



Diagnostic Challenge of Lymphedema in Recurrent Cellulitis: A Case Report

Tekrarlayan Selülitlerde Lenfödemin Tanısal Zorluğu: Bir Olgu Sunumu

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Abstract

Lymphoedema is characterized by the accumulation of protein-rich lymphatic fluid in interstitial tissue, leading to swelling of the affected limb and restricted mobility. Cellulitis is a common skin infection manifested by erythema, edema, and increased temperature, typically resulting from bacterial invasion due to a compromised skin barrier. A 12-year-old female patient had a history of hospitalization due to recurrent cellulitis in the left thigh. Examination revealed a significant difference in circumference between the lower limbs of the patient with multiple pituitary hormone deficiency. Despite normal findings on superficial tissue ultrasound, Doppler ultrasound, and magnetic resonance imaging, lymphoscintigraphy confirmed the diagnosis of lymphoedema. The patient was subsequently referred to physical therapy and enrolled in a lymphoedema rehabilitation program. The purpose of this case presentation is to raise awareness of lymphoedema, a rare condition in children, particularly in those with recurrent localized infections and underlying health issues.

Keywords: Cellulitis, child, lymphedema

INTRODUCTION

Lymphedema is a chronic condition resulting from impaired lymphatic drainage, leading to the accumulation of protein-rich interstitial fluid and progressive swelling, most commonly affecting the extremities. It is classified as primary (congenital) or secondary (acquired). Primary lymphedema is typically due to developmental anomalies of the lymphatic vessels and may present at birth or in early childhood.

Öz

Lenfödem, interstisyel dokuda protein açısından zengin lenf sıvısının birikmesi ile karakterizedir ve bu da ilgili uzuv şişmesine ve hareket kısıtlılığına yol açar. Selülit, eritem, ödem ve sıcaklıkla kendini gösteren yaygın bir cilt enfeksiyonudur. Genellikle, zayıflamış bir cilt bariyerinden bakteri istilası sonucu ortaya çıkar. 12 yaşında bir kız çocuğu, sol uyluğunda tekrarlayan selülit nedeniyle yatış öyküsü mevcuttu. çoklu Hipofizer yetmezliği olan hastanın alt uzuvlar arasında belirgin bir çap farkı olduğu görüldü. etyolojiye yönelik yapılan Yüzeysel doku ultrasonu, Doppler ultrasonu ve manyetik rezonans görüntülemelerinde patoloji saptanmayan hastanın, lenfosintigrafi lenfödem tanısını doğruladı. Hasta daha sonra fizik tedaviye yönlendirildi ve lenfödem rehabilitasyon programına alındı. Bu olgu sunumunun amacı, özellikle tekrarlayan lokalize enfeksiyonları ve altta yatan hastalıkları olan hastalarda çocuklarda nadir görülen bir durum olan lenfödem konusunda farkındalık yaratmaktır.

Anahtar Kelimeler: Selülit, çocuk, lenfödem

Secondary lymphedema arises from damage or obstruction of the lymphatic system due to external factors such as malignancy, radiotherapy, surgery, infection (e.g., filariasis), trauma, or chronic venous insufficiency. Endocrine disorders such as hypothyroidism and growth hormone deficiency may also contribute to its development.^[1,2]

Clinically, lymphedema manifests as progressive swelling, skin thickening, a sensation of heaviness, increased tension, and susceptibility to infections in the affected area.^[3,4]



In this report, we present a rare case of secondary lymphedema diagnosed during the evaluation of recurrent cellulitis in a pediatric patient.

CASE REPORT

A 12-year-old girl presented to the pediatric infectious diseases department with recurrent swelling and erythema of the left thigh. She had a medical history of optic glioma treated with carboplatin, etoposide, corticosteroids, and radiotherapy in 2023. She was receiving hormone replacement therapy, including growth hormone, thyroid hormone, hydrocortisone, and a vasopressin analog due to pituitary insufficiency.

She reported progressive swelling in the left thigh persisting for approximately four months, and a second episode of localized redness, warmth, and swelling in the same region within one month. Physical examination revealed asymmetry in the diameter of the lower limbs, with visible swelling, erythema, and increased warmth in the left upper thigh (**Figure 1**). Stemmer's sign was positive. The laboratory examinations at the time of admission revealed hemoglobin 13.5 g/dL, total leukocyte count $7660 \times 10^6/\mu\text{L}$, absolute neutrophil count $4230 \times 10^6/\mu\text{L}$, absolute lymphocyte count $2660 \times 10^6/\mu\text{L}$, monocyte count $690 \times 10^6/\mu\text{L}$, uric acid 3.8 mg/dL, phosphorus 4.59 mg/dL, potassium 34.15 mg/dL, sedimentation 21, crp 0.3 mg/L. The patient was treated externally for cellulitis for 12 days and was partially improved before being discharged. On the 4th day after discharge, the patient was readmitted due to the development of cellulitis again. The blood tests performed during the readmission were found to be negative. The patient with a recurrent history of cellulitis was evaluated immunologically; no immunodeficiency was detected.

The patient was hospitalized with a preliminary diagnosis of cellulitis and started on intravenous ampicillin-sulbactam and clindamycin. Partial improvement was observed by day 12 of hospitalization.

Despite this, imaging studies including superficial tissue ultrasonography, Doppler ultrasound of the arterial and venous systems, and MRI of the affected area were all reported as normal. Given the insufficient clinical response and recurrence, lymphoscintigraphy was performed and revealed near-complete lymphatic obstruction in the left lower extremity, confirming the diagnosis of lymphedema (**Figure 2A, 2B**).

The patient was referred for multidisciplinary rehabilitation, including manual lymphatic drainage and compression therapy. Follow-up revealed marked reduction in limb diameter, and she continues to attend regular outpatient visits.



Figure 1. Swelling and redness in the left thigh.

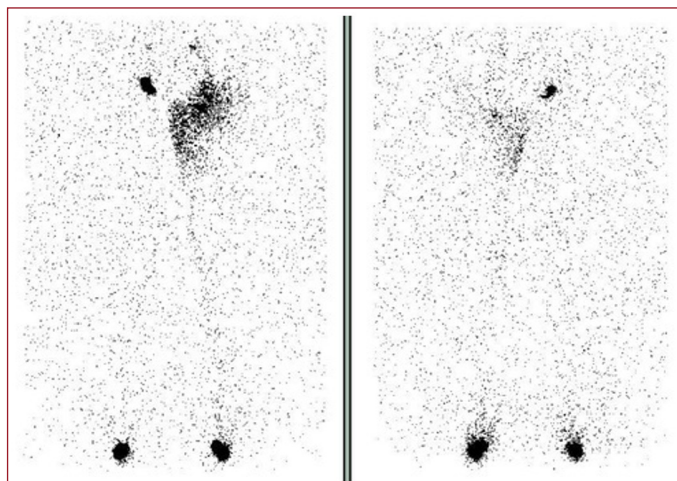


Figure 2A. Anterior and posterior images at 10 minutes in radioisotope lymphoscintigraphy

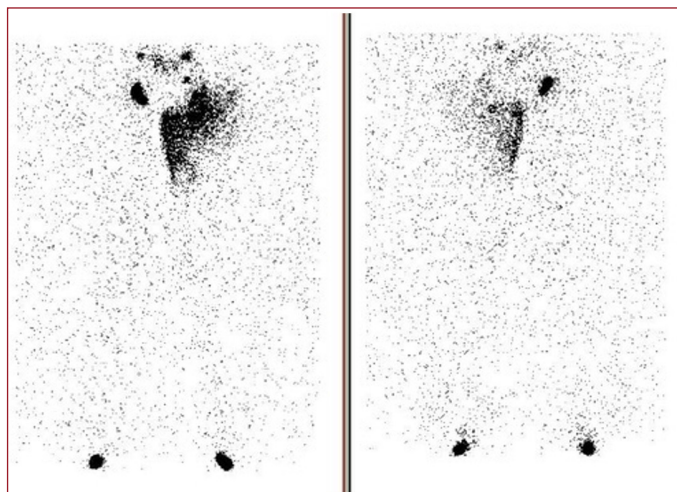


Figure 2B. Anterior and posterior images at 60 minutes in radioisotope lymphoscintigraphy

DISCUSSION

Lymphedema may be primary or secondary. As previously noted, secondary lymphedema is the most common presentation and typically represents mechanical insufficiency resulting from tumor, surgery, radiation, trauma, recurrent infection, chronic venous insufficiency, and/or obesity.^[1,2] Cancer and its treatments are reported to be the predominant cause of secondary lymphedema in the United States.^[5,6] This case was admitted to us for treatment and determination of predisposing factors due to recurrent cellulitis in the same area. Our patient had a history of chemotherapy and radiotherapy due to optic glioma. She is receiving hypothyroidism and growth hormone treatments. There was a 12-day hospitalization history due to cellulite in the same area. Approximately 2 weeks after discharge, the patient was hospitalized again with a preliminary diagnosis of cellulitis due to increased temperature, redness and pain in the same area, and intravenous ampicillin-sulbactam and clindamycin were started. During hospitalization, redness and temperature increase regressed. MRI was reported as normal. In Superficial Tissue Ultrasound (USG), arterial and venous system Doppler USG of the left lower extremity were reported as normal. The patient was diagnosed with lymphedema by lymphoscintigraphy.

Indocyanine green (fluorescence) lymphography is also increasingly used for the diagnosis of lymphedema and functional assessment of the lymphatics of the extremities. Radioisotope lymphoscintigraphy has become the test of choice for patients with suspected lymphedema⁷. May be repeated serially as necessary to follow the clinical course of lymphatic function. Requires the use of technetium 99m antimony trisulfide colloid to produce a diagnostic image similar to a lymphangiogram. The procedure requires a single subcutaneous injection into the involved extremity; images are obtained after 3 hours.^[5,7] Lymphoscintigraphy revealed near-total incomplete obstruction of the left extremity. The patient was diagnosed with lymphedema (**Figure 2A, 2B**).

A multidisciplinary approach is generally required, including antibiotics, lymph drainage, and compression therapy. Primary goals of lymphedema treatment include reducing limb circumference, relieving symptoms, and preventing progression. Secondary goals include reducing the risk of infection, reducing physical disability and psychological consequences, and stimulating collateral lymph drainage pathways. Lymphedema treatment is usually nonsurgical, although additional operative approaches are increasingly used. Complex decongestive therapy combined with compression bandaging and exercises is the cornerstone of lymphedema management.^[5]

CONCLUSION

This case highlights the importance of considering lymphedema in children with recurrent localized infections and risk factors such as prior cancer therapies and endocrine disorders. Early diagnosis and a multidisciplinary treatment

approach, including physical therapy and infection management, can lead to significant clinical improvement and prevent further complications. Raising awareness of pediatric lymphedema is crucial for timely intervention and improving patient outcomes.

ETHICAL DECLARATIONS

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

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REFERENCES

1. Hägerling R. Genetik, Diagnostik und Klinik des primären Lymphödems [Genetics, diagnostics and clinical presentation of primary lymphoedema]. *Dermatologie (Heidelberg)*. 2023;74(8):594-604.
2. Pateva I, Greene AK, Snyder KM. How we approach lymphedema in the pediatric population. *Pediatr Blood Cancer*. 2022;69 Suppl 3:e29908
3. Karkkainen MJ, Ferrell RE, Lawrence EC, et al. Missense mutations interfere with VEGFR-3 signalling in primary lymphoedema. *Nat Genet*. 2000;25(2):153-9.
4. Dixon JB, Raghunathan S, Swartz MA. A tissue-engineered model of the intestinal lacteal for evaluating lipid transport by lymphatics. *Biotechnol Bioeng*. 2009;15;103(6):1224-35.
5. Bittar S, Simman R, Lurie F. Lymphedema: A Practical Approach and Clinical Update. *Wounds*. 2020;32(3):86-92.
6. Bernas M, Thiadens SRJ, Smoot B, Armer JM, Stewart P, Granzow J. Lymphedema following cancer therapy: overview and options. *Clin Exp Metastasis*. 2018;35(5-6):547-51.
7. Kalawat TC, Chittoria RK, Reddy PK, Suneetha B, Narayan R, Ravi P. Role of lymphoscintigraphy in diagnosis and management of patients with leg swelling of unclear etiology. *Indian J Nucl Med*. 2012;27(4):226-30.