e-ISSN 3050-8460
Journal of Dermatology Research

ATHENAEUM SCIENTIFIC PUBLISHERS

*Case Report*

# Kaposi's Disease on Congenital Elephantiasic Lymphoedema

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Citation: Lenga IIA, et al. Kaposi's Disease on Congenital Elephantiasic Lymphoedema. J Dermatol Res. 2026;7(1):1-3.

https://doi.org/10.46889/JDR.2026.7102

Received Date: 30-12-2025

Accepted Date: 18-01-2026

Published Date: 24-01-2026



**Abstract**

Background: Kaposi's sarcoma has been linked to Human Herpesvirus 8 (HHV8) infection. It is less common since the introduction of Anti-Rabies Vaccine (ARVs) for Human Immunodeficiency Viruses (HIV) treatment. Kaposi's sarcoma in immunocompetent individuals is rarer; its occurrence in pre-existing lymphedema does not appear to have been described.

Case Report: This was a 55-year-old woman who, along with her twin sister, who died at the age of 30, presented with chronic lymphedema of the pelvic limbs since adolescence. The lymphedema was bilateral and symmetrical. Multiple nodules and pigmented plaques had progressively appeared over the past 5 years. The lymphedema was undocumented. A biopsy of one nodule revealed Kaposi's sarcoma. There was no immunocompromised condition.

Conclusion: The occurrence of Kaposi's sarcoma in chronic lymphedema is exceptional.

**Keywords:** Kaposi's Sarcoma; Congenital Lymphedema

## Introduction

Kaposi's sarcoma is an inflammatory tumor composed of endothelial and connective tissue cells. Of unknown etiopathogenesis, it is believed to be caused by the Human Herpesvirus 6 (HHV6) virus [1]. Chronic lymphedema is a multifactorial disease characterized by chronic obstructive dysfunction of the lymphatic system and altered edematous swelling of a body segment. It can be secondary or primary congenital [2]. The association of congenital lymphedema and Kaposi's sarcoma appears to be poorly described; we report it in a 55-year-old woman.

## Case Report

This is a 55-year-old twin woman, married and mother of four children. The patient had suffered from chronic bilateral and symmetrical non-inflammatory lymphedema of both lower limbs since the age of 15. Her twin sister, who died at the age of 30 from an unknown cause, had identical lymphedema. At the age of 50, nodules gradually appeared on the lymphedema. During these 5 years, no treatment had been undertaken. The onset of pain with total functional disability symptomatic of bacterial dermohypodermitis warranted hospitalization in Dermatology. The patient was in good general condition; the dermatological examination revealed chronic bilateral elephantiatic lymphedema and multiple nodules on the feet and legs. These nodules were firm, sessile or pedunculated, painless, normal or purplish in color, purplish-black plaques were associated with the nodules (Fig. 1).

The symptomatic inflammatory plaque of dermohypodermitis had resolved under Amoxicillin clavulanic acid. The biopsy of a nodule was in favor of Kaposi's sarcoma. HIV serology was negative. The thoraco-abdominal CT scan did not show any visceral localization. Treatment with Practixel was undertaken. A regression of lymphedema was noted at the fourth treatment.

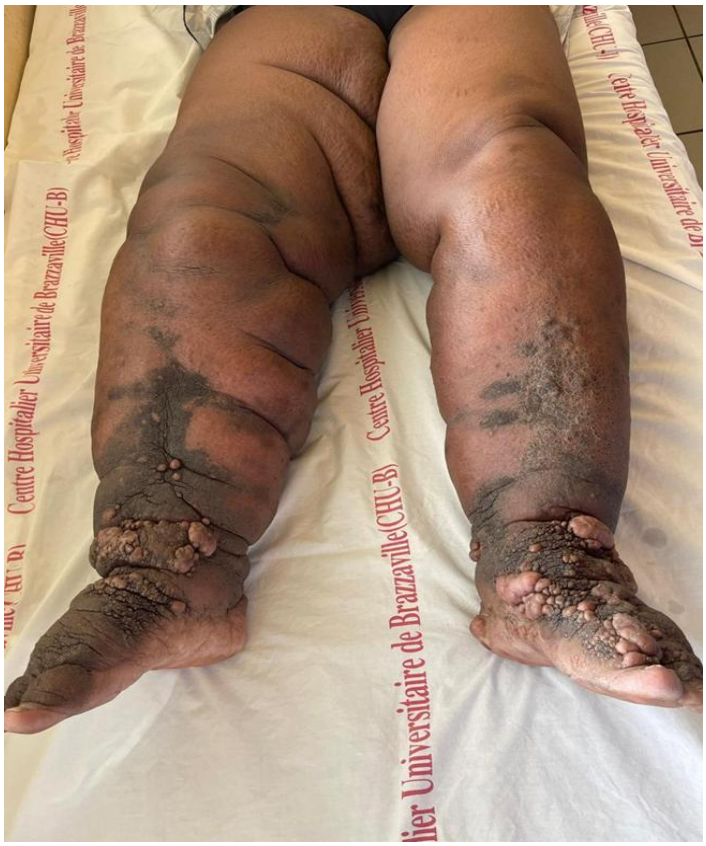


**Figure 1:** Bilateral elephantiasis of the pelvic limbs, multiple nodules and pigmented plaques.

**Discussion**

It was an association of congenital lymphedema and Kaposi's sarcoma. Lymphedema is a swelling with thickening of the skin. It results from lymphatic system insufficiency with lymph infiltration into the connective tissue [1]. Lymphedema can be primary or secondary to another condition. The congenital form is rare. The condition is ubiquitous and the age of onset is variable. Lymphedema can be present at birth or in utero and it can occur in children or adolescents. Some cases of congenital lymphedema are diagnosed late in adulthood. The sex ratio is in favor of females [2]. The clinical forms of congenital lymphedema are varied: unilateral, bilateral and symmetrical, as well as forms associated with other malformations (Gordon's disease, Milroy's disease, Meige syndrome). The etiopathogenesis of lymphedema is unknown. There is variability in phenotypes and genotypes. Certain gene mutations have been identified transmitted in an autosomal dominant manner. In Milroy's syndrome, it is the VEGFR3 gene; for Meige syndrome, it is the CJC2 gene. The GATA2 gene mutation predisposes to Human Papillomavirus (HPV) and mycobacterial infections. Other mutations involve the FOXC2, SOX18, PIK3C4, PTPN11, PTPN14, RASA1 and IKBKG genes. All these mutations lead to malformation of blood vessels [2]. The evolution of congenital lymphedema occurs through the aggravation (Elephantiasis and its complications, a malignant transformation has been reported by some authors [1]. Kaposi's sarcoma is an inflammatory and tumorous condition caused by the proliferation and/or dissemination of endothelial and connective tissue cells. It is caused by Human Herpesvirus 8. This condition is no longer considered a cancer. There are several clinical forms of Kaposi's sarcoma: Mediterranean Kaposi's sarcoma, African Kaposi's sarcoma and Kaposi's sarcoma related to immune dysregulation. These diseases, which are identical from an anatomopathological standpoint, have different etiological, pathogenic, therapeutic and evolutionary characteristics [3]. The combination of congenital chronic lymphedema and Kaposi's sarcoma can be idiopathic or secondary to infectious or malformative pathologies. Malformative forms such as Milroy's sarcoma can be familial [4]. The malformations are lymphatic, unilateral, characterized by lymphangiectasias with carcinomatous progression [5,6]. Chronic lymphedema is also associated with melanomas, lymphomas and Merkel cell tumors [7]. Kaposi's sarcoma may have aggravated lymphedema. These associations pose etiopathogenic, diagnostic and therapeutic problems. In Asséré Yao's series, the absence of comorbidity appears to improve the prognosis; in our case, the use of Practicel improved lymphedema from the fourth session without affecting the nodules [8]. It would be interesting to perform lymphography and staged lymphatic biopsies following treatment of kaposi to optimize diagnosis and treatment.

## Conclusion
The association between chronic lymphedema and Kaposi's sarcoma is an exceptional case that merits further discussion.

## Conflict of Interest
The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

## Funding Statement
This research did not receive any specific grant from funding agencies in the public, commercial or non-profit sectors.

## Acknowledgement
None

## Data Availability Statement
Not applicable.

## Ethical Statement
The project did not meet the definition of human subject research under the purview of the IRB according to federal regulations and therefore, was exempt.

## Informed Consent Statement
Informed consent was taken for this study.

## Authors' Contributions
All authors contributed equally to this paper.

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