

Haemorrhage into Non-functioning Adrenal Cysts-Report of Two Cases and Review of the Literature

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Abstract

Adrenal cysts are a rare condition and are usually non-functioning and asymptomatic. Most of the reported cases were incidental findings or discovered at autopsy. However, large cysts have a tendency to develop complications such as intracystic haemorrhage and rupture, which can present as an acute surgical emergency. We report two cases of adrenal cysts with intracystic haemorrhage. One patient presented with persistent non-specific upper abdominal pain, investigations with ultrasound (US) scan and computed tomographic (CT) scan revealed a left adrenal cyst and gallstones. Simultaneous cholecystectomy and adrenalectomy was performed with resultant relief of symptoms. The second patient presented with acute abdominal pain simulating acute surgical abdomen. Preoperative CT scan showed a large cystic lesion in the region of the tail of the pancreas with radiological evidence of haemorrhage but was unable to confirm its origin. The cyst was found to have arisen from the left adrenal gland at laparotomy; left adrenalectomy with complete excision of the cyst was done. Histology showed pseudocyst with haemorrhage in both cases. Pseudocyst is the commonest histological type encountered clinically. We believe the second case is related to pregnancy and childbirth as the patient presented during puerperium and the cyst, even though very large in size (25 x 15 x 15 cm), was not noted during antenatal screening with US scan.

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Introduction

Doran, in 1903, reported the first case of adrenal cyst described by Greiseliuss in 1670.¹ It was not until 1966 that Foster reported the 220th case.² By 1989, there were only about 300 reported cases.³ The incidence of adrenal cysts is low and estimated to be about 1 in 1500. Wahl⁴ in 1951 found 9 unsuspected adrenal cysts in 13,996 autopsies and Hodges and Ellis⁵ reported 2 cases in 1100 autopsies. The low incidence of reported adrenal cysts is due partly to their rarity and partly to the asymptomatic nature of these lesions. When asymptomatic, treatment depends on the underlying pathology and size. New generation ultrasound (US) scan and computed tomographic (CT) scan can detect and localised these lesions with high accuracy, but are unable to differentiate cystic neoplasm from benign cysts. Many authors^{2,6-8} thus recommend surgical exploration and excision for accurate histologic diagnosis. Selected small cysts with obvious benign features on radiological imagings can be aspirated percutaneously and observed.⁹⁻¹¹ Adrenal cysts, especially when large in size, can develop complications such as intracystic haemorrhage and rupture which can

simulate acute surgical abdomen and require urgent surgical treatment.

Case Reports

Case 1

A 57-year-old woman presented to her general practitioner (GP) complaining of vague upper abdominal discomfort for about one month. She was previously healthy, her blood pressure was normal and there was no significant abnormality detected during abdominal examination. Ultrasound scan by her GP revealed a 5 cm in diameter septated cystic lesion in the left supra-renal region. Incidentally, gallstones were noted in her gallbladder. CT scan was done and this showed a 5-cm thick-walled cystic lesion in the left adrenal with features suggestive of recent bleeding into the cyst. The right adrenal and 24-hour urinary vanillylmandelic acid (VMA) were normal.

Surgical exploration through bilateral subcostal incisions was done. A thick-walled cyst, 5 cm in diameter, was found in the left adrenal gland, very adherent to the

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remnant adrenal tissue, which was thin and atrophic. A cystic tumour cannot be excluded by gross examination. Left adrenalectomy and cholecystectomy were performed. Histology showed "the cyst contains blood clot and debris; its wall is composed of granulation tissue infiltrated with hemosiderophages, foamy macrophages, lymphocytes and plasma cells and is in turn surrounded by a thick fibrous wall; consistent with benign haemorrhagic pseudocyst of the adrenal". Her recovery after the operation was uneventful.

Case 2

A healthy 31-year-old first-time mother presented with acute abdominal pain 6 weeks after normal vaginal delivery of her baby. She was in pain and in distress. Blood pressure was 100/60 mmHg but the pulse was 100/min. She was mildly anaemic. There was a very large swelling in the left side of her abdomen extending from the left hypochondrium to the lower abdomen. This was very tense and acutely tender. Her haemoglobin was 10 g/dl, white cell count was raised at 10,700/ul, while serum electrolytes and amylase level were normal. An urgent CT scan of the abdomen was done which revealed a huge 15 x 24 cm cystic lesion in the region of the pancreatic tail with evidence of recent haemorrhage (Fig. 1). It was difficult to be certain whether the cyst arose from the tail of pancreas or the left adrenal because of the large size, which distorted the normal anatomy.

Exploration through a midline incision was done. The cyst was found to arise from the left adrenal, measuring 25 x 15 x 15 cm and contained more than 1.5 litres of haemorrhagic fluid and blood clots. The remnant adrenal tissue was very thin and haemorrhagic. Complete excision of the cyst and left adrenalectomy was performed. Histology showed a benign cyst with no epithelial lining or evidence of malignancy, consistent with a pseudocyst of the adrenal gland. Her postoperative recovery was uneventful.

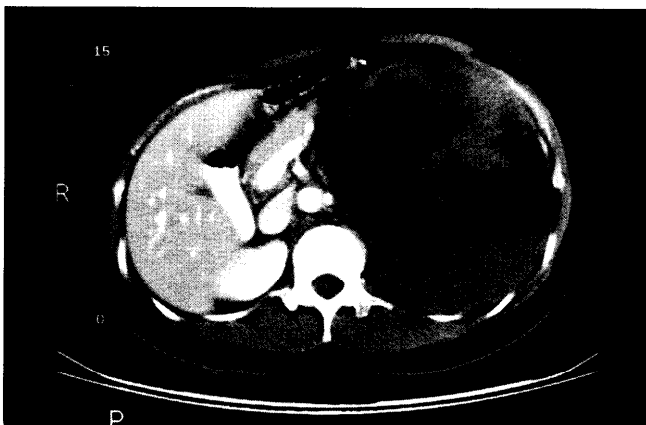


Fig. 1. Huge cystic lesion of the (L) adrenal gland in the region of the pancreatic tail with evidence of recent haemorrhage.

Discussion

Adrenal cysts occur in all ages, with a peak in the third to sixth decades^{2,9,12-15} and slight female preponderance of 2:1 to 3:1 female to male ratio.^{2,9,12,15} Majority of adrenal cysts are asymptomatic and are discovered incidentally or at autopsies.^{2,4,5,12} Adrenal cysts are rarely associated with adrenal hyperfunction, which is suggestive of a cystic cortical adenoma. Bilateral adrenal cysts occur in 8% to 10% of cases.²

Non-functioning adrenal cysts do not have a characteristic symptom-complex. The most prominent clinical features include dull pain in the adrenal regions, gastrointestinal symptoms due to compression on the bowel and a palpable mass.^{2,6,12} The cysts can attain a large size as in Case 2. Armitage¹⁶ reported a 30-cm cyst in 1959. The pain associated with adrenal cysts is usually dull and persistent, however, it may become acute and intense and associated with shock when the cysts rupture or when there is acute intracystic haemorrhage^{2,6,17,18} as in Case 2. Rarely adrenal cysts may get infected and produce constitutional symptoms of fever and leukocytosis.^{19,20}

Radiological studies remain the most important tools in the diagnosis of adrenal cysts. Prior to the advent of US scan and CT scan, combinations of plain films, gastrointestinal contrast study, urography and angiography were used, but the yields were poor and correct preoperative diagnoses of adrenal cysts were reached in only 3.2% to 7.2% of patients.^{2,12} New generation US and CT scans have greatly enhanced the visualisation of the adrenals and localisation of lesions arising from them. The incidence of adrenal incidentaloma has also increased with wider availability of US and CT scans.^{9,17,18,21,22} However, the difficulty in differentiating a benign cyst from an cystic adenoma or a malignant lesion remains. Rozenblit et al¹⁰ in their review suggested that a small adrenal cyst with near water attenuation and a thin wall (<3 mm) is likely to be benign. Selective biochemical studies should be carried out when a functioning adrenal cyst is suspected.

The first classification of adrenal cyst was proposed by Terrier and Lecene²³ in 1906. This was later modified by Abeshouse et al¹² and Barron and Emanuel.²⁴ It is now well accepted that adrenal cysts may be classified into 4 categories:

1. Parasitic cysts (7%):^{2,6,25} Most of these are echinococcal in origin and diagnosis is made by identifying the parasites or its constituents in the cysts.
2. Epithelial cysts (9%):^{2,6,25} These are uncommon and can be either a true retention cyst, embryonal cyst or a cystic adenoma.
3. Endothelial cysts (45%):^{2,6,25} This category includes the commoner lymphangiomatous cyst (42%) and

the rare haemangiomas cyst which makes up only 3% in this group.

4. Pseudocysts (32% to 39%) account for more than 60% of all clinically encountered adrenal cysts. Both our cases are of this variety. They are results of cystic resolution of haematoma in normal adrenal glands, which may follow trauma, bleeding diathesis or severe infection. Pseudocysts arising from haemorrhagic cystic degeneration of adrenal tumours have been reported but rare.^{7,12,19} Adrenal cyst occurring during pregnancy is rare, with only two other reported cases to the best of our knowledge.^{26,27} We believe the second case of pseudocyst in our report occurred during pregnancy secondary to adrenal haemorrhage, and subsequently complicated by intracystic haemorrhage during puerperium.

Treatment of adrenal cysts depends on the underlying pathology, size of the cysts, associated symptoms and the occurrence of complications.⁹ Many authors recommend surgical exploration for accurate histological diagnosis to rule out malignancy and functioning tumour.^{2,6-8} However, with high resolution CT scan, accurate localisation of the adrenal cysts and assessment of their contents as well as the thickness of the cysts wall can be achieved. Some authors suggest that small cysts with thin wall and contents attenuation of near water density are likely benign and can be managed by percutaneous aspiration.⁹⁻¹¹ Surgically, the adrenal glands can be accessed via the lumbar approach, transabdominally or occasionally via a thoraco-abdominal approach when the cysts are very big. Both our cases were approached transabdominally, the first with a bilateral subcostal incision as cholecystectomy was planned at the same time and in the second with a longitudinal midline incision. Since the early 1990s, laparoscopic adrenalectomy has been shown to be safe and offers faster recovery and shorter hospital stay.^{28,29} Laparoscopic approach to the adrenal can be anterior (transperitoneal) or posterior (retroperitoneal).³⁰ Needlescopic adrenalectomy has also been reported.³¹ The objective of surgical treatment of adrenal cysts is complete excision with preservation of the remnant adrenal tissue. Foster² and Cerise et al⁶ advocate surgical enucleation as the treatment of choice. Ghandur-Mnaymneh et al²⁵ has pointed out that pseudocysts have the tendency to become adherent to surrounding structures mimicking malignant tumour. This was also the reason for performing adrenalectomy in the first case. When complicated by haemorrhage, rupture or infection and presented with acute abdominal pain, a high index of suspicion, aided by CT scan, can help in reaching an accurate preoperative diagnosis, albeit difficult. These usually require urgent surgical exploration and excision of the cyst with or without adrenalectomy.^{17,18,20}

Conclusion

Adrenal cysts are uncommon clinical occurrences, majority being incidental findings. Usual symptoms are related to the size and local pressure effect of the cysts or occasionally secondary to complications such as intracystic haemorrhage, rupture or infection. In the majority of cases, the cysts should be excised for accurate diagnosis or relief of symptoms and prevention of complications. Those few cases that are asymptomatic and have CT features highly suggestive of a benign cyst can be managed by percutaneous aspiration.

Addendum

We encountered a third case of adrenal cyst during revision of this manuscript. A healthy 58-year-old man presented with gradual onset right-sided abdomen pain over one week. Clinically he was afebrile; examination of the abdomen revealed a mildly tender swelling in the right hypochondrium suspicious of acute cholecystitis or liver abscess. CT scan of the abdomen showed a large cyst (20 cm in size) in the right lobe of the liver. This was punctured percutaneously under CT scan guidance, draining more than 2.5 litres of straw-coloured fluid. He subsequently developed fever, raised white cell count and increased abdominal pain over the following few days. A repeat CT scan of the abdomen showed recollection of the liver cyst back to its original size and the content was radiologically more dense than previously, suggestive of infection or haemorrhage into the cyst. He was then referred for surgical drainage. At laparotomy, the cyst was found to have arisen from the right adrenal gland; it measured 24 cm in diameter and contained more than 3 litres of dark brown fluid; consistent with recent haemorrhage into the cyst. There was minimal residual normal adrenal gland visible. Right adrenalectomy with complete excision of the cyst was done. Histology confirmed inflammatory pseudocyst of the adrenal with foci of haemorrhage. He was well after the operation and went home on 6th postoperative day.

REFERENCES

1. Greiseliuss (cited by Doran A H G). Cystic tumor of the suprarenal body successfully removed by operation. *Br Med J* 1903; 1:1558.
2. Foster D G. Adrenal cysts. Review of literature and report of case. *Arch Surg* 1966; 92:131-43.
3. Medeiros L J, Weiss L M, Vickery A L. Epithelial-lined (true) cyst of the adrenal gland: a case report. *Hum Pathol* 1989; 20:491-2.
4. Wahl H R. Adrenal cysts. *Am J Pathol* 1951; 27:758.
5. Hodges F V, Ellis F R. Cystic lesions of the adrenal gland. *Arch Pathol* 1958; 66:53.
6. Cerise E J, Hammon J W. Adrenal cyst. *Surgery* 1968; 63:903-10.
7. Kearney G I, Mahoney E M. Adrenal cysts. *Urol Clin North Am* 1977; 4:273-83.
8. Kearney G I, Mahoney E M, Maher E, Harrison J H. Functioning

- and nonfunctioning cysts of the adrenal cortex and medulla. *Am J Surg* 1977; 134:363-8.
9. Sroujeh A S, Farah G R, Haddad M J, Abu-Khalaf M M. Adrenal cysts: Diagnosis and management. *Br J Urol* 1990; 65:570-5.
 10. Rozenblit A, Morehouse, Amis ES Jr. Cystic adrenal lesions: CT features. *Radiology* 1996; 210:541-8.
 11. Stewart B H. Adrenal cysts: Pathogenesis and histological identification with a report of 6 cases [editorial comment]. *J Urol* 1979; 122:87.
 12. Abeshouse G A, Goldstein R B, Abeshouse B S. Adrenal cysts: review of literature and report of three cases. *J Urol* 1959; 81:711.
 13. Esquivel E, Grabstald H. Giant adrenal cyst. *J Urol* 1965; 94:635-8.
 14. Tehrani M, Wolgin W, Niedwich A. Adrenal cysts: report of two cases. *J Urol* 1965; 93:427.
 15. Cerney J C, Warhowsky A, Hall J. The preoperative diagnosis of adrenal cysts. *J Urol* 1970; 104:787.
 16. Armitage H V. Benign cyst of adrenal gland. *Arch Surg* 1959; 79:160.
 17. Pasciak R M, Cook W A. Massive retroperitoneal haemorrhage owing to a ruptured adrenal cyst. *J Urol* 1988; 139:98-100.
 18. Keane P F, Hyland J M. Haemorrhage into an adrenal cyst. *Br J Clin Prac* 1990; 44:527-8.
 19. Ellis F H, Dawe C J, Clagett O T. Cysts of the adrenal glands. *Ann Surg* 1952; 136:217.
 20. Naess H, Svendsen E, Varhaug J E. Infected pseudocyst of adrenal gland. *Eur J Surg* 1991; 157:237.
 21. Copeland P M. The incidentally discovered adrenal mass. *Ann Intern Med* 1983; 98:940-5.
 22. Wood D E, Delbridge L, Reeve T S. Surgery for adrenal tumors: is operation for small incidental tumor appropriate? *Aust N Z J Surg* 1987; 57:739-42.
 23. Terrier F, Lecene P. *Rev Chir* 1906; 34: 321. In: Zuckner J. Adrenal cyst. *Arch Pathol* 1950; 50:468.
 24. Barron S H, Emanuel B. Adrenal cyst: a case report and a review of the pediatric literature. *J Pediatr* 1961; 59:592.
 25. Ghandur-Mnaymneh L, Slim M, Muakasa K. Adrenal cysts: pathogenesis and histological identification with a report of 6 cases. *J Urol* 1979; 122:87-91.
 26. Tait D L, Williams J, Sandstad J, Lucci J A III. Benign adrenal cyst presenting in a pregnant patient. *Am J Perinatol* 1997; 14:461-4.
 27. Trauffer P M, Malee M P. Adrenal pseudocyst in pregnancy. A case report. *J Reprod Med* 1996; 41:195-7.
 28. Winfield H N, Hamilton B D, Bravo E L, Novick A C. Laparoscopic adrenalectomy: the preferred choice? A comparison to open adrenalectomy. *J Urol* 1998; 160:325-9.
 29. Filippini S, Guerrieri M, Arnaldi G, Giovagnetti M, Masini A M, Lezoché E, et al. Laparoscopic adrenalectomy: a report on 50 operations. *Eur J Endocrinol* 1998; 138:584-93.
 30. Miyake O, Yoshimura K, Yoshioka T, Honda M, Kokado Y, Miki T, et al. Laparoscopic adrenalectomy. Comparison of the transperitoneal and retroperitoneal approach. *Eur Urol* 1998; 33:303-7.
 31. Gill I S, Soble J J, Sung G T, Winfield H N, Bravo E L, Novick A C. Needlescopic adrenalectomy—the initial series: comparison with conventional laparoscopic adrenalectomy. *Urology* 1998; 52:180-6.