A patient-based self-examination survey for staging the severity of lymphoedema

Carol M Morris, Teresa S Lee, Sharon A Czerniec, Andrea J Mangion

Key words

Bioimpedance, cancer, lymphoedema, lymphoedema staging, self-report

Carol M Morris and Teresa S Lee are lymphoedema physiotherapists at the Melanoma Institute Australia, Sydney, Australia, Teresa Lee is also a physiotherapist at the Northern Sydney Cancer Centre, Royal North Shore Hospital, Sydney, Australia; Sharon A Czerniec is senior lecturer in physiotherapy at Australian Catholic University, Sydney, Australia; and Andrea J Mangion is a physiotherapist at Northern Sydney Lymphoedema Institute, Australia

Abstract

Background: A lymphoedema self-assessment tool can help therapists deliver appropriate advice, and prioritise patients who are on waiting lists or who cannot access a lymphoedema clinic. **Aims:** This pilot study aims to assess the validity and reliability of the Lymphoedema Self-Examination Survey (LYSES) and the relationship between outcomes obtained through LYSES and healthcare professionals' lymphoedema assessment. **Methods:** Participants with lymphoedema (n=54) completed the LYSES, with the same survey repeated 1 week later. Bioimpedance measurement and International Society of Lymphology (ISL) staging were conducted by lymphoedema healthcare professionals, blinded to each other. **Results:** There was little agreement between self-reports of elevation being helpful and ISL staging of ≤I (κ= −0.154), substantial agreement between self-reports of indentation and ISL staging of II (κ=0.631), and poor agreement between self-reports and healthcare professionals' reports of skin firmness and skin changes (>ISL stage IIb) (κ=0.089 and κ=0.466, respectively). Lymphoedema severity based on ISL staging was able to account for 31% of the variability in L-Dex rank scores (p=0.005). **Conclusion:** LYSES cannot be used as an accurate self-report survey.

Lymphoedema is swelling caused by the failure of the lymphatic system to remove the normal accumulation of proteins and fluid from body tissues (Saito et al, 2013). Left untreated, it is a condition that can negatively impact upon a patient's physical and psychological health, and their quality of life (Franks et al, 2006; Girgis et al, 2011). It is commonly described as an unmet need for cancer survivors, with previous studies calling for better acknowledgement of this condition by healthcare professionals (Beesley et al, 2007; Bernas et al, 2010; Paskett et al, 2012).

Previously published lymphoedema self-examination tools focus on breast cancer-related arm lymphoedema symptoms, which may be less relevant to those patients with lymphoedema affecting other body parts. These surveys directed at breast cancer patients include the Lymphedema Symptom Intensity and Distress Survey –Arm (LSIDS-A) and a telephone survey developed by Norman et al (2001). The LSIDS-A evaluates symptoms and calculates symptom burden (Ridner et al, 2009). It consists of items that ask patients to rate symptoms such as swelling, fatigue, tightness and aching on a ten-point scale. The Norman et al (2001) survey asked breast cancer participants whether there was a difference between the sizes of their affected and unaffected hand or arm, and to assess the degree of difference as 'very slight', 'noticeable' or 'very noticeable'.

The patient's experience of his or her symptoms is important but first the healthcare professional has to be convinced that swelling exists and the patient must be able to accurately communicate this to his or her healthcare professional. This is not always easy, particularly for patients who may be triaged on the telephone, or for rural and remote patients who are managed via telehealth because they cannot physically attend a lymphoedema clinic. Development of patient registries for the purpose of government lobbying or funding often include survey items documenting signs of lymphoedema including response to elevation, presence of indentation and skin changes, because it helps confirm the existence of swelling and its severity (Australasian Lymphology Association, 2015).

The Lymphoedema Self-Examination Survey (LYSES) was developed by experienced lymphoedema healthcare professionals in response to the need for a quick, effective and accurate tool for patients to report the presence and severity of their lymphoedema. The LYSES questions reflect the signs of lymphoedema as outlined by the International Society of Lymphology (ISL) lymphoedema staging classification (ISL, 2013). The ISL provides an appropriate framework that allows systematic categorisation of clinical observations. It identifies lymphoedema as more than an accumulation of fluid in the tissues and acknowledges the dynamic, active and progressive changes in the tissues that accompany this disease (Morgan and Lee, 2008). This system identifies five stages (0, I, IIa, IIb and III) of lymphoedema progression by clinically examining swelling, response to elevation, skin and tissue changes to assess the

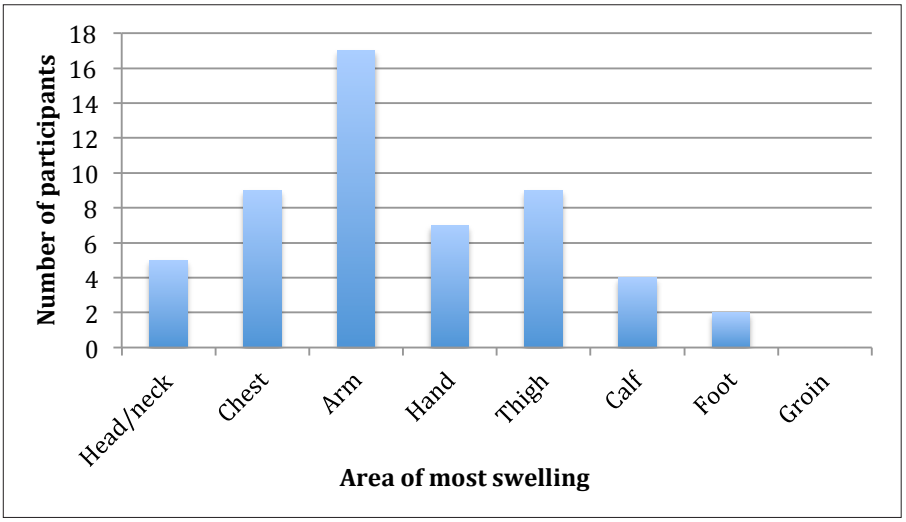

Figure 1. The area of most swelling as reported in the questionnaire by participants.

severity of lymphoedema. The validity and reliability of ISL staging has never previously been published and no prior studies have examined its relationship with bioimpedance measurement, which has been established as an accurate tool for assessing extracellular fluid changes present in patients with lymphoedema (Ward, 2006).

The primary aim of this pilot study was to assess the validity and reliability of the LYSES as reported by lymphoedema patients. We also assessed the relationship between outcomes obtained through LYSES, healthcare professionals' classification of ISL stage, and the results of bioimpedance analysis.

Methods

Participants

Participants were recruited from two different lymphoedema clinics in Sydney (the Royal North Shore Hospital and the Melanoma Institute Australia). Ethics approval was obtained through Sydney Local Health District RPAH Zone (X13-0016) and the Northern Sydney Local Health District Human Research Ethics Committee (1305-178M) in accordance with the Helsinki Declaration (General Assembly of the World Medical Association, 2014). Both male and female patients were invited to participate. Participants had to be >18 years old, identify themselves as having lymphoedema from any cause, in any area of their body, and be able to read and communicate in English for the completion of the required surveys. Of the patients who reported arm or leg lymphoedema, only those with unilateral limb lymphoedema were recruited, as an unaffected contralateral side was required for bioimpedance measurements.

Eligible patients identified from the clinic database were posted a letter of invitation and a participant information sheet. Expressions of interest were followed up with a phone call from one of the investigating healthcare professionals to confirm eligibility and organise a suitable time to attend the clinic to give written consent, complete the surveys and for clinical examination.

Lymphoedema self-examination survey

Participants completed the LYSES, which is a five-question survey that asked for a yes/no response to questions about the characteristics of their swelling. The first question asked them to identify their area of "most" swelling. The other questions were specific to the characteristics of their swelling and included:

- The response of their swelling to elevation (Does elevation reduce this swelling?)
- The presence of skin indentation (Are indentations obvious on this swollen area when firm pressure has been applied to the area for a period of time?)
- Tissue firmness (Does this swollen area feel firmer or thicker than other areas?)
- Skin changes (Is the skin over this swollen area leathery, wrinkled, discoloured, have abnormal folds or wart-like growths and/or leaking fluid?).

Clinical examination

Each participant was assessed separately by two lymphoedema physiotherapists. The first physiotherapist completed a data collection form that recorded information about the participant's age, sex, body mass index, type of lymphoedema (primary or secondary) and medical history including previous surgery, radiation and chemotherapy.

The first physiotherapist then examined the participant's area of 'most swelling', as identified on LYSES, and staged the severity of his or her lymphoedema using ISL staging. Bioimpedance measurement was also performed using the L-Dex® U400 or Imp XCA Extracellular Fluid Analysis (both ImpediMed, Brisbane, Australia) to measure extracellular fluid impedance by following a standardised measurement protocol and passing a small electrical current through adhesive skin electrodes (Ward et al, 2009). A lymphoedema index (L-Dex) was produced by the Imp® XCA or U400, which is reflective of the impedance ratio between the affected and unaffected limbs. Previous research has shown that the results from single-frequency and multifrequency devices are comparable (York et al, 2009).

The second physiotherapist was blinded to the results obtained by the first physiotherapist, and again staged the participant's lymphoedema using the ISL classification. One week following the clinical assessment, participants were mailed or emailed a second LYSES to complete to ensure the test–retest reliability of the LYSES.

Statistical analysis

Cohen's Kappa coefficient was used to assess the relationship between self-report (LYSES) and clinical examination (ISL staging). Cohen's Kappa coefficient is a statistical measure of inter-rater agreement and takes into account the agreement occurring by chance. A Kappa of 1.0 represents perfect agreement, while a Kappa <0.4 is considered only fair to poor agreement (Viera and Garrett, 2005). The relationships between ISL staging and bioimpedance were analysed using the Kruskal–Wallis test.

Results

A total of 54 participants were recruited to this pilot study. Of these participants, 45 were female and nine were male. The age range was from 40 to 87 years old (mean age 63.24 ± 13.23 years). One patient had

primary and 53 patients had secondary lymphoedema. The time since surgery for secondary lymphoedema ranged from 7 months to 17 years (median 1.5 years; interquartile range 4.98). The area of 'most' swelling for participants is shown in *Figure 1*. The arm was the most frequently reported area of swelling by our study sample.

Agreement between LYSES

Participants were able to identify their area of 'most' swelling with excellent consistency in the two LYSES administered 1 week apart. The test–retest reliability of the LYSES displayed agreement for area of swelling, with Kappa ranging from 0.797 for the hand and 1.0 for the groin and head and neck (*Figure 2a*).

Participants were also able to consistently answer questions regarding the response of their swelling to elevation, and whether or not indentations were apparent, between the two LYSES administered 1 week apart (κ=0.731 and κ=0.631, respectively). Participants' response to questions on skin firmness and skin changes were not consistent (κ=0.089 and κ=0.466, respectively) between the two LYSES (*Figure 2b*).

Agreement between lymphoedema healthcare professionals

ISL stages 0 and I were combined for analyses because the participants (n=12) in these groups would have 'mild' lymphoedema that would respond to elevation. Stages IIb and III were combined because the participants (n=15) in these groups would have 'severe' lymphoedema where pitting is minimal or absent. Stage IIa remained as a single category for analyses because participants (n=27) in this stage would have 'moderate' lymphoedema with significant pitting. There was substantial agreement between ISL stages determined by the two healthcare professionals (κ=0.630).

LYSES and ISL stage agreement

Our ability to determine agreement between self-report and ISL stages was confounded by the poor reliability of the self-report items relating to skin firmness and skin changes. Hence, only self-report items relating to response to elevation and the presence of indentations were used.

There was no agreement between self-reported benefits of elevation and clinical reported benefits of elevation in the ISL stage 0–I group (κ=–0.154). There was also no agreement between self-reported presence of indentation/pitting and clinical examination in the ISL stage IIa group (κ=0.282).

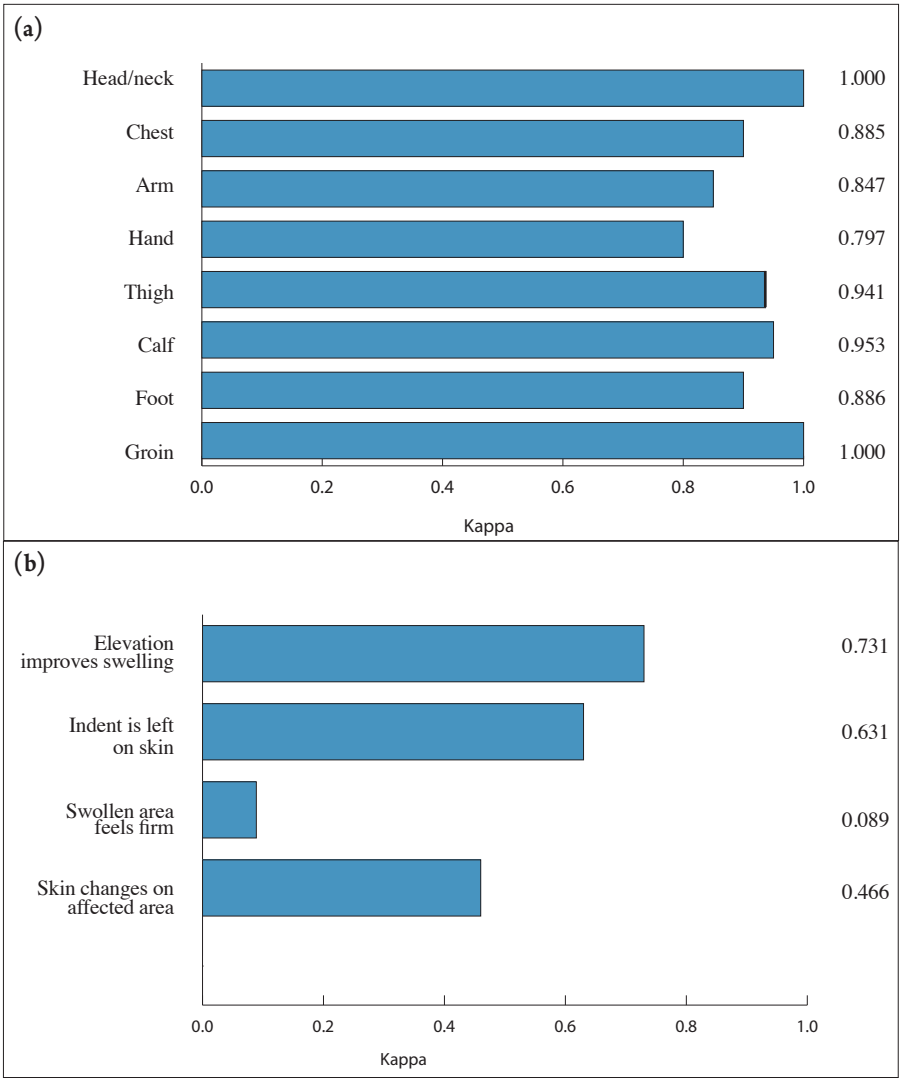

Figure 2. Test–retest reliability of participants' reported area of (a) most swelling and (b) clinical signs.

ISL stage and bioimpedance agreement

Median L-Dex scores for the mild, moderate and severe lymphoedema groups were 6.9, 32.6 and 55.0, respectively. Thirty-one per cent of the L-Dex rank score was accounted for by lymphoedema severity based on ISL staging (p=0.005). *Post-hoc* testing revealed a significant difference in L-Dex rank between mild and moderate lymphoedema groups and between mild and severe groups. No difference in L-Dex rank was found between moderate and severe lymphoedema groups.

Discussion

This pilot study gave an insight to the complexities involved in patients' understanding of lymphoedema signs. Patients with lymphoedema are unable to assess signs of swelling, such as tissue firmness and skin changes, in the same way as lymphoedema healthcare professionals. Our results showed great disparity between self-report and the ISL staging of these items.

Healthcare professionals use palpation to assess tissue firmness, as required by ISL staging. Using the fingertips, light pressure is applied to the affected body part to assess the pliability, texture and condition of the subcutaneous tissue as compared to an unaffected area. To an experienced lymphoedema healthcare professional, firmness as stated by ISL classification would be considered tissue fibrosis associated with long-standing lymphoedema.

Patients rely upon their sensation and perception of 'firmness' with respect to their own experience with swelling. Given that many of the participants in the study have early lymphoedema and have never seen or experienced firmness associated with long-standing lymphoedema, over-reporting of this item was seen in our LYSES results.

Similarly, healthcare professionals were in search of lymphoedema-related skin changes such as keratosis, papillomatosis, fibrotic breakdown and lymphorrhoea, which are associated with stage III of the ISL classification. Participants did not interpret the term 'skin change' in the same way as healthcare professionals, as they were observed to report any skin difference including dryness, follicle irritation, eczema or scarring as a symptom on LYSES, irrespective of whether or not it was a lymphoedema-associated change.

Participants did not do a 1-minute pitting test (Brorson et al, 2008) on themselves and relied upon their memory to assess whether or not they had experienced indentations on their skin after firm pressure had been applied. Their interpretation of indentation included any mark left on the skin, including that of their compression garment, rings or watches, irrespective of whether or not pitting or a ridge could be felt around the mark.

When assessing ISL stage, healthcare professionals do not leave a participant's limb in an elevated position for extended periods to observe its response, so the rating of this stage is actually based on asking the participant about whether or not his or her swelling is reduced with elevation. There was no agreement between participants' response to the LYSES question on elevation and that of ISL stages 0 or I, where elevation would be considered helpful to swelling. Similarly, there was no agreement between participants' response to the LYSES question on indentation on their area of most swelling and that of ISL stage IIa, where pitting would be considered apparent.

Although we specifically asked patients and lymphoedema therapists to answer the survey questions in relation to the 'area of most swelling', this area may be so large that it extends beyond lymphatic territories, where there may be differences in tissue composition and its response to elevation and palpation. It is also possible that the problem lies in the division of ISL categories. The ISL staging system has been criticised by other authors as being too simple and too crude for accurate assessment of lymphoedema patients (Lee et al, 2011). The categories are not distinct, with overlapping of signs such as elevation and pitting between categories. Patients may experience acute on chronic lymphoedema, so that there may be components of their swelling that are alleviated by elevation, and demonstrate pitting in the presence of tissue fibrosis or skin changes. These signs are challenging to classify as they span three categories of ISL staging. There may also be mixed aetiology, with vascular components contributing to lymphatic overload, making fibrotic swelling more amenable to elevation. Although our experienced healthcare professionals demonstrated good inter-rater reliability in their ISL staging of participants, novice therapists may find this system ambiguous and open to interpretation.

The relationship between lymphoedema staging and L-Dex reflects increasing extracellular fluid. This suggests that ISL staging could be used to triage patients on the basis of lymphoedema severity in general medical practice and in lymphoedema clinics where bioimpedance may not be readily available. ISL staging does not, however, facilitate detection of lymphoedema prior to the manifestation of swelling, which is a clinical advantage of bioimpedance analysis (Cornish et al, 2001; Ward, 2006; Ridner et al, 2009).

Unfortunately, survey questions that include self-report of lymphoedema signs without prior instruction do not appear to be valid or reliable. This may present a problem for the current Australian and New Zealand Lymphoedema Registry, which uses three very similar questions to the LYSES in assessing patient self-report of response to elevation, presence of skin indentation and skin changes in their online survey of lymphoedema patients (Australasian Lymphology Association, 2015). A larger sample may yield a different outcome but results from this pilot study would suggest that lymphoedema signs should only be included in surveys that are completed by the treating healthcare professionals. Patient self-report surveys are best used for symptom description and burden.

Future directions

Of the four clinical signs described in the ISL staging, we believe that there is good potential for the pitting test being a useful self-assessment tool for patients, but it will require instruction to yield more reliable agreement. Trayes et al (2013) recommend the location, time of pitting resolution and depth of pitting be recorded to determine the extent of oedema and treatment response. In the telehealth situation, instructions can be provided for patients to administer a pitting test on themselves.

The outcome of this study suggests that it may be worthwhile reverting to patient-based surveys that require no instruction. For example, asking patients to rate the severity of their lymphoedema by rating whether their lymphoedema is noticeable only to themselves, family and friends who know them well, or to strangers (Norman et al, 2001). Although, these may seem like crude categories of lymphoedema severity, it may serve the simple purpose of triaging patients over the telephone in regional and remote areas and/or for research or patient registry purposes.

Conclusion

LYSES was not a valid and reliable self-report survey for patients with lymphoedema. Patients were not able to identify lymphoedema signs, hence there was little correlation between the LYSES and ISL staging. When used by experienced lymphoedema therapists, the ISL staging classification is a valid and reliable tool in assessing the severity of lymphoedema.

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