Indian J Surg (December 2015) 77(Suppl 3):S1334–S1342
DOI 10.1007/s12262-015-1206-y

REVIEW ARTICLE

Adrenal Gland Lymphangiomas

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Received: 1 October 2014 / Accepted: 5 January 2015 / Published online: 23 January 2015

Abstract Lymphangiomas of the adrenal glands (ALs) are benign vascular lesions. Approximately, 53 cases have been reported in the literature. The current study reviews and analyzes the clinical and pathologic features of all reported ALs and additionally illustrates a typical case of adrenal lymphangioma (AL). In order to perform the review analysis, a search of the international literature for ALs in adults was conducted. Thirty-eight related articles were found. Clinical and pathological information were obtained for all the reported cases and a database was created. ALs were detected more frequently in women than men. The mean age of occurrence was 39.5 years, while their mean size was 8.86 cm. Fifty-nine percent of ALs were right-sided. Size and localization were responsible for the presenting symptoms, though 30.4 % were asymptomatic. Diagnosis was made postoperatively in all cases by histological results. ALs are rare and benign lesions. They usually present as an incidental finding after abdominal imaging. The diagnosis is made after the surgical removal by histological and immunohistochemical examinations.

Keywords Lymphangioma · Adrenal tumor · Incidentaloma · Adrenal cyst

Abbreviations

AL	Adrenal lymphangioma
F	Female
M	Male
US	Ultrasonography
CT	Computed tomography
MRI	Magnetic resonance imaging

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Introduction

Adrenal cysts are rare lesions. They can be classified as endothelial cysts (45 %), pseudocysts (39 %), epithelial cysts (9 %), and parasitic cysts (7 %) [1]. Endothelial cysts include hemangiomas, hamartomas, and lymphangiomas [1]. Adrenal lymphangiomas (ALs) are rare benign vascular lesions that usually remain asymptomatic throughout life [2]. They are usually unilateral, variable in size, with smooth borders, and pure cystic internal structure [2]. To date, only a few cases have been reported in the literature.

The aim of this study is to review the literature in order to determine clinical features, pathological findings, and management of these lesions. Additionally, we present a patient with a typical AL in the right adrenal gland.

Materials and Methods

An electronic search of PubMed/Medline database using the medical subjected headings (MeSH) was performed, using combinations of the following keywords/phrases: adrenal cystic tumors; adrenal tumor; incidentalomas; adrenal cyst; adrenal lymphangiomas. Original research papers, case reports, and review articles were included.

Searching the international literature since 1965, when Lynn [3] reported one of the first known AL, to 2014, 38 articles describing 53 cases of ALs in adults (>16 years) were found [1–39]. The majority of these articles were single case reports. It is noteworthy that 15 cases were reported in two

Table 1 Adrenal lymphangiomas (ALs)

Case	Author/year	Sex/age	Site	Size	Symptoms & signs	Hormone secreting	Imaging modalities	Treatment	Pathology/immunohistochemistry	Follow-up (months)
1	Lynn [3]/1965	M/50	Left	5.0	Hypertension, headaches, dizziness, nausea, lassitude	No	Urography, angiography	Open adrenalectomy	CL	24
2	Boscaino [5]/1977	F/21	Right	NR	Urinary infection symptoms, Palpable mass	No	Urography, angiography	Open adrenalectomy	CL	NR
3	Georgi [7]/1982	F/48	Right	12.0	Pain in right upper abdomen, hypertension	No	U/S, angiography	Open adrenalectomy	CL	NR
4	Millon [8]/1982	M/45	NR	NR	Hypertension	NR	U/S, angiography	Open adrenalectomy	CL	NR
5	Pfister [10]/1987	F/51	Right	5.0	Lumbar pain	Iodocholesterol	Angiography	Open adrenalectomy	CL	NR
6	Gossot [11]/1987	F/39	Left	10.0	Fever, incidental adrenal mass	No	U/S, CT, angiography	Open adrenalectomy	CL	3
7	Sanromá [12]/1988	F/20	Right	2.5	Epigastric & upper right quadrant pain, nausea, vomiting	No	U/S, CT	Open adrenalectomy	CL	NR
8	Gleeson [13]/1988	F/38	Right	7.0	Right loin pain, weight loss	No	U/S, CT, urography, angiography	Open adrenalectomy	Multiple CL	NR
9	Costantino [14]/1993	M/16	Left	10.0	Epigastric pain	No	U/S, CT, urography	Open adrenalectomy	CL	NR
10	Berthet [15]/1993	M/57	Right	10.0	Hypertension	No	U/S, CT, urography	Open resection	CL	NR
11	Berthet [15]/1993	F/32	Right	30.0	Right hypochondrium mass	No	U/S, CT, angiography	Open adrenalectomy	CL	NR
12	Berthet [15]/1993	F/33	Right	13.0	Incidental adrenal mass haemolymphangioma	No NR	U/S, CT	Open resection	Cystic	
13	Camara [16]/1994	F/25	Right	14.0	Right upper quadrant pain, nausea	No	U/S	Open adrenalectomy	CL	NR
14	Iderne [17]/1995	F/52	Right	12.0	Right upper quadrant pain, nausea, vomiting	Aldosterone	U/S, CT, urography	Open partial adrenalectomy	CL	NR
15	Alapont Pérez [19]/1996	F/34	NR	NR	Lower urinary tract infection, continuous and aggravating pain in left lumbar area	No	U/S, CT, urography	Open adrenalectomy	CL	NR
16	Mortelé [21]/1996	M/58	Bilateral	R:21, L:4	Diffuse abdominal pain, nausea, vomiting (syndrome Gorlin-Goltz)	No	U/S, CT, MRI	Open resection	CL	NR
17	Hoeffel [22]/1999	F/22	Bilateral	R:7.8, L:8.7	Left upper abdominal pain, palpated mass	No	U/S, CT, MRI, urography	Open resection	CLs	NR
18	Longo [23]/2000	F/30	Right	4.0	Right flank discomfort	No	U/S, CT, MRI, urography	Open adrenalectomy	CL	24
19	Trojan [24]/2000	M/40	Right	5.7	Partial small-bowel obstruction	No	U/S, CT	Open resection	CL	NR
20	Yokota [25]/2000	F/53	Left	NR	Residual urine feeling	Catecholamines and cortisol	U/S, CT, MRI, urography, angiography	Open adrenalectomy	CL	NR
21	Touiti [26]/2003	F/38	Right	9.4	Chronic right lumbar pain	Aldosterone	U/S, CT, MRI, urography	Open subtotal adrenalectomy	CL	3
22	Satou [27]/2003	M/46	Left	2.0	Palpitation and siderosis	No	U/S, CT, MRI			24

Table 1 (continued)

Case	Author/year	Sex/age	Site	Size	Symptoms & signs	Hormone secreting	Imaging modalities	Treatment	Pathology/immunohistochemistry	Follow-up (months)
								Laparoscopic adrenalectomy	Multiple CL/LCA, CD20, factor VIII and CD34 IN endothelium	
23	Luncă [28]/2004	F/47	Left	12.0	Incidental adrenal mass	No	U/S	Open resection	CL	NR
24	Robledo-Ogazón [29]/2004	F/21	Right	7.0	Hypertension, frontal headache, nausea, anxiety, irritability, depression	No	U/S, CT	Open adrenalectomy	Multiple CL	NR
25	Garcia [30]/2004	F/22	Left	3.5	Right lower quadrant abdominal pain	NR	U/S, CT, MRI	Open partial adrenalectomy	CL/CD 31	NR
26	Ates [1]/2005	F/26	Right	9.0	Weakness, put weight, lumbago	No	U/S, CT, MRI	Open adrenalectomy	Multiple CL/CD31, CD34, smooth muscle actin	NR
27	Nouira [31]/2007	F/30	Left	NR	Incidental adrenal mass	NR	U/S, CT	Laparoscopic excision	CL	NR
28	Bettaïeb [32]/2007	F/22	Left	35	Left flank pain, nausea, vomiting, constipation	No	U/S, CT	Open subtotal adrenalectomy	CL/CD34, factor VIII	NR
29	Pereira [33]/2007	F/41	Left	3.0	Hypertension	Catecholamines	U/S, CT	Open resection	Multiple CL/CD31, CD34	NR
30	Chien [34]/2008	F/59	Right	6.0	Incidental adrenal mass	Catecholamines	U/S, CT	Open adrenalectomy	CL/D2-40, CD34	16
31	Chien [34]/2008	F/43	Right	5.0	Hypertension, dizziness	No	U/S, CT	Open adrenalectomy	CL/D2-40, CD34	26
32	Chien [34]/2008	M/50	Left	15.0	Left flank soreness	No	U/S, CT	Open adrenalectomy	CL/D2-40, CD34	21
33	Chien [34]/2008	F/53	Right	7.0	Flank pain	No	U/S, CT	Open adrenalectomy	CL/D2-40, CD-34	191
34	Chien [34]/2008	F/31	Right	18.0	Painless abdominal mass	No	U/S, CT	Open adrenalectomy	CL/D2-40, CD-34	1
35	Chien [34]/2008	M/58	Left	5.7	Incidental adrenal mass	No	U/S, CT	Open adrenalectomy	CL/D2-40, CD34	23
36	Ait Ali [35]/2009	F/33	Right	6.5	Right flank pain, inferior edemas	NR	U/S, CT, MRI	Open resection	CL	NR
37	Cutaja [36]/2009	F/60	Left	4.0	Recurrent abdominal pain	NR	U/S, CT, MRI	Open adrenalectomy	CL	NR
38	Ellis [2]/2010	F/51	Right	3.2	Flank pain, hypertension	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40	NR
39	Ellis [2]/2010	M/38	Right	2.0	Morbid obesity, hypertension	NR	U/S, CT, MRI	Partial adrenalectomy	CL/D2-40, CD31	NR
40	Ellis [2]/2010	F/28	Left	13.5	Back pain, increasing adrenal mass	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40, CD31, VIMENTIN	NR
41	Ellis [2]/2010	M/46	Right	4.0	Urethral bleeding	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40, CD34	NR
42	Ellis [2]/2010	F/32	Left	6.5	Abdominal pain, early satiety	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40	NR
43	Ellis [2]/2010	M/46	Right	4.0	Incidental adrenal mass	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40, CD31	NR
44	Ellis [2]/2010	F/42	Right	6.5	Increasing back pain, increasing size of adrenal mass	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40	NR
45	Ellis [2]/2010	F/39	Right	NR	Florid endometriosis, incidental adrenal mass	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40, CD31, CD34, factor VIII	NR
46	Ellis [2]/2010	F/56	Left	2.5	Chest pain	NR	U/S, CT, MRI	Open adrenalectomy	CL/D2-40	NR

Table 1 (continued)

Case	Author/year	Sex/age	Site	Size	Symptoms & signs	Hormone secreting	Imaging modalities	Treatment	Pathology/immunohistochemistry	Follow-up (months)
47	Sallami [38]/2013	F/29	Right	6.0	Lumbar pain	No	Pyelography, U/S, CT	Open adrenalectomy	CL/CD31, CD34	17
48	Liu [39]/2013	F/45	Left	3.0	Incidental adrenal mass	No	U/S, CT	Retroperitoneoscopic adrenalectomy	CL/CD31, CD34	NR

M male, *F* female, *NR* not reported, *CL* cystic lymphangioma, *U/S* ultrasonography, *CT* computed tomography, *MRI* magnetic resonance imaging

articles, one by Ellis et al. [2] (nine cases) and the other by Chien et al. [34] (six cases). Berthet et al. [15] mentioned three cases; however, one of the described patients was previously reported by the same authors [40]. Constantino et al. [14, 41] reported the same patient twice, whereas Bisceglia et al. [37] described a case of lymphangioma like adenomatoid tumor and was excluded from our analysis. In five cases, we were unable to obtain the data and were also excluded from the analysis [4, 6, 9, 18, 20].

The review analysis included 31 articles which describe 48 AL cases. Table 1 shows in details all the reported ALs. Each article was carefully studied and a database was created. This database included 13 characteristics of ALs such as gender, age, presenting symptoms, tumor size and localization, laboratory findings, diagnostic imaging modalities, treatment, malignancy, pathological and immunohistochemical findings, and follow-up surveillance.

Results

Table 2 summarizes all ALs features. Thirty-six female and 12 male patients were reported (F/M: 3/1), and all patients were adults (>16 years) at the time of diagnosis. The most common symptoms were pain (47.9 %) and hypertension (14.6 %). Pain was localized in the back, flanks, right upper quadrant, or was generalized as abdominal pain. Hypertension seems to be unrelated with ALs. It is remarkable that 15 ALs were diagnosed incidentally, as patients were presented with unrelated or no complaints. Laboratory findings in six ALs cases showed hormones over secretion [10, 17, 25, 26, 33, 34]. The detected hormones were catecholamines, aldosterone, cortisol, and iodocholesterol. The majority of ALs (58.7 %) were right-sided. Mortele et al. [21] and Hoeffel et al. [22] reported two cases of bilateral ALs. The mean size of ALs was 8.7 cm (maximum diameter was used to determine the size of the cyst). Imaging modalities have misdiagnosed all the reported ALs. To date, ALs have been characterized mostly as incidentalomas (56.52 %) and rarely as adrenal adenomas [2, 26, 36], pheochromocytomas [2, 29, 33], or adrenal cysts [2, 5, 7]. Moreover, five ALs have been characterized as non-adrenal lesions [2, 14, 15, 17, 34]. Accurate diagnosis for all ALs was made postoperatively by pathology reports. Surgical excision was considered as the appropriate treatment in all reported ALs. Open transabdominal adrenalectomy was the procedure of choice. Satou et al. [27] and Nouira et al. [31] performed laparoscopic adrenalectomy, while Liu et al. [39] has performed recently a retroperitoneoscopic procedure.

Macroscopically, the lesions were mostly unilocal and described as cystic, while two lesions were characterized as cystic haemolymphangiomas [11, 15]. Potency of malignant transformation was described in two cases [2, 27] with atypical lymphocytes and marked degenerative changes. Data

Table 2 Characteristics of adrenal lymphangiomas (ALs)

		N of patients	Percentage (%)	Mean	Range
Gender	Male	12	25.0		
	Female	36	75.0		
Age (years)				39.5	16–60
Symptoms and signs	Pain	23	47.9		
	Hypertension	7	14.6		
	Palpable mass	2	4.1		
	Partial small-bowel obstruction	1	2.1		
	Unrelated or no complaints	15	31.3		
Tumor size (diameter in cm)				8.86	2.0–35.0
Location	Right	27	58.7		
	Left	17	36.9		
	Bilateral	2	4.4		
Type of cyst	Unilocular	25	53.2		
	Multilocular	22	46.8		
Treatment	Open resection	8	16.6		
	Open partial adrenalectomy	5	10.4		
	Open adrenalectomy	32	66.7		
	Laparoscopic excision	1	2.1		
	Laparoscopic adrenalectomy	1	2.1		
	Retroperitoneoscopic adrenalectomy	1	2.1		
Malignancy	No	46	95.9		
	Possible	2	4.1		
Follow up (months)				32.4	1–191

concerning the immunohistochemistry were available in 20 patients. Immunostains included CD31, CD34, factor VIII, and D2-40. Regarding the two ALs associated with lymphocyte proliferation, the one was found positive to CD20 and LCA stains [27], while the other had positive reactivity for CD20, CD79a, CD43, and B cell lymphoma-2 stains [2]. Overall, for both cases, the cells were regarded as atypical, but the lymphoid process could not be further characterized because of the diffused degenerative cytologic changes [2, 27]. In all patients having available follow-up surveillance data, the postoperative period was uneventful. Lymphoproliferative process was not detected in the two ALs associated with atypical cells [2, 27].

Case Presentation (Illustrative Case of AL)

We report a 39-year-old woman with a previous history of head melanoma excision. Staging abdominal computed tomography (CT) revealed a lesion of 9.0×6.5×4.0 cm located on the right adrenal gland. It appeared to be of water density with loculi lacking of enhancement (Fig. 1a, b). The multicystic lesion seemed to displace the right liver lobe as shown in magnetic resonance imaging (MRI) (Fig. 1c, d). Serum and urine analyses were not indicative of a functioning adrenal tumor. The patient's medical history of melanoma and the size of the lesion (>6 cm) were considered as risk factors for malignancy and therefore the surgical removal of right adrenal gland was considered appropriate. The patient was scheduled for open adrenalectomy via transabdominal approach. The duration of operation was 85 min, and the cystic lesion was excised en-block with the right adrenal gland, while macroscopically no infiltration of the surrounding tissues was observed (Fig. 2). The postoperative period was uneventful, and the patient was discharged on the fifth postoperative day.

Histological examination revealed a multi-cystic lesion with flat endothelial cells adjacent to the normal-appearing adrenal cortex (Fig. 3). Immunohistochemical staining results showed that the cells were positive for CD31, CD34, and D2-40 and negative for cytokeratines (Fig. 3). Based on histopathological results, the diagnosis of cystic AL was made.

The patient's follow up included CT on the sixth postoperative month and afterwards annually. Two years after surgery, the patient remains free of symptoms.

Discussion

Lymphangiomas are benign malformations of vessels, most frequently discovered in childhood [1]. The cause of

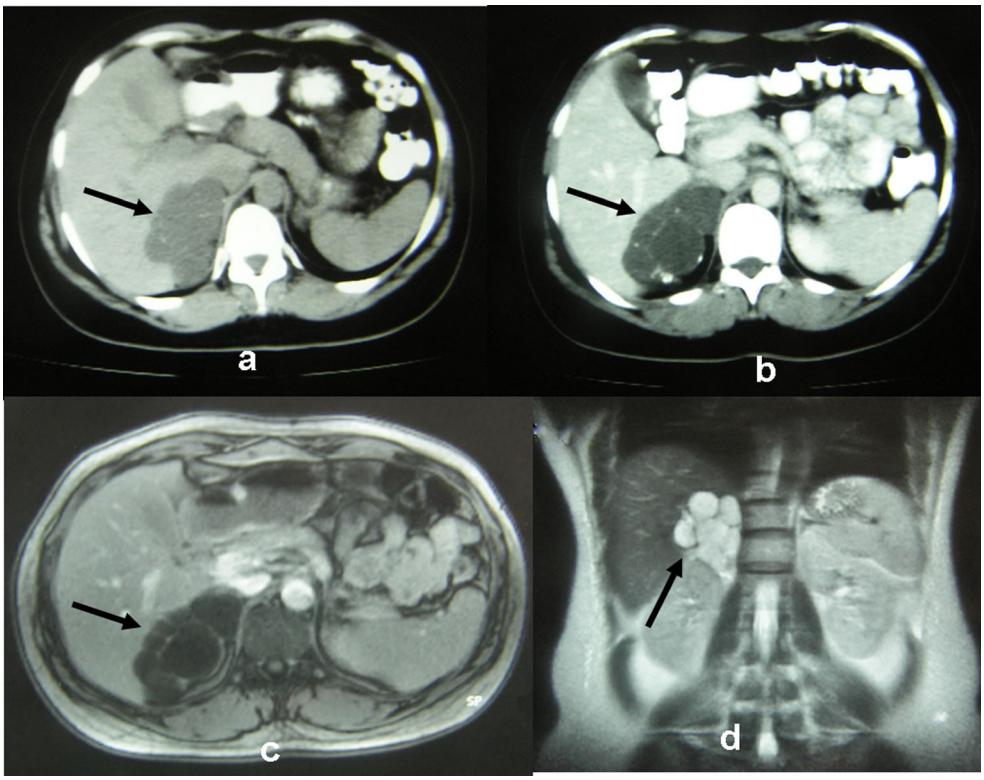

Fig. 1 **a**, **b** CT images are showing a lesion of 9.0×6.5×4.0 cm located on the right adrenal gland. **c**, **d** MRI images are showing a multi-cystic lesion on the right adrenal gland. All *arrows* are indicating the cystic lesion

lymphangiomas has not been established. They are generally regarded as congenital malformations in which obstruction or agenesis of lymphatic tissue results in lymphangiectasia due to lack of normal communication of the lymphatic system [42]. Other authors believe that these tumors arise from continuous growth of ectopic or malformed lymphatic tissue, or represent a hyperplastic reaction to inflammation, or a lymphatic hamartoma [22]. They are usually located in the neck, axillary region, and mediastinum (95 %) [43], whereas the remaining 5 % appear in other parts of the body such as lungs, pleura, pericardium, gastrointestinal system, pancreas, liver, gallbladder, mesentery kidney, and adrenal glands [44]. ALs are uncommon lesions, and their pathogenesis is the same as in the other lymphangiomas [1].

Clinical presentation is not typical for ALs, whereas one third of the cases were incidentally detected. They are usually presented in young women, but there is no formal explanation for that female predominance. Laboratory findings are also nonspecific and usually not helpful as a diagnostic tool. In six cases, hormone hypersecretion was reported [10, 17, 25, 26, 33, 34]. Since ALs are not hormone-productive lesions, it is remarkable that hypersecretion findings were presented in these cases. Nevertheless, pathology reports were not indicative of functional adrenal tumors.

The widespread use of diagnostic images modalities such as CT and MRI has facilitated the diagnosis of adrenal neoplasms including ALs. However, accurate diagnosis was not made preoperatively and they were characterized as

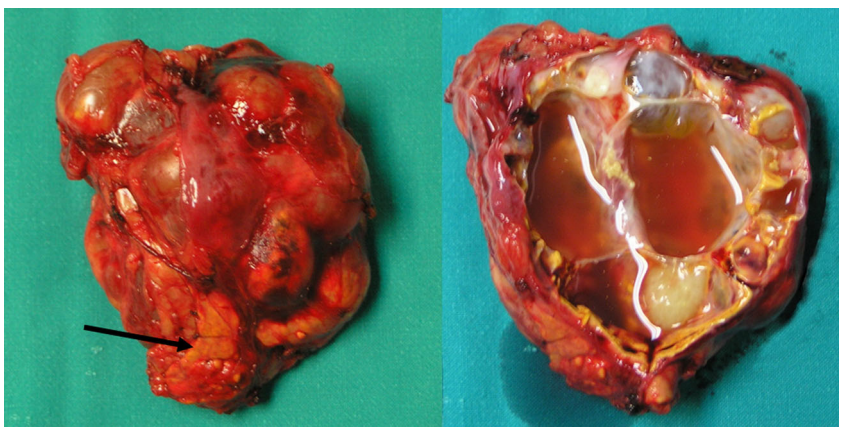

Fig. 2 Surgical specimen, cystic tumor of the right adrenal gland. *Arrow* is indicating the adrenal tissue

incidentalomas. Ultrasonography, CT, and MRI are the modalities used in the evaluation of adrenal cystic lesions [45]. Ultrasound often demonstrates a well-marginated lesion. If calcification or internal debris is present, ultrasound may show more complicated appearance with acoustic shadowing and internal echoes [23]. On CT, adrenal cystic lesions are characterized by lack of enhancement with intravenous contrast. The cystic fluid measures water density or higher if hemorrhagic or protein component is presented [46]. On MRI, they are low in signal intensity on T1-weighted images and high on T2-weighted images [23, 47]. Pheochromocytoma, neuroblastoma, adenomatoid tumor, schwannoma, rare cases of metastasis in adrenal glands (i.e., breast cancer), and rare cases of primary adrenal gland malignant lymphoma can present as cystic lesions and should be included into the differential diagnosis of ALs [45]. Cyst's internal texture, wall thickness, calcification pattern, absence of a true contrast enhancement, positive clinical/laboratory findings, and patient history should all be considered in making the diagnosis [44].

Clinical management of ALs can be aided by the imaging findings. Lesion diameter is the main indication for AL removal. However, there is no consensus in the literature regarding the optimal size of the tumor in need of surgical intervention [48]. Small asymptomatic ALs with clear fluid can simply be observed, whereas large symptomatic lesions should be excised [32]. Some authors recommend aspiration of the contents of adrenal cysts both for diagnosis and management instead of surgical excision, if the suspicion of malignancy is low, or the lesion is non-functional and asymptomatic [1]. This method, though, is characterized by the high re-accumulation of the cyst fluid and probably the dispersion of malignant cells in the peritoneal cavity, while its ability to determine the histology of cyst is limited. However, it may be the best and only option in a patient who is at high surgical risk [23].

Surgery should be the appropriate treatment for large ALs. For many years, open transabdominal adrenalectomy had been applied for the surgical removal of these neoplasms. This procedure requires a relatively large incision due to the deep retroperitoneal location of the adrenal glands [48]. Laparoscopic excision, though, seems to be particularly suitable as most adrenal tumors are small and pathologically benign [49]. Laparoscopic surgery is feasible for all benign adrenal tumors even in those with large size (>8 cm) [50]. The benefits of laparoscopic adrenalectomy are reduced blood loss, lower requirements for analgesia, shorter hospital stay, and quicker recovery. In view of this, it is suggested that minimally invasive adrenalectomy might lead to a better clinical outcome, as compared to the open surgery [46, 48].

At pathological studies, ALs have a recognizable endothelial lining. Multiloculated cystic and endothelial lined cavities with lymphocyte aggregation are focally observed on the cyst's wall [27]. The content of the cyst is mainly proteinaceous [27]. Immunohistochemical studies are also used to establish the diagnosis. The endothelial lining stains positive for factor VIII-related antigen, CD31 and CD34, while it demonstrates a lack of staining for cytokeratin. These findings confirm the lymphatic rather than the mesothelial nature of the

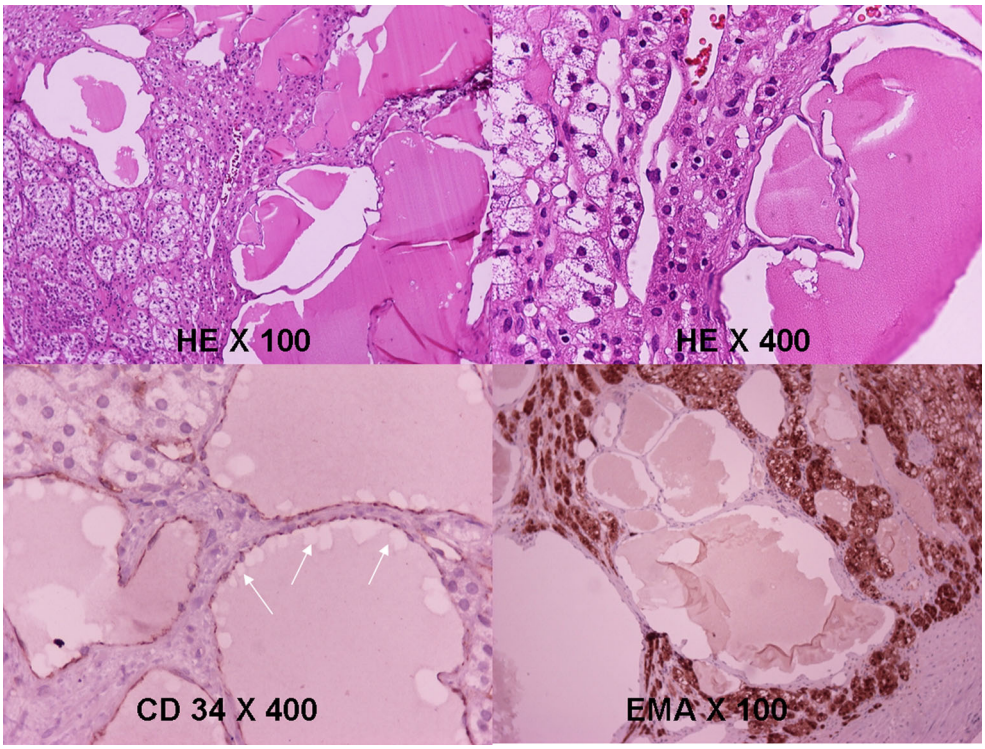

Fig. 3 The mass consists of dilated eosinophilic material-containing spaces. A layer of cells cover the inner surface (HE ×100, HE ×400). Cells stain positive for CD 34 and negative for EMA. *HE* hematoxylin–eosin, *EMA* epithelial membrane antigen

lining [1]. ALs are also stained positive by the relatively novel D2-40. This is a monoclonal antibody directed against the human podoplanin, a transmembrane mucoprotein expressed by lymphatic endothelial cells among others [51]. Unlike other vascular markers, such as CD31 and CD34, that label both blood vessel and lymphatic endothelium, D2-40 immunoreactivity is restricted to lymphatic endothelium, thus, it is as a more specific marker of lymphatic lineage [2]. Our patient's AL followed this pattern.

In conclusion, ALs are rare, cystic, benign lesions. The diagnosis is established after surgery by pathological report. Adrenalectomy is the appropriate treatment of large symptomatic ALs, while small asymptomatic cysts may be treated conservatively under observation with radiological surveillance. To date, long-term results are excellent with good prognosis.

Conflict of Interest The authors declare that they have no conflicts of interest to report.

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