Novel treatment (new drug/intervention; established drug/procedure in new situation)

CASE REPORT

Postsurgical large adrenal cyst recurrence: treatment by means of percutaneous alcohol ablation

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SUMMARY
We describe a case of a 28-year-old man who presented with symptomatic, right-sided, large adrenal cyst recurrence 9 months after laparoscopic decortication. Final treatment was achieved by means of percutaneous aspiration and ethanol ablation. On 6-month follow-up the patient was asymptomatic and the cyst remained minimised. In our opinion, percutaneous treatment with alcohol ablation of primary benign symptomatic or recurrent uncomplicated adrenal cysts should be considered as an effective alternative method when patients are frail or surgery fails to resolve the problem.

BACKGROUND
We think this is an important case presentation because it offers a therapeutic alternative for minimal invasive treatment of adrenal cysts.

CASE PRESENTATION
A 28-year-old man presented with abdominal discomfort and right flank pain. He had a history of laparoscopic decortication of a large right adrenal cyst 9 months earlier. Physical examination was unremarkable and initial laboratory tests were normal.

Preoperative non-enhanced CT imaging revealed a large cyst of 12 cm diameter in the right retroperitoneal space, originating from the right adrenal gland (figure 1). The right adrenal gland was indiscernible, the inferior vena cava was medially displaced and the right kidney had migrated caudally with a twist. All serological and hormonal secretion tests were negative. Although the serological antihydatid tests might be false negative in numerous cases, sectional imaging was not in favour of Echinococcus disease, so the final diagnosis was a simple right adrenal cyst.

The patient was laparoscopically operated on and the diagnosis of a simple adrenal cyst was intraoperatively confirmed. The cyst was immediately evacuated by means of decortication. Two months later, postoperative MRI revealed cyst recurrence with minimal shrinkage of the cyst (figure 2).

Six months later, the patient presented to us with right flank pain and a feeling of abdominal weight. Ultrasound revealed the presence of a large cystic, unilocular mass with a diameter of slightly over 12 cm, residing between the right kidney and the liver. The patient refused additional surgery but consented for the less invasive treatment option of percutaneous drainage with cyst evacuation and alcohol ablation. Two months later, the cyst was successfully percutaneously punctured under ultrasound and fluoroscopic guidance with the patient under conscious sedation. After placement of an 8 Fr catheter, 900 ml of clear fluid was evacuated. Filling of the lesion with 1:1 diluted contrast medium followed and no leakage or communication with other anatomic structures was noted (figure 3). Subsequently, 300 mL of 98% ethanol was slowly injected into the cyst while the patient remained in prone position and then in supine position at 20 min intervals. Finally, the cyst was re-evacuated and the catheter removed. The patient was admitted for 12 h monitoring and was then discharged in good condition.

One, 3 and 6 months later, abdominal ultrasound follow-up revealed adequate cyst shrinkage down to 2.8×6.9 cm (figure 4). The patient remains symptom free to date.

DISCUSSION
Adrenal cysts are uncommon lesions (0.064–0.18% in autopsy series) for which a 3:1 female predominance is reported.\(^1\)\(^2\) These lesions rarely produce any clinical symptoms yet they have been associated with abdominal discomfort, feeling of abdominal ‘weight’ or ‘fullness’, flank pain and arterial hypertension. If very large in size, they can be seen as a palpable mass or, more frequently, discovered as incidental findings during diagnostic imaging examinations of the abdomen performed for other purposes. Occasionally, complications occur due to infection, haemorrhage, rupture and compression of adjacent structures.\(^3\)\(^4\)

According to histopathological classification, four types of cysts are recognised: parasitic, epithelial, endothelial and pseudocysts.\(^5\)\(^6\) Hydatid cysts of the adrenal gland account for only 7% of all adrenal cysts and occur in cases of disseminated infection by Echinococcus granulosus.\(^5\) Epithelial
cysts are also rare, representing about 9% of cystic lesions of the adrenal gland and, since they lack characteristic imaging findings, differential diagnosis from other adrenal cystic lesions may be difficult to make. Endothelial cysts comprise the majority of adrenal cystic lesions and are further divided into two categories: lymphangiomatous (42%) and haemangiomatous cysts (3%). Finally, adrenal pseudocysts are considered the second most common pathological subtype of adrenal cystic lesions accounting for approximately 39% of all adrenal cysts. They are formed secondary to intraglandular haemorrhage or infarction and consist of a fibrous capsule without an epithelial lining.

Adrenal cysts, as imaged with ultrasonography, are usually anechoic, with or without internal septations. About 15% of them contain wall calcification, which appears as high-echogenicity foci with acoustic shadowing. On CT imaging, findings of a non-enhancing mass with near-water attenuation can differentiate adrenal cysts from adenomas and multiplanar reconstructions can prove helpful in defining the origin of the lesion and its association with neighbouring anatomical structures, such as kidneys or pancreas. In case of intracystic complications such as haemorrhage or infection, the adrenal cyst may lose its homogeneous attenuation rendering CT imaging non-diagnostic. MRI can be useful in such cases, minimising the differential diagnosis. On MRI, simple cysts typically demonstrate homogeneously low-signal intensity on T1-weighted images and high-signal intensity on T2-weighted images, with no internal contrast enhancement. If haemorrhage is present, this will appear hyperintense on both, T1-weighted and T2-weighted images. Peripheral calcification is, however, difficult to appreciate on MRIs.

On follow-up imaging, adrenal cysts can remain stable in size, decrease or even enlarge over time. In a series of 20 patients, Ricci et al reported that lesion enlargement was actually a frequent finding on follow-up and as an isolated manifestation should not be considered as suspicious for malignancy. There is not a generally accepted consensus on management. According to the literature, most authors agree that intervention is indicated only for symptomatic, large (>5 cm) or potentially malignant cysts. However, it is important to mention that less than 7% of all adrenal cystic lesions are malignant. Under this scope, an acceptable practice for small, asymptomatic and hormonally inactive adrenal cysts is expectant surveillance with interval follow-up imaging, without surgery or other kind of intervention. Non-functional, large or symptomatic lesions are usually managed by aspiration to rule out malignancy, followed by open surgical or laparoscopic excision in order to alleviate symptomatology. Cyst evacuation by aspiration alone has a high probability of fluid reaccumulation. In experienced centres, laparoscopic decortication and marsupialisation should be the preferred treatment option followed by laparoscopic partial or total adrenalectomy. With this approach, better cosmetic results, less blood loss, fewer postoperative complications and shorter hospitalisation have been reported. Open surgery should be reserved for very large or complicated cysts, parasitic cysts and functioning or potentially malignant cystic lesions together with appropriate follow-up studies.

A combination of percutaneous evacuation and alcohol ablation is a well established treatment option for simple renal and liver cysts. This is one of few reports to the best of our knowledge, to apply the method for a simple adrenal cyst. It is a simple and easy way to reduce the patient’s symptoms but the authors agree that it deserves consideration only after malignant or parasitic causes have been excluded, as in our case.

In conclusion, percutaneous treatment with alcohol ablation of primary benign symptomatic or recurrent uncomplicated adrenal cysts should be considered as an effective alternative method when patients are frail or surgery fails to resolve the problem.
Learning points

- Percutaneous treatment with alcohol ablation of primary benign symptomatic or recurrent uncomplicated adrenal cysts should be considered as an effective alternative method when patients are frail or surgery fails to resolve the problem.
- Special attention must be paid to exclude a malignant cystic tumor.
- Follow-up is important in order to diagnose re-occurrence of the cyst.

Contributors AH performed the interventions; AK edited the text and reviewed the literature; IP reviewed the case report and added surgical aspects; CM was responsible for the patient’s care and also edited the text.

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REFERENCES