



Original research

Cystic adrenal lesions: Clinical and surgical management. The experience of a referral centre



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HIGHLIGHTS

- Management and surgical treatment of adrenal cysts.
- Focusing on the importance of extensive clinical evaluation as a diagnostic tool.
- Guide for the correct indication and timing of surgical resection.

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ABSTRACT

Background: Cystic adrenal lesions (CALs) represent a rare entity having heterogeneity in etiology and clinical manifestations. Due to their very low incidence and heterogeneity in clinical aspects, many controversies still exist about their management. **Methods:** From 1984 to 2012, 21 patients (7 M, 14 F, mean age 48.2 years) underwent adrenalectomy for CALs. 9 patients suffered from hypertension, and 7 were affected by thyroid disorders. **Results:** 4 patients presented with vague abdominal pain, while in 17 patients the CAL was incidentally identified during imaging examinations. All patients underwent evaluation of adrenal functionality and imaging study. We found 1 case of cystic pheochromocytoma (confirmed by urinary and blood sampling, and MIBG-scan). All patients underwent adrenalectomy (open approach in the first 10 patients treated from 1984 to 1996, laparoscopic lateral transabdominal approach in the other 11 cases). Indication to surgery included: size over 4.5 cm in 16 cases, suspected malignancy at imaging evaluation (not confirmed by histology) in 4 cases, cystic pheochromocytoma in 1 case. Histology revealed 11 endothelial cysts, 3 pseudocysts, 6 epithelial cysts and 1 cystic pheochromocytoma. **Conclusions:** The presence of CAL, even asymptomatic, requires complete endocrinological evaluation and imaging study. In the presence of large size, endocrine activity or any suspicion of malignancy, patients must be referred to surgery.

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1. Introduction

Cystic adrenal lesions (CALs), first described by Greiseliuss in 1670 [1], represent a rare entity with heterogeneity in etiology and clinical manifestations.

Due to the rarity of these lesions, only few case-series have been reported in literature [2–7].

Smaller CALs are often detected incidentally during US, CT scan or MRI evaluations, whereas larger cysts may present with pain, gastrointestinal symptoms or palpable masses [6,7].

After the first histological classification scheme (based on autoptic examinations) proposed in 1959 by Abeshouse [8] and revised by Foster in 1966 [9], CALs have been divided into four categories: endothelial cysts, epithelial cysts, pseudocysts, and parasitic cysts. In 1999, Neri and Nance identified pseudocysts as the most common subtype in their extensive review on surgically treated cases [10].

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CALs are often unilateral, even if bilateral lesions have been found in 8–15% of cases [3]; they are more common from the 3rd to the 5th decade, and may range from few millimeters to 50 or more cm in diameter [2,4,11,12]. For this heterogeneity in clinical aspects, many controversies still exist about their management.

The Authors report their 28-year experience, focusing on clinical evaluation and surgical treatment.

2. Patients and methods

From 1984 to 2012, 432 adrenalectomies have been performed in 411 patients at our Institution. Among these patients (M:F ratio 191:220, mean age 45.6 years) 21 (7 M, 14 F, mean age 48.2 years) were affected by CALs. 9 on these 21 patients suffered from primary hypertension, and 7 (3 M, 4 F) were affected by other endocrine disorders (multinodular goiter and/or hypothyroidism). No patient was admitted in emergency setting, no previous trauma history or surgery were reported.

Demographics and characteristics of patients affected by CALs are summarized on Table 1.

3. Results

Clinical, functional, histological features and surgical outcomes are reported on Table 2.

All patients were evaluated by the same clinical team having expertise in adrenal disorders, as well as in endocrine-related hypertension. Among these patients, 4 referred only vague abdominal pain and had CAL larger than 10 cm. In the other 17 cases CAL was and incidentally found during imaging studies carried out for different indications. In these cases size varied from 4 to 8.2 cm.

All patients received evaluation of adrenal function in order to identify any possible adrenal disorder:

- serum sodium and potassium levels and the ratio between morning plasma aldosterone and plasma renin activity to identify primary hyperaldosteronism;
- 24-h cortisol urinary excretion and overnight 1 mg dexamethasone suppression test to identify hypercortisolism (even subclinical);
- 24-h metanephrine urinary excretion to identify the presence of pheochromocytoma;
- hypertensive patients received ambulatory blood pressure monitoring (ABPM) to identify any possible lack in physiological night reduction in blood pressure levels (non-dipper), that is considered suspect for endocrine-related hypertension [13].

Table 1

Patient demographics, clinical symptoms, hypertension, diagnostic work-up, cysts size and side.

Mean age (range)	48.2 (31–70)
Sex (M/F)	7/14
Hypertension	9/21
Symptoms	
Asymptomatic/incidental	17
Abdominal pain	4
Gastrointestinal symptoms	–
Diagnostic work-up	
Endocrine evaluation	21
CT scan	19
MRI	15
MIBG-scan	1
Mean cyst size (range)	5.8 cm (4.4–13)
Cyst side (left/right)	12/9

Table 2

Surgical indications, approach and outcome, histology.

Surgical indications	
Size	16
Function	1
Suspicion of malignancy	4
Open approach	10
Midline incision	6
Extraperitoneal flank approach	4
Laparoscopic approach	11
Mean operating time	
Open approach	92.4 min
Laparoscopy	86.2 min
Mean post-operative stay	
Open approach	7.5 days
Laparoscopy	3.2 days
Complications ^a	
Grade I	3 patients
Grade II	2 patients
Grade IIIa	–
Grade IIIb	1 patient
Grade IVa	–
Grade IVb	–
Grade V	–
Histology	
Vascular/endothelial cysts	11
Epithelial cysts	6
Pseudocysts	3
Cystic pheochromocytoma	1

^a According to Clavien-Dindo Classification [16].

In one case we found increased levels of urinary metanephrines in a hypertensive patient with. This patient was then fully evaluated for pheochromocytoma (blood sampling and MIBG-scan), and he was found to have hypercaptation on the right CAL, so he was diagnosed as having a right adrenal cystic pheochromocytoma. So the patient received pharmacologic alfa-blockage for four weeks prior to surgery.

After accurate clinical and functional evaluation, all patients underwent imaging studies (CT-scan or MRI). In 9 cases, due to uncertain diagnosis, patients were submitted to both studies. Large CAL size (over 6 cm in larger diameter) in 16 cases, and uncertain radiological features (complex mass, intracystic septa with contrast enhancement, increased wall thickness) in 4 cases were the indications to surgical excision for patients having non-functioning CALs.

Percutaneous CAL fluid sampling or percutaneous drain prior to surgery were never carried out, since all patients were fit for surgery and general anesthesia, according to ASA risk classification.

All patients received adrenalectomy as treatment of choice. No extended surgery was needed and no adrenal-sparing procedures were performed. 10 patients, treated from 1984 to 1996, underwent “open” surgical adrenalectomy (by midline incision or extraperitoneal flank approach, depending on cyst size), while the other 18 patients underwent laparoscopic adrenalectomy always by lateral transabdominal approach, as previously described [14,15].

Early post-operative outcomes and complications, according to Clavien–Dindo classification [16], are summarized on Table 2. One patient required re-laparotomy to revise hemostasis on the first post-operative day. No visceral injuries were reported. All patients were discharged within the 8th post-operative day, and the mean post-operative stay was 3.3 days for patients submitted to laparoscopic adrenalectomy, and 6.1 days for patients submitted to open surgery.

All surgical specimens were evaluated by conventional histology with immunohistochemistry, and the older specimens were newly revised and reconsidered during 2001 [4]. No case of malignancy was identified. According to Abeshouse and Foster classification, 11

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