



Patient-Reported Outcome Measures in Lymphedema: A Systematic Review and COSMIN Analysis

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The article entitled “Patient-Reported Outcome Measures in Lymphedema: A Systematic Review and COSMIN Analysis” by Dr. Beelen and colleagues reports outcomes of a systematic literature review guided by the PRISMA criteria of all patient-reported outcome measures (PROMs) used to assess health-related quality of life for patients with lymphedema.¹

The authors used the Consensus-Based Standards for the Selection of Health Measurement Instruments (COSMIN) checklist, a framework for evaluating the methodologic quality of PROM development. They found that none of the lymphedema-specific PROMs met all of the COSMIN quality standards for development and that a major shortcoming in the development of these tools was the lack of substantial open-ended patient input during the development phase. The authors therefore concluded that the currently available instruments are inadequate and may not represent the totality of the patient’s quality of life.

With the number of cancer survivors increasing, the treatment of secondary lymphedema is a growing health care need. Lymphedema has a profound negative impact on patients’ quality of life, and PROMs provide insight from patients about their quality of life associated with their condition and how this changes with time and the treatment they have received.^{2–4} It is therefore essential that the

development of PROMs involve patients, and as this systematic review highlights, only Upper Limb Lymphedema (ULL)-27 was developed with patient interviews. Six of the tools involved patient surveys during the development phase, which also may provide validity if the surveys were conducted in an open-question format.

Although no questionnaire met all of the COSMIN criteria, which were developed in an international expert opinion Delphi study, it is not clear whether this means that a tool is not valid or reliable. For example, no study was scored as adequate or greater on more than 4 of the 11 COSMIN domains. No information was available in any study for cross-cultural validity, and responsiveness was almost never addressed. As demonstrated in this report, a wide variety of PROMs have been used across published studies of lymphedema, indicating a lack of consensus among investigators about the most suitable lymphedema-specific PROM to use.

The lack of a universally accepted and applied lymphedema-specific PROM, such as the Breast-Q in breast reconstruction, limits the comparisons that can be made between different studies and the ability to combine outcomes data from different centers performing lymphedema surgery for meta-analysis. Previous systematic reviews have lacked a consensus regarding the most suitable PROM to use.^{2–4} Although one review found the ULL-27 to have strong psychometric properties,³ compared with the Lymphedema Life Impact Scale (LLIS) questionnaire, a clinical study of secondary upper extremity lymphedema found the LLIS to be more sensitive for measuring the degree of physical and functional disability.⁵ The authors did not conclude which of questionnaires currently available is the

most suitable. However, this information can help guide the choice of a questionnaire during this time of waiting for the development and validation of new tools.

From this study, the authors make the case that a current need exists for a valid and reliable lymphedema-specific PROM that adheres to modern development and validation guidelines, including extensive qualitative input from patients. Any new PROM needs to be valid for both upper- and lower-extremity lymphedema as a measurement for different domains of the lymphedema condition, clinical change over time, and effectiveness of treatments.

The new PROM also must demonstrate better reliability than the questionnaires currently used in high-volume lymphedema surgery centers.^{5–7} In addition, to allow meaningful interpretation, minimally important differences that patients perceive as beneficial need to be identified to guide treatment decisions, direct research in evaluating the comparative effectiveness of different treatments, and to design statistically well-constructed studies.^{8–10}

In summary, the findings of this study demonstrate the need for the development of a new lymphedema-specific PROM that will be accepted for widespread use to guide treatment decisions. Dr. Beelen and colleagues are to be congratulated for their significant contribution to the literature in this important clinical area.

DISCLOSURE There are no conflicts of interest.

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