the code was shown under column 2. In column 3 the total frequency was provided and in column 4 the ranking of the total. The frequency of the ICF categories, as well as the rankings found in each separate study (empirical study, expert survey, systematic review and qualitative study) were also presented (columns 5 to 12).

Recruitment of Conference Participants

Experts in the field of lymphedema were invited to participate in the consensus. No remuneration or honorarium was paid to the experts who agreed to attend. In order to identify the most active experts, a steering committee formed before development of ICF Core sets was consulted. Furthermore, invitation was based on scientific publications, professional background, informal network, and region of residence. (13).

Consensus Process

Four weeks preceding the conference, the expert participants were provided with information on the ICF and the Excel documents were sent by email. At the beginning of the conference, results of the four preparatory studies were presented and printed copies were given to the participants. Afterwards, the procedural steps of the consensus meeting were explained and the participants were divided into three expert groups (i.e. upper limb, lower limb, and midline). Each group was moderated by a member of the Steering Group and a secretary was allocated to each group. Both the moderator and the secretary were not allowed to participate in the discussion and voting process. In each group, experts openly discussed arguments in favor of or against the uptake of the ICF category in the Comprehensive Core Set. In a first step, each group had to come to a Comprehensive Core Set. Experts had to choose from the first 100 rankings (above the red line) those ICF categories that they considered essential. They could argue to add ICF categories from the second 100 rankings (above the blue line) and - only with very strong arguments - from the categories below the blue line. When the group came to consensus (i.e., when more than 50% of the members of the working group were in favor), a first version of the Comprehensive Core Set was defined. In a second step, experts individually had to choose 10 categories from the Comprehensive Core Set which they felt to be important in lymphedema, being a part of the Brief Core Set. Afterwards, these categories were discussed in the working group and consensus had to be reached. When both the Comprehensive and the Brief Core Set were defined in each working group, a plenary session took place. In this session, experts had the opportunity to discuss the Core Sets findings of the other working groups as the final consensus discussion. After the conference, the Steering Group discussed both the process as well as the results and had the opportunity for small alterations to the final versions.

RESULTS

Twenty-three experts participated in the consensus conference. Six were physicians, seven physical therapists, three nurses, five stocking suppliers, one occupational therapist, and one skin therapist. The countries where they practiced were: Australia, Belgium, Canada, Germany, Israel, Italy, The Netherlands, Sweden, the United Kingdom, and United States. These countries represent WHO regions: North America, Europe, and Western Pacific. Based on results for the three lymphedema regions using first, second, third, and fourth level classifications from the four preparatory studies, Comprehensive and Brief ICF Core Set for lymphedema were developed.

The numbers of categories in the Comprehensive Core Sets were for the upper and lower extremity and midline 101, 111 and 89 respectively. For **the** Brief Core Sets they were 23, 18 and 20, respectively. *Table 1* shows the ICF categories that were included in the Comprehensive and Brief Core Sets.

Table 1 Comprehensive and Brief Core Sets for the upper limb (UL in black colours), lower limb (LL in red colours) and midline region (M in green colours).In bold = categories of the common part of the Comprehensive Core Set; Arabic = categories of the common part of the Brief Core Set; (B) = Part of the Brief Core Set; (M) = categories of the common part of the midline region; (G) = categories of the genital region; (H) = categories of the head and neck region; (Breast) = category of the breast region.

ICF code	Description	UL	LL	M
b1	CHAPTER 1 MENTAL FUNCTIONS	X		
b126	Temperament and personality functions			X (M)
b1261	Agreeableness		X	
b1263	Psychic stability	X (B)	X	
b1264	Openness to experience	X		
b1265	Optimism	X		
b1266	Confidence	X	X	
b130	Energy and drive functions			X (M)
b1301	Motivation	X	X (B)	
b134	**Sleep functions**	X	X	X
b152	**Emotional functions**	X	X (B)	X
b164	Higher-level cognitive functions	X (B)		X
b1644	**Insight**	X	X	X
b1646	Problem-solving		X	
b1670	Reception of written language		X	
b180	Experience of self and time functions			X (M)
b1801	***Body image***	X(B)	X(B)	X(B)
b260	Proprioceptive function	X	X	
b265	Touch function	X		X (M)
b2702	Sensitivity to pressure	X	X	
b280	***Sensation of pain***	X(B)	X(B)	X(B)
b415	Blood vessel functions		X	
b4150	Functions of arteries		X	
b435	**Immunological system functions**	X	X (B)	X (B)(M)
b4350	Immune response		X	X (M)
b4352	**Functions of lymphatic vessels**	X	X	X
b4353	Functions of lymph nodes		X	
b440	Respiration functions			X (M)
b455	Exercise tolerance functions		X (B)	X (B)(M)
b4550	General physical endurance	X (B)		
b4552	**Fatiguability**	X (B)	X	X
b5104	Salivation			X (H)

b5105	Swallowing			X (H)
b530	**Weight maintenance functions**	X	X	X
b540	General metabolic functions		X	X (M)
b5403	Fat metabolism	X		
b550	Thermoregulatory functions		X	
b620	Urination functions			X (G)
b640	**Sexual functions**	X	X	X (B)(M)
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS	X		
b710	Mobility of joint functions	X (B)		X (M)
b7101	Mobility of several joints		X (B)	
b730	Muscle power functions	X (B)		X (M)
b7303	Power of muscles in lower half of the body		X (B)	
b770	Gait pattern functions		X	X (G)
b780	**Sensations related to muscles and movement functions**	X	X	X
b810	**Protective functions of the skin**	X	X	X
b820	**Repair functions of the skin**	X	X	X
b840	**Sensation related to the skin**	X	X	X
d155	**Acquiring skills**	X	X	X (B)(M)
d170	Writing	X		
d230	***Carrying out daily routine***	X(B)	X(B)	X(B)
d240	Handling stress and other psychological demands			X (B)(M)
d2400	Handling responsibilities	X		
d3	CHAPTER 3 COMMUNICATION	X		
d330	Speaking			X (H)
d350	Conversation			X (H)
d360	Using communication devices and techniques	X		
d4	CHAPTER 4 MOBILITY	X		
d410	**Changing basic body position**	X	X	X
d4100	Lying down		X	X (G)
d4101	Squatting		X	
d4102	Kneeling		X	

d4103	Sitting		X	
d4104	Standing		X	
d4105	Bending		X	
d415	Maintaining a body position	X		
d4153	Maintaining a sitting position		X	X (G)
d4154	Maintaining a standing position		X	X (G)
d430	**Lifting and carrying objects**	X	X	X
d4401	Grasping	X		
d445	Hand and arm use	X (B)		
d4452	Reaching	X		
d450	**Walking**	X	X (B)	X
d4500	Walking short distances		X	
d4501	Walking long distances		X	
d455	Moving around			X (B)(G)
d4551	Climbing		X	
d4552	Running		X	
d4554	Swimming	X		
d4600	Moving around within the home		X	
d4602	Moving around outside the home and other buildings		X	
d470	Using transportation			X (G)
d4702	Using public motorized transportation	X	X	
d475	Driving	X		X (G)
d4750	Driving human-powered transportation		X	
d4751	Driving motorized vehicles		X	
d5	CHAPTER 5 SELF-CARE	X (B)		
d510	**Washing oneself**	X	X	X
d520	Caring for body parts		X	X (B)(M)
d5200	Caring for skin	X	X	
d5202	Caring for hair	X		
d530	**Toileting**	X	X	X
d540	**Dressing**	X	X	X
d5402	Putting on footwear		X	
d5404	Choosing appropriate clothing	X		

d550	Eating			X (H)
d560	Drinking			X (H)
d570	***Looking after one’s health***	X(B)	X(B)	X(B)
d5700	Ensuring one’s physical comfort	X		
d5701	Managing diet and fitness	X (B)	X	
d5702	Maintaining one's health	X	X	
d620	Acquisition of goods and services			X (M)
d6200	Shopping	X	X	
d630	Preparing meals	X	X	
d640	**Doing housework**	X (B)	X	X
d6400	Washing and drying clothes and garments	X		
d6505	Taking care of plants, indoors and outdoors	X		
d660	Assisting others	X	X	
d7	CHAPTER 7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS	X		
d750	Informal social relationships			X (M)
d770	**Intimate relationships**	X (B)	X	X
d845	Acquiring, keeping and terminating a job		X (B)	
d850	**Remunerative employment**	X (B)	X	X (B)(M)
d870	Economic self-sufficiency	X		X (M)
d920	**Recreation and leisure**	X (B)	X	X (B)(M)
d9201	Sports	X		X (M)
d9205	**Socializing**	X	X	X
e1101	**Drugs**	X	X	X
e1150	**General products and technology for personal use in daily living**	X	X	X
e1151	***Assistive products and technology for personal use in daily living***	X(B)	X(B)	X(B)
e1201	Assistive products and technology for personal indoor and outdoor mobility and transportation		X	
e1300	General products and technology for education	X		
e135	Products and technology for employment		X	
e150	Design, construction and building products and technology of buildings for public use	X	X	

e155	**Design, construction and building products and technology of buildings for private use**	X	X	X
e1650	Financial assets		X	
e210	Physical geography		X	
e225	**Climate**	X	X	X
e2250	Temperature	X		X (M)
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS	X		
e310	***Immediate family***	X(B)	X(B)	X(B)
e320	**Friends**	X (B)	X	X (B)(M)
e325	**Acquaintances, peers, colleagues, neighbours and community members**	X	X	X
e330	**People in positions of authority**	X	X	X
e340	**Personal care providers and personal assistants**	X	X	X
e355	***Health professionals***	X(B)	X(B)	X (B)
e410	Individual attitudes of immediate family members		X	X (M)
e420	**Individual attitudes of friends**	X	X	X
e425	**Individual attitudes of acquaintances, peers, colleagues, neighbours and community members**	X (B)	X	X
e450	Individual attitudes of health professionals	X		X (M)
e460	**Societal attitudes**	X	X	X
e465	Social norms, practices and ideologies			X (M)
e555	Associations and organizational services, systems and policies			X (M)
e5550	Associations and organizational services	X	X	
e5600	Media services		X	
e570	Social security services, systems and policies	X		
e580	Health services, systems and policies		X (B)	X (M)
e5800	Health services		X	
e5801	Health systems	X		
e5850	**Education and training services**	X	X (B)	X
s330	Structure of pharynx			X (H)
s340	Structure of larynx			X (H)
s420	**Structure of immune system**	X	X	X
s4200	Lymphatic vessels	X	X	

s4201	Lymphatic nodes	X	X	
s630	Structure of reproductive system		X	X (M)
s6303	Structure of vagina and external genitalia		X	
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT	X		
s710	Structure of head and neck region	X		X (B)(H)
s720	Structure of shoulder region	X		X (H)
s730	Structure of upper extremity	X (B)		
s7300	Structure of upper arm	X		
s7302	Structure of hand	X		
s740	Structure of pelvic region		X	X (B)(G)
s750	Structure of lower extremity		X (B)	
s7500	Structure of thigh		X	
s7501	Structure of lower leg		X	
s75011	Knee joint		X	
s7502	Structure of ankle and foot		X	
s760	**Structure of trunk**	X	X	X (B)(Breast)
s7701	Joints		X	
s810	**Structure of areas of skin**	X	X	X

In *Table 2* the division of the ICF components is shown. *Table 3* shows the division of ICF levels in the different Core Sets. No first levels were selected, except for the Core Sets for the upper limb. Most of the categories came from the second and third level. Only 1 fourth level (in the Comprehensive Core Set for the lower limb) was included.

Table 2. Division of ICF components in the Core Sets for lymphedema(UL = Upper Limb, LL = Lower Limb, M = Midline)

	ICF components				
Core Set for lymphedema	Body functions	Body structures	Activities and Participation	Environmental factors	Total
Comprehensive UL	29	8	41	23	101
Comprehensive LL	32	14	40	25	111
Comprehensive M	29	9	30	21	89
Brief UL	8	2	9	4	23
Brief LL	8	1	4	5	18
Brief M	5	3	8	4	20
Total	111	37	132	82	362

Table 3. Division of ICF levels in the Core Sets for lymphedema (UL = upper limb, LL = lower limb, M = midline)

	Number of levels of the ICF				
Core Set for lymphedema	1	2	3	4	Total
Comprehensive UL	8	56	40	0	104
Comprehensive LL	0	55	55	1	111
Comprehensive M	0	71	17	0	88
Brief UL	1	16	6	0	23
Brief LL	0	12	6	0	18
Brief M	0	18	2	0	20
Total	9	228	126	1	364

Concerning the Comprehensive Core Sets, some categories are mentioned in all Core Sets: 14 from the component Body Functions, three from Body Structure, 14 from Activities and Participation, and 15 from Environmental Factors. For the Brief Core Set the division is: two from Body Functions, two from Activities and Participation, and three from Environmental Factors. Because midline lymphedema can occur in different body parts (i.e. head and neck , breast and genital region), some categories for the Midline Core Sets concern all body parts and some are only applicable for those particular regions. For the Comprehensive Core Sets, 23 categories are applicable for all regions, 11 for the head and neck, and nine for the genital region. For the Brief Core Set, nine are applicable for all regions, one for the head and neck, one for the breast, and two for the genital region (*see Fig. 2*).

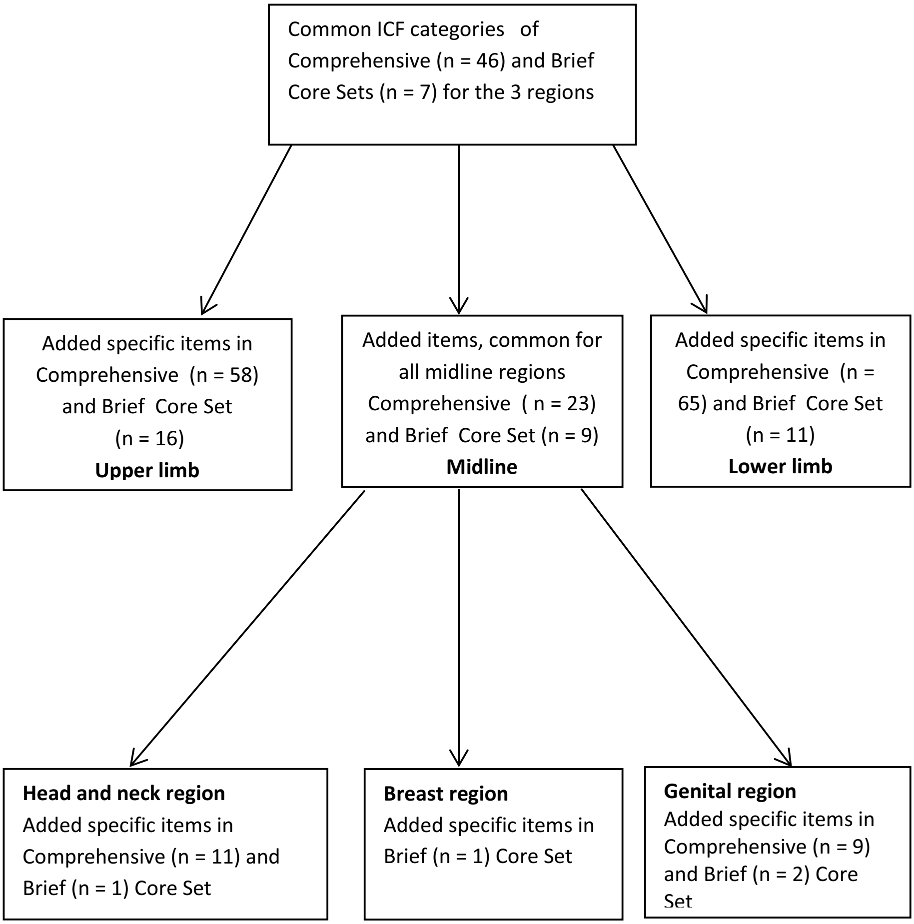

Figure 2 Flow chart of the Comprehensive and Brief core sets

DISCUSSION

Lymphedema is a complex health condition which presents itself in several ways. It can affect functioning of a patient in various ways, areas, and levels, and it involves different health professions, organizations, and policies. The ICF is a conceptual framework and classification system by the WHO with which this complexity can be addressed. With the use of the ICF, the consensus conference succeeded to identify Core Sets of variables that are important in lymphedema, with integrated data from preparatory studies. The results of this international consensus were: Comprehensive Core Sets for Upper limb lymphedema (104 ICF categories), Lower limb lymphedema (111 ICF categories), and Midline lymphedema (89 ICF categories), and Brief Core Sets for upper limb (23 categories), Lower limb (8 categories), and Midline (20 categories) lymphedema.

The use of the ICF in patients with lymphedema is challenging because the ICF, which is based on the bio-psycho-social model, contains terms which can be used to describe not only functioning of the individual itself, but also the context in which he or she is living (3). Furthermore, the ICF provides a universal language and since lymphedema is a health condition which presents itself both in developed and on a large scale also in under-developed countries (14), its applicability in both cases is multifaceted.

The Core Sets contain all ICF components, stressing the variety and complexity of lymphedema. Unfortunately, personal factors, as part of the contextual factors, are not yet classified within the ICF, although various attempts have been made recently (15,16). Seven categories were mentioned in all Core Sets leading to the most relevant topics in lymphedema, according to this group of experts: Body Image, Pain, Daily Routine, Hosiery, Family, Health Professionals, and Self-management. These items contain two categories from Body Functions, one from Activities and Participation, four from Environmental Factors and none of Body Structures. Although the shape of extremities and the midline region is important in lymphedema, experts seem to value the function of the lymphatics more than the form, which evidently is the outcome of the malfunction of the lymphatics. It is obvious that Environmental Factors are important, although the outcomes of the literature review show that these items are not examined on a large scale and are scarcely used in lymphedema specific questionnaires (10).

The Brief Core Sets are developed to define a standard that can be used as the minimum dataset to be collected in patient evaluation or in clinical studies. When a minimum dataset in lymphedema is available, this could help in performing comparison across settings which would eventually enhance standardization of reporting evidence (e.g., treatment outcomes) (4). Currently the International Lymphedema Framework (ILF) is running a study to develop a minimal dataset as well (17). The Brief Core Sets could be implemented in this study.

Compared to other consensus conferences to develop ICF Core Sets (4,5), there are some points of special interest. Whereas other conferences lasted two or three days, this conference lasted only one day because of lack of financial support. The organization managed to collect a representative group of experts for one day, preceding the 5th congress of the ILF in Glasgow. Despite the short time frame, it is expected that the developed Core Sets encompass a large proportion of the problems that patients with lymphedema encounter. Although other consensus procedures excluded first level categories in the Core Sets, in this Core Set first level categories appear in the Upper limb Core Sets. It was the definite will of this working group to include these first level categories. Future validation research will determine whether these categories will keep their place in the Core Sets. Another issue concerns the sample of experts being composed only from developed countries. This distribution of experts was quite similar to the pool of experts from the worldwide expert survey which was also conducted as part of the Core Set development. The implication is that the Core Sets are developed with the focus on the health care systems in these countries. Future validation is needed in the under-developed countries to examine the usability of the Core Sets in these regions. Furthermore, the country of origin and skew distribution of the professional experts could be a confounder. Perhaps a greater number of skin therapists and occupational therapists could have led to more contribution from this point of view. Another limitation is the potential bias regarding patient selection in the preparatory qualitative and cross-sectional studies in The Netherlands (11,12). To compensate for this problem somewhat, one of the studies was also performed in Australia. Finally, a last bias can be the fact that quite a large proportion (9% of the Comprehensive Core Sets and 0.6% of the Brief Core Sets) of the ICF categories of below the blue line (rankings higher than 200, meaning these categories were scarcely mentioned in the preparatory studies), were included in the Core Sets. This could be due to the individual attitudes of experts in the working groups or to the fact that the working group decided to choose a higher level category to include a broader scope related to a health problem, leading to a subjective impact in these Core Sets. The Comprehensive ICF Core Sets for lymphedema integrate the experiences and needs of different health professions. The domains of the Core Sets can guide the patient-centered goal setting process in multidisciplinary settings, where resources and problems could be identified and profession-specific treatment goals can be identified. The identified categories help in multidisciplinary settings to assign patients to specific areas of expertise for evaluation and treatment, and for clinical-decision making (4). The categories of the Brief ICF Core Set can be used as a minimum set of domains to report in trials or intervention studies. It can also be used in independent clinical practice setting which would help clinicians compare results and weigh the evidence of their results. (4). In the future, validation studies will be helpful in determining if there are subsets of the Core Sets that could be more meaningful given a certain setting and to investigate the Core Set's applicability across a range of health professions and cultures. International and national frameworks in lymphology should be involved to implement the ICF Core Sets for lymphedema.

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Chapter 10

General Discussion

Introduction

The purpose of this thesis is to develop International Classification of Functioning, Disability and Health (ICF) Core Sets for lymphedema. Specific core sets for lymphedema should result in a better understanding of the problems of the individual lymphedema patient and a more tailor made therapy.
Lymphedema is a chronic health condition, which gives sincere problems in daily life and has a negative effect on the health-related quality of life of these patients. In many cases lymphedema is a result of cancer treatment and as such lymphedema can be seen as the 'price of survival'. In the under-developed countries lymphedema can be caused by parasitic infections. The problems associated with lymphedema can be divided in impairments (like pain, swelling, all kinds of sensitivity problems, skin problems and problems in emotional functions), limitations in activities (like walking and household), and restrictions in participation (such as employment and sports). The occurrence and severity of these functioning problems are influenced by environmental factors (such as health care systems, treatments and attitudes of others) and personal factors (such as age, comorbidity, coping, and life style).

The project and its overall results

The development of the ICF Core Sets for lymphedema was performed according to the rules of a WHO (World Health Organization) accepted system (1) and contained several phases (2). In the preparatory phase five studies were performed from four perspectives; the researchers' perspective (two studies), the clients' perspective (one study), the health professionals' clinical perspective (one study), and the mondial experts' perspective (one study). The results of the preparatory studies were expressed in meaningful concepts and these concepts were linked to ICF categories. The final results of these studies are lists of items within: Body Structures, Body Functions, Activities and Participation, Environmental Factors, and Personal factors.

Body Functions

All preparatory studies reveal the same results with respect to the categories most mentioned. These were always ‘b435, Immunological system functions’ (because swelling is one of the major problems in lymphedema and lymph vessels and nodes are part of the immunological system), ‘b280, Sensation of pain’ (most of the patients complain about pain; patients with lower limb lymphedema more than patients with upper limb lymphedema), ‘b265,Touch functions’ (the sensation of the skin is often altered because of the edema), ‘b152, Emotional functions’, and ‘b126, Temperament and personality functions’ (both categories are important because of the psycho-social impact of lymphedema). There is one exception: in the cross-sectional study, representing the health professionals’ clinical perspective, ‘b710, Mobility of joint functions’, ‘b730, Muscle power functions’, and ‘b530, Weight maintenance functions’ were most frequently mentioned. Probably this is a result of the policy (more focus on self-management) of the hospital where the study was performed. Currently, risk-factors, muscle power and mobility are being integrated in treatment more and more. Although a substantial proportion of the literature describes these items, most of the literature we found focused on swelling and pain.

Body Structures

In all studies ‘s730, Structure of upper extremity’, ‘s750, Structure of lower extremity’, and ‘s630, Structure of reproductive system’ were the most mentioned categories. Category ’s810, Structure of areas of skin’ was only mentioned in the cross-sectional study representing the health professionals’ clinical perspective and in the literature study concerning lymphedema-specific questionnaires (one of the two studies representing the researchers’ perspective). The questionnaires often are developed by health professionals and one of the conclusions of the cross-sectional health professionals’ perspective study was that health professionals lay more emphasis on body structures than the patients themselves. However, in the qualitative research with patient focus groups (representing the clients’ perspective), the structure of the skin area is mentioned in the genital lymphedema group as an item of major concern.

Activities and Participation

Most mentioned categories for all studies were: ‘d415, Maintaining a body position’, ‘d570, Looking after one’s health’, ‘d450, Walking’, ‘d640, Doing housework’, and ‘d850, Remunerative employment’. Patients themselves as well as health professionals consider ‘d920, Recreation and leisure’ as of great importance, whereas in the lymphedema-specific questionnaires this category is only mentioned in questionnaires concerning the upper limb.

Environmental Factors

In all studies ‘e580, Health services, systems and policies’, ‘e355, Health professionals’, ‘e310, Immediate family’, and ‘e115, Products and technology for personal use in daily life’ were mentioned most frequently. The last category relates dominantly to the prescribed use of hosiery and bandages, being a substantial part of the treatment of lymphedema. In the questionnaires this is only a topic in those concerning the upper limb, although stockings have a high influence on the quality of life of patients with lower limb lymphedema as well (8-11). So there is a need to involve questions on this topic in future questionnaires for lower limb lymphedema.

Environmental factors can act as a barrier or a facilitator. The categories ‘e580, Health services, systems and policies’ and ‘e355, Health professionals’ are of great importance, but quite often it appears that they are predominantly named as a barrier. Especially the qualitative research revealed the ambivalence of the patient with respect to the health professional. Often seen as of major assistance, but many times also someone who underestimates the impact of lymphedema and the related prescribed lifestyle for the patient. These problems might be attenuated by providing more qualitative patient information about the impact of lymphedema to the health professionals.

Personal factors

Personal factors are not part of the ICF yet; however, several suggestions on the subdivision and classification of personal factors have been made (12-14). In the various surveys of this thesis, items on personal factors have been listed. Coping strategies of lymphedema patients, comorbidity, lifestyle, age, and race were the most frequently mentioned personal factors. Coping strategies are the most important personal factor, because the lymphedema patient, as every patient, seeks the best way to deal with his/her health condition. Coping strategies vary from mental solutions (thinking of nice things, being pro-active, and trying to act as common as they did before the lymphedema) to (self-constructed) material (for example for support of urination by genital lymphedema patients, or using a hoola-hoop for more drainage of lymphedema).

Consensus conference

An international consensus conference was organized for which various health professionals, being lymphedema experts from different countries were invited. The data of four (one literature review excluded, because we only used the data of the lymphedema-specific questionnaires and followed the method proposed by Cieza and Stucki (1)) preparatory studies were subjects for consensus on the most important categories. The consensus process resulted in the first versions of six ICF Core Sets for lymphedema. A Comprehensive (C) and a Brief (B) Core Set were defined, each for three regions: upper limb (101 (C) and 23 (B) items), lower limb (111 (C) and 18 (B) items) and midline (89 (C) and 20 (B) items) lymphedema. These Core Sets appeared to contain different items in relation with the region of the edema. However, the comprehensive core sets of the three regions had 46 categories in common and the Brief Core Sets seven categories. These categories appear to represent the core problems of the lymphedema patient. As the Brief Core Sets are derived from the Comprehensive sets, it can be concluded that the following seven items are the most important for the lymphedema patient: Body Image, Pain, Daily Routine, Hosiery, Family, Health Professionals, and Self-management. These items contain two categories from Body Functions, two from the Activities and Participation domain and three from Environmental Factors.

ICF Core Sets for lymphedema

The ICF offers a conceptual framework to describe the functioning of a patient and the factors influencing his/her functioning. To make the connections between the different components during the process of clinical reasoning more visible, Steiner et al. constructed a so called Rehabilitation Problem Solving (RPS) form as a tool for physical therapists (3). Although already published in 2002, not many Dutch physical therapists are using this form in daily practice. The administration of the data of clinical examination and treatment mostly happens with the use of electronic patient files. Although these files address the division of patient problems as proposed by the ICF, the use in daily practice is not user-friendly. Many items have to be filled in, which makes this administration very time-consuming. Moreover, the examination and results of the treatment of lymphedema patients sometimes requires a special way of administrating, which cannot easily be performed with the general format of physical therapy administration. For instance, palpation and judgement of the swelling need a more detailed description than is possible with the present format and the volume measurements are not implemented in the electronic patient files. Despite the comprehensive format, there is often no space for clinical reasoning, let alone a RPS form. With the introduction of ICF Core Sets for lymphedema, there is an opportunity for physical therapists to streamline both the administration and clinical reasoning of the lymphedema patient. By offering more clarity in the possible problems in functioning and the factors influencing the functioning of the lymphedema patient as well as the use of a decision tree structure, the Core Sets can act as a tool for the clinical administration. By using the presented sets these will come to more refinement and eventually reduction. So in fact it is not a stable tool, but an active tool, which will constantly has to be evaluated and adapted in the course of time. ICF Core Sets can already be helpful in the diagnostic phase: during anamnesis and examination the ICF Core Sets can be used as a checklist of items to be asked for or examined. By analysing the results of the anamnesis and the clinical examination, a physical therapist's diagnosis and a prognosis can be formulated. Also in this phase of analysis the ICF Core Sets can act as a checklist. When it is decided that physical therapy is a useful intervention, a treatment plan is developed together with the patient based on the physical therapist's diagnosis, starting with the formulation of the treatment goals. In this stage, the ICF Core Sets are useful in discussing with the patient the possible short and long term goals for the treatment (4-8). To evaluate the treatment goals, outcomes have to be formulated in connection to the treatment goals; during the evaluation phase it must be possible to determine – based on the outcomes - whether the treatment goals are reached. Subsequently appropriate measurement instruments have to be chosen (9). Following the outcomes of the measurements before the treatment, the treatment goals, and the prognosis the intervention and the frequency of treatment can be determined (10). And finally, the results of re-examination can provide feedback about the effect of the treatment.

All these steps are already implemented in electronic patient files and the diagnostic process (i.e. anamnesis, examination and measurements) can serve as a basis for the defining of the contents of the RPS form. In this way the ICF Core Sets for lymphedema provide a less time-consuming administration and a transparency of the clinical reasoning (11). This is achieved by the fact that the Core Sets already offer a format of categories to be examined and so more or less direct the anamnesis and clinical examination although sound clinical reasoning must always be the basis for therapeutic decisions.

Unfortunately, knowledge on lymphedema epidemiology is scarce because good registration is absent. However the number of patients worldwide is estimated to be approximately 140-250 million (12). In the Netherlands information about the incidence of lymphedema is hard to find, because of the great variance in studies, expressions, the way lymphedema is diagnosed, and the fact that there is no sufficient registration of lymphedema patients, since patients are predominantly classified in hospital registration systems according to the 'first' disease such as cancer. Besides, in most patients lymphedema is developing after hospital discharge. For this reasons the incidence of lymphedema can only be estimated.

For a proper description of the problems in (daily) functioning of the patient the ICF is needed. ICF Core Sets for lymphedema offer a way to document these problems and facilitating uniformity of data (by using the codes that are specific for the ICF category), not only in the Netherlands but worldwide. Clinicians and researchers are probably more willing to document these items, because the Core

Sets are compact and easier to apply than the total ICF. Also the selection of measurement instruments is easier when Core Sets are available , creating an increase of the relevance of these measurement instruments and increasing the chance that researchers use the same measurement instruments in different studies. In this way a basic set of measurement instruments can be designed which creates an opportunity to compare the results of different studies using these instruments, increasing the knowledge of lymphedema. Implementation of the ICF Core Sets in the Dutch guideline for lymphedema in the Netherlands (13), the consensus document of the International Society for Lymphedema (ISL) (14), as well as the best practice document of the International Lymphedema Framework (ILF) (15) is a way to achieve uniformity in several ways such as administrating, describing, and classifying the problems of lymphedema patients. Currently patient education, including self-management, is becoming an increasingly important part of holistic patient treatment. In lymphedema treatment this is also the case. The ICF offers the opportunity to facilitate patient education, but is too comprehensive to handle in daily practice. ICF Core Sets are better suitable to apply in daily practice, because of their compactness. Also they could serve as a stepping-stone to discuss functioning and treatment with the lymphedema patient. In the Dutch guideline for lymphedema, the possibility for the use of ICF Core Sets in patient education is already mentioned (13). ICF Core Sets can be incorporated in the guideline, so that the guideline also makes use of an internationally broadly accepted framework.

Comments on the project

Every project has its strengths and weaknesses. The outcome of the project with six solid Core Sets was overall successful, although there are some flaws and we will discuss these here.

Qualitative research

In the qualitative research a great deal of patients with upper limb lymphedema were included and only a relatively small group of patients with genital and head and neck (midline) lymphedema. Not only is the latter group of patients relatively small, but often they are not willing to talk about their problems. This is especially true for patients with genital lymphedema. These circumstances may have had some effects on the kind of items that the total group of interviewed patients indicated as important. However this distribution reflects the composition of the lymphedema patient group in total.

Cross-sectional study

For the development of the Core Sets we followed a systematic method as indicated by the WHO and described by the group of Stucki et al. (16, 17). According to that method a cross-sectional survey among health professionals had to be performed using a multi-center approach. However, in The Netherlands there is only one hospital really specialized in the treatment of lymphedema patients. This centre attracts diverse patients with lymphedema from all over the country, with a good mix of in- and outpatients, and thereby covering the total spectrum of the lymphedema patient, as well in clinical presentation, as genotype and geographical distribution. To compensate this, a study was done in Australia as well, but only the results of the first group of patients could be used during the consensus conference. The results of the Australian study will be published separately. In the cross-sectional study we asked the patients for only specific categories as mentioned in the literature reviews, whereas the original method makes use of the ICF in total.

International perspective

The project was initiated and performed in The Netherlands with international contributions in the expert research and the cross sectional clinical study in Australia, of which a part of the data were used for the consensus conference. There were also international contributions in the consensus conference itself. Although the literature review concerns world-wide articles and there were contributions from India in the expert research, much of the other parts of the development process had contributions from Western countries only. Future validation research in other, non-Western countries, including developing countries will reveal if the Core Sets are fully applicable in these countries, or may need extensions or additions. Circumstances are so different between the developed and underdeveloped world that special projects should be run for validation in the different regions.

Patient involvement and perspective

Patients were involved in the preparatory studies (19). In the study about the health professionals' clinical perspective the health professionals had to rank the patients' health on a scale from 0 to 10 and the patients had to do that also. It came forward that health professionals judge the health of the patients lower than the patients themselves. It could well be that most of the patients have adapted to the problems caused by the lymphedema and therefore judge themselves at a higher level. One of the results of the validation study on the ULL27 was that there was no correlation between swelling of the arm and health-related quality of life (18). This is in line with the reflections on adaptation to the lymphedema problems. Another reason for this higher level judgement could be that patients categorize problems like lymphedema not as part of their health, since cancer, being the basic reason of lymphedema, has been cured what makes that patients judge their health higher.

Although not included in the WHO procedure for developing Core Sets, it would have been an attractive option to involve patients in the consensus conference as well. Although we have considered this possibility, but it appeared to create problems with respect to (universal conference) language, costs, and travelling possibilities. We recommend to involve the patient perspective in the validation studies for the Core Sets for lymphedema, as was done already in the validation studies for other Core Sets (4-6). Hieblinger et al. examined the patient point of view in fibromyalgia patients and concluded that most of the categories of the ICF Core Set were relevant, but a few new categories emerged and these should be considered to be incorporated in the Core Sets for fibromyalgia (19). Validation studies point at the fact that Core Sets themselves as well as the criteria for the development process need updates at a regular basis.

The development process of the Core Sets for lymphedema in comparison to other Core Sets

This project followed a structured development process (1, 20, 21). In the preparatory phase five studies were conducted. Originally there is a set of four studies, including one literature review on disease-specific questionnaires. In this project a second literature review was performed, linking meaningful concepts from the Method and Results sections of articles about lymphedema to ICF categories. The purpose was to compare the items which were discussed in these sections with the content of the questionnaires. In other words, did the questions in the used questionnaires connect to the concepts in the text of the article. The results indicate that for upper limb lymphedema the ICF categories in the text were similar to those in the questionnaires, whereas for lower limb lymphedema this was not the case. The addition of this second literature review was not done previously.

When overviewing the various processes in the development of Core Sets, one can say that in the course of time the initial format has been abandoned by the developers (22). For instance the development of Core Sets for Low Back Pain, Acute hospital, and Rheumatoid Arthritis consisted of three preliminary studies, lacking the qualitative research (17, 23, 24). In the process to come to Core Sets for Ankylosing Spondylitis the cross-sectional study is absent (25). In a systematic review about ICF Core Sets developing from 2001 to 2012 Yen et al. (26) came to what they call the 'most commonly adopted approach'. This approach contains two preliminary studies - the cross-sectional study followed by a systematic literature review - followed by a consensus procedure, as the final phase. When comparing the preliminary phase of the development of ICF Core Sets for lymphedema with previous developments of other Core Sets, one can say that with five studies a comprehensive picture of important categories for the lymphedema patient has been achieved.

Compared with the average final consensus conference this project used a one-day conference instead of a two- or even three-days conference (26). Much of the preparation was done preceding the conference by sending information on the ICF and all the data that were gathered in the preparatory phase to the participants of the conference. By planning the consensus conference on the day before the start of an international conference on lymphedema, we were able to gather an interesting group of 23 experts. These experts represented six different health professions, whereas four are obligatory (26). The division of these professionals was slightly different compared with the overall division in the review of Yen et al. (26). In most other ICF Core Sets development processes, physicians were most represented, whereas in this conference physical therapists were maybe over represented (30%). Representatives of stocking suppliers and skin therapists were not participating in other Core Set projects, but were contributing for 22% and 4% respectively in this conference. This is because lymphedema requires a very specialized approach of specific health professionals. Six Core Sets were produced at the end of the consensus conference (all other Core Sets contain only two sets, namely one Comprehensive and one Brief Core Set). The choice for six Core Sets was made because of the different ways of functioning in the three different lymphedema regions, i.e. upper limb, lower limb and midline region. From the study of Yen et al. (26) it was learned that Comprehensive Core Sets had an average of 92.5 categories and Brief Core Sets 26 categories. The Comprehensive Core Sets for lymphedema (Upper limb, Lower limb and Midline) consist of 101, 111 and 89 categories respectively; the Brief Core Sets of 23, 18 and 20 categories. It can be concluded that the amount of categories of all six Core Sets for lymphedema are within the ‘normal’ range.

In earlier Core Set development studies it was expected that the focus would lie on domains within Body Functions and Activities and Participation, but it turned out that Environmental Factors were of equal importance (27, 28). The Core Sets for stroke, for instance, contained 25% environmental factors (27). For the Core Sets for lymphedema the Comprehensive Core Sets for upper limb, lower limb, and midline contain 22%, 23%, 24% Environmental Factors, respectively. The Brief Core Sets contain 17%, 27%, 20% Environmental Factors respectively. These percentages stress the importance of environmental factors for the functioning of people with lymphedema.

ICF Core Sets in a broader way

Currently a lot of disease-specific ICF Core Sets already exist (1, 17, 19, 23, 24, 29-33), but Core Sets for lymphedema were not yet composed. As stated in the introduction, the ICF provides a comprehensive framework of human functioning as well as a classification system (21). The ICF offers a universal language understood by health professionals, researchers, policy makers, patients, and patient organizations. By adding environmental and personal factors and a mutual influence between the different components, to its predecessor the International Classification of Impairments, Disabilities and Handicaps (ICIDH), the ICF moved away from a framework describing the consequences of a disease to a bio-psycho-social framework (34). The reason for constructing disease-specific Core Sets is that the ICF itself contains more than 1400 categories and is not quite workable in daily practice. Core Sets will make the ICF useful for health care providers (35), because they contain the most important items for patients with a specific health condition, collected from the perspective of all persons involved. The Core Sets facilitate the description of the functioning of patients and the formulation of treatment goals. Description uniformity is needed to compare data for the evaluation of treatment efficacy and to collect data for research. ICF Core Sets will be more inviting for health care providers, researchers and all other professionals or institutes to use them for these purposes. The ICF Core Sets may be used as a guide for clinical reasoning and to record the results of clinical reasoning in electronic patient files. By being more ‘user-friendly’, ICF Core Sets could help raise the profile and impact of specific diseases on functioning for a particular client group to a larger audience (36).

Comprehensive ICF Core Sets have proved to be beneficial to clarify team roles and facilitate clinical reasoning (36, 37). We hope that the introduction of the ICF Core Sets for lymphedema can further guide multidisciplinary cooperation and individual practitioners in service delivery. The Core Sets can serve as an eye-opener for the risk that health professionals tend to use only the components they are comfortable with, i.e. body functions and structures. The inclusion of environmental factors, for instance, will facilitate consideration of these concepts by professionals, who traditionally would not do so (36).

McIntyre and Tempest (36) proclaim a possible disadvantage of Core Sets, namely that health professionals are often working with people with co-morbid conditions and multiple pathologies. So, in fact more than one disease-specific Core Set should be necessary to ensure a correct description of the impact on functioning of the patient. For instance, patients with breast-cancer related lymphedema have functioning problems which are the result of both the lymphedema as the consequences of the cancer itself. These two conditions are closely interwoven and there is no getting away from that. During the construction of the project this problem is encountered by the fact that in many cases the consequences of cancer, being the most important cause of the lymphedema, were in fact incorporated in the different studies of the development process, and therefore the Core Sets are in fact 'real life' sets. When the Core Sets for upper limb lymphedema are compared with the already existing Core Sets for breast cancer (consisting of 78 items for the Comprehensive and 29 items for the Brief Core Set) (38), there is an overlap of 16 items of the Brief Core Sets. Compared with the Comprehensive Core Set there is an overlap of 45 items. It is obvious that both Core Sets have similar items, but on the other hand are complementary.

McIntyre and Tempest also discuss the fact that Core Sets could be a step backwards towards the medical model (36). Utilizing a core set would imply the focus on the disease and its consequences, rather than the bio-psycho-social impact on the individual or group. With the introduction of Environmental Factors and the focus on Activities and Participation, the ICF and the ICF Core Sets are well prepared to tackle this possible relapse to ICF's predecessor the ICIDH, in which this medical model was more prominent.

Opponents also claim that the ICF only addresses the negative aspects of having a specific health condition (i.e. what the patient cannot do), instead of the positive aspects (what the patient still can do despite of his health condition) (42). However the wording of the ICF is neutral (instead of the negative wording in the ICIDH) and addressing the limitations and impairments of the patient does not exclude having attention for the aspects which are without problems.

A shortcoming of the ICF system and subsequently the ICF Core Sets, is that personal factors are not included. Some attempts to resolve this problem were done (39-41) in the past, but the inclusion of personal factors is still experimental.

Therefore until further notice some space for personal factors has to be available when using the ICF Core Sets.

Finally, developed as a means to facilitate clinical reasoning, Core Sets should not feel like ‘corsets’, stifling reasoning and act like recipes in a cookbook. Space to describe the functioning of an individual patient is always required, also when using Core Sets. Core Sets are aids for the treatment and coaching of the patient besides many other tools used by health care workers. For example, Core Sets can help choosing patient related outcome measures (PROMs), because the Core Sets also contain the patients’ perspective. By using the Core Sets in selecting PROMs you know that relevant aspects are measured, leading to a better understanding of the experienced health and more insight in the quality of care given.

ICF Core Sets for lymphedema and the Dutch guideline for lymphedema

The Dutch guideline for lymphedema mentions the ICF Core Sets for lymphedema (13) as being under development. With the results presented in this thesis it is possible to integrate the Core Sets in the guideline from now on. For example, the conclusions of Chapter H3 ‘Consequences of lymphedema for the patient’ can be compared with the Core Sets. These conclusions were based on several reviews (42, 43). The consequences of lymphedema mentioned were: pain (part of all Core Sets), functional limitations (not included in the Core Sets because it is no ICF category, but when interpreted as limitations in activities part of all Core Sets), fatigue (present in all Comprehensive Core Sets, but only mentioned in the Brief Core Set for the upper limb), decreased physical activity (not included in the Core Sets because the description is too broad), decreased adaptation to illness (being a personal factor, which is not included in the present Core Sets), decreased social support (again this is a rather broad term, but all Core Sets have categories concerning this social support (chapter 3 of the list of environmental factors is about support)), loss of self-confidence (part of the Comprehensive Core Sets for upper and lower limb, but lacking in midline and in all Brief Core Sets), depression (only mentioned in the Comprehensive Core Set for upper limb lymphedema), and disturbed self-image (the term ‘self-image’ is only part of the Comprehensive Core

Set for midline lymphedema, but when interpreted as ‘body-image’ part of all Core Sets).

Integration of the ICF Core Sets into the Dutch guideline also provides the possibility to analyze the measurement instruments, advised by the guideline. As an example, two health-related quality of life questionnaires for lymphedema in the upper limb are compared with the Brief Core Set for upper limb lymphedema, namely the Lymph-ICF for the upper limb (44) and the Upper Limb Lymphedema 27 (ULL27) (18). The ULL27 is linked to the ICF following existing rules (45, 46), the Lymph-ICF is not linked according these rules, but translated by the authors, following their own interpretation and based on the results of the preparatory studies for the construction of this questionnaire (Table 1).

Table 1. HRQOL questionnaires and the Brief Core Set for upper limb lymphedema.

(+) = Code d4250 is no ICF code but added by the composers of the measurement instrument.

ICF code	Description (English / Dutch)	Lymph-ICF upper limb	ULL27	Brief ICF Core Set upper limb
b1263	Psychic stability / Psychische stabiliteit		**x**	**x**
b1265	Optimism / Optimisme		x	
b1266	Confidence / Vertrouwen	x	x	
b1301	Motivation / Motivatie		x	
b134	Sleep functions / Slaap		x	
b1341	Onset of sleep / Inslapen		x	
b1342	Maintenance of sleep / Doorslapen		x	
b152	Emotional functions / Stemming	x		
b1522	Range of emotion / Bereik van stemmingen		x	
b164	Higher level cognitive functions / Hogere cognitieve functies			x
b1801	Experience of self / Lichaamsbeeld			x
b265	Touch function / Tast		x	
b280	Sensation of pain / Pijngewaarwording		x	x
b18014	Pain in upper limb / Pijn in bovenste extremiteit	x		
b4352	Functions of lymphatic vessels / Functies van lymfevaten		x	
b439	Functions of the haematological and immunological systems, other specified and unspecified / Functies van hematologisch systeem en afweersysteem, anders gespecificeerd en niet gespecificeerd	x		
b4550	General physical endurance / Algemeen fysiek uithoudingsvermogen			x
b4552	Fatiguability / Vermoeibaarheid	x		x
b710	Mobility of joint functions / Mobiliteit van gewrichten			x
b7101	Mobility of several joints / Mobiliteit van meerdere gewrichten	x		
b730	Muscle power functions / Spiersterkte			x
b7301	Power of muscles of one limb / Sterkte van spieren in één ledemaat	x		
b840	Sensation related to the skin / Gewaarwording verband houdend met huid	x	x	
d230	Carrying out daily routine / Uitvoeren van dagelijkse routinehandelingen			x

d415	Maintaining a body position / Handhaven van lichaamshouding		x	
d4150	Maintaining a lying position / Handhaven van liggende houding	x		
d4250	Sunbathe / Zonnebaden (+)	x		
d4301	Carrying in the hands / Dragen in handen	x		
d4401	Grasping / Grijpen		x	
d445	Hand and arm use / Gebruiken van hand en arm		x	x
d4452	Reaching / Reiken	x	x	
d4453	Turning or twisting the hands or arms / Om- of ronddraaien met armen of handen		x	
d4458	Hand and arm use, other specified / Gebruiken van hand en arm, anders gespecificeerd	x		
d450	Walking / Lopen		x	
d4501	Walking long distances / Lopen van lange afstanden	x		
d4702	Using public motorized transportation / Gebruiken van gemotoriseerd openbaar vervoer		x	
d4750	Driving human-powered transportation / Besturen van door menskracht voortbewogen vervoermiddel	x		
d4751	Driving motorized vehicles / Besturen van gemotoriseerde voertuigen	x		
d5	SELF-CARE / ZELFVERZORGING			x
d5200	Caring for skin / Verzorgen van huid		x	
d5202	Caring for hair / Verzorgen van het haar		x	
d540	Dressing / Zich kleden	x	x	
d5404	Choosing appropriate clothing / Kiezen van juiste kleding			
d570	Looking after one’s health / Zorgdragen voor eigen gezondheid			x
d5701	Managing diet and fitness / Zorgdragen voor dieet en fitheid			x
d5702	Maintaining one’s health / Behouden van eigen gezondheid		x	
d6200	Shopping / Boodschappen doen		x	
d6300	Preparing simple meals / Bereiden van eenvoudige maaltijden	x		
d640	Doing housework / Huishouden doen			x
d6400	Washing and drying clothes and garments / Wassen en drogen van kleding		x	
d6402	Cleaning living area / Schoonmaken van woonruimte	x		
d6403	Using household appliances / Bedienen van huishoudelijke apparatuur	x		

d6505	Taking care of plants, indoors and outdoors / Verzorgen van planten, binnens- en buitenshuis	x		
d770	Intimate relationships / Intieme relaties			x
d7701	Spousal relationships / Samenlevingsrelaties		x	
d850	Remunerative employment / Betaald werk	x	x	x
d920	Recreation and leisure / Recreatie en vrije tijd		x	x
d9201	Sports / Sport	x		
d9202	Arts and culture / Kunst en cultuur		x	
d9204	Hobbies / Hobby's	x		
d9205	Socializing / Sociale activiteiten	x	x	
d9209	Recreation and leisure, unspecified / Recreatie en vrije tijd, niet gespecificeerd	x		
e1151	Assistive products and technology for personal use in daily living / Ondersteunende producten en technologie voor persoonlijk gebruik in het dagelijks leven			x
e310	Immediate family / Naaste familie		x	x
e320	Friends / Vrienden			x
e355	Health professionals / Hulpverleners in de gezondheidszorg			x
s420	Structure of immune system / Anatomische eigenschappen van afweersysteem			x
s730	Structure of upper extremity / Anatomische eigenschappen van bovenste extremiteit			x

As shown in table 1 it is possible to compare the similarities as well as the differences between the two questionnaires and the ICF Core Sets. It is also possible to compare the representation of the components of the ICF (Body Functions, Body Structures, Activities and Participation, and Environmental Factors) in the two questionnaires, in order to get an impression of which questionnaire has the broadest coverage of the ICF. In this case, for instance, the Lymph-ICF has no coverage of Environmental Factors.

Conclusion and recommendations

A consensus process integrating evidence and expert opinion contributed to the development of six solid ICF Core Sets for lymphedema. The Core Sets reflect the multidisciplinary and complex characteristics of treatment of patients with lymphedema. This development is a first step towards an internationally accepted and standardized framework when collecting and reporting variables or measures in the field and practice of lymphedema.

Since the ICF Core Sets for lymphedema are now available, it is possible to look at possibilities for implementation and for further research / validation.

1) The present versions are first versions, i.e. they need adaptation in daily practice to find out if the selected categories of the Core Sets are really that important and if other categories need to be added. The best way for health professionals to participate in the further development of the Core Sets is to seize this opportunity to debate and influence the direction of the next steps in this development. Since the project was initiated in The Netherlands, it would be advisable to start the implementation also in The Netherlands. The best way is to start top-down as well as bottom-up. If the Core Sets will be integrated in the education courses for lymphedema treatment and, on the other hand, the Core Sets are integrated in the Dutch guideline for lymphedema and the Dutch umbrella organizations concerning lymphedema (such as the Nederlandse Vereniging voor Dermatologie en Venereologie (NVDV), Nederlandse Vereniging voor Fysiotherapie binnen de Lymfologie (NVFL), and het Nederlands Lymfoedeem Netwerk (NLNet)) embrace them, this can be achieved easier. Moreover, also international organisations, like the International Lymphedema Framework (ILF) and the International Society for Lymphedema (ISL), can contribute to the implementation by including the Core Sets in their Best Practice Documents and education programs.

2) After the implementation of the Core Sets, a re-evaluation has to take place while considering the results of the validation and testing of the first

versions of the ICF Core Sets, preferably after about five years (phase III) (Figure 1).

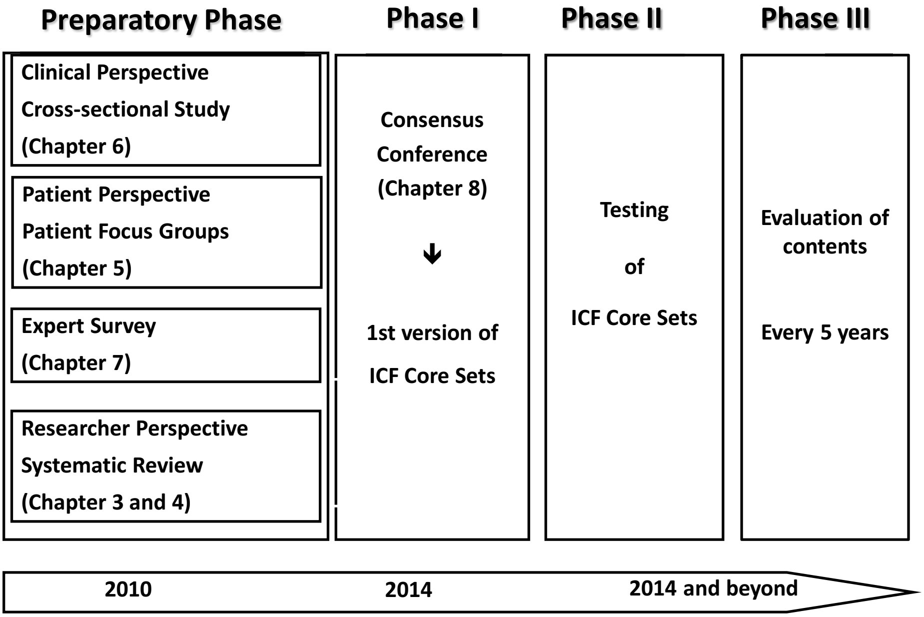

Fig 1. Addition of phase III in the schedule of the development of the ICF Core Sets for lymphedema.

3) It would also be advisable to implement the RPS form together with the ICF Core Sets in the electronic patient files, because of the compatibility between the two.

   Like all other ICF Core Sets, the Core Sets for lymphedema need adaptation for specific health professionals and countries. For instance physical therapists use other categories of the ICF than physicians or nurses. Cross-validation for different countries is needed, because of the differences in social and cultural aspects. For example, Zeldenryk et al found out that current generic disability measurement instruments used within the lymphatic filariasis field of Bangladesh, fail to adequately measure the known impact of lymphatic filariasis-related disability (47-

49). They conclude that there remains a need for a lymphatic filariasis-specific quality of life/disability measurement instrument, to adequately measure the impact of disability for individuals and communities.

4) Finally, with the Core Sets as a basis, basic sets of measurement instruments in lymphedema can be developed or chosen from the existing instruments.

All the above mentioned recommendations serve only one purpose, and that is to collect more information about the health condition and to facilitate and improve the lymphedema care for the individual patient. With this thesis being the start of it, the development of ICF Core Sets for lymphedema should be a continuing process.

“All ends with beginning”.

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Chapter 11

Summary/Samenvatting

Summary

This thesis describes a project on the development of ICF Core Sets for lymphedema. The motivation for this project is the fact that within lymphology little structure and unity can be found in the area of clinical reasoning, the use of measurement instruments and registration data sets, and patient education in mono- and multidisciplinary settings. ICF Core Sets are considered to offer an appropriate tool in this respect.

Chapter 1 is a general introduction of this thesis. Lymphedema is defined as an abnormal accumulation of fluid and protein in the body tissues as a result of a dis balance between the flow and the discharge of fluid. Lymphedema is a chronic condition and its influence on physical functioning can result in distress and loss of quality of life. Altered body shape and the necessity for daily use of specialized garments or bandaging as a standard treatment for stabilized lymphedema make acceptance of the condition difficult for the patient as well as for his or her family and environment. In addition, lymphedema requires lifelong self-care: patients must follow prescribed regimens, such as daily bandaging or the use of special garments, utilize skin care regimens, and exercise caution with open wounds. Generally, the activity and participation levels of persons with lymphedema are lower than those of their healthy peers. The ICF is accepted by the World Health Assembly as a universal framework for classifying and describing functioning in people with specific health conditions. Although the ICF classification system with more than 1400 categories can serve as a reference, the ICF is not easily applicable in clinical practice. Therefore, tools such as ICF Core Sets (health-specific selections of ICF categories) are helpful to make the ICF more appealing for implementation in medical/allied health care. The advantages of the use of ICF Core Sets are in the field of: determining relevant outcome measures, making a proper choice of measurement instruments, offering a framework for guidelines, giving structure in reporting and clinical reasoning, and giving the possibility to determine the severity of certain components of functioning, to generate more knowledge about lymphedema, to help in communicating with patients, and to provide structure in multidisciplinary settings.

In **chapter 2** we give an overview of the development process to come to ICF Core Sets for lymphedema. The process contains two phases: a preparatory (developmental) phase and a validation (implementation) phase. In this thesis the developmental phase will be described. In this phase five preparatory studies were performed: a literature review from the researchers' point of view (this study existed of two separate researches), a qualitative research from the patients' perspective, a cross-sectional clinical study to reveal the health professionals' point of view, and an expert research among experts in the field of lymphedema. The data of four of the five preparatory studies were presented at an international consensus conference resulting in the first versions of the Core Sets. The second phase (validation and implementation) is beyond this thesis.

Chapter 3 discusses the first literature review. In this research we explored the measurement instruments for lymphedema and especially the lymphedema-specific questionnaires. The meaningful concepts in these questionnaires were linked to the ICF in order to obtain frequency tables of the most occurring ICF categories. Of the - by means of a structured search procedure - 2381 abstracts retrieved, 136 studies were included, based on the in- and exclusion criteria. The selected studies were divided in 88 about lymphedema in the upper limb (65%), 34 about the lower limb (25%), five about the midline (3%) and nine about combinations of these areas (7%). Considering the questionnaires, it is striking that there are 51 different questionnaires in the included studies, so it seems that every researcher used his/her own questionnaire. In total, 12 lymphedema-specific questionnaires were found (nine for the upper limb, two for the lower limb and one for lymphedema in general). A total of 301 'meaningful concepts' was extracted from the questionnaires, of which 271 (90%) could be linked to the ICF. There were 45 two-level ICF categories linked to concepts in more than two questionnaires; 13 in Body Functions, six in Body Structures, 16 in Activities and Participation, and 10 in Environmental Factors. The most frequently measured categories were 'Structure of upper extremity', 'Immunological system functions', 'Looking after one's health', 'Sensation of pain', 'Touch functions', 'Dressing', and 'Health services, systems and policies'. The ICF appeared a valuable means to point out items from questionnaires, which are important for patients with lymphedema.

Chapter 4 describes a second analysis of the literature. In this review the Methods and Results section of the 136 selected articles were explored for ‘meaningful concepts’, which were linked to the ICF, resulting in frequency tables of the most occurring ICF categories. The ICF categories of the studies 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. By examining both the questionnaires and the studies themselves, a useful selection of ICF categories has been identified. We recommend the use of both questionnaires and studies because these approaches are complementary and because the use of both methods yields more data than can be obtained by using either method alone.

In **chapter 5** we describe the translation and cross-validation (i.e. the applicability of the questions in the Dutch culture and society) from French into Dutch of one of the analyzed health-related quality of life questionnaires for upper limb lymphedema; the Upper Limb Lymphedema 27 (ULL27). In chapter 3 this questionnaire is linked to the ICF. Considering the Dutch version the internal consistency of the domains of the questionnaire (physical, psychological and social dimensions) was good as were the convergent validity and discriminant validity. Upper limb volume and the domains of the ULL27 were not correlated. The Dutch translation of the ULL27 questionnaire has good internal consistency and validity but further research is needed to determine its responsiveness.

The study to explore the patients’ perspective is described in **chapter 6**. We performed a qualitative research with focus groups of lymphedema patients to identify and quantify ‘meaningful concepts’ from the patients’ perspective about the most important factors, which influence functioning and health-related quality of life. This was done identifying and subsequently quantifying ‘meaningful concepts’ about lymphedema. Six focus group interviews in five different centers were organized, audiotaped, transcribed verbatim and analyzed. A total of 2681 relevant ICF linkings were performed with the focus group data, resulting in 130 different second-level categories. Of these 130 second-level categories, 41 (31.5%) categories were categorized as Body Functions, 20 (15.5%) as Body

Structures, 41 (31.5%) as Activities and Participation, and 28 (21.5%) as Environmental Factors. Overall, the most important issues according to the patients were the use of hosiery and bandages, support and relationships, and the fact that their arms and legs were so malformed. Based on their experiences with lymphedema, patients reported activity limitations and participation restrictions combined with impaired body functions. Anatomical changes (Body Structures) were also often mentioned as a problem in daily life. Environmental factors such as health professionals, hosiery and family members are reported to act as a barrier or facilitator for patient functioning.

Chapter 7 reports the results of a cross-sectional survey in which lymphedema patients (n = 200) were interviewed by their health professionals on the basis of a specific questionnaire. This questionnaire contained the items that resulted from the literature review of the questionnaires. By means of the Parantion database the severity of specific ICF categories could be determined for each patient, making it possible to describe the functioning of lymphedema patients and to identify their problems. The most frequent mentioned items were in the Body Functions component: muscle power and mobility of joints, in the Activities and Participation component: doing housework, and changing and maintaining a body position, and in the Environmental Factors component: Health professionals, who can act as both facilitators and barriers. Interestingly, patients assessed their health more positively than health professionals do.

Chapter 8 describes the results of an email based worldwide survey among experts in the field of lymphedema with questions based on components of the ICF. The purpose was to identify and quantify meaningful concepts in lymphedema from health professionals' perspectives. In total, 142 health professionals from seven different health professions and 20 different countries answered the questions. The answers of the health professionals included 7373 aspects of functioning, of which 6296 could be linked to 359 different ICF categories. Of these categories, 109 belonged to body functions (30.4%), 55 to body structures (15.3%), 121 to activities and participation (33.7%), and 74 to environmental factors (20.6%). Overall, the most mentioned items were: ' Health services, systems and policies' (686 times), 'Immunological system functions' (563 times), ‘Looking after one’s health’ (385 times), ‘Products and technology for personal use in daily life’ (299 times), and ‘Dressing’ (285 times).

Chapter 9 presents the outcomes of the consensus conference to determine the first ICF Core Sets for lymphedema, based on the preparatory studies. In order to achieve this, frequency rankings were made of the ICF categories derived from these studies. By means of discussions in working groups and plenary sessions with invited international experts in the field of lymphedema from different professions and countries, final consensus on ICF categories was achieved. This resulted in Comprehensive and Brief Core Sets for lymphedema, each for three regions: upper limb, lower limb and midline lymphedema. ICF Core Sets for lymphedema appeared to contain different items in relation with the region of the edema. For instance in midline lymphedema urinating and sexual functions are important and in lymphedema of the upper or lower limb the mobility of joints. In the future these Core Sets need to be validated for different groups of health professions and for different countries.

In **Chapter 10** the main findings from the studies presented in this thesis are discussed and placed into a broader perspective. Thus, a comparison is made with the development of other Core Sets and certain criticisms discussed about the Core Sets. In addition, the Core Sets for lymphedema are compared with the Dutch Guideline for Lymphedema. It appears that the ICF Core Sets can be properly implemented in certain parts of the guideline. The distinction between measurement instruments recommended by the guideline with respect to the Core Sets can be made. As an example the ULL27 and Lymph-ICF for the upper extremity questionnaire are discussed. Furthermore, the limitations of the various studies and the project in total are discussed and suggestions about validation and implementation procedures are given.

Samenvatting

Dit proefschrift beschrijft een project over de ontwikkeling van ICF Core Sets voor lymfoedeem. De motivatie voor dit project komt voort uit het feit dat er binnen de lymfologie weinig structuur en eenheid is op het gebied van: klinisch redeneren, het gebruik van meetinstrumenten, het vastleggen van datasets en voorlichting van de patiënt in mono- en multidisciplinaire omgevingen. In dit opzicht worden ICF Core Sets gezien als een geschikt instrument.

Hoofdstuk 1 is een algemene introductie van dit proefschrift. Lymfoedeem wordt gedefinieerd als een abnormale ophoping van vocht en eiwit in de lichaamsweefsels als gevolg van een disbalans tussen aanvoer en afvoer van vocht. Lymfoedeem is een chronische aandoening en heeft invloed op het fysiek functioneren. Dit kan resulteren in pijn, angst en verlies van kwaliteit van leven. De veranderde lichaamsvorm maakt het noodzakelijk dagelijks gebruik te moeten maken van speciale kleding of zwachtels (standaardbehandeling voor gestabiliseerd lymfoedeem). Dit maakt de acceptatie van de aandoening moeilijk voor de patiënt, familie en omgeving. Daarnaast vereist lymfoedeem levenslange zelfzorg: patiënten moeten voorgeschreven adviezen opvolgen, zoals het dagelijks zwachtelen of het gebruik van speciale kledingstukken, huidverzorging en het voorzichtig zijn met wondjes. In het algemeen zijn de activiteiten en participatie van mensen met lymfoedeem minder dan die van hun gezonde leeftijdsgenoten. De ICF is door de World Health Assembly erkend als een universeel raamwerk voor de classificatie en beschrijving van de functie van mensen met specifieke gezondheidsproblemen. Hoewel de ICF als classificatiesysteem met meer dan 1400 categorieën kan dienen als naslagwerk, is de ICF niet eenvoudig toepasbaar in de klinische praktijk. Daarom zijn hulpmiddelen zoals ICF Core Sets (gezondheid-specifieke selecties van ICF categorieën) nuttig om de ICF makkelijker toepasbaar te maken voor het gebruik in de medische- en paramedische zorg. De voordelen van het gebruik van ICF Core Sets liggen op het gebied van: het bepalen van relevante uitkomstmaten, het maken van een geschikte keuze van meetinstrumenten, het bieden van een raamwerk voor richtlijnen, het bieden van structuur in rapportage en klinisch redeneren, de mogelijkheid om de ernst te bepalen van bepaalde onderdelen van de functie, het genereren van meer kennis over lymfoedeem, de communicatie met de patiënt en het bieden van structuur in de multidisciplinaire samenwerking.

In **hoofdstuk 2** geven we een overzicht van het ontwikkelingsproces om te komen tot ICF Core Sets voor lymfoedeem. Het proces bestaat uit twee fasen: een voorbereidende (ontwikkelings) fase en een validatie (implementatie) fase. In dit proefschrift wordt de ontwikkelingsfase beschreven. In deze fase werden vijf voorbereidende studies uitgevoerd: een literatuurstudie vanuit het perspectief van de onderzoeker (deze studie bestond uit twee afzonderlijke onderzoeken), een kwalitatief onderzoek vanuit het standpunt van de patiënt, een cross-sectionele klinische studie om het beeld van de hulpverlener te bepalen en een expert onderzoek onder deskundigen op het gebied van lymfoedeem. De gegevens van vier van de vijf voorbereidende studies werden gepresenteerd op een internationale consensus conferentie, hetgeen resulteerde in de eerste versies van de Core Sets. De tweede fase (validatie en implementatie) valt buiten dit proefschrift.

Hoofdstuk 3 beschrijft het eerste literatuuronderzoek. **I**n dit onderzoek onderzochten we de meetinstrumenten voor lymfoedeem en met name de lymfoedeem-specifieke vragenlijsten. De ‘meaningful concepts’ (items die van betekenis zijn) in deze vragenlijsten werden gecodeerd m.b.v. de ICF, teneinde frequentietabellen te krijgen van de meest voorkomende ICF categorieën. Van de, via een gestructureerde zoekprocedure, 2381 gevonden abstracts, werden 136 studies opgenomen op basis van de in- en uitsluitingscriteria. De geselecteerde studies waren als volgt verdeeld: 88 over lymfoedeem in de bovenste extremiteit (65%), 34 betreffende de onderste extremiteit (25%), vijf over midline (3%) en negen over combinaties van deze gebieden (7%). Bij nadere beschouwing van de vragenlijsten valt op dat er in totaal 51 verschillende vragenlijsten in de studies voorkomen. Het lijkt erop dat elke onderzoeker zijn eigen vragenlijst gebruikt. In totaal werden 12 lymfoedeem-specifieke vragenlijsten aangetroffen (negen voor de bovenste extremiteit, twee voor de onderste extremiteit en een voor lymfoedeem in het algemeen). Een totaal van 301 'meaningful concepts’ werd gevonden in de vragenlijsten, waarvan 271 (90%) konden worden gekoppeld aan de ICF. Er werden 45 tweede-level ICF categorieën gekoppeld in meer dan twee vragenlijsten; 13 behorend tot Functies, zes tot Anatomische eigenschappen, 16 tot Activiteiten en Participatie en 10 tot Externe factoren. De meest frequente categorieën waren “Anatomische eigenschappen van de bovenste extremiteit", "Functies van het afweersysteem”, “Zorgdragen voor eigen gezondheid”, “Pijngewaarwording”, “Tast functies”, “Zich kleden” en “Voorzieningen, systemen en beleid met betrekking tot gezondheidszorg”. De ICF bleek een waardevol middel te zijn om uit vragenlijsten items te bepalen, die belangrijk zijn voor patiënten met lymfoedeem.

Hoofdstuk 4 beschrijft een tweede analyse van de literatuur. In deze studie werden de Methode en Resultaten secties van de 136 geselecteerde artikelen onderzocht op ‘meaningful concepts’. Deze werden gecodeerd met de ICF, waardoor frequentietabellen van de meest voorkomende ICF categorieën ontstonden. De ICF categorieën van de studies werden vervolgens vergeleken met de ICF categorieën uit het onderzoek over de vragenlijsten om te bepalen of onderzoekers deze items ook daadwerkelijk onderzochten. Door zowel de vragenlijsten als de studies zelf te onderzoeken, kwam een bruikbare selectie van ICF categorieën naar voren. We raden het gebruik van zowel vragenlijsten als studies aan. Het gebruik van beide methoden samen levert meer data op dan afzonderlijk, daar zij complementair zijn.

In **hoofdstuk 5** beschrijven we de vertaling en cross-validatie (de toepasbaarheid van de vragen in de Nederlandse cultuur en samenleving) van een van de geanalyseerde gezondheid gerelateerde kwaliteit van leven vragenlijsten voor lymfoedeem van de bovenste extremiteit; de Upper Limb lymphedema 27 (ULL27) vanuit het Frans naar het Nederlands. In hoofdstuk 3 is deze vragenlijst gecodeerd met de ICF. De interne consistentie van de domeinen van de Nederlandse versie van deze vragenlijst (fysieke, psychologische en sociale dimensies) was goed net als de convergente en discriminante validiteit. Het volume van de bovenste extremiteit en de domeinen van de ULL27 waren niet gecorreleerd. De Nederlandse vertaling van de ULL27 vragenlijst heeft een goede interne consistentie en validiteit, maar verder onderzoek is nodig om de responsiviteit te bepalen.

De studie om het perspectief van de patiënten te onderzoeken is beschreven in **hoofdstuk 6.** We hebben een kwalitatief onderzoek gedaan met focusgroepen van lymfoedeem patiënten om 'meaningful concepts' te identificeren en te kwantificeren vanuit het perspectief van de patiënt over de belangrijkste factoren die van invloed zijn op de functie en de gezondheid gerelateerde kwaliteit van leven. Dit werd gedaan door 'meaningful concepts' over lymfoedeem te identificeren en vervolgens te kwantificeren. Zes focusgroep interviews in vijf verschillende centra werden georganiseerd, op band opgenomen, uitgetypt en geanalyseerd. Met de focus groep gegevens werd een totaal van 2681 relevante ICF categorieën gevonden. Dit resulteerde in 130 verschillende categorieën op het tweede niveau. Van deze 130 tweede niveau categorieën, werden 41 (31,5%) categorieën gecategoriseerd als Functies, 20 (15,5%) als Anatomische eigenschappen, 41 (31,5%) als behorend bij Activiteiten en Participatie en 28 (21,5%) als Externe factoren. In het algemeen waren, volgens de patiënten, de belangrijkste problemen: misvormde ledematen, gebruik moeten maken van kousen en bandages, het nodig hebben van ondersteuning en nadelige invloed op relaties. Op basis van hun ervaringen met lymfoedeem gaven patiënten aan dat ze beperkingen in activiteiten en restricties in participatie hadden, in combinatie met stoornissen in lichaamsfuncties. Anatomische veranderingen (Anatomische eigenschappen) werden vaak beschouwd als een probleem in het dagelijks leven. Verder kunnen omgevingsfactoren zoals hulpverleners, kousen en familieleden een herstel belemmerende of bevorderende factor voor de patiënt zijn.

Hoofdstuk 7 vermeldt de resultaten van een cross-sectioneel onderzoek waarin lymfoedeem patiënten (n = 200) werden geïnterviewd door hun hulpverleners op basis van een specifieke vragenlijst. Deze vragenlijst bevatte de items die voortvloeiden uit het literatuuronderzoek over de vragenlijsten. Via de Parantion database kon de ernst van specifieke ICF categorieën worden bepaald voor elke patiënt, waardoor het mogelijk was om de functie van patiënten met lymfoedeem te beschrijven en hun problemen te identificeren. De meest voorkomende items waren in de Functies component: spierkracht en mobiliteit van de gewrichten, in de Activiteiten en Participatie component: het doen van huishoudelijk werk en het veranderen en onderhouden van bepaalde lichaamshoudingen en in de Externe factoren component: zorgverleners, die zowel kunnen optreden als herstel belemmerende als herstel bevorderende factoren. Interessant is dat patiënten hun gezondheid positiever beoordelen dan hulpverleners dat doen.

Hoofdstuk 8 beschrijft de resultaten n.a.v. een wereldwijd e-mail onderzoek onder lymfoedeem deskundigen met vragen op het gebied van lymfoedeem gebaseerd op onderdelen van de ICF. Het doel was om ‘meaningful concepts’ over lymfoedeem te identificeren en te kwantificeren vanuit het perspectief van de hulpverlener. In totaal 142 hulpverleners van zeven verschillende beroepen in de gezondheidszorg en uit 20 verschillende landen beantwoordden de vragen. De antwoorden van de hulpverleners bevatten 7373 aspecten over de functie, waarvan 6296 konden worden gekoppeld aan 359 verschillende ICF categorieën. Van deze categorieën, waren er 109 betreffende Functies (30,4%), 55 over Anatomische eigenschappen (15,3%), 121 over Activiteiten en Participatie (33,7%) en 74 betreffende Externe factoren (20,6%). De meest genoemde punten waren over het algemeen: “Voorzieningen, systemen en beleid met betrekking tot gezondheidszorg” (686 keer), "Functies van het afweersysteem” (563 keer), “Zorgdragen voor eigen gezondheid” (385 keer), “Producten en technologie voor persoonlijk gebruik in het dagelijks leven” (299 keer) en “Zich kleden” (285 keer).

Hoofdstuk 9 beschrijft de uitkomsten van de consensusconferentie om de eerste ICF Core Sets voor lymfoedeem te bepalen, op basis van de voorbereidende studies. Om dit te bereiken werden van deze studies frequentie rangordes van de ICF categorieën gemaakt. Door middel van discussies in werkgroepen en plenaire sessies met uitgenodigde internationale experts op het gebied van lymfoedeem uit verschillende beroepen en landen, werd definitieve consensus over ICF categorieën bereikt. Dit resulteerde in uitgebreide en korte Core Sets voor lymfoedeem, elk voor drie regio's: de bovenste ledematen, onderste ledematen en midline lymfoedeem. ICF Core Sets voor lymfoedeem bleken verschillende items in relatie tot de regio van het oedeem te bevatten. Bijvoorbeeld bij midline lymfoedeem zijn urineren en seksuele functies belangrijk en bij lymfoedeem van de bovenste of onderste ledematen de mobiliteit van de gewrichten. In de toekomst is het noodzakelijk dat deze Core Sets worden gevalideerd voor de verschillende beroepen in de gezondheidszorg en verschillende landen.

In **hoofdstuk 10** worden de belangrijkste bevindingen uit de studies in dit proefschrift besproken en geplaatst in een breder perspectief. Zo wordt een vergelijking gemaakt met de ontwikkeling van andere Core Sets en bestaande kritiek over de Core Sets besproken. Daarnaast worden de Core Sets voor lymfoedeem vergeleken met de Nederlandse Richtlijn voor lymfoedeem. Het blijkt dat de ICF Core Sets goed in bepaalde delen van de richtlijn kunnen worden geïmplementeerd. Het onderscheid tussen meetinstrumenten aanbevolen door de richtlijn ten opzichte van de Core Sets kan worden gemaakt. Als voorbeeld worden de ULL27 en de Lymph-ICF (vragenlijsten voor de bovenste extremiteit) besproken. Bovendien worden de beperkingen van de diverse studies en het project in totaal besproken en suggesties over de validatie en implementatie gedaan.

Chapter 12

Appendix Australian Data
Appendix ICF Core Sets
List of co-authors
List of publications
Curriculum Vitae
PhD Portfolio
Dankwoord

Appendix Australian Data

Table 1. Total amount of ICF categories mentioned by patients with upper limb lymphedema (n = 42).

ID_ICF	Title	Clinical research Australia
b	BODY FUNCTIONS	
b1	CHAPTER 1 MENTAL FUNCTIONS	
b126	Temperament and personality functions	4
b130	Energy and drive functions	12
b152	Emotional functions	7
b160	Thought functions	2
b164	Higher-level cognitive functions	2
b180	Experience of self and time functions	1
b2	CHAPTER 2 SENSORY FUNCTIONS AND PAIN	
b210	Seeing functions	1
b265	Touch function	13
b270	Sensory functions related to temperature and other stimuli	11
b280	Sensation of pain	21
b4	CHAPTER 4 FUNCTIONS OF THE CARDIOVASCULAR, HAEMATOLOGICAL, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS	
b415	Blood vessel functions	4
b435	Immunological system functions	15
b455	Exercise tolerance functions	15
b5	CHAPTER 5 FUNCTIONS OF THE DIGESTIVE, METABOLIC AND ENDOCRINE SYSTEMS	
b530	Weight maintenance functions	9
b540	General metabolic functions	2
b550	Thermoregulatory functions	8
b6	CHAPTER 6 GENITOURINARY AND REPRODUCTIVE FUNCTIONS	
b640	Sexual functions	1
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS	
b710	Mobility of joint functions	11
b730	Muscle power functions	12
b8	CHAPTER 8 FUNCTIONS OF THE SKIN AND RELATED STRUCTURES	
b810	Protective functions of the skin	21
b820	Repair functions of the skin	20
b830	Other functions of the skin	13
d	ACTIVITIES AND PARTICIPATION	
d1	CHAPTER 1 LEARNING AND APPLYING KNOWLEDGE	
d155	Acquiring skills	2

d170	Writing	4
d2	CHAPTER 2 GENERAL TASKS AND DEMANDS	
d230	Carrying out daily routine	18
d3	CHAPTER 3 COMMUNICATION	
d325	Communicating with - receiving - written messages	1
d345	Writing messages	3
d360	Using communication devices and techniques	3
d4	CHAPTER 4 MOBILITY	
d410	Changing basic body position	15
d415	Maintaining a body position	13
d430	Lifting and carrying objects	27
d445	Hand and arm use	23
d450	Walking	8
d460	Moving around in different locations	7
d5	CHAPTER 5 SELF-CARE	
d510	Washing oneself	2
d520	Caring for body parts	5
d540	Dressing	9
d550	Eating	2
d570	Looking after one’s health	1
d6	CHAPTER 6 DOMESTIC LIFE	
d640	Doing housework	17
d660	Assisting others	4
d7	CHAPTER 7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS	
d710	Basic interpersonal interactions	2
d720	Complex interpersonal interactions	1
d750	Informal social relationships	2
d770	Intimate relationships	3
d8	CHAPTER 8 MAJOR LIFE AREAS	
d845	Acquiring, keeping and terminating a job	4
d850	Remunerative employment	5
d870	Economic self-sufficiency	6
d9	CHAPTER 9 COMMUNITY, SOCIAL AND CIVIC LIFE	
d910	Community life	5
d920	Recreation and leisure	17
e	ENVIRONMENTAL FACTORS	
e1	CHAPTER 1 PRODUCTS AND TECHNOLOGY	
e110	Products or substances for personal consumption	5

e115	Products and technology for personal use in daily living	28
e125	Products and technology for communication	11
e130	Products and technology for education	22
e135	Products and technology for employment	3
e140	Products and technology for culture, recreation and sport	9
e150	Design, construction and building products and technology of buildings for public use	5
e155	Design, construction and building products and technology of buildings for private use	8
e165	Assets	15
e2	CHAPTER 2 NATURAL ENVIRONMENT AND HUMAN-MADE CHANGES TO ENVIRONMENT	
e210	Physical geography	4
e215	Population	3
e225	Climate	33
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS	
e310	Immediate family	33
e315	Extended family	17
e320	Friends	30
e325	Acquaintances, peers, colleagues, neighbours and community members	19
e330	People in positions of authority	18
e355	Health professionals	33
e4	CHAPTER 4 ATTITUDES	
e410	Individual attitudes of immediate family members	14
e415	Individual attitudes of extended family members	13
e420	Individual attitudes of friends	18
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	18
e460	Societal attitudes	10
e465	Social norms, practices and ideologies	2
e5	CHAPTER 5 SERVICES, SYSTEMS AND POLICIES	
e535	Communication services, systems and policies	14
e555	Associations and organizational services, systems and policies	16
e570	Social security services, systems and policies	19
e580	Health services, systems and policies	33
e585	Education and training services, systems and policies	15
s	BODY STRUCTURES	
s4	CHAPTER 4 STRUCTURES OF THE CARDIOVASCULAR, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS	

s420	Structure of immune system	13
s6	CHAPTER 6 STRUCTURES RELATED TO THE GENITOURINARY AND REPRODUCTIVE SYSTEMS	
s630	Structure of reproductive system	3
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT	
s720	Structure of shoulder region	15
s730	Structure of upper extremity	15
s750	Structure of lower extremity	2
s760	Structure of trunk	9
s8	CHAPTER 8 SKIN AND RELATED STRUCTURES	
s810	Structure of areas of skin	14

Table 2. Total amount of ICF categories mentioned by patients with midline lymphedema (n = 1).

ID_ICF	Title	Clinical research Australia
b	BODY FUNCTIONS	
b1	CHAPTER 1 MENTAL FUNCTIONS	
b126	Temperament and personality functions	1
b130	Energy and drive functions	1
b2	CHAPTER 2 SENSORY FUNCTIONS AND PAIN	
b280	Sensation of pain	1
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS	
b730	Muscle power functions	1
d4	CHAPTER 4 MOBILITY	
d410	Changing basic body position	1
d415	Maintaining a body position	1
d430	Lifting and carrying objects	1
d5	CHAPTER 5 SELF-CARE	
d510	Washing oneself	1
d520	Caring for body parts	1
d540	Dressing	1
d6	CHAPTER 6 DOMESTIC LIFE	
d640	Doing housework	1
d8	CHAPTER 8 MAJOR LIFE AREAS	
d845	Acquiring, keeping and terminating a job	1
d850	Remunerative employment	1
d9	CHAPTER 9 COMMUNITY, SOCIAL AND CIVIC LIFE	
d920	Recreation and leisure	1
e	ENVIRONMENTAL FACTORS	
e1	CHAPTER 1 PRODUCTS AND TECHNOLOGY	
e110	Products or substances for personal consumption	1
e115	Products and technology for personal use in daily living	1
e155	Design, construction and building products and technology of buildings for private use	1
e2	CHAPTER 2 NATURAL ENVIRONMENT AND HUMAN-MADE CHANGES TO ENVIRONMENT	
e225	Climate	1
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS	
e310	Immediate family	1
e315	Extended family	1

e320	Friends	1
e325	Acquaintances, peers, colleagues, neighbours and community members	1
e330	People in positions of authority	1
e355	Health professionals	1
e4	CHAPTER 4 ATTITUDES	
e410	Individual attitudes of immediate family members	1
e415	Individual attitudes of extended family members	1
e420	Individual attitudes of friends	1
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	1
e460	Societal attitudes	1
e5	CHAPTER 5 SERVICES, SYSTEMS AND POLICIES	
e555	Associations and organizational services, systems and policies	1
e570	Social security services, systems and policies	1
e580	Health services, systems and policies	1
s4	CHAPTER 4 STRUCTURES OF THE CARDIOVASCULAR, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS	
s420	Structure of immune system	1
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT	
s720	Structure of shoulder region	1
s760	Structure of trunk	1
s8	CHAPTER 8 SKIN AND RELATED STRUCTURES	
s810	Structure of areas of skin	1

Table 3. Total amount of ICF categories mentioned by patients with lower limb lymphedema (n = 47).

ID_ICF	Title	Clinical research Australia
b	BODY FUNCTIONS	
b1	CHAPTER 1 MENTAL FUNCTIONS	
b114	Orientation functions	1
b126	Temperament and personality functions	13
b130	Energy and drive functions	20
b152	Emotional functions	13
b160	Thought functions	1
b164	Higher-level cognitive functions	3
b180	Experience of self and time functions	4
b2	CHAPTER 2 SENSORY FUNCTIONS AND PAIN	
b210	Seeing functions	0
b265	Touch function	11
b270	Sensory functions related to temperature and other stimuli	18
b280	Sensation of pain	24
b4	CHAPTER 4 FUNCTIONS OF THE CARDIOVASCULAR, HAEMATOLOGICAL, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS	
b415	Blood vessel functions	5
b435	Immunological system functions	22
b455	Exercise tolerance functions	27
b5	CHAPTER 5 FUNCTIONS OF THE DIGESTIVE, METABOLIC AND ENDOCRINE SYSTEMS	
b530	Weight maintenance functions	16
b540	General metabolic functions	8
b550	Thermoregulatory functions	17
b6	CHAPTER 6 GENITOURINARY AND REPRODUCTIVE FUNCTIONS	
b640	Sexual functions	4
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS	
b710	Mobility of joint functions	20
b730	Muscle power functions	22
b8	CHAPTER 8 FUNCTIONS OF THE SKIN AND RELATED STRUCTURES	
b810	Protective functions of the skin	26
b820	Repair functions of the skin	20
b830	Other functions of the skin	19
d	ACTIVITIES AND PARTICIPATION	
d2	CHAPTER 2 GENERAL TASKS AND DEMANDS	

d230	Carrying out daily routine	10
d3	CHAPTER 3 COMMUNICATION	
d330	Speaking	1
d4	CHAPTER 4 MOBILITY	
d410	Changing basic body position	26
d415	Maintaining a body position	21
d430	Lifting and carrying objects	8
d445	Hand and arm use	1
d450	Walking	26
d460	Moving around in different locations	20
d5	CHAPTER 5 SELF-CARE	
d510	Washing oneself	8
d520	Caring for body parts	9
d540	Dressing	13
d570	Looking after one’s health	2
d6	CHAPTER 6 DOMESTIC LIFE	
d640	Doing housework	15
d660	Assisting others	10
d7	CHAPTER 7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS	
d710	Basic interpersonal interactions	3
d720	Complex interpersonal interactions	3
d750	Informal social relationships	8
d770	Intimate relationships	5
d8	CHAPTER 8 MAJOR LIFE AREAS	
d845	Acquiring, keeping and terminating a job	10
d850	Remunerative employment	7
d870	Economic self-sufficiency	5
d9	CHAPTER 9 COMMUNITY, SOCIAL AND CIVIC LIFE	
d910	Community life	12
d920	Recreation and leisure	21
d930	Religion and spirituality	3
e	ENVIRONMENTAL FACTORS	
e1	CHAPTER 1 PRODUCTS AND TECHNOLOGY	
e110	Products or substances for personal consumption	14
e115	Products and technology for personal use in daily living	31
e125	Products and technology for communication	11
e130	Products and technology for education	18
e135	Products and technology for employment	2

e140	Products and technology for culture, recreation and sport	8
e150	Design, construction and building products and technology of buildings for public use	22
e155	Design, construction and building products and technology of buildings for private use	12
e165	Assets	6
e2	CHAPTER 2 NATURAL ENVIRONMENT AND HUMAN-MADE CHANGES TO ENVIRONMENT	
e210	Physical geography	9
e215	Population	1
e225	Climate	32
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS	
e310	Immediate family	34
e315	Extended family	24
e320	Friends	27
e325	Acquaintances, peers, colleagues, neighbours and community members	22
e330	People in positions of authority	19
e355	Health professionals	37
e4	CHAPTER 4 ATTITUDES	
e410	Individual attitudes of immediate family members	21
e415	Individual attitudes of extended family members	16
e420	Individual attitudes of friends	16
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members	13
e460	Societal attitudes	15
e465	Social norms, practices and ideologies	3
e5	CHAPTER 5 SERVICES, SYSTEMS AND POLICIES	
e535	Communication services, systems and policies	9
e555	Associations and organizational services, systems and policies	13
e570	Social security services, systems and policies	17
e580	Health services, systems and policies	31
e585	Education and training services, systems and policies	10
s	BODY STRUCTURES	
s4	CHAPTER 4 STRUCTURES OF THE CARDIOVASCULAR, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS	
s410	Structure of cardiovascular system	3
s420	Structure of immune system	26
s6	CHAPTER 6 STRUCTURES RELATED TO THE GENITOURINARY AND REPRODUCTIVE SYSTEMS	
s630	Structure of reproductive system	4

s630	Structure of reproductive system	4
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT	
s740	Structure of pelvic region	3
s750	Structure of lower extremity	34
s760	Structure of trunk	1
s8	CHAPTER 8 SKIN AND RELATED STRUCTURES	
s810	Structure of areas of skin	29

Appendix ICF Core Sets

Table 1. Comprehensive ICF Core Set for lymphedema in upper extremity.

b1	CHAPTER 1 MENTAL FUNCTIONS
b1263	Psychic stability
b1264	Openness to experience
b1265	Optimism
b1266	Confidence
b1301	Motivation
b134	Sleep functions
b152	Emotional functions
b164	Higher-level cognitive functions
b1644	Insight
b1801	Body image
b260	Proprioceptive function
b265	Touch function
b2702	Sensitivity to pressure
b280	Sensation of pain
b435	Immunological system functions
b4352	Functions of lymphatic vessels
b4550	General physical endurance
b4552	Fatiguability
b530	Weight maintenance functions
b5403	Fat metabolism
b640	Sexual functions
b7	CHAPTER 7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS
b710	Mobility of joint functions
b730	Muscle power functions
b780	Sensations related to muscles and movement functions
b810	Protective functions of the skin
b820	Repair functions of the skin
b840	Sensation related to the skin
d155	Acquiring skills
d170	Writing
d230	Carrying out daily routine

d2400	Handling responsibilities
d3	CHAPTER 3 COMMUNICATION
d360	Using communication devices and techniques
d4	CHAPTER 4 MOBILITY
d410	Changing basic body position
d415	Maintaining a body position
d430	Lifting and carrying objects
d4401	Grasping
d445	Hand and arm use
d4452	Reaching
d450	Walking
d4554	Swimming
d4702	Using public motorized transportation
d475	Driving
d5	CHAPTER 5 SELF-CARE
d510	Washing oneself
d5200	Caring for skin
d5202	Caring for hair
d530	Toileting
d540	Dressing
d5404	Choosing appropriate clothing
d570	Looking after one’s health
d5700	Ensuring one’s physical comfort
d5701	Managing diet and fitness
d5702	Maintaining one's health
d6200	Shopping
d630	Preparing meals
d640	Doing housework
d6400	Washing and drying clothes and garments
d6505	Taking care of plants, indoors and outdoors
d660	Assisting others
d7	CHAPTER 7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS

d770	Intimate relationships
d850	Remunerative employment
d870	Economic self-sufficiency
d920	Recreation and leisure
d9201	Sports
d9205	Socializing
e1101	Drugs
e1150	General products and technology for personal use in daily living
e1151	Assistive products and technology for personal use in daily living
e1300	General products and technology for education
e150	Design, construction and building products and technology of buildings for public use
e155	Design, construction and building products and technology of buildings for private use
e225	Climate
e2250	Temperature
e3	CHAPTER 3 SUPPORT AND RELATIONSHIPS
e310	Immediate family
e320	Friends
e325	Acquaintances, peers, colleagues, neighbours and community members
e330	People in positions of authority
e340	Personal care providers and personal assistants
e355	Health professionals
e420	Individual attitudes of friends
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members
e450	Individual attitudes of health professionals
e460	Societal attitudes
e5550	Associations and organizational services
e570	Social security services, systems and policies
e5801	Health systems
e5850	Education and training services
s420	Structure of immune system

s4200	Lymphatic vessels
s4201	Lymphatic nodes
s7	CHAPTER 7 STRUCTURES RELATED TO MOVEMENT
s710	Structure of head and neck region
s720	Structure of shoulder region
s730	Structure of upper extremity
s7300	Structure of upper arm
s7302	Structure of hand
s760	Structure of trunk
s810	Structure of areas of skin

Table 2. Comprehensive ICF Core Set for lymphedema in midline.

b126	Temperament and personality functions
b130	Energy and drive functions
b134	Sleep functions
b152	Emotional functions
b164	Higher-level cognitive functions
b1644	Insight
b180	Experience of self and time functions
b1801	Body image
b265	Touch function
b280	Sensation of pain
b435	Immunological system functions
b4350	Immune response
b4352	Functions of lymphatic vessels
b440	Respiration functions
b455	Exercise tolerance functions
b4552	Fatiguability
b5104	Salivation
b5105	Swallowing
b530	Weight maintenance functions
b540	General metabolic functions
b620	Urination functions
b640	Sexual functions
b710	Mobility of joint functions
b730	Muscle power functions
b770	Gait pattern functions
b780	Sensations related to muscles and movement functions

b810	Protective functions of the skin
b820	Repair functions of the skin
b840	Sensation related to the skin
d155	Acquiring skills
d230	Carrying out daily routine
d240	Handling stress and other psychological demands
d330	Speaking
d350	Conversation
d410	Changing basic body position
d4100	Lying down
d4153	Maintaining a sitting position
d4154	Maintaining a standing position
d430	Lifting and carrying objects
d450	Walking
d455	Moving around
d470	Using transportation
d475	Driving
d510	Washing oneself
d520	Caring for body parts
d530	Toileting
d540	Dressing
d550	Eating
d560	Drinking
d570	Looking after one’s health
d620	Acquisition of goods and services
d640	Doing housework

d750	Informal social relationships
d770	Intimate relationships
d850	Remunerative employment
d870	Economic self-sufficiency
d920	Recreation and leisure
d9201	Sports
d9205	Socializing
e1101	Drugs
e1150	General products and technology for personal use in daily living
e1151	Assistive products and technology for personal use in daily living
e155	Design, construction and building products and technology of buildings for private use
e225	Climate
e2250	Temperature
e310	Immediate family
e320	Friends
e325	Acquaintances, peers, colleagues, neighbours and community members
e330	People in positions of authority
e340	Personal care providers and personal assistants
e355	Health professionals
e410	Individual attitudes of immediate family members
e420	Individual attitudes of friends
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members
e450	Individual attitudes of health professionals
e460	Societal attitudes

e465	Social norms, practices and ideologies
e555	Associations and organizational services, systems and policies
e580	Health services, systems and policies
e5850	Education and training services
s330	Structure of pharynx
s340	Structure of larynx
s420	Structure of immune system
s630	Structure of reproductive system
s710	Structure of head and neck region
s720	Structure of shoulder region
s740	Structure of pelvic region
s760	Structure of trunk
s810	Structure of areas of skin

Table 3. Comprehensive ICF Core Set for lymphedema in lower extremity.

b1261	Agreeableness
b1263	Psychic stability
b1266	Confidence
b1301	Motivation
b134	Sleep functions
b152	Emotional functions
b1644	Insight
b1646	Problem-solving
b1670	Reception of written language
b1801	Body image
b260	Proprioceptive function
b2702	Sensitivity to pressure
b280	Sensation of pain
b415	Blood vessel functions
b4150	Functions of arteries
b435	Immunological system functions
b4350	Immune response
b4352	Functions of lymphatic vessels
b4353	Functions of lymph nodes
b455	Exercise tolerance functions
b4552	Fatiguability
b530	Weight maintenance functions
b540	General metabolic functions
b550	Thermoregulatory functions
b640	Sexual functions
b7101	Mobility of several joints
b7303	Power of muscles in lower half of the body
b770	Gait pattern functions
b780	Sensations related to muscles and movement functions
b810	Protective functions of the skin
b820	Repair functions of the skin
b840	Sensation related to the skin

d155	Acquiring skills
d230	Carrying out daily routine
d410	Changing basic body position
d4100	Lying down
d4101	Squatting
d4102	Kneeling
d4103	Sitting
d4104	Standing
d4105	Bending
d4153	Maintaining a sitting position
d4154	Maintaining a standing position
d430	Lifting and carrying objects
d450	Walking
d4500	Walking short distances
d4501	Walking long distances
d4551	Climbing
d4552	Running
d4600	Moving around within the home
d4602	Moving around outside the home and other buildings
d4702	Using public motorized transportation
d4750	Driving human-powered transportation
d4751	Driving motorized vehicles
d510	Washing oneself
d520	Caring for body parts
d5200	Caring for skin
d530	Toileting
d540	Dressing
d5402	Putting on footwear
d570	Looking after one’s health
d5701	Managing diet and fitness
d5702	Maintaining one's health
d6200	Shopping

d630	Preparing meals
d640	Doing housework
d660	Assisting others
d770	Intimate relationships
d845	Acquiring, keeping and terminating a job
d850	Remunerative employment
d920	Recreation and leisure
d9205	Socializing
e1101	Drugs
e1150	General products and technology for personal use in daily living
e1151	Assistive products and technology for personal use in daily living
e1201	Assistive products and technology for personal indoor and outdoor mobility and transportation
e135	Products and technology for employment
e150	Design, construction and building products and technology of buildings for public use
e155	Design, construction and building products and technology of buildings for private use
e1650	Financial assets
e210	Physical geography
e225	Climate
e310	Immediate family
e320	Friends
e325	Acquaintances, peers, colleagues, neighbours and community members
e330	People in positions of authority
e340	Personal care providers and personal assistants
e355	Health professionals
e410	Individual attitudes of immediate family members
e420	Individual attitudes of friends
e425	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members
e460	Societal attitudes
e5550	Associations and organizational services

e5600	Media services
e580	Health services, systems and policies
e5800	Health services
e5850	Education and training services
s420	Structure of immune system
s4200	Lymphatic vessels
s4201	Lymphatic nodes
s630	Structure of reproductive system
s6303	Structure of vagina and external genitalia
s740	Structure of pelvic region
s750	Structure of lower extremity
s7500	Structure of thigh
s7501	Structure of lower leg
s75011	Knee joint
s7502	Structure of ankle and foot
s760	Structure of trunk
s7701	Joints
s810	Structure of areas of skin

Table 4. Brief ICF Core Set for lymphedema in upper extremity.

b1263	Psychic stability
b164	Higher-level cognitive functions
b1801	Body image
b280	Sensation of pain
b4550	General physical endurance
b4552	Fatiguability
b710	Mobility of joint functions
b730	Muscle power functions
d230	Carrying out daily routine
d445	Hand and arm use
d5	CHAPTER 5 SELF-CARE
d570	Looking after one's health
d5701	Managing diet and fitness
d640	Doing housework
d770	Intimate relationships
d850	Remunerative employment
d920	Recreation and leisure
e1151	Assistive products and technology for personal use in daily living
e310	Immediate family
e320	Friends
e355	Health professionals
s420	Structure of immune system
s730	Structure of upper extremity

Table 5. Brief ICF Core Set for lymphedema in midline.

b1801	Body image
b280	Sensation of pain
b435	Immunological system functions
b455	Exercise tolerance functions
b640	Sexual functions
d155	Acquiring skills
d230	Carrying out daily routine
d240	Handling stress and other psychological demands
d455	Moving around
d520	Caring for body parts
d570	Looking after one’s health
d850	Remunerative employment
d920	Recreation and leisure
e1151	Assistive products and technology for personal use in daily living
e310	Immediate family
e320	Friends
e355	Health professionals
s710	Structure of head and neck region
s740	Structure of pelvic region
s760	Structure of trunk

Table 6. Brief ICF Core Set for lymphedema in lower extremity.

b1301	Motivation
b152	Emotional functions
b1801	Body image
b280	Sensation of pain
b435	Immunological system functions
b455	Exercise tolerance functions
b7101	Mobility of several joints
b7303	Power of muscles in lower half of the body
d230	Carrying out daily routine
d450	Walking
d570	Looking after one’s health
d845	Acquiring, keeping and terminating a job
e1151	Assistive products and technology for personal use in daily living
e310	Immediate family
e355	Health professionals
e580	Health services, systems and policies
e5850	Education and training services
s750	Structure of lower extremity

Curriculum Vitae

Curriculum vitae

In 1958 werd Peter Viehoff in Utrecht geboren op 29 november. Hij behaalde zijn Atheneum in 1977 en in datzelfde jaar begon hij met de opleiding Fysiotherapie aan de toenmalige Stichting Utrechtse Paramedische Academie (SUPA). Na het behalen van zijn diploma in 1981 moest hij in militaire dienst. Hij vervulde deze als sergeant-fysiotherapeut in het Militair Hospitaal te Utrecht. De verplichte dienstplicht werd omgezet in een Kort Verband Vrijwilliger (KVV) contract. Vervolgens bleef hij tot 1987 in het Militair Hospitaal en behaalde hij in die tijd de diploma's Sportfysiotherapie en Oedeemtherapie. In 1985 trouwde hij met Irma Versteeg en ging hij in Nieuwegein wonen, waar hij nog steeds woont. Door de verslechterde arbeidsmarkt in Nederland week hij in 1988 uit naar Duitsland. Daar heeft hij 1 jaar gewerkt als "krankengymnast" in de Praxis Hanus in Düsseldorf. In 1989 werd dochter Roxanne geboren en was het beter om dichter bij huis te gaan werken. Daarom ging hij vanaf 1990 werken in de Groepspraktijk voor fysio- en manuele therapie 't Gilde in Gorinchem. Tot op heden werkt hij daar nog als 1 van de praktijkhouders. In de tussentijd werd in 1991 zoon Mitchell geboren en behaalde hij ook het diploma Manuele Therapie in 1998. Zijn interesse voor wetenschappelijk onderzoek werd gewekt door het volgen van de cursus Scholing in Wetenschap van het Nederlands Paramedisch Instituut. Toen dan ook de mogelijkheid bestond voor het volgen van de universitaire opleiding Fysiotherapiewetenschap in Utrecht was de stap om die opleiding te gaan volgen niet moeilijk. In 2006 behaalde hij de drs titel met onderzoek naar het vertalen en valideren van een Franse vragenlijst op het gebied van lymfoedeem. Dit smaakte naar meer en vanaf 2008 had hij de eer om in het Erasmus Medisch Centrum Rotterdam promotieonderzoek te gaan doen op het gebied van lymfoedeem. Dit deed hij naast zijn werk als fysiotherapeut en in de weekenden. Naast zijn werk heeft hij ook verschillende nevenfuncties bekleed. Zo was hij lid van de wetenschappelijke commissie van de Nederlandse Vereniging voor fysiotherapeuten in de Lymfologie (NVFL)(2004-2011) en bestuurslid van de studentenvereniging Scientia Fundus (2002-2009). Op dit moment is hij als docent verbonden aan de opleidingen "Fysiotherapie Master Lymfologie en Oncologie" van instituut De Berekuyl en "Master oncologiefysiotherapie" van Avans+.

Curriculum vitae

Peter Viehoff was born in 1958 in Utrecht on the 29th of November. He received his secondary school diploma in 1977 and in that same year he began education in Physiotherapy at the Foundation Utrecht Paramedical College (SUPA). After graduating in 1981 he was in military service. He passed this as a sergeant physiotherapist at the Military Hospital in Utrecht. The mandatory conscription was converted into a Short Volunteer Relationship (KVV) contract. He remained in the Military Hospital until 1987 and during that time he earned the diplomas Sport Physiotherapy and Edema Therapy. In 1985 he married Irma Versteeg and he went to live in Nieuwegein, where he still lives. Due to the deteriorating labor market in the Netherlands he went to Germany in 1988. There he worked one year as "Krankengymnast" in Praxis Hanus in Düsseldorf. Daughter Roxanne was born in 1989 and it was better to work closer to home. Therefore, from 1990, he went to work in the Group Practice for Physiotherapy and Manual therapy 't Gilde in Gorinchem. To date, he's still working there as one of the practice holders. In the meantime, in 1991, son Mitchell was born and he also graduated Manual Therapy in 1998. His interest in research was woken by following the course Education in Science at the Dutch Institute of Allied Health Care. And then there was the possibility to follow university education in Physiotherapy Science in Utrecht. The step to follow that course was not difficult. In 2006 he achieved his Master of Science degree with research on the translation and validation of a French questionnaire in the field of lymphedema. This tasted for more and he had the honour to do PhD research in the field of lymphedema at the Erasmus Medical Center, Rotterdam from 2008. He did this in addition to his work as a physiotherapist and at weekends. Besides his work he also held several other positions. He was a member of the scientific committee of the Dutch Association for Physiotherapists in Lymphology (NVFL) (2004-2011) and member of the student association Scientia Fundus at Utrecht University (2002-2009). Currently he is a teacher related to the study " Master Lymphology and Oncology Physiotherapy " of the Berekuyl institute and "Master oncology physiotherapy" at Avans +.

List of publications

List of publications

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Dankwoord

Dankwoord

Een doctorandus is letterlijk “iemand die doctor aan het worden is”. Een metamorfose, die niet elke doctorandus daadwerkelijk ondergaat. Toen ik in 2006 de drs titel behaalde was mijn idee hierover echter duidelijk: Ik wilde graag verder in het onderzoek doen, maar ik wist alleen niet hoe. Na 2 jaar kwamen in 2008 de ICF Core Sets op mijn pad na een bezoek aan een Internationaal Manueel Therapie congres. En opeens ontstond het idee om deze, via een PhD traject, te gaan ontwikkelen voor lymfoedeem.

Een uitspraak luidt: “Een traject zonder obstakels leidt doorgaans nergens toe”, maar obstakels op je pad zijn er om te ontdekken of je ergens werkelijk voor wil gaan. Nou, gedurende de jaren die ik aan het project heb besteed, zijn de obstakels er veelvuldig geweest en weet ik ook waar de afkorting dr. voor staat. Wanneer deze afkorting voor iemands naam staat, kun je er zeker van zijn dat het een doorzetter is. Want dat is wel een van de competenties die je moet hebben, ofwel verkrijgen. Weliswaar is het de bedoeling dat je laat zien dat je zelfstandig onderzoek kunt doen, maar je staat er niet alleen voor. Ik prijs mij gelukkig dat ik van vele mensen ondersteuning heb gekregen. Een aantal wil ik hierbij graag in het bijzonder bedanken.

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Voor het ontwikkelen van ICF Core Sets is het gebruikelijk dat deze wordt ondersteund door een stuurgroep. Naast de boven genoemde personen bestond deze uit:
Dr. Robert Damstra, beste Robert, bedankt dat je zo achter de ICF en het project staat. Je hebt me geholpen op diverse manieren. Uiteraard met je kijk op de lymfoedeempatiënt, de mogelijkheid om in Nij Smellinghe het cross-sectionele onderzoek te gaan doen en je medewerking aan andere onderzoeken.
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Alle stuurgroep leden bedankt dat jullie naar Glasgow wilden komen voor de consensus conferentie om daar een positieve bijdrage te leveren in welke functie dan ook.

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PhD Portfolio

PhD Portfolio

Summary of PhD training and teaching activities

Name PhD student	: Petrus Bernardus Viehoff
PhD period	: 2010- 2015
Erasmus MC Department	: Dermatology
Promotor	: Prof.dr. H.A.M. Neumann
Supervisor	: Dr. Y.F. Heerkens
Supervisor	: Dr. C.D. van Ravensberg

	Year	Workload (Hours/ ECTS)
PhD training		
General skills		
- NPI Course: Physical therapy for peripheral arterial suffering.	2009	24 hours
- NPI Masterclass: Bringing lymphology into holistic lymphedema management.	2010	8 hours
- Course medical library Erasmus Medical Centre: Endnote X3/X4.	2011	4 hours
Presentations		
- ISL conference, Sydney. ‘ULL27: Dutch Translation and Validation of an Illness-Specific Health-related Quality of Life questionnaire for Patients with Upper Limb lymphedema’ and ‘The development of ICF Core Sets for lymphedema’.	2009	1 ECTS
- ILF conference, Toronto. Workshop: ‘The use of ICF Core Sets for lymphedema in daily practice’.	2011	1 ECTS
- ISL conference, Malmo. ‘Using ICF Core Sets for lymphedema in daily practice’ and ‘The development of ICF Core Sets for lymphedema: Literature review’.	2011	1 ECTS

- NVFL conference, Rotterdam. ‘De ontwikkeling van ICF Core Sets voor lymfoedeem’.	2012	1 ECTS
- ILF conference, Montpellier. ‘The development of ICF Core Sets for lymphedema: Qualitative research’.	2012	1 ECTS
- ISL conference, Rome. ‘The development of ICF Core Sets for lymphedema: Expert research’ and ‘The development of ICF Core Sets for lymphedema: Qualitative research’.	2013	1 ECTS
- ILF conference, Glasgow. ‘The development of ICF Core Sets for lymphedema’.	2014	1 ECTS
- BSL conference, Birmingham. ‘Developing ICF Core Sets for lymphedema and what can you do with it’ and ‘ICF Core Sets for lymphedema: The quest for qualitative results’.	2014	1 ECTS
International conferences		
- International Society of Lymphology, Sydney, Australia.	2009	1 ECTS
- World Confederation for Physical Therapy, Amsterdam, The Netherlands.	2011	1 ECTS
- International Lymphedema Framework, Toronto, Canada.	2011	1 ECTS
- International Society of Lymphology, Malmo, Sweden.	2011	1 ECTS
- International Lymphedema Framework, Montpellier, France.	2012	1 ECTS
- International Society of Lymphology, Rome, Italy.	2013	1 ECTS
- International Lymphedema Framework, Glasgow, England.	2014	1 ECTS
- British Society of Lymphoedema, Birmingham, England.	2014	1 ECTS

National conferences		
- Nascholing oedeemtherapeuten en fysiotherapeuten ‘Flebologie en lymfologie’, Utrecht, The Netherlands.	2010	1 ECTS
- Conference Lymphedema and Gynaecologic cancer, Utrecht, The Netherlands.	2011	1 ECTS
- Conference on lymphedema, Rotterdam, The Netherlands.	2012	1 ECTS
Other		
- Organization and presentation International Consensus Conference on the development of ICF Core Sets for lymphedema, Glasgow, England	2014	0
- Poster ‘The development of ICF Core Sets for lymphedema’. WHO-FIC congress, Barcelona, Spain.	2014	1 ECTS
Teaching activities		
- Oncological physical therapy ‘Thesis coach’, Avans+, Breda, The Netherlands.	2006	80 hours
- Oncological physical therapy ‘Masterclass Clinical Reasoning’, Avans+, Breda, The Netherlands.	2012	32 hours
- Physical therapy Master Lymphology and Oncology ‘ICF’, de Berekuyl, Hierden, The Netherlands.	2013	40 hours

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Oldekamp Medisch
V Wijngaarden Medisch
Haddenham Health Care
Lohmann and Rauscher
VvAA

The cover photograph was taken in 2008 in National Park Krka in Croatia. This photo was chosen, because the waterfalls symbolize the opportunities the ICF Core Sets offer, to eventually improve the discharge of lymphedema in lymphedema patients.