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# Uncommon Presentation of Adrenal Cystic Lymphangioma: A Case Report and Review of literature

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***Authors' contributions***

*This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.*

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***Case Report***

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## ABSTRACT

Cystic lymphangioma of the adrenal gland is a rare benign tumor, typically discovered incidentally due to its asymptomatic nature. Imaging techniques such as ultrasound and CT scans play a critical role in suggesting the diagnosis, but definitive confirmation is achieved through histopathological analysis of the surgical specimen. The preferred treatment is surgical excision, with complete resection offering an excellent prognosis. However, incomplete removal may lead to recurrence, underscoring the importance of regular postoperative monitoring. We present a case of a 55 year old man diagnosed with cystic lymphangioma, successfully treated through surgical resection, with plans for ongoing follow-up.

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## 1. INTRODUCTION

Cystic lymphangioma is a rare benign malformative tumor of the lymphatic vessels with various locations, encountered in both adults and children. The lymphangioma was first described by Redenbacher [1] in 1828, but its connection to the lymphatic system was established half a century later by Koester [2]. The exact origin of cystic lymphangiomas is still debated, although the congenital malformation theory is the most widely accepted; however, the congenital malformative theory is currently the most accepted.

Typically, the lesion is singular. When the involvement is multifocal, it is referred to as lymphangiomatosis. Cystic lymphangiomas can be found in any anatomical site with lymphatic circulation, but they most commonly occur in the cervical and axillary regions.

Adrenal cystic lymphangiomas have been recognized as rare benign tumors since the late 17th century. Greiselius made the first description during an autopsy for massive cystic hemorrhage. The largest series was published by Abeshouse et al. [3] in 1959, which included 155 cases, 57% of which were discovered during autopsies [4]. Adrenal cystic lesions are rare, with an incidence of approximately 0.06% in the general population. Adrenal cysts are classified as endothelial cysts, pseudocysts, epithelial cysts, and parasitic cysts. Endothelial cysts (20% to 32% of all adrenal cysts) are further subdivided into lymphangiomatous and angiomatous subgroups [5].

Adrenal cystic lymphangiomas are often asymptomatic and are difficult to diagnose preoperatively. Ultrasound and CT scans play a significant role in the exploration of these tumors. Diagnosis relies on histology. Surgical exploration is often indicated in cases of diagnostic uncertainty. The treatment of choice is surgical.

We report a case of an adrenal cystic lymphangioma with a review of the literature.

## 2. CASE PRESENTATION

A 55-year-old patient, with no medical history, was admitted for the management of an abdominal mass. Blood pressure was at 150/90 mmHg. No fever.

The abdominal examination revealed a mobile, palpable, painless mass in the right hypochondrium with a cystic appearance and well-defined edges. The rest of the physical examination was unremarkable.

Abdominal ultrasound showed a large multicystic mass with a thickened wall and a honeycomb appearance, suggesting a hydatid cyst of the liver, classified as stage III according to GHARBI's classification.

A CT scan of the abdomen and pelvis revealed a large cystic mass measuring 180 x 140 mm, displacing the right hemidiaphragm and pushing the right kidney downward, consistent with either a hydatid cyst of the liver or a cystic liver tumor (Fig. 1).

Abdominal MRI confirmed the diagnosis of a multicystic mass of the right adrenal gland (Fig. 2).

The patient underwent surgery using a right subcostal approach with complete excision of the right adrenal cystic mass (Figs. 3 and 4).

The pathological report favored a cystic lymphangioma of the adrenal gland.

Postoperative recovery was uncomplicated, with good clinical and radiological progress. Two years later, there was no sign of recurrence.

## 3. DISCUSSION

Cystic lymphangiomas are rare benign tumors, with an incidence estimated at 1 in 100,000 hospitalizations. [6]. They typically occur near lymphatic regions, such as the cervical and axillary areas (95%) [7]. The remaining 5% are found in the abdominal, mediastinal, or thoracic cavities [7]. Cystic adrenal lymphangiomas account for 45% of benign cystic adrenal tumors according to Ghandur-Mnayney [8]. Adrenal cystic lesions are rare, with an incidence of about 0.06% in the general population [9]. The onset of symptoms usually occurs between the ages of 30 and 50, with peak incidence in the fourth decade of life [10].

Regarding etiology and pathogenesis, a congenital origin is currently considered the most likely by most authors. Cystic adrenal lymphangiomas are generally asymptomatic and

discovered incidentally, as in the case of our patient. Observed functional signs are non-specific.

Computed tomography (CT) is an excellent initial diagnostic tool in adults. It confirms the mass syndrome, localization, size, and relationships with neighboring organs. It has the advantage of differentiating between fluid-filled and solid tumors. There is no enhancement after contrast injection. On CT, the cystic adrenal lymphangioma is identified as a homogeneous hypodense lesion, non-enhancing, with smooth borders and a thin wall (<3.5 mm) [11].

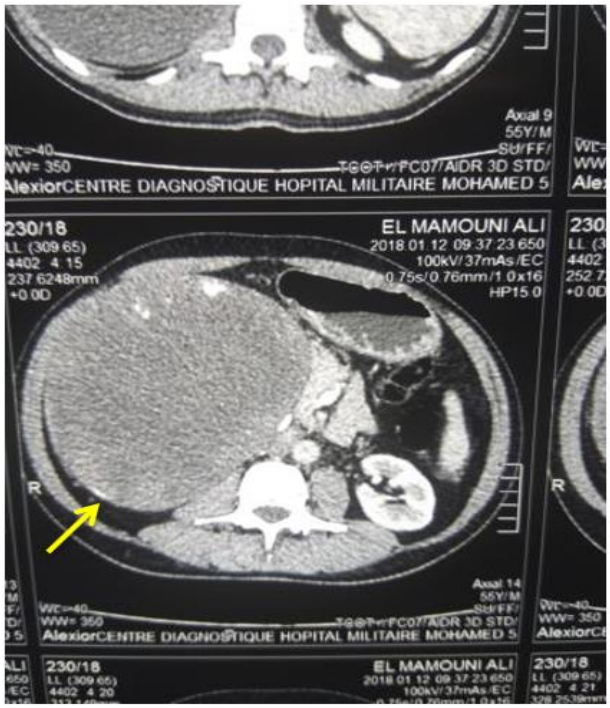


**Fig. 1. Abdominal scan showing a large right adrenal mass displacing the right kidney downward**

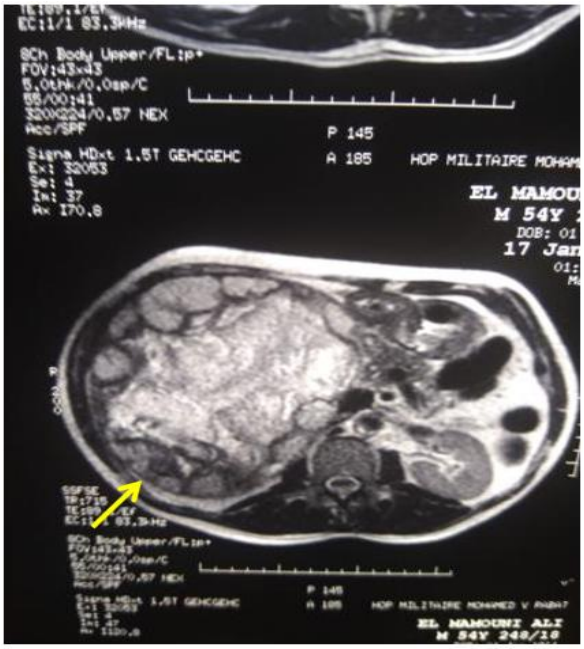


**Fig. 2. Abdominal MRI showing a large multilocular cystic mass in the right adrenal gland**

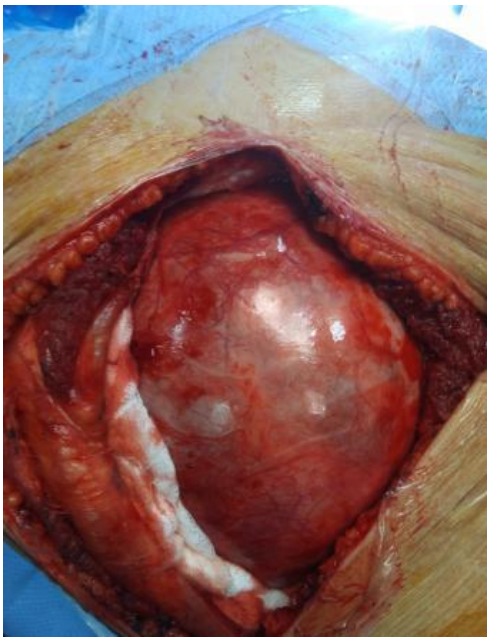

**Fig. 3. Operative view showing the right adrenal cystic mass**

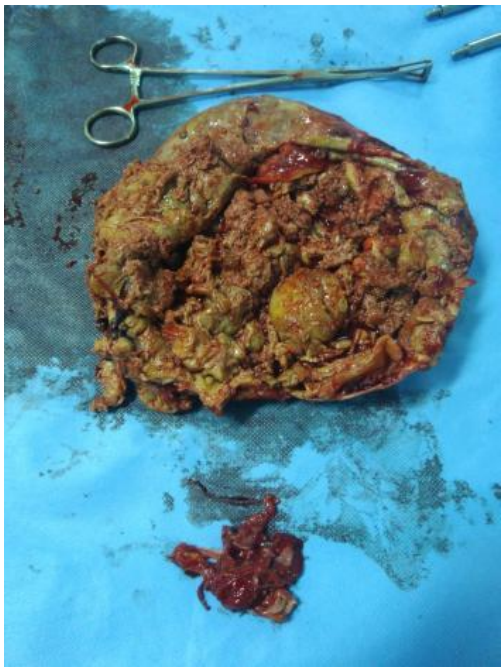

**Fig. 4. Image showing the adrenal mass after opening the the operative specimen**

MRI is particularly useful for better defining the cyst's characteristics and its relationship with surrounding structures of the cyst's relationships and content. On MRI, cystic adrenal lymphangiomas are generally homogeneous, with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, without internal enhancement. MRI is more sensitive for detecting intracystic hemorrhages and complicated cysts, which show high signal intensity on both T1 and T2-weighted images [11].

Surgical treatment is the preferred approach, consisting of complete resection of the lesion, especially for masses larger than 5 cm. Adrenalectomy can be performed via laparotomy or laparoscopy [12]. For our patient, who had an

18 cm adrenal mass, adrenalectomy was performed via laparotomy.

Complications can occur, including intrakystic hemorrhage, infection, and cyst rupture, although malignant transformation is rare.

The prognosis for abdominal cystic lymphangiomas is excellent when resection is complete. Recurrence is the main complication if the resection is partial. Some authors have reported a recurrence rate ranging from 10 to 15% (9.5% in the STEYAERT series) in cases of incomplete cyst resection, which necessitates regular ultrasound monitoring for these patients. In our case, with a two-year follow-up, there has been no recurrence.

## 4. CONCLUSION

Cystic lymphangioma of the adrenal gland is a rare benign tumor. These tumors are often asymptomatic and are usually discovered incidentally during imaging for unrelated conditions. Diagnosis can be suggested by imaging, particularly ultrasound and computed tomography (CT). Definitive diagnosis is achieved through pathological examination of the surgical specimen.

Surgical removal, with complete excision of the lesion, remains the treatment of choice. With the advent of laparoscopy and mastery of its techniques, resection of this cyst using this method may be more beneficial.

The prognosis is excellent; however, recurrences can occur in cases of incomplete cyst resection, necessitating regular monitoring of these patients.

## DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of manuscripts.

## CONSENT

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

## ETHICAL APPROVAL

As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

## REFERENCES

1. Miloundjaa J, Manfoumbi ngomaa AB, Mba Ellab R, Nguema B, dzangc E, N'zoubaa Au Gabon L. Lymphangiomes kystiques cervicofaciaux de l'enfant. Annales d'otorhinolaryngologie et chirurgie cervico-faciale. 2007;277-84.
2. Lingemean RE. Cystic hygroma of the parotid region. Laryngoscope. 1960;70: 983.
3. Steyaert H, Guitard J, Moscovici J, Jurcic M, Vaysse P, Juskiewenski S. Abdominal cystic Lymphangioma in children: Benign lesions that can have a proliferative course journal of pediatric surgery. 1996;31(5): 677-680.
4. Abeshouse GA, Goldstein RB, Abesluous BS. Adrenal cyst : review of the literature and report of three cases. J Urol. 1981: 711-9.
5. Erickson LA, Lloyd RV, Hartman R, Thompson G. Cystic adrenal neoplasms. Cancer. 2004; 101:1537-44.
6. Feldberg MA, Hendriks AV, Van Leeuwen MS,Witkamp TD, Obertop H. Retroperitoneal cystic lymphangiom-asection imaging in two cases, and review of the literature. Clin Imaging 1990;14: 26-30.
7. Khoda J, Hertzanu Y, Sebbag G, Lantsberg L, Barky Y. Adrenal cysts: diagnosis and therapeutic approach. Int Surg. 1993;(78):239-242.
8. Ghandur 1 m, SlimM, Mvakassa K. Adrenal cysts: pathogenis and histological identification with a report of 6 cases. J Urol. 1979;122:87-91.
9. MilstonMS, Minielly JA, Van Schoyck P, Forrest JB. Abdominal pain secondary to a lyphangiomatous cyst of the adrenal: case report and review of literature, J akla estate Med Assoc. 1994;87:225-7.
10. Garcia M, Louis 4th LB, Vernon S. Cystic adrenal lymphangioma. Arch Pathol Lab Med. 2004 ;128:713-4
11. Guo YK, Yang ZG, Li Y. et al. Uncommon adrenal masses: CT and MRI features with histopathologic correlation. European Journal of Radiology. 2007;(62): 359-370.

12. Kasperlik-Załuska AA, Otto M, Cichocki A. et al.1, 161 patients with adrenal incidentalomas: indications for surgery. Langenbeck's Archives of Surgery. 2008; (393):121-126.



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