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Case Study

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ADRENAL CYSTIC LYMPHANGIOMA: A REPORT OF TWO CASES AND LITERATURE REVIEW

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ABSTRACT

Adrenal cystic lymphangioma (ACL) is an exceptionally rare benign tumor resulting from a developmental malformation of lymphatic vessels within the adrenal gland. We report two cases managed in our department, highlighting the clinical, radiological, histopathological, and therapeutic features of this unusual entity. Diagnosis relies on imaging and is confirmed by histopathological examination. Complete surgical excision remains the treatment of choice, ensuring a favorable outcome and excellent prognosis.

KEYWORDS: Adrenal gland, cystic lymphangioma, benign tumor, imaging, adrenalectomy, retroperitoneum.

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INTRODUCTION

Adrenal cystic lymphangioma (ACL) is a rare benign lesion, representing less than 1% of all adrenal cystic masses.[1,2] It arises from a congenital or acquired malformation of lymphatic channels within the adrenal gland, leading to the formation of clear, multiloculated cystic cavities. This condition most frequently affects adult women and is usually discovered incidentally during imaging studies performed for unrelated reasons.[3,4]

Clinically, most ACLs are non-functional and asymptomatic. When symptoms are present, they are generally related to mass effect—such as abdominal discomfort, pain, or a palpable lump.[5] Diagnosis is primarily based on cross-sectional imaging (CT, MRI), and confirmation relies on histopathological analysis. Surgical removal ensures complete recovery and prevents complications.

CASE REPORTS

Case 1

A 52-year-old woman with a medical history of cervical and bronchial carcinoma was found to have a left adrenal mass incidentally during an oncologic evaluation. She was asymptomatic, and hormonal assessment was within normal limits.

Abdominal CT revealed a well-defined, multiloculated cystic lesion measuring 6 cm, with no contrast enhancement or local invasion. MRI confirmed a thin-walled, hypointense T1 and hyperintense T2 lesion.

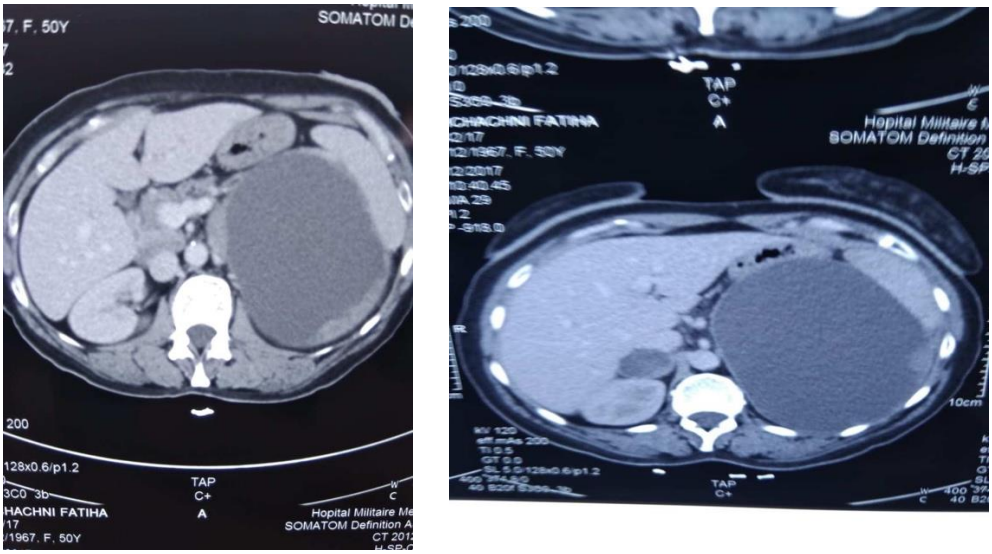

Figure 1 and 2: Abdominal CT scan showing a large left adrenal cystic mass displacing the left kidney and the stomach.

The patient underwent open left adrenalectomy. Macroscopic examination revealed a multiloculated cyst filled with clear fluid. Histopathological analysis confirmed adrenal cystic lymphangioma, with endothelial positivity for D2-40, CD31, and CD34. The postoperative course was uneventful, and no recurrence was noted at six months.

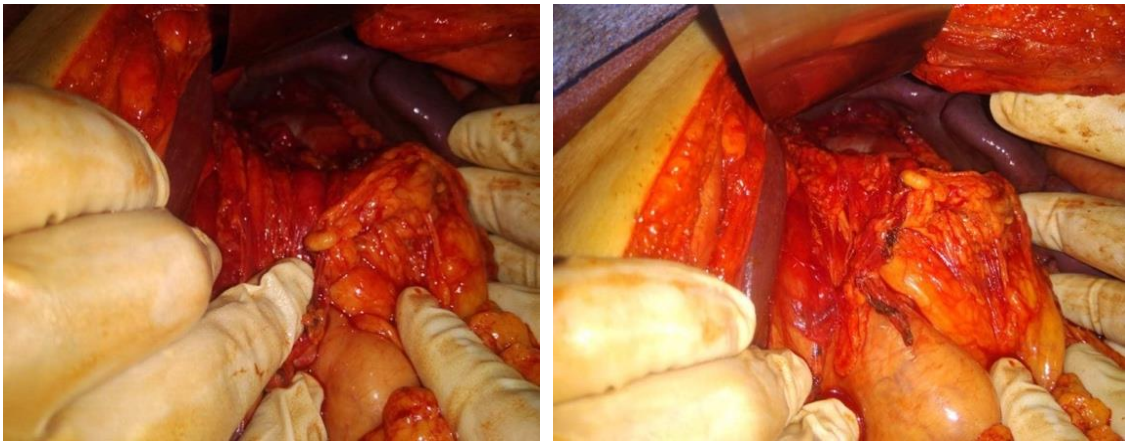

Figure 3 and 4: Intraoperative exposure of the left adrenal mass.

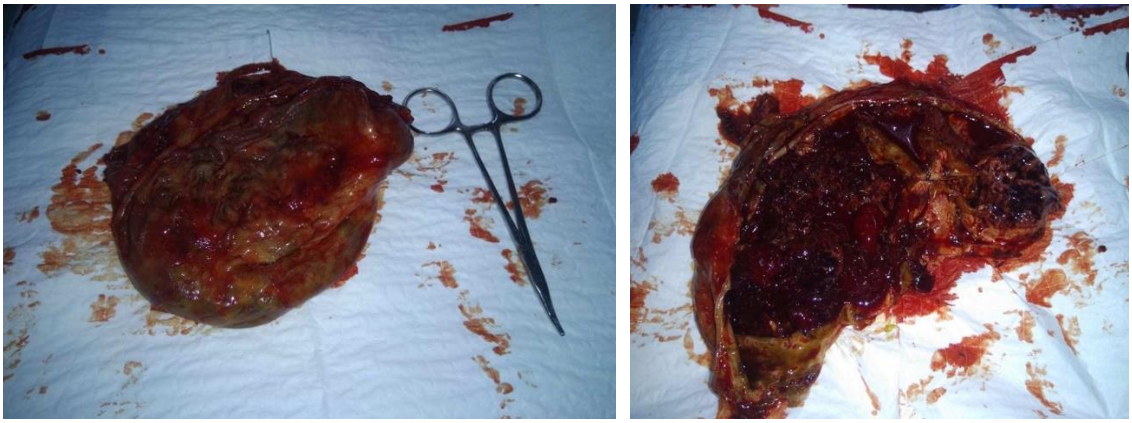

Figure 5: (A) Resected specimen of the left adrenal mass; (B) Opened surgical specimen showing the cystic cavity.

Case 2

A 47-year-old woman presented with intermittent diffuse abdominal pain. Ultrasound examination revealed a right adrenal cystic mass measuring 5.5 cm. MRI showed a thin-walled, multiloculated cystic lesion, hypointense on T1 and hyperintense on T2 sequences, without invasion of adjacent structures. Hormonal evaluation was normal.

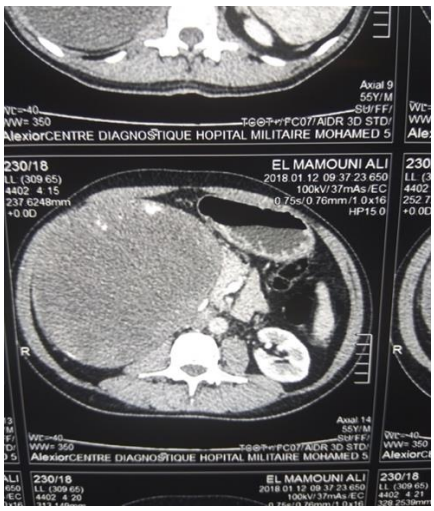

Figure 6: Abdominal CT scan showing a large right adrenal cystic mass displacing the right kidney downward.

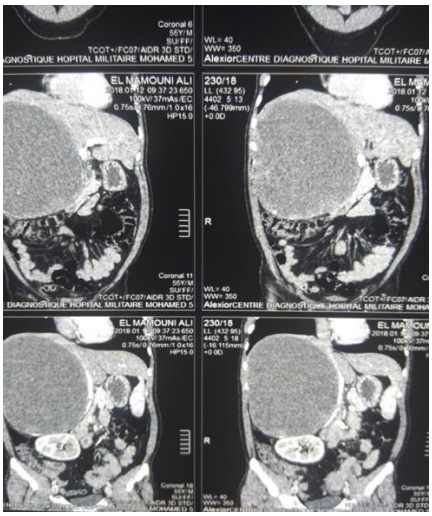

Figure 7: Sagittal CT section showing the right adrenal mass.

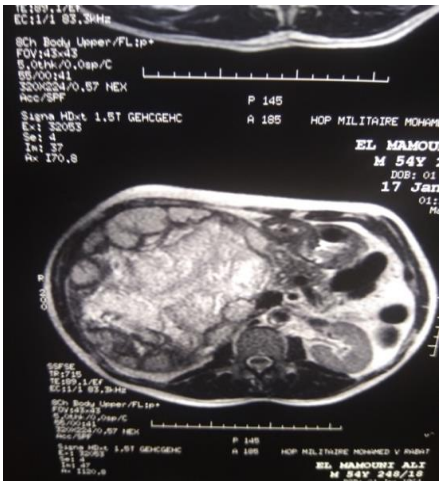

Figure 8: Abdominal MRI showing a large multiloculated right adrenal cystic mass.

An open right adrenalectomy was performed. Histopathological examination confirmed adrenal cystic lymphangioma. The postoperative outcome was uneventful, and no recurrence was observed at one-year follow-up.

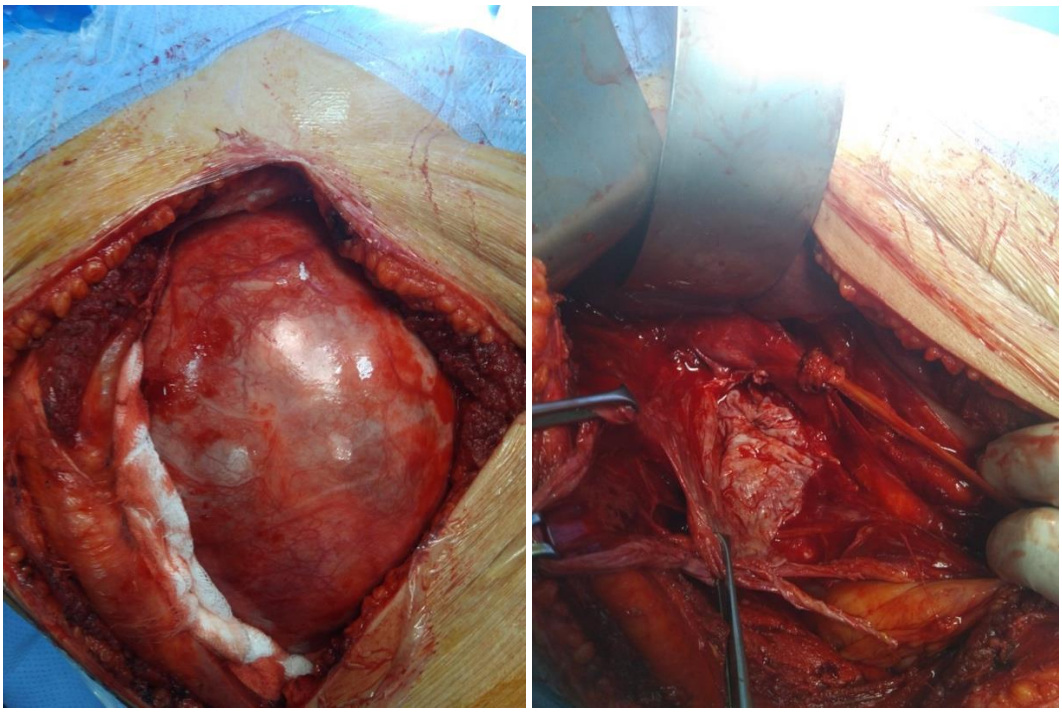

Figure 9: (A) Intraoperative view showing the right adrenal cystic mass; (B) Operative field after resection of the specimen.

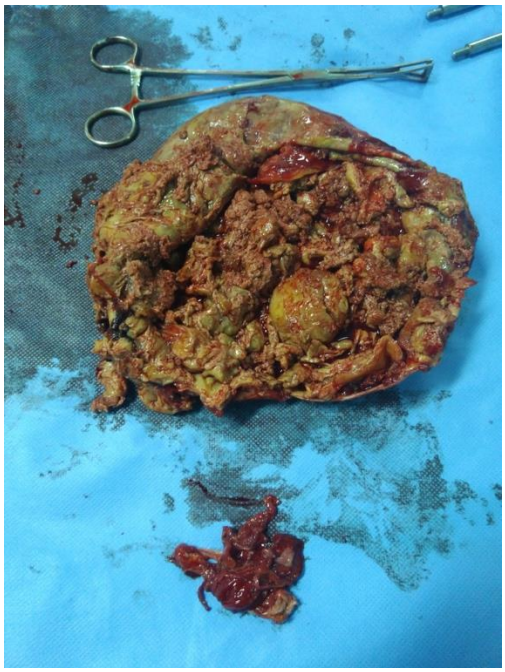

Figure 10: Image showing the adrenal mass after opening of the surgical specimen.

These two cases illustrate the typical presentation and favorable prognosis of this rare adrenal lesion following complete surgical excision.

DISCUSSION

Adrenal cystic lymphangioma is a rare benign tumor, most often discovered incidentally in adults. It arises from malformations of the lymphatic vessels within

the adrenal gland, leading to multiloculated cysts filled with clear fluid. Histologically, the cyst wall is lined by lymphatic endothelium expressing markers such as D2-40 (podoplanin), CD31, and CD34.[1,6-8]

The pathogenesis of adrenal lymphangiomas remains controversial. Some authors suggest a congenital origin, resulting from sequestration of embryonic lymphatic channels, whereas others propose an acquired mechanism secondary to lymphatic obstruction due to inflammation or trauma.[9] Recent reports suggest a potential continuum between congenital malformations and adult-onset lesions, highlighting the need for further molecular studies.

Epidemiologically, most cases occur in women aged 30–60 years, with around two-thirds discovered incidentally during imaging studies.[4,10] Symptomatic patients may present with non-specific abdominal pain, a palpable mass, or signs of compression. Rare complications include intracystic hemorrhage, infection, or hypertension.[11]

Recent advances in imaging have improved lesion characterization. High-resolution MRI and dual-energy CT allow better distinction between adrenal cystic lymphangioma and cystic pheochromocytoma or cystic metastases. Diffusion-weighted imaging and 3D reconstruction can aid in preoperative planning and surgical approach.[19] Nevertheless, histopathology remains the definitive method for diagnosis.[12-14]

The differential diagnosis remains broad, including epithelial cysts, pseudocysts, parasitic cysts, and adrenal tumors such as pheochromocytomas, adrenocortical carcinomas, or metastases.[15] Immunohistochemistry is essential for confirming the lymphatic origin of the lesion.

Surgical excision remains the treatment of choice. Complete removal—either partial or total adrenalectomy—prevents recurrence and provides definitive diagnosis. Laparoscopic and robotic adrenalectomy have become standard for benign adrenal cystic lesions, demonstrating safety and low recurrence rates, even for lesions up to 8–10 cm, provided careful dissection and preoperative imaging planning.[20] Open surgery is reserved for large, invasive lesions, while robotic-assisted adrenalectomy is increasingly reported in specialized centers.[16-17]

The prognosis after complete resection is excellent, with negligible recurrence and extremely rare malignant transformation.[2,18] Regular follow-up is advised in cases of incomplete excision. A recent case series confirms that minimally invasive approaches are safe and effective even for large lesions, supporting their growing adoption.[21]

Clinical implications and perspectives

Given its rarity, adrenal cystic lymphangioma should always be included in the differential diagnosis of adrenal incidentalomas, particularly in women.[19] The increasing adoption of minimally invasive and image-guided approaches, including laparoscopic and robotic adrenalectomy, may help avoid unnecessary adrenalectomies in purely cystic, nonfunctional lesions.[20,21] Future research should focus on molecular and genetic characterization of these lesions, as well as the long-term outcomes of minimally invasive techniques.[19,20] Establishing multicenter registries could improve understanding of prevalence, growth risk factors, and the development of standardized follow-up strategies.[21]

CONCLUSION

Adrenal cystic lymphangioma is a rare benign lesion, most often discovered incidentally. Imaging—especially CT and MRI—plays a central role in diagnosis, but histological confirmation is mandatory. Complete surgical excision, preferably via a laparoscopic approach, provides definitive treatment and an excellent prognosis. This entity should be considered in the differential diagnosis of any well-defined cystic adrenal lesion to ensure appropriate management and avoid unnecessary radical procedures.

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