Cystic lymphangiomas: A rare axillary presentation in a young adult patient: A Clinical Overview and literature review

Abstract:
Cystic lymphangiomas are rare congenital malformations of the lymphatic system, characterized by benign dysembryoplastic lesions. These anomalies predominantly occur in the cervicofacial and axillary regions, with most cases diagnosed during childhood. Adult presentations are exceedingly uncommon.

We present a case of axillary cystic lymphangioma in a 32-year-old patient, highlighting its atypical adult onset and clinical implications.

Key words: cystic lymphangioma; malformation; lymphatic system, axillary mass, adult case.

Introduction :

Cystic lymphangiomas are rare benign dysembryoplastic lesions of the lymphovascular system, resulting in a tumoral syndrome due to abnormal proliferation of lymphatic vessels [1]. Predominantly localized in the cervicofacial and axillary regions [2], these malformations typically present during early childhood [1], with adult-onset cases remaining exceptionally rare.

This congenital anomaly is attributed to abnormal sequestration of lymphatic tissue that retains its proliferative potential. Three principal histological subtypes are recognized: capillary, cavernous, and cystic lymphangiomas [3].

We report a case of cystic lymphangioma with delayed diagnosis in a 32-year-old adult male, emphasizing the clinical challenges and management considerations of this exceptionally rare presentation in adulthood.

Case Presentation

A 32-year-old male patient, with a documented history of left axillary cystic lymphangioma since 2011, underwent two prior surgical resections (2011 and 2014).

⇒ **Clinical Examination**
The patient presented with multiple subcutaneous nodular lesions in the left axillary region. The lesions were:

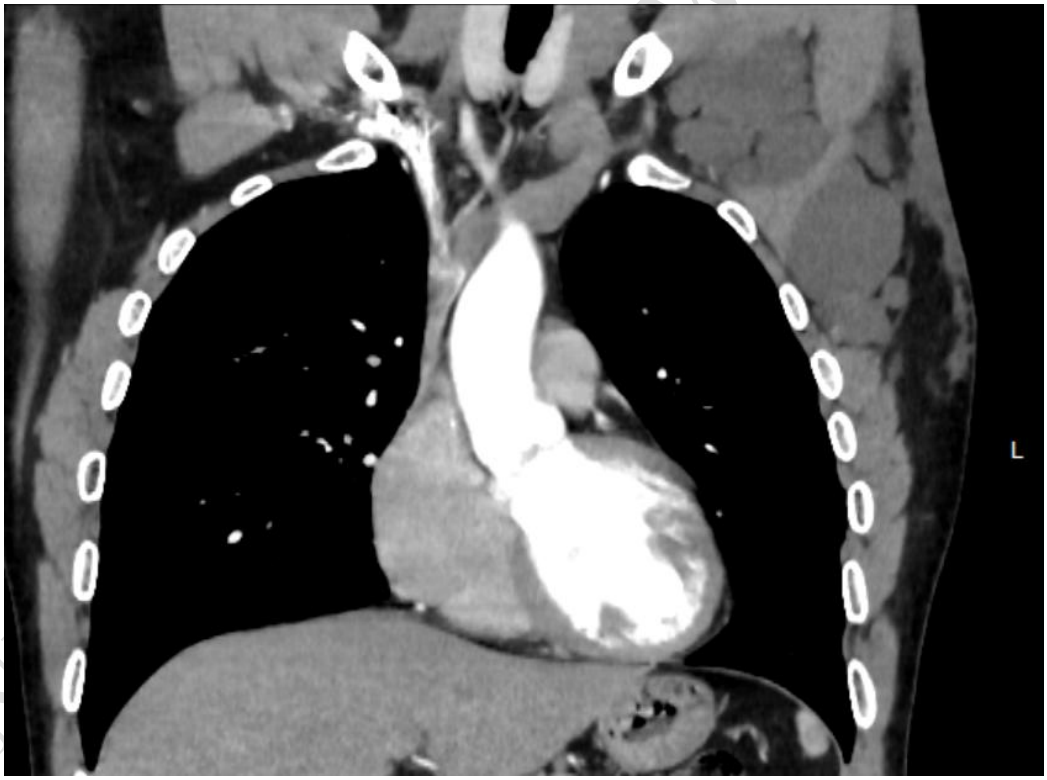
- Palpable, painless, and well-circumscribed
- Firm, mobile, and non-pulsatile
- Without overlying inflammatory signs

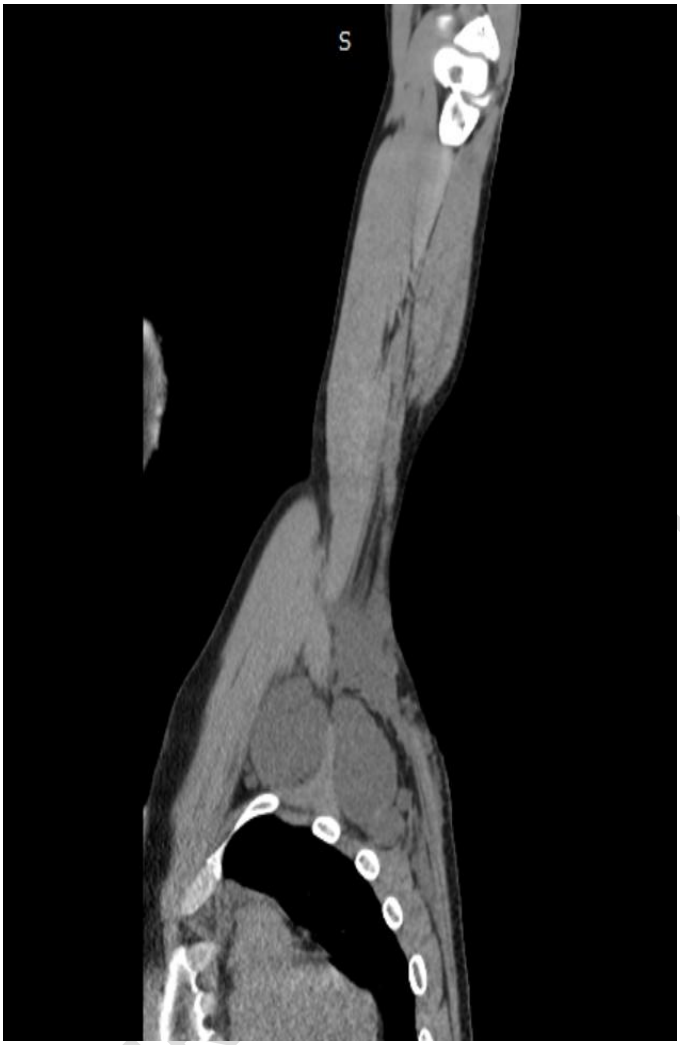
- No sensorimotor deficits were observed in the ipsilateral upper limb, and radial/cubital pulses were preserved.

⇒ **Imaging Findings**

- **Ultrasound**: Revealed a multiloculated anechoic cystic mass.
- **CT:** CT scan showing fluid collections within the deep soft tissues of the left axillary region, closely associated with collateral branches of the axillary artery, consistent with recurrence of a known cystic lymphangioma. Post-therapeutic changes in the skin and subcutaneous tissues.
- **MRI**: Demonstrated two well-defined, thin-walled, multiloculated cystic formations:
    - *Larger lesion*: Left axillary region (114 × 37 × 33 mm)
    - *Smaller lesion*: Ipsilateral lateral thoracic wall

    Both exhibited:
    - Marked T2 hyperintensity and T1 hypointensity
    - No gadolinium enhancement (post-contrast)
    - Delicate hypointense T1/T2 septa with mild contrast uptake

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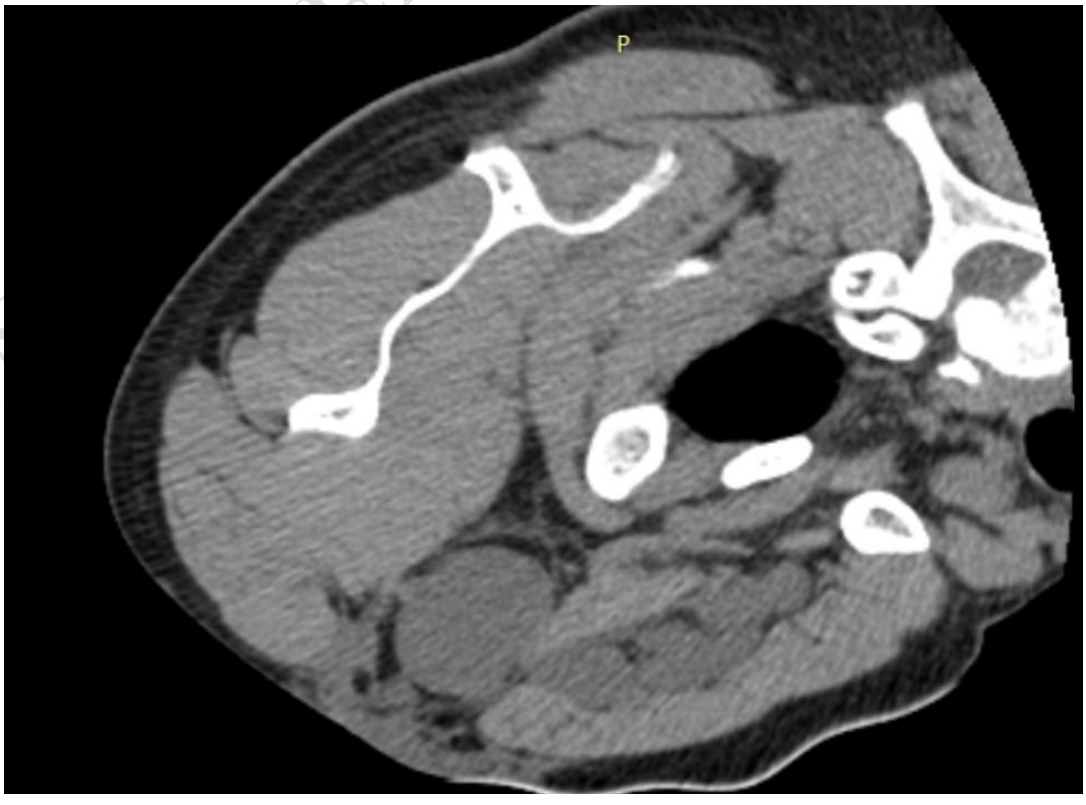
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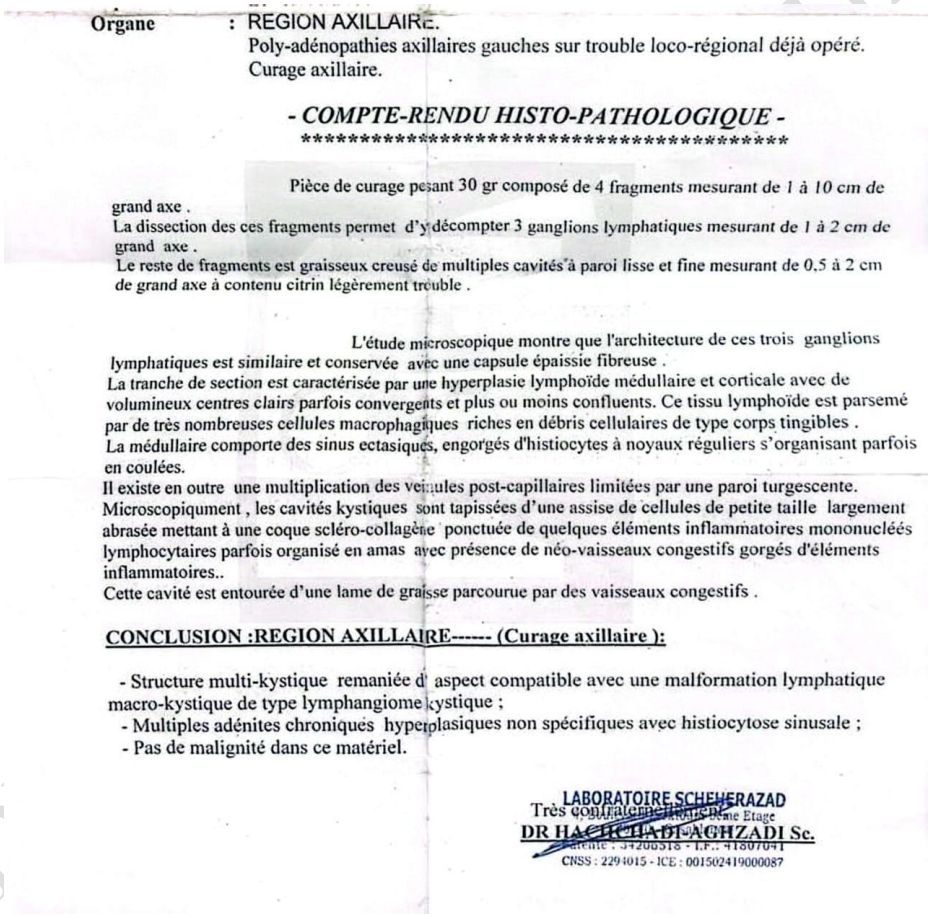
Radiological images show a multiloculated cystic mass in the left axilla without detectable solid components, associated with satellite nodules exhibiting identical characteristics. Overall findings are consistent with cystic lymphangiomatosis.

⇒ **Diagnosis**

Imaging findings were consistent with **cystic lymphangioma**, later confirmed by histopathological analysis of excised axillary specimens.

⇒ **Pathology report**

The anatomic pathology results confirmed the diagnosis, as shown in the following report.



Organe : REGION AXILLAIRE.
Poly-adénopathies axillaires gauches sur trouble loco-régional déjà opéré.
Curage axillaire.

- COMPTE-RENDU HISTO-PATHOLOGIQUE -
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Pièce de curage pesant 30 gr composé de 4 fragments mesurant de 1 à 10 cm de grand axe .
La dissection des ces fragments permet d'y décompter 3 ganglions lymphatiques mesurant de 1 à 2 cm de grand axe .
Le reste de fragments est graisseux creusé de multiples cavités à paroi lisse et fine mesurant de 0,5 à 2 cm de grand axe à contenu citrin légèrement trouble .

L'étude microscopique montre que l'architecture de ces trois ganglions lymphatiques est similaire et conservée avec une capsule épaissie fibreuse .
La tranche de section est caractérisée par une hyperplasie lymphoïde médullaire et corticale avec de volumineux centres clairs parfois convergents et plus ou moins confluents. Ce tissu lymphoïde est parsemé par de très nombreuses cellules macrophagiques riches en débris cellulaires de type corps tingibles .
La médullaire comporte des sinus ectasiqués, engorgés d'histiocytes à noyaux réguliers s'organisant parfois en coulées.
Il existe en outre une multiplication des vaisseaux post-capillaires limitées par une paroi turgescente.
Microscopiquement , les cavités kystiques sont tapissées d'une assise de cellules de petite taille largement abrasée mettant à une coque scléro-collagène ponctuée de quelques éléments inflammatoires mononucléés lymphocytaires parfois organisé en amas avec présence de néo-vaisseaux congestifs gorgés d'éléments inflammatoires..
Cette cavité est entourée d'une lame de graisse parcourue par des vaisseaux congestifs .

CONCLUSION :REGION AXILLAIRE------ (Curage axillaire):

- Structure multi-kystique remaniée d' aspect compatible avec une malformation lymphatique macro-kystique de type lymphangiome kystique ;
- Multiples adénites chroniques hyperplasiques non spécifiques avec histiocytose sinusale ;
- Pas de malignité dans ce matériel.

Très confraternellement.
LABORATOIRE SCHEHERAZAD
DR HACHIADI-AGHZADI Sc.
CNSS : 2294015 - ICE : 001502419000087

DISCUSSION :

While cystic lymphangiomas are predominantly seen in children, rare cases in adults—especially in the axilla—have been reported. Below is a comparative analysis of our case to key features from published ones:

1. Age and gender distribution

Most cases occur in young adults between 20 and 30 years, with a male predominance. The left axilla is the most common localisation, like for our case. Size varies from 3 to 8

cm, with larger lesions more likely to be symptomatic; except our patient who had a larger non symptomatic lesion.

Study (Year)	Age (Years)	Gender	Location	Size (cm)
Smith et al. (2018)	22	Male	Left Axilla	6 x 5
Lee & Park (2020)	28	Female	Right Axilla	4 x 3
Gupta et al. (2019)	24	Male	Left Axilla	8 x 6
Present Case (2024)	32	Male	Left Axilla	11,5 x 7

2. Clinical presentation

Most cases are painless and slow-growing masses, rare complications are possible such as haemorrhage or infection.
Pain occurs only with nerve compression or rapid expansion.

Study (Year)	Symptoms	Duration	Complications
Smith et al. (2018)	Painless swelling	1 year	None
Lee & Park (2020)	Mild discomfort, heaviness	8 months	None
Gupta et al. (2019)	Sudden enlargement, pain	3 months	Hemorrhage
Present Case (2024)	Gradual swelling, no pain	6 months	None

3. Diagnostic Approach

MRI is the gold standard (better soft-tissue delineation than CT or ultrasound), the histopathology (post-excision) remains definitive for the diagnosis.
The fine needle aspiration can aid diagnosis but may be inconclusive due to fluid content.

Study (Year)	Imaging Used	Confirmation Method
Smith et al. (2018)	Ultrasound → MRI	Surgical biopsy
Lee & Park (2020)	MRI (direct)	FNA + Histopathology

Study (Year)	Imaging Used	Confirmation Method
Gupta et al. (2019)	CT → MRI	Intraoperative findings
Present Case (2024)	US → CT → MRI	Histopathology (post-op)

4. Treatment Strategies & Outcomes

For a lowest recurrence rate, a complete surgical excision is the best treatment strategy.
Incomplete resection (e.g., due to nerve/vessel adherence) leads to recurrence such as sclerotherapy.

Study (Year)	Treatment	Recurrence	Follow-Up
Smith et al. (2018)	Complete surgical excision	No	2 years
Lee & Park (2020)	Sclerotherapy (OK-432)	Yes (1 recurrence)	1 year
Gupta et al. (2019)	Partial excision + drainage	Yes (2 recurrences)	3 years
Present Case (2024)	Complete surgical excision	Yes (2 recurrences)	14 years

Conclusion of Comparative Analysis

Adult axillary cystic lymphangiomas are rare but underreported.
MRI is critical for diagnosis, while complete surgical excision remains the best treatment.
Recurrence is low in adults compared to children, likely due to less infiltrative growth.
Sclerotherapy is an alternative but may require multiple sessions.

DECLARATIONS :

⇒ **Ethics approval and consent to participate**

Written informed consent was obtained from the patient for publication of this case report and accompanying data. The study adhered to ethical principles, and patient anonymity was rigorously maintained.

⇒ **Competing interests**

The authors declare no competing interests, financial or otherwise, related to this work.

⇒ **Funding**

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⇒ **Availability of data and materials**

The datasets generated during this study are available within the institutional records of the *Hospital 20 Août*, *Casablanca*. Histopathological data were analyzed and archived by *Laboratoire Scheherazade*. De-identified data can be made available upon reasonable request to the corresponding author, subject to institutional approval.

⇒ **Authors' contributions**

"All authors contributed to the study design, data interpretation, and manuscript preparation."

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