CLINICAL CASE

Adrenal cyst: our experience in its laparoscopic management

Quiste suprarrenal: nuestra experiencia en su manejo laparoscópico

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Abstract

Background: *Adrenal cysts are a rare entity; their diagnosis and treatment are challenging due to the lack of information, generating uncertainty regarding the best management.* **Case reports:** *We present three cases of adrenal cysts that received minimally invasive treatment with laparoscopic surgery, without complications and with a favorable evolution.* **Discussion:** *The new minimally invasive surgical techniques offer a therapeutic opportunity that allows preserving structures and obtaining the necessary material for histopathological diagnosis, as well as a resolution of the symptoms.*

Keywords: *Adrenal cyst. Laparoscopic de-roofing. Laparoscopy.*

Resumen

Antecedentes: *Los quistes suprarrenales son una afección rara. Su diagnóstico y su tratamiento son un desafío debido a la escasa información, lo que genera incertidumbre respecto al mejor manejo.* **Casos clínicos:** *Presentamos tres casos de quistes suprarrenales que recibieron tratamiento de mínima invasión con cirugía laparoscópica, sin presentar complicaciones y con una evolución favorable.* **Discusión:** *Las nuevas técnicas quirúrgicas de mínima invasión ofrecen una oportunidad terapéutica que permite preservar estructuras y obtener material necesario para su diagnóstico histopatológico, así como la resolución de la sintomatología.*

Palabras clave: *Quiste suprarrenal. Destechamiento laparoscópico. Laparoscopía.*

Introduction

Adrenal cysts were first described in 1670 by Greiselius[1]. So far, more than 600 cases have been reported in the literature, with an incidence in autopsy studies carried out in 1951 and 1958 of 0.064% to 0.18%[1-3], respectively, while in a 2005 clinical series, which examined 149 laparoscopic adrenalectomies, an incidence of 5.4% was reported[4].

Most adrenal cysts are unilateral and there is no laterality predominance, although 8% to 15% occur bilaterally[1,3]. They are more common in women, with a ratio of 2-3:1, and predominate at between the third and fifth

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Date of reception: 04-04-2020
Date of acceptance: 17-09-2020
DOI: 10.24875/CIRUE.M20000239

Cir Cir (Eng). 2021;89(2):247-252
Contents available at PubMed
www.cirugiaycirujanos.com

decades of life[1-3,5]. As for their size, it varies between several millimeters to up to 50 cm in diameter[1,6,7]. Regarding their clinical presentation, most are asymptomatic and benign; however, it is essential to bear in mind that adrenal cysts are a heterogeneous group whose diagnosis and management are often unclear. When they occur with symptoms, they do it with abdominal pain, gastrointestinal symptoms, or a palpable tumor.

Adrenal cysts classification was initially carried out by Abeshouse in 1959, and subsequently was modified by Foster in 1966, who divided them into four histological groups: endothelial cysts, pseudocysts, epithelial cysts and parasitic cysts[1,5,8]. In the study by Abeshouse and Foster in 1966[5], with 115 autopsies, endothelial cysts were the most common subtype, with 45%. On the other hand, Neri and Nance[1], in 1999, with a group of 613 patients, reported that pseudocysts were the most common subtype, with 56%. This difference can be explained by the fact that while the former represent lesions found in autopsies, the latter were surgically removed lesions; it is possible that Abeshouse and Foster reports represent a more real overall incidence. The incidence rates reported by Abeshouse and Foster, and by Neri and Nance[1,5], for (simple) endothelial cysts were 45% and 24%; for pseudocysts, 39% and 56%; for epithelial (true) cysts, 9% and 6%; for parasitic (hydatid) cysts, 7% and 2%; and for unspecified cysts, 0% and 12%, respectively[1,5]. A 2004 Mayo Clinic study reported that 78% were pseudocysts, 20% endothelial cysts and 2% epithelial cysts[9].

It is important for a comprehensive evaluation to be available, which may include a determination of their functional status and malignant potential. Although it is uncommon for them to be functional, it is a possibility that should be borne in mind, since hormonally active cystic adrenal neoplasms have been described. Only 15% of all incidentalomas are functional, and for cystic incidentalomas this percentage is much lower[1,3,10]. Tests for their evaluation include serum potassium and aldosterone determination, dexamethasone suppression test, and dehydroepiandrosterone sulfate (DHEA-S) and metanephrines determination in plasma and urine[1,3,7,11]. Once histopathological diagnosis is established, association of pseudocysts with neoplasms should be considered. Both the studies by Foster and by Neri and Nance[1,5] described a 7% association with malignancy, and more recent studies have described that the association of pseudocysts with neoplasms is 18.7% to 44%; however, in general, up to 95% of cases of malignant adrenal cysts are metastatic[1,3,5,8,9]. Imaging studies that are useful for diagnosis include computed tomography (CT), magnetic resonance imaging and ultrasonography, which can serve for malignancy evaluation. Benign adrenal cysts have a density of 0 to 20 HU on CT, with thin, smooth walls that lack enhancement after contrast medium administration. If they exhibit higher attenuation values, hemorrhage, intra-cystic residues or calcification should be considered, with the latter being found in approximately 15% of cases and not implying malignancy. On the other hand, lesions with more than 10 HU can be observed in lipid adenomas, pheochromocytomas, metastases and adrenal cortical carcinomas. A wall thickness of more than 5 mm with enhancement or stippled central calcification may suggest malignancy[1,2,6,12]. Adrenal cysts size should not be regarded as a criterion for malignancy[1,3].

Once the diagnosis is established, we are dealing with the ideal therapeutic decision for each case. Currently, there is no general agreement on which the best therapeutic option is, and treatment is divided into surgical approach and a more conservative surveillance approach. Among the possible management strategies, percutaneous needle aspiration, sclerotherapy, surgical resection, and cyst de-roofing have been described. It has been proposed that cysts > 5 cm should be surgically approached due to the risk of bleeding and other secondary complications. Each case should be individualized and related conditions be evaluated, including findings in other organs, associated anomalies, functional status of the cyst, symptoms, size, images suggestive of hemorrhage or heterogeneous images, the possibility of malignancy and the risk of potential complications such as bleeding or infection[1,3,7]. If parasitic, functional, or malignant cysts are identified, surgical excision should be considered. Whenever possible, the kidney and adrenal gland should be preserved, which has been favored with minimally-invasive techniques, such as laparoscopic and robotic-assisted surgery, which achieve a decrease in morbidity, shorter hospital stays, less blood loss and better cosmetic results[1,13].

Clinical cases

Case 1

Twenty-two-year-old woman with a history of cesarean section. She referred a 5-year history of colic-type pain with an intensity of 8 on visual analogue scale for pain, located on left lumbar dorsal region with irradiation to ipsilateral flank, which increased with physical activity and improved with rest. She denied urinary

symptoms. On physical examination, no anomalies were observed. A CT scan without and with intravenous iodinated contrast (Fig. 1) reported a left adrenal cystic tumor with thin, well-defined borders, and homogeneous content within fluid range (18 HU), which showed no enhancement with contrast medium, with an approximate size of 7.7 x 6.7 x 6.1 cm, for a volume of 164.5 cm^3. Since the images were characteristic of a simple cyst, and the patient had no data consistent with a metabolically active cyst, no functionality tests were performed. In view of the symptoms and cyst dimensions, performing a laparoscopic de-roofing of the left adrenal cyst was decided (Fig. 2). Surgical time was 2 hours and 45 minutes, with 100-mL bleeding. There were no complications during surgery. Post-surgical evolution was favorable and she was discharged from hospital at 48 hours. The histopathological report indicated that it was a simple adrenal cyst; cytology indicated a sample with clean background and sparse fibrillar fragments, where no inflammatory or atypical cells were observed. At her first follow-up appointment, the patient reported pain only on surgical wounds, of mild intensity. At 5 months of follow-up, the patient remains asymptomatic.

Case 2

Thirty-five-year-old male with a 12-month history of moderate-intensity left lumbar colic-like pain. Ultrasound reported a left pararenal cyst, and management with non-steroidal anti-inflammatory drugs was therefore started, with partial symptom improvement. Subsequently, a CT scan without and with intravenous iodinated contrast reported a 5 x 4-cm left adrenal tumor, with 4-HU homogeneous content, which showed no enhancement with contrast medium administration. Urinary metanephrines determination and pre-surgical laboratory tests showed no data suggestive of functional adrenal tumor, and performing a left laparoscopic adrenalectomy was therefore decided. The procedure was uneventful. Post-surgical evolution was satisfactory, and discharge was therefore decided at 48 hours. During follow-up, the patient referred pain improvement. Currently, after 7 months of follow-up, he remains free of pain.

Case 3

Forty-two-year-old male, diagnosed with hypertension one year earlier. He referred that one year prior, during hypertension management, he had a renal ultrasound performed, in which a lesion, probably dependent on the right adrenal gland, was found. Physical examination showed no abnormalities. CT scan without and with intravenous iodinated contrast (Fig. 3) identified a 8.6 x 7.6 x 6.9-cm right adrenal cyst with regular borders, with hypodense content within fluid range (14 HU), which did not change with contrast medium administration. Due to the size of the cyst, laparoscopic right adrenal cyst de-roofing was decided. Surgical time was 62 minutes and anesthetic time was 75 minutes, with-20 mL bleeding. There were no complications during surgery. Post-surgical evolution was favorable and he was discharged from hospital at 24 hours. Histopathology report indicated a simple adrenal gland cyst. The patient is on follow-up at outpatient services, and 20 days after surgery, he remained asymptomatic.

Discussion

The information published so far corresponds to small case series. Adrenal cysts low incidence limits the evidence and recommendations, which is why their diagnosis, management and follow-up remain without consensus. Further research is needed to aid in decision-making in order for the best patient outcomes to be achieved.

Our patients, a 22-year-old woman and two men aged 35 and 42 years, are within the age range reported in the literature[1,2]. Two presented with pain and all three had a normal physical examination. In two of the patients, ultrasound was initially carried out, and all underwent contrast-enhanced CT, with simple adrenal cyst characteristics being observed. Laterality was left in two cases and right in the other. Functionally tests for adrenal cystic neoplasm were performed only in one patient, which were negative; however, none had clinical data consistent with adrenal tumor with endocrine activity. It is important to remember that, although there is consensus about the performance of screening tests to detect catecholamine and cortisol excess in most patients with adrenal incidentaloma, this does not apply to those with adrenal tumors whose imaging characteristics are typical of myelolipoma or cysts[14]. The decision on whether or not to perform functionality tests for simple adrenal cysts will be by the physician's choice according to the characteristics of each case. In addition, most adrenal cysts will be benign and hormonally non-functional tumors[15].

In two of the three cases, surgical management was decided due to patient symptoms, while in the third, it

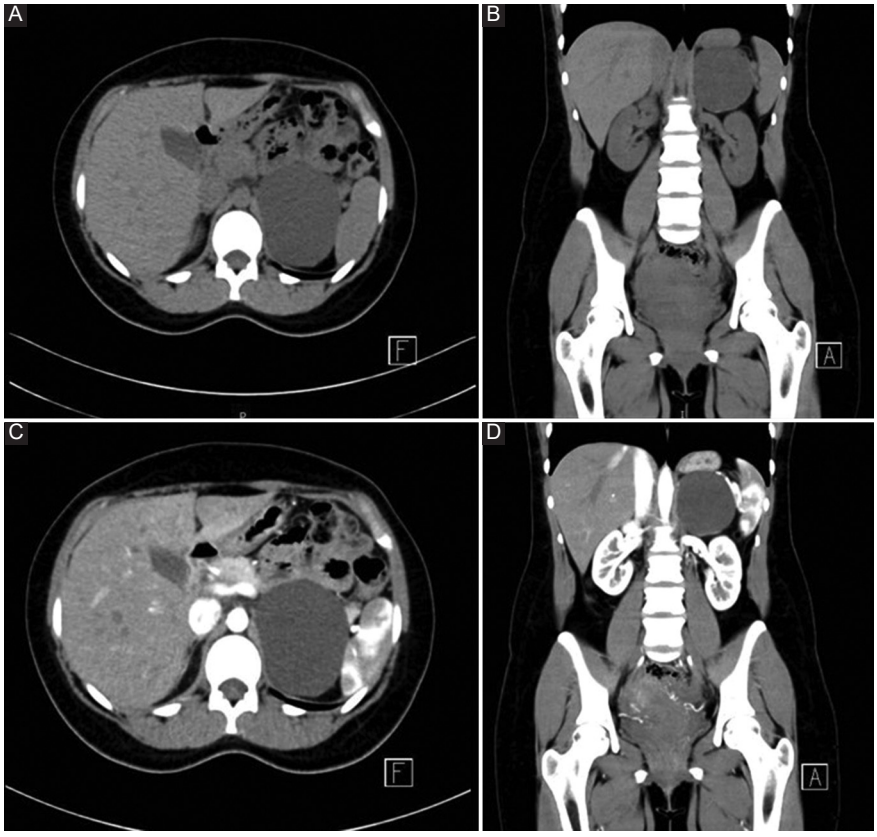

Figure 1. *Computed tomography.* **A:** *Axial section without contrast.* **B:** *Coronal section without contrast.* **C:** *Axial section with intravenous iodinated contrast; no enhancement is observed.* **D:** *Coronal section with intravenous iodinated contrast; no enhancement is observed.*

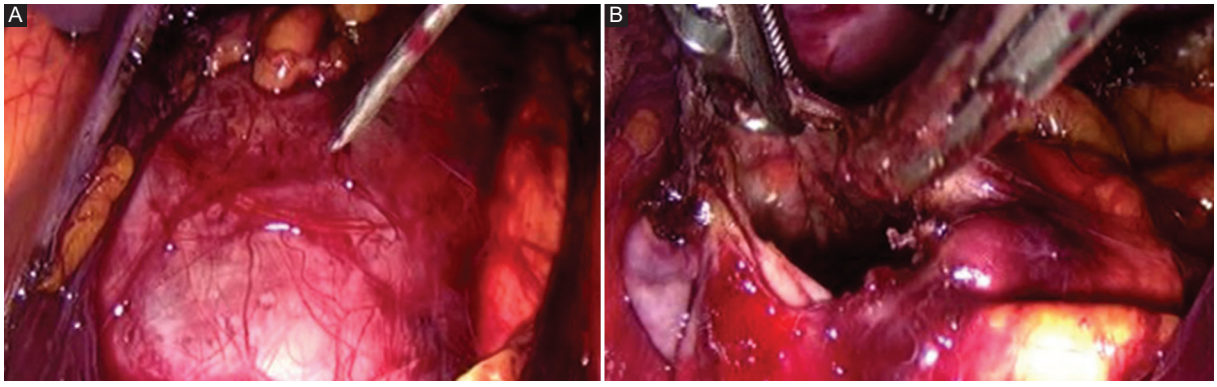

Figure 2. *Intraoperative images.* **A:** *The left adrenal cyst is observed.* **B:** *Adrenal cyst de-roofing and resection are performed.*

was due to the dimensions of the cyst. In two cases, de-roofing was performed, while in the other, adrenalectomy was carried out; in all patients, the approach was via laparoscopic route

We should point out that adrenal cysts treatment continues to be the subject of discussion, and there is still no clear consensus on their surgical management, partly due to the rareness of this condition, which is why there is uncertainty as to whether recommendations for the management of adrenal incidentalomas should be extrapolated to adrenal cysts, considering that cysts are much less common than other adrenal lesions, and that there is scarce evidence on their management. Symptoms, size and functionality or malignancy suspicion have been proposed as suggestions for surgical management to be carried out; however,

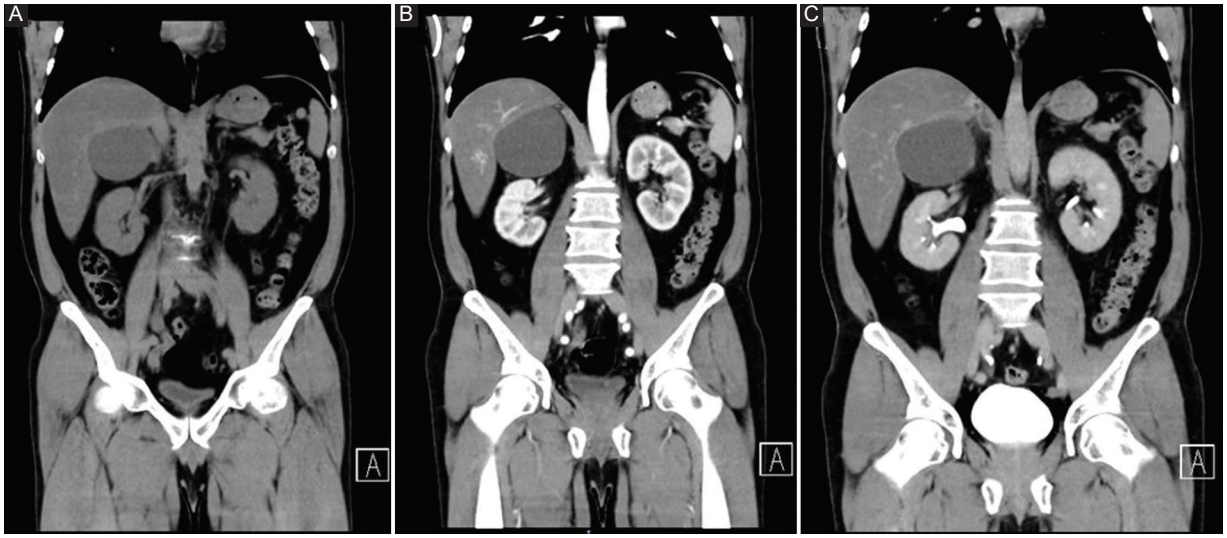

Figure 3. *Computed tomography.* **A:** *Coronal section without contrast.* **B:** *Coronal section with intravenous iodinated contrast; no enhancement is observed.* **C:** *Coronal section at elimination phase.*

even when surgical treatment is considered, there is no consensus on the best option between enucleation, decortication and marsupialization, or en bloc adrenalectomy. Although the need for clear guidelines that allow deciding which adrenal cysts require surgical treatment is evident, the safety of laparoscopic management has been well documented[15]. Whenever possible, procedures should be aimed at preserving the adrenal gland, by resecting the cystic wall and sparing adrenal tissue. The decision on the extent of surgery will depend on each case, surgeon's choice and intraoperative presentation. Laparoscopic surgery can be considered current method of choice owing to its unquestionable advantages, and because adrenal gland preservation is justified in most patients[16].

In those patients who had experienced symptoms, pain improvement was documented during their follow-up, with the purpose of surgery thus being met, and agreeing with observations described in the literature about surgical and minimally-invasive management[1,17]. Although there is still no consensus establishing the best therapeutic behavior, minimally-invasive surgeries, such as laparoscopic and robotic-assisted approaches, provide an opportunity for adrenal cysts effective treatment.

Acknowledgements

Adrián A. Ramírez-Beltrán is resident physician of the urology department at Hospital General de Mexico Dr. Eduardo Liceaga, for the 2017-2021 period.

Funding

This research has not received any specific grant from agencies of the public, commercial or non-profit sectors.

Conflicts of interest

The authors declare that there are no conflicts of interest.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this research.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained informed consent from the patients and/or subjects referred to in the article. This document is in the possession of the corresponding author.

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