

# Aggravated Primary Lymphedema of Contralateral Upper Limb after the Radical Mastectomy: A Case Report

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## Case report

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# Abstract

**Background:** Breast cancer-related lymphedema is usually characterized by edema of the affected the ipsilateral upper limb and trunk. We report a case of aggravated primary lymphedema in the contralateral limb and trunk after breast cancer resection and axillary lymph node dissection.

**Case presentation:** A 63-year-old female developed right thorax-back and upper limb swelling since childhood. After the modified mastectomy, the swelling of her right chest, back and upper limb increased. While she had no edema of left torso and limb. There was no relevant supplementary examination data because she refused to take lymphoscintigraphy or MRI scan. However, according to her medical history and physical examination, she was preliminarily diagnosed as primary lymphedema, International Society of lymphedema stage 2.

## Conclusions:

Primary lymphedema and secondary lymphedema may be both results of the interaction of multiple factors and can be induced or aggravated by trauma, surgery or other reasons besides the abnormal lymphatic development of individuals.

## Background

Lymphedema can be classified into primary and secondary types. Primary lymphedema (PLE) is due to a developmental defect of (malformation, dysplasia) of the lymph vessels and/or lymph nodes. PLE can be developed if minor surgery is performed in a region of the body in which the transport capacity is subnormal because of dysplasia of the lymph vessels. Breast cancer-related lymphedema (BCRL) is a common secondary lymphedema (SLE). It can be seen in the affected upper limb often after axillary lymph nodes dissection (ALND)(28.2%)or sometimes after sentinel lymph node biopsy (SLNB) (5.6%) [1]. Although there have been a few reports indicating that BCRL may affect the clinically “normal” contralateral arm, only exacerbation of lymphedema in the contralateral trunk and upper limb while no obvious edema in the affected upper limb was seldom addressed before. In this report, we describe a patient who underwent progressive PLE in the right trunk and upper limb after left radical mastectomy.

## Case Presentation

A 63-year-old female visited the clinic of the Peking Union Medical College Hospital for her right thoracic back and upper limb edema. She developed right thorax-back and upper limb swelling since childhood, and upper limb swelling was confined to her right hand and wrist. She received modified mastectomy for left breast cancer one year ago in a local hospital. Her left axillary lymph node was removed. Six months after the operation, the swelling of her right chest, back and upper limb gradually increased, and the swelling of her right forearm and upper arm appeared. In recent one month, the edema was markedly aggravated, and the skin of upper arm and forearm became stiff with exudation of right nipple. She had no complaints of swelling of left torso and limb, pain and numbness of right limb. Her past medical

history did not reveal any relevant disease related to arm swelling. Her family history was unremarkable for any similar illness. On physical examination, the right thorax-back, upper limb and hand were markedly swollen. The skin of the right upper limb was stiff, and yellowish transparent water-like exudate could be seen in the right nipple. There was no visible swelling in the left chest, back and upper limb, as shown in the following figure.

Examination of bilateral lower extremities were completely normal.

She refused to take lymphoscintigraphy or MRI scan as a provincial patient without medical insurance. So we haven't got the relevant supplementary examination data. However, according to her medical history and physical examination, she was preliminarily diagnosed as PLE, International Society of lymphedema (ISL) stage 2.

## Discussion And Conclusion

PLE can occur at an early age and can also be triggered by trauma, surgery or other reasons. BCRL is a common SLE, usually confined to the affected upper limb or and thorax-back. The traditional view of BCRL pathophysiology is that removal of axillary nodes results in obstruction to the flow of lymph from the arm, leading to accumulation of protein-rich fluid in the interstitium: the 'stopcock' hypothesis. But some women develop BCRL after sentinel node biopsy, whereas others do not after clearance surgery, which suggest that a simple 'stopcock' mechanism does not explain many clinical aspects, including the delayed onset and selective sparing of some regions, e.g, hand. BCRL is a result of interaction between several pathophysiological processes, and is not simply a 'stopcock' effect resulting from removal of axillary lymph nodes<sup>[2]</sup>. Predisposition hypothesis has therefore attracted the attention of some researchers. Aldrich found the appearance of vessel abnormalities on unaffected arms with increasing time which indicated that BCRL might progress to affect the clinically "normal" arm and become a systemic, rather than local, malady. So care should be given to both arms after cancer treatment<sup>[3]</sup>. In other two studies, rates of depot clearance after subcutaneous injection of radiolabeled protein in the hands and subsequent appearance in venous blood suggested lymphatic dysfunction in both affected and contralateral "normal" arms<sup>[4, 5]</sup>. Bains found that women with upper-limb BCRL had reduced lower-limb lymphatic function and a large proportion of women with breast cancer had abnormal lower-limb lymphatic function, irrespective of whether they had symptoms of upper-limb BCRL or not. Possible explanations could be a systemic effect of breast cancer or its treatment, or an unidentified association between breast cancer and lymphatic dysfunction. There is a constitutional 'global' lymphatic dysfunction in patients who develop BCRL<sup>[6]</sup>. Furthermore, the risk factors of BCRL are multiple. A genetic predisposition for lymphatic dysfunction, other surgical or injury damage to lymphatics, body mass index, medications, and fitness level are all suspected systemic factors contributing to the development of lymphedema<sup>[7, 8]</sup>.

In addition, WeiRen Pan found that the individualization of lymphatic system development varies greatly<sup>[9]</sup>. The clinical symptoms and signs of lymphedema are related to the anatomical differences of

individual lymphatic system.

The special point of this case was that radical mastectomy aggravated the original lymphedema of trunk and upper limb, but it only occurred in the opposite side of the operation. In addition to the above predisposing hypothesis and cancer treatment could trigger or aggravate systemic edema, the individual differences of lymphatic dysplasia and the resection or damage of lymphatic vessels and lymph nodes in the affected trunk and upper extremities after surgery should also be taken into account. And the lymph flow of the affected side might be past sagittal watershed and be reversed toward the opposite side, thus leading to the progressive PLE of the unaffected trunk and upper limb. Bobbio had ever reported the similar situation, as showed in the following figure.

To summarize, PLE and SLE may be both results of the interaction of multiple factors. Tumor itself or cancer treatment may induce or aggravate SLE in susceptible patients. PLE can be induced or aggravated by trauma, surgery or other reasons besides the abnormal lymphatic development of individuals, and may be manifested differently according to the specific conditions of individual lymphatic abnormalities. Further studies need to determine whether systemic manifestation of lymphatic abnormalities in cancer-related lymphedema are indicative of progressive disease and their relationship with cancer treatment.

## Abbreviations

PLE

Primary lymphedema

BCRL

Breast cancer-related lymphedema

SLE

Secondary lymphedema

ALND

Axillary lymph nodes dissection

SLNB

Sentinel lymph node biopsy

ISL:

International Society of lymphedema

# Declarations

## Availability of data and materials

N/A.

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### Contributions

Ying Liu had substantial contributions to the work including the acquisition, analysis, and interpretation of data, drafting and revising the manuscript. Weiren pan aided in revising the manuscript critically for important intellectual content and polishing the language of English. Jinghong Guan was in charge of clinical diagnosis and evaluation of breast cancer of this case and polishing the language of English. Xiao Long was responsible for diagnosis and evaluation of lymphedema of this case, design of the work, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved, And all authors have read and approved the manuscript.

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### Ethics declarations

Ethics approval and consent to participate

Written and informed consent was obtained from the patient for the publication of this study; moreover, no patient health identification (PHI) was recorded.

### Consent for publication

The patient gave her written informed consent for the publication of her data.

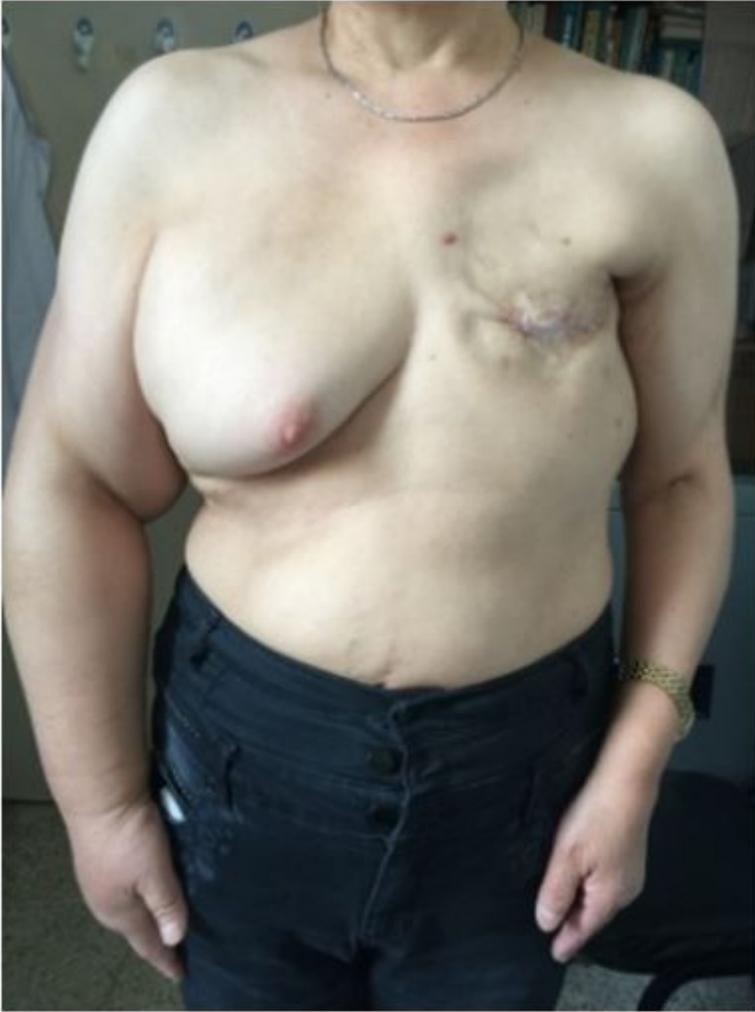
## Competing interests

Drs. Ying Liu, Weiren pan, Jinghong Guan and Xiao Long had no competing interests or financial ties to disclose.

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## Figures



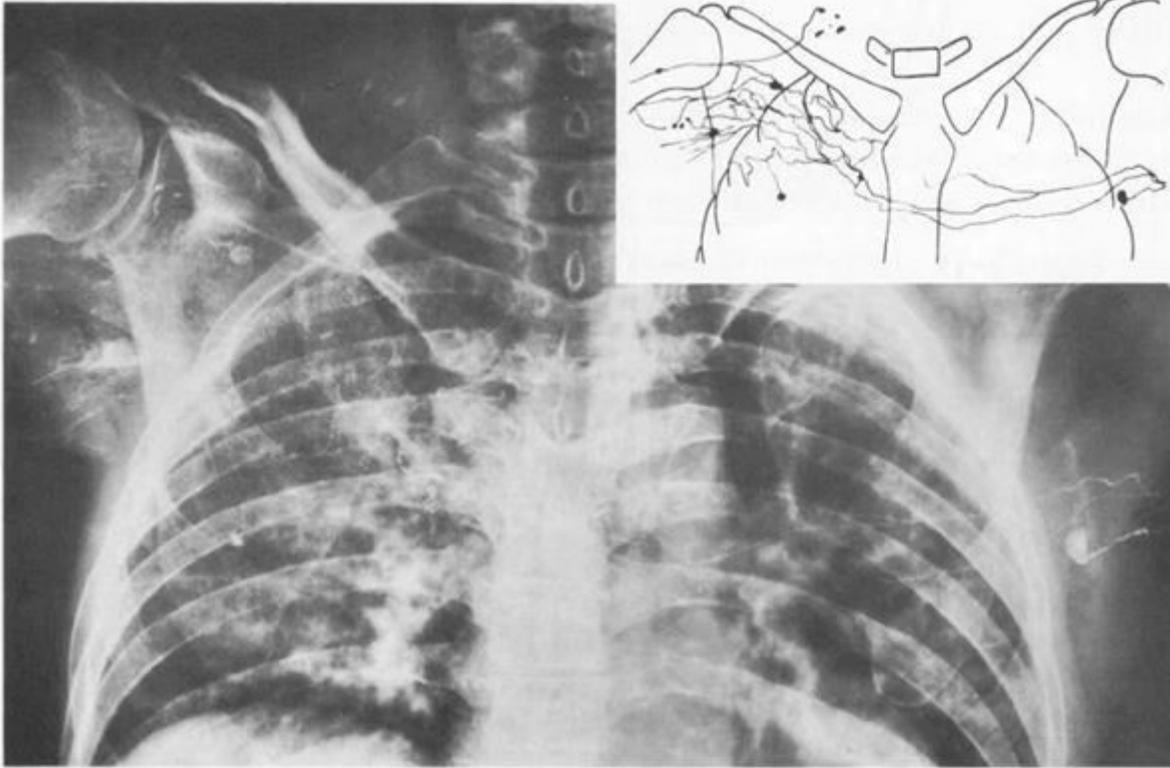
**Figure 1**

Anterior view of trunk and upper limb. Postoperative changes of left chest after mastectomy. Swelling of the right thorax, upper limb and hand and normal left upper limb.



**Figure 2**

Posterior view of trunk and upper limb. Swelling of the right back, upper limb and hand and normal left back and upper limb.



**Figure 3**

External connections between the mammary regions on both sides. P.Bobbio,G. Peracchia, F. Pellegrino, Connessioni linfatiche presternalifra le regioni mammary dei due lati, *AteneoParmense* 33 (supp) (1962)95-109.