

Research

Health-Related Quality of Life and Coping Style in Patients with Primary and Cancer-Related Lower Limb Lymphedema in an Outpatient Clinic in Japan

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ABSTRACT

AIM : Patients with primary lymphedema have different issues to patients with cancer-related lymphedema because they each have a distinct pathogenesis even though they both display edema in their lower limbs. The purpose of this study was to investigate health related quality of life (HRQOL) and the psychosocial state of primary lymphedema patients by comparing them with cancer-related lymphedema patients.

METHODS : The Medical Outcomes Study 36-Item Short Form Survey (SF-36), the Japanese version of the Hospital Anxiety and Depression Scale (HADS), and a short version of the Coping Orientation to Problems Experienced scale (Brief-COPE) were administered to patients with primary and cancer-related lower limb lymphedema (LLL) in an outpatient clinic. The demographic data and lymphedema conditions were obtained from medical records and physical assessments.

RESULTS : Seventy-eight patients with primary LLL and 63 patients with cancer-related LLL were included in this study. In the SF-36, the primary LLL patients had lower general health than the normative value, but the other health statuses were not lower than the normative value from the Japanese population. Furthermore, significant differences were not found in comparison with cancer-related LLL patients. In the HADS, the proportion of anxiety and depression was not high in patients with primary LLL. The higher active coping and self-blame scores in the Brief-COPE were distinctive in patients with primary LLL after adjusting for age.

CONCLUSIONS : This study found a significant difference in coping styles between patients with primary lymphedema and those with cancer-related lymphedema, while there was no significant difference in HRQOL between the groups.

KEY WORDS : primary congenital lymphedema, coping behavior, anxiety, depression

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Introduction

Primary lymphedema results from an intrinsic abnormality of the lymphatic system¹⁾. According to the time of onset, primary lymphedema is classified into congenital, praecox (initial manifestation before the age of 35), and tarda (initial manifestation after the age of 35), and the most common presentation is at onset of puberty and in females¹⁾. While there are no clear epidemiological investigations into primary lymphedema, the prevalence of primary lymphedema ranges from 6% to 12% according to past studies²⁻⁴⁾. The lower limb is most commonly affected in primary lymphedema⁵⁾.

Although there are many reports of health related quality of life (HRQOL) for secondary lymphedema, few HRQOL studies have been reported for primary lymphedema patients. Previous studies have reported that patients with primary lymphedema suffer from mobility impairment and comorbidity caused by cellulitis⁶⁾. They also experience a long waiting time for referral, poor body image, and clothing issues related to the swollen leg⁷⁾. Consequently, they have not only physical but also psychological impairments. Patients with primary lymphedema have different issues to those with secondary lymphedema, such as cancer-related lymphedema, because primary lymphedema and cancer-related lymphedema have a distinct pathogenesis even though they both display edema in the lower limbs. Equally, there is a possibility of a difference in HRQOL between the two. There is a limited number of studies comparing patients with primary lymphedema and secondary lymphedema, and the distinctiveness of the primary lymphedema patient is not clear. By comparing the HRQOL between the two, we can investigate whether it differs or is the same and the reasons, and can consider the appropriate approach for patients with primary lymphedema.

In addition, the psychosocial aspect of primary lymphedema patients should be examined. It is reported that psychological damage can affect compliance with lymphedema treatment⁸⁾. As one solution, we can prevent them from experiencing a stressful condition by using suitable coping styles⁹⁾. Coping is defined as "cognitive and behavioral efforts to manage specific

external and/or internal demands that are appraised as onerous or exceeding the resources of a person"¹⁰⁾. In other words, coping is a strategy by which people manage stress. When encountering similar stressful events, individuals tend to employ the same coping style¹¹⁾ although coping styles might vary within an individual¹⁰⁾. Previous research into patients' coping ability has been conducted in patients with chronic illness and lymphedema. The results showed that they tend to use emotional-focused coping rather than problem-focused coping techniques¹²⁾¹³⁾. However, most of this research has focused on patients with secondary lymphedema, such as breast cancer-related upper limb lymphedema. Few previous studies have investigated specifically the coping styles of patients with primary lymphedema, and their tendencies for coping strategies are unidentified.

We focused on differences in HRQOL between primary and cancer-related lymphedema patients and psychosocial state due to differences in etiology. We targeted patients with primary lower limb lymphedema (LLL) and patients with cancer-related LLL as a comparison because most patients with secondary lymphedema in Japan have cancer-related lymphedema. The purpose of this study was to investigate HRQOL and the psychosocial state of primary LLL patients in comparison with patients with cancer-related LLL.

Methods

1. Setting and participants

This cross-sectional study was executed at an outpatient clinic for patients with lymphedema in Tokyo from November 2009 to November 2010. This clinic was chosen from medical facilities in Japan dealing with a variety of lymphedema, and the average number of patients seen per week was approximately 150. In the clinic, conservative treatments are used with most patients, including compression garments or bandages, or other management aids, such as manual lymphatic drainage, are provided if necessary.

This study is part of a series of research studies with lymphedema patients. All patients who met the following inclusion criteria were recruited: (1) they were diagnosed with primary lymphedema or can-

cer-related LLL by a physician specializing in lymphedema (i.e., stage I or over of the lymphedema stages of the International Society of Lymphology ; ISL), based on medical history and physical observations ; and (2) they visited the clinic at least twice and underwent conservative treatment. As most of the patients with primary lymphedema have edema in the lower limbs, comparison participants were selected from patients with cancer-related lower limb edema. Patients younger than 16 years of age or those who were unable to fill in the questionnaire on their own were excluded. The Ethics Committee of the Graduate School of Medicine, the University of Tokyo, approved the study protocol (#2763-1). Written informed consent was obtained from all patients.

2. Procedure

After confirming that all participants met the criteria, the data of the patients' demographic details and lymphedema status, including time since onset of edema, edematous sites, and lymphedema stage, were collected from medical records. The participants then completed a self-administered, structured questionnaire within the clinic that included questions related to lymphedema onset, coping style, and states of anxiety and depression. The participants were asked to mail the questionnaire back to the researcher within a week using a prepaid envelope to ensure that participants were given a limited time in which to answer.

3. Measurements

1) Demographics

Age, sex, body mass index, and job status were included in the demographic data.

2) Lymphedema status

The edematous sites were classified as unilateral or bilateral. Edema was categorized using the five lymphedema stages of the ISL as follows : stage 0, where there are sub-clinical conditions despite impaired lymph transport ; stage I, where there is an early accumulation of fluid that subsides with limb elevation ; stage II, where limb elevation alone rarely reduces swelling and pitting is manifest ; late stage II, where there may or may not be pitting as tissue fibrosis supervenes ; and stage III, which encompasses lymphostatic elephantiasis where pitting is absent and trophic skin changes develop, such as acanthosis, fat deposits, and warty

overgrowths¹⁴⁾. The time since onset of edema, suffering from cellulitis within 30 days, history of hospital admission due to cellulitis, support from family, and belonging to the patients' association were evaluated. Self-care comprised of compression garments, bandaging, skincare, self-lymphatic massage, exercise, and intermittent pneumatic compression were also checked.

3) The Medical Outcomes Study 36-Item Short Form Survey

The Medical Outcomes Study 36-Item Short Form Survey (SF-36) was used to assess HRQOL. The SF-36, which was developed to measure the state of general health, consists of eight subscales : physical functioning (PF) ; role-physical (RP) ; bodily pain (BP) ; general health perception (GH) ; vitality (VT) ; social functioning ; role-emotional (RE) ; and mental health (MH)¹⁵⁾. The raw score on each scale ranges from 0 to 100 with a lower score indicating a more severe health status. The Japanese version of the SF-36 was used in this study, which is confirmed for the Japanese general population and lymphedema patients¹⁶⁾¹⁷⁾. First, using the SF-36 manual, the z-scores of each subscale were calculated as a norm based score (NBS) from the Japanese population¹⁸⁾. Then, the scores of the physical component summary (PCS) and mental component summary (MCS) were calculated using factor loadings for the Japanese population¹⁸⁾. The average of the NBS was set at 50, and every 10 points showed 1 SD. The Cronbach's alpha for each subscale and summary score in our study ranged from .88 to .90.

4) Coping Orientation to Problems Experienced scale

A short version of the Coping Orientation to Problems Experienced scale (Brief-COPE) was used to assess coping style¹⁹⁾²⁰⁾. The questionnaire consists of 14 subscales, with two items each, and it measures conceptually differentiable coping reactions. Scores range from 2 to 8 for each subscale, with higher scores representing a greater use of the coping styles. In the Japanese version of the Brief-COPE, criterion-related and construct validity have been confirmed by previous research²¹⁾. The Cronbach's alpha for each subscale in our study ranged from .63 to .7.

5) Hospital Anxiety and Depression Scale

The Japanese version of the Hospital Anxiety and Depression Scale (HADS) was used to assess psycholo-

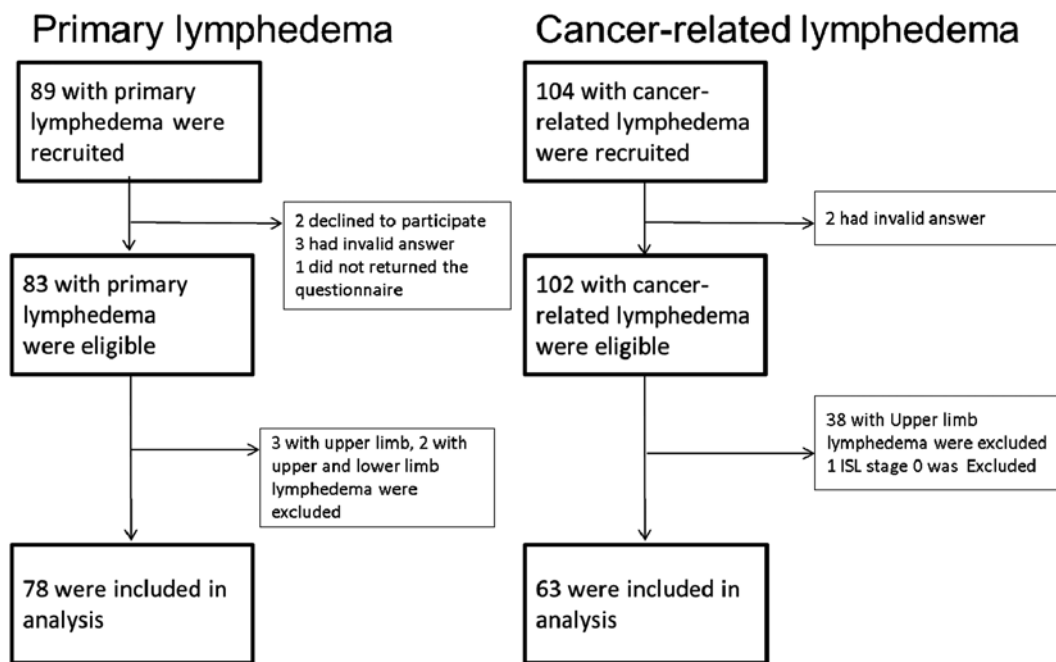


Figure 1 Study Participants

gical state^{22/23}. This consists of seven questions relating to anxiety and seven relating to depression, in which patients rate each item on a four-point scale. Higher scores indicate a greater prevalence of each respective issue. Scores from 0 to 7 are typically thought to indicate no distress, 8 to 10 to indicate doubtful distress, and 11 to 21 to indicate definite distress. Total scores ≥ 8 were taken to be indicative of distress in this study. The Japanese version of the scale has been validated among the Japanese general population²⁴.

4. Data analysis

Descriptive data are expressed as n (%) for categorical variables, and means (SD) or medians (25th and 75th percentiles) for continuous variables. Differences between groups were evaluated using t-tests, χ^2 tests, Fisher’s exact test, or Mann-Whitney U tests. We tested the difference between the NBS of the SF-36 and age- and sex-stratified national norm (ASNN) scores. Analysis of covariance (ANCOVA) for adjusting the effect of age and sex was used to compare the SF-36 scores between patients with primary and cancer-related limb lymphedema. ANCOVA for adjusting the effect of age was used to compare the Brief-COPE scores between the groups. Age-stratified analysis was also conducted where an interaction existed between age and lymphedema classification. Statistical signifi-

cance was set at $p < .05$. All statistical analyses were performed using the Statistical Analysis System software, version 9.2 (SAS Institute Inc., Cary, NC, USA).

Results

1. Recruitment results

Figure 1 summarizes the study participants. A total of 89 patients with primary lymphedema and 104 patients with cancer-related lymphedema were recruited. Regarding primary lymphedema, two patients chose not to participate, one did not return the questionnaire by mail, and three provided invalid answers for more than half of the questionnaire items. In addition, three with upper limb lymphedema, and two with upper and LLL, were excluded. Regarding cancer-related lymphedema, two patients gave invalid answers, and 38 upper limb lymphedema patients and one patient with ISL stage 0 were excluded. A total of 78 patients with primary LLL and 63 patients with cancer-related LLL were subjected to the analyses.

2. Patient characteristics

Demographic characteristics, lymphedema status, and the results of t-tests and χ^2 tests by lymphedema classification are shown in Table 1. The age of patients with primary LLL was 42.5 ± 14.0 years and the age of patients with cancer-related LLL was 61.5 ± 11.9 years

Table 1 Demographics and Lymphedema status of patients

	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value
Characteristics			
Age (y), mean (SD)	42.5 (14.0)	61.5 (11.9)	<0.001 ^{a)}
Sex, n (%)			
Male	22 (28.2)	3 (4.8)	<0.001 ^{b)}
Female	56 (71.8)	60 (95.2)	
BMI (kg/m ²), mean (SD)	21.8 (3.3)	22.7 (3.5)	0.115 ^{a)}
Time since onset of edema (y), mean (SD)	14.6 (12.3)	5.8 (5.3)	<0.001 ^{a)}
Unilateral/bilateral edema, n (%)			
Unilateral	63 (80.7)	45 (71.4)	0.193 ^{b)}
Bilateral	15 (19.3)	18 (28.6)	
ISL lymphedema staging, n (%)			0.017 ^{b)}
Stage I	2 (02.6)	9 (14.3)	
Stage II	42 (53.8)	36 (57.1)	
Late stage II	22 (28.2)	15 (23.8)	
Stage III	12 (15.4)	3 (04.8)	
Cellulitis within 30 days, n (%)	24 (30.8)	18 (28.6)	0.777 ^{a)}
History of hospital admission for cellulitis, n (%)	22 (28.2)	12 (19.1)	0.206 ^{a)}
Self-care			
Compression garments, n (%)	71 (91.3)	58 (92.1)	0.826 ^{a)}
Bandaging, n (%)	20 (25.6)	11 (17.5)	0.244 ^{a)}
Skin care, n (%)	25 (32.1)	19 (30.2)	0.809 ^{a)}
Self-lymphatic massage, n (%)	45 (57.7)	46 (73.0)	0.059 ^{a)}
Exercise, n (%)	30 (38.5)	25 (39.7)	0.882 ^{a)}
Intermittent pneumatic compression, n (%)	11 (14.1)	6 (9.5)	0.407 ^{a)}
Having a job, n (%)	45 (57.7)	19 (30.2)	0.001 ^{a)}
Support from family, n (%)	55 (70.5)	46 (73.0)	0.743 ^{a)}
Belonging to the patients' association of lymphedema, n (%)	6 (7.7)	2 (3.2)	0.298 ^{c)}

N (%) or mean (SD), a) t-test b) χ^2 test c) Fisher's exact test
 LLL, lower limb lymphedema

($p < .001$). The proportion of females was 71.8% for patients with primary LLL and 95.2% for patients with cancer-related LLL ($p < .001$). Among primary lymphedema patients, the duration of suffering from lymphedema was longer than for patients with cancer-related LLL (14.6 years vs 5.8 years, $p < .001$). More than 80% of patients belonged to the middle stage, such as ISL stage II or late stage II, and there were

significant differences between patients with primary LLL and cancer-related LLL ($p < .017$). Patients with primary LLL were more likely to have job (57.7% vs 30.2%, $p = .001$). There were no significant differences between them in terms of suffering from cellulitis ($p = .777$), self-care (compression garments $p = .826$, bandaging $p = .244$, skin care $p = .809$, self-lymphatic massage $p = .059$, exercise $p = .882$, intermittent pneumatic com-

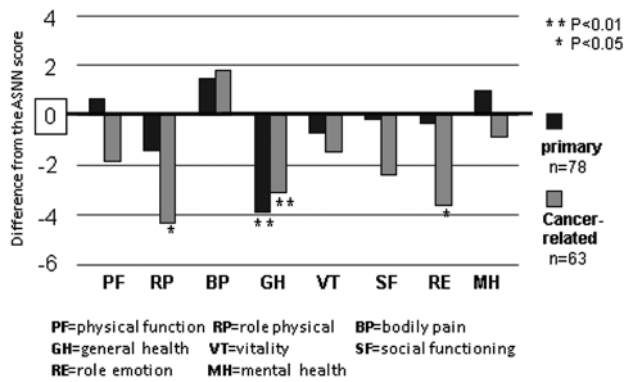


Figure 2 SF-36 scores of patients with primary and cancer-related lower limb lymphedema compared to the ASNN scores

pression $p=.407$), support from family ($p=.743$), or belonging to the patients' lymphedema association ($p=.298$).

3. SF-36 scores

Of the subscales of the SF-36, the mean GH score was lower than the ASNN score, shown as 0, in primary LLL patients ($p<.01$), and not only the GH score ($p<.01$) but also the RP and RE scores ($p<.05$) were significantly lower than the ASNN score in cancer-related LLL patients. There were no significant differences in PF, BP, or MH in primary LLL patients and no significant difference in BP in cancer-related LLL patients, although they had a higher score than the ASNN score (Figure 2). In the comparison of the scores between primary LLL patients and cancer-related LLL patients, all scores except MCS of primary LLL patients were higher than those of cancer-related LLL patients (Table 2). There was a significant difference in PF ($p<.001$), RP ($p<.002$), RE ($p=.011$), and PCS ($p<.001$). However, there was no difference in RP and RE after adjusting for age and sex. There was an interaction between age and classification of lymphedema in PF and age-stratified analyses were conducted. There tended to be a higher PF score for the primary LLL patients (Least Square Means 43.3-52.2) than the cancer-related LLL patients (Least Square Means 39.6-45.6) in the 48 years old and above group ($p=.0071$).

4. HADS scores

A total of 27 (19.1%) patients demonstrated an anxiety score greater than 8, and 15 (10.6%) patients had a depression score greater than 8. In addition, 15

(19.1%) patients with primary LLL and 12 (19.1%) patients with cancer-related lymphedema had anxiety scores ≥ 8 , while 7 (9.0%) and 8 (12.7%) patients had depression scores ≥ 8 . There were no differences in the proportion of patients with anxiety or depression scores of ≥ 8 between patients with primary LLL and those with cancer-related LLL. Although the median scores and 25th and 75th percentiles of both groups' scores were within normal ranges, there was a significant difference in depression score between them. The depression score of patients with cancer-related LLL was higher than those with primary LLL ($p=.026$) (Table 3).

5. Brief-COPE scores

Regarding the comparison of patients with primary LLL and cancer-related LLL for the Brief-COPE, there were significant differences in planning, behavioral disengagement, use of instrumental support, use of emotional support, acceptance, active coping, denial, and self-blame. Planning, use of instrumental support, use of emotional support, acceptance, active coping, and self-blame were higher in patients with primary LLL. Denial and behavioral disengagement were higher in patients with cancer-related LLL. Scores of active coping and self-blame coping styles were higher among primary LLL patients than among those suffering from cancer-related LLL ($p<.001$, $p=.021$) after adjusting for age. Positive reframing coping tended to be more prevalent among patients with primary leg lymphedema ($p=.069$) (Table 4). Since there were interactions between age and classification of lymphedema in planning and acceptance coping styles, age-stratified analyses were conducted. There were higher scores for planning coping and acceptance coping styles for the primary LLL patients in the 48 years old and above group ($p=.004$, $p=.041$) (Table 5).

Discussion

In this research, we compared the HRQOL and coping styles of LLL patients who had a different etiology: primary and cancer-related lymphedema. There were four remarkable findings in this study. First, the primary LLL patients had lower GH than the normative value, but the other health statuses were not lower than the standard value. Second, as for the HRQOL of the

Table 2 Comparison of SF-36 scores in patients with primary and cancer-related lower limb lymphedema

Variables	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value ^{a)}	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value ^{b)}
	mean (SD)	mean (SD)		Least Square Means (95%CI)	Least Square Means (95%CI)	
Physical functioning (PF)	51.7 (8.2)	42.8 (12.6)	<.0001	49.4 (46.9-51.9)	45.7 (42.8-48.5)	*
Role-physical (RP)	48.9 (10.5)	42.2 (13.5)	0.002	47.0 (44.0-50.0)	44.6 (41.2-47.9)	0.325
Bodily pain (BP)	51.6 (11.2)	50.3 (10.1)	0.452	50.8 (48.1-53.5)	51.3 (48.2-54.3)	0.849
General health perception (GH)	46.5 (9.8)	45.2 (8.0)	0.394	46.6 (44.3-48.8)	45.2 (42.6-47.8)	0.491
Vitality (VT)	48.9 (9.3)	48.4 (8.0)	0.774	48.4 (46.2-50.6)	49.0 (46.5-51.5)	0.755
Social functioning (SF)	49.6 (10.0)	46.6 (10.5)	0.083	48.8 (46.2-51.4)	47.5 (44.6-50.5)	0.554
Role-emotional (RE)	49.8 (10.6)	44.4 (13.6)	0.011	47.6 (44.7-50.6)	47.0 (43.7-50.4)	0.809
Mental health (MH)	50.7 (9.1)	49.4 (7.8)	0.383	50.0 (47.8-52.1)	50.3 (47.9-52.7)	0.849
Physical component summary (PCS)	50.4 (11.0)	41.5 (14.3)	<.0001	47.9 (44.9-50.9)	44.6 (41.2-48.1)	0.208
Mental component summary (MCS)	49.0 (9.3)	50.8 (6.8)	0.201	49.3 (47.2-51.3)	50.5 (48.1-52.8)	0.502

a) t-test b) ANCOVA for adjusting for age and sex *Interaction between age and classification

Table 3 Psychological status of primary and cancer-related lower limb lymphedema patients

	Total (n=141)	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value
HADS anxiety score (from 0 to 21)	median (P25-P75) 4 (2-6)	4 (2-7)	5 (3-6)	0.275 ^{a)}
HADS depression score (from 0 to 21)	median (P25-P75) 3 (2-6)	3 (1-5)	4 (3-6)	0.026 ^{a)}
HADS anxiety score ≥ 8	n (%) 27 (19.1)	15 (19.2)	12 (19.0)	0.478 ^{b)}
HADS depression score ≥ 8	n (%) 15 (10.6)	7 (9.0)	8 (12.7)	0.476 ^{b)}

a) χ^2 test b) Mann-Whitney U test
HADS, Hospital Anxiety and Depression Scale.

primary LLL patients, significant differences were not identified in comparison with cancer-related LLL patients after adjusting for age and sex. Third, we revealed the psychological status of primary LLL patients for the first time using the HADS, and the proportion of anxiety and depression was not high in patients with primary LLL. Finally, there was a significant difference in the Brief-COPE between primary LLL and cancer-related LLL patients. The higher active coping and self-blame scores in the Brief-COPE were distinctive in patients with primary lymphedema after adjusting for age. Additionally, the

higher scores of planning and acceptance coping were also distinctive in patients over 48 years old.

Only GH was significantly lower for patients with primary LLL than the normative value and there were no statistical differences in the other dimensions of the SF-36. A low GH in the SF-36 means the “recognition of not being in good health and gradually worsening.”¹⁸⁾ As lymphedema is a chronic condition with no cure at the moment, its progress is unavoidable if it is neglected²⁵⁾. In a study by Huggenberger et al.²⁶⁾, which studied the HRQOL of primary LLL patients, there were no significant differences compared to normative data

Table 4 Brief-COPE scores between patients with primary and cancer-related lower limb lymphedema

Variables	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value ^{a)}	Primary LLL (n=78)	Cancer-related LLL (n=63)	P-value ^{b)}
	Mean (SD)	Mean (SD)		Least Square Means (95%CI)	Least Square Means (95%CI)	
Subscale of the Brief-COPE						
Self-distraction	5.2 (1.1)	5.0 (1.1)	0.173	5.2 (5.0-5.5)	5.0 (4.7-5.3)	0.246
Active coping	6.0 (1.0)	5.6 (1.0)	0.014	6.2 (5.9-6.4)	5.3 (5.1-5.6)	<0.001
Denial	3.4 (1.3)	4.0 (1.3)	0.020	3.7 (3.4-4.0)	3.7 (3.3-4.0)	0.993
Substance use	3.2 (1.5)	3.0 (1.5)	0.466	3.3 (2.9-3.6)	2.9 (2.5-3.4)	0.331
Use of emotional support	5.4 (1.2)	4.8 (1.4)	0.012	5.3 (5.0-5.6)	4.9 (4.6-5.3)	0.201
Use of instrumental support	5.3 (1.2)	4.8 (1.3)	0.009	5.2 (4.9-5.6)	4.9 (4.6-5.3)	0.236
Behavioral disengagement	3.8 (1.0)	4.3 (1.2)	0.005	3.9 (3.7-4.2)	4.1 (3.8-4.4)	0.509
Venting	4.6 (1.3)	4.4 (1.1)	0.456	4.5 (4.2-4.8)	4.6 (4.3-5.0)	0.495
Positive reframing	5.6 (1.2)	5.4 (1.3)	0.455	5.7 (5.4-6.1)	5.3 (4.9-5.6)	0.069
Planning	6.0 (1.1)	5.4 (1.4)	0.003	–	–	*
Humor	4.3 (1.3)	4.3 (1.2)	0.831	4.3 (4.0-4.7)	4.2 (3.8-4.6)	0.57
Acceptance	6.0 (0.9)	5.6 (1.2)	0.013	–	–	*
Religion	3.6 (1.4)	3.7 (1.6)	0.750	3.8 (3.5-4.2)	3.4 (3.0-3.8)	0.175
Self-blame	4.8 (1.2)	4.4 (1.2)	0.038	4.9 (4.6-5.2)	4.3 (4.0-4.7)	0.021

a) t-test b) ANCOVA for adjusting for age *Interaction between age and classification

Table 5 Age-stratified analyses of subscales of the Brief-COPE

	<48 years old			≥48 years old		
	Primary LLL (n=52)	Cancer-related LLL (n=10)	P-value ^{a)}	Primary LLL (n=26)	Cancer-related LLL (n=53)	P-value ^{a)}
Planning	5.9 (5.6-6.2)	6.2 (5.4-6.9)	0.514	6.2 (5.7-6.7)	5.2 (4.9-5.6)	0.004
Acceptance	6.0 (5.7-6.3)	6.5 (5.9-7.1)	0.158	6.0 (5.6-6.5)	5.4 (5.1-5.8)	0.041

Least Square Means (95%CI). a) P-value adjusted for age

although the proportion of ISL stage III patients in the study was higher than in the present study (38.5% vs 15.4%). From these findings, although physical state has been relatively preserved and the degree of lymphedema is in the moderate stage in this study's primary LLL patients, they might have fears about their edema becoming worse associated with a change in appearance. In this study, the SF-36 scores, except for GH, were relatively high in primary LLL patients. Research that has compared the HRQOL of lymphedema patients before and after treatment showed an improvement in physical activity, pain, and vitality²⁷⁾. In our study, as participants visited the clinic at least twice,

we assumed that appropriate care including obtaining diagnosis and adequate information in the clinic had an effect on the HRQOL.

The HRQOL of patients with primary LLL was not significantly different from that of patients with cancer-related LLL after adjusting for age and sex. In this study, we controlled age and sex, which were considered to have an impact on the HRQOL, and tried to find that the difference in the etiology influenced the HRQOL. As a result, it was found that there was no difference in the outcomes of the HRQOL. Even though the etiology is different, when the patient's feeling of burden to the symptoms of lymphedema does not

change, the difference would not be observed in the HRQOL, especially regarding the physical aspects. The research by Huggenberger et al. concluded that the VT of patients with secondary LLL was lower than that of the patients with primary LLL but a clear reason was not mentioned²⁶⁾.

Focusing on the mental aspect of the primary LLL patients, the percentage of patients with anxiety or depression in the HADS was not low compared with the results of investigations of depression in lower leg ulcer patients²⁸⁾. As such, it can be said that the psychological state of primary lymphedema patients is relatively stable. It is reported that physical factors, such as pain or the smell, are related to the psychological distress of leg ulcer patients. The score of BP in the SF-36 was above the standard in the primary LLL patients in this study. This specific feature of lymphedema influenced the HADS score. The difference of depression score between patients with primary and cancer-related LLL is supported by the study that cancer-survivors show a high prevalence of depression²⁹⁾.

The patients with primary lymphedema used more active coping and self-blame than the cancer-related LLL patients in the Brief-COPE. Active coping refers to taking a proactive approach in solution finding²⁰⁾. As many primary lymphedema patients spend a long time on a waiting list before a consultation in the hospital and receiving a diagnosis³⁰⁾, the feeling of relief after beginning appropriate care might be related to their positive behavior. This conclusion is in line with the findings of another study that investigated patients who had just received comprehensive information and had feelings of being supported and motivated in the treatment after referral⁷⁾. Although the self-blame score was not very high among the Brief-COPE scores, the reason why the self-blame score in primary LLL patients was higher than that of cancer-related LLL may be the unknown cause of the impairment, and primary LLL patients may be attributing the cause to themselves. Previous studies showed that self-blame is associated with psychological distress³¹⁾³²⁾. Regarding that the planning and acceptance scores of patients with primary LLL were higher than those of cancer-related LLL patients over 48 years old, we assume that coping strategies might be more established due to the duration

of the disease compared to that of the cancer-related LLL patients who were thought to have suffered from the disease in middle age or later. This is consistent with a case study of primary LLL patients by Symvoulakis⁶⁾.

In this study, we found that coping styles differed between the groups. We need to be aware of their different coping styles and to use specific physical and mental approaches to achieve a desirable coping style as well as HRQOL. In one study in our series, we reported that approaching coping was positively associated with the mental component summary (MCS) score and distractive coping was negatively associated with the MCS score in the SF-36³³⁾. Thus, coping affects HRQOL, especially psychological status. Considering the coping property of primary LLL patients that was revealed in this study, sharing information and encouraging patients to develop an understanding of lymphedema are important factors that lead to a feeling of relief and support approaching coping strategies, including active coping, planning, and acceptance. Moreover, the approval of patients' efforts to self-manage is important for avoiding self-blame coping. The provision of care to improve general health is necessary for patients in the moderate stage of primary LLL, whose decrease in body function is not noticeable. Hence, physical self-management, such as the prevention of cellulitis that causes difficulty in working or everyday life, must be undertaken so as not to let the existing edema deteriorate.

There are limitations to this study. As the SF-36 is a general HRQOL scale, there is a possibility that physical function and physical role peculiar to the LLL patients have not been reflected. In addition, as we conducted a cross-sectional study, we cannot infer cause and effect and grasp the changes over time. This study focused on the participants after referral to the clinic. However, the HRQOL of patients with primary LLL before referral or first admission to the clinic might be lower than found in this study when the characteristic of primary lymphedema patients is considered. As we measured the differences and tendencies of coping styles for patients with primary and cancer-related lymphedema, we were unable to investigate intrapersonal changes in coping style for primary lymphedema patients from the early onset of the condition to its maturity. Future research needs to pay more attention to the intrapersonal

changes in HRQOL and coping style to discover the specific psychosocial aspects of primary LLL patients.

Conclusion

This study found a significant difference in coping styles between patients with primary lymphedema and those with cancer-related lymphedema while there was no significant difference in HRQOL between the groups. Our study highlights the importance of providing proper support according to their coping disposition, which leads to the maintenance of HRQOL. Sharing information about lymphedema and enhancing physical self-management is necessary for patients with primary LLL.

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原発性下肢リンパ浮腫患者とがん関連下肢リンパ浮腫患者における健康関連 QOL とコーピングスタイルの比較

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要旨

背景：原発性リンパ浮腫患者とがん関連リンパ浮腫患者では疾患の成り立ちやその背景の違いから健康関連 QOL (HRQOL) やコーピングスタイルが異なることが予測される。本研究は、原発性下肢リンパ浮腫患者とがん関連下肢リンパ浮腫患者を比較することにより、原発性リンパ浮腫患者の HRQOL とコーピングスタイルを明らかにすることを目的とする。

方法：HRQOL を測定するスケールとして SF-36 日本語版、日本語版 HAD 尺度 (HADS) を用い、コーピングスタイルの測定には日本語版 Brief-COPE を用いた。基本属性、リンパ浮腫の状態は診療録、フィジカルアセスメントより情報を得た。

結果：原発性下肢リンパ浮腫患者 78 名、がん関連下肢リンパ浮腫患者 63 名が対象となった。年齢、性別をマッチングさせた国民標準値と比較すると原発性下肢リンパ浮腫患者における HRQOL は全般的健康感 (GH) のみ低い結果であった。さらに、共分散分析を用い年齢、性別を調整した両群の HRQOL の比較においては、有意差は認められなかった。HADS では、不安、抑うつともに点数 8 (不安、抑うつの疑いのあるスコア) 以上の患者の割合は低い結果であった。Brief-COPE の項目では、原発性下肢リンパ浮腫患者における積極的コーピングと自己非難の得点が、がん関連下肢リンパ浮腫患者よりも高い結果であった。

結論：原発性下肢リンパ浮腫患者とがん関連下肢リンパ浮腫患者の HRQOL の違いは確認されなかったが、コーピングスタイルの違いが明らかとなった。

キーワード：先天性リンパ浮腫、ストレス対処行動、不安、抑うつ