Adrenal Pseudocyst with Atypical Imaging Appearance: two cases report

JUI-LUNG FANG1  JER-SHYUNG HUANG1  HUAY-BEN PAN1  I-YIN LU2  SHIH-CHENG CHOU3

Department of Radiology1, Surgery2, Pathology3 Kaohsiung Veterans General Hospital

Adrenal pseudocysts are rare adrenal lesions with reported typical and atypical imaging appearance. We report two adult cases and both of them have atypical appearance. Especially in our first case, the huge low attenuation mass has a non-enhanced irregular thick wall, calcified spots and multiple enhancing mural nodules, and occupies the space between liver and right kidney. Lipiodol embolization and surgical resection are performed in two medical centers respectively under the imaging diagnosis of hepatoma. The unusual presentations deserve our careful discussion.

Key words: Adrenal pseudocyst; Hepatocellular carcinoma

Adrenal pseudocysts are rare adrenal lesions. Both typical and atypical appearances have been reported in the literatures. We report 2 cases of adrenal pseudocysts. One of them showed very unusual appearance with thick cystic wall, enhancing mural nodules and indistinct fat plane between the mass and liver. The other one is a thin-walled, huge right adrenal cystic lesion. The pathology, etiology, typical and atypical imaging appearances of adrenal pseudocysts are discussed.

Case 1

A 71-year-old male is a patient with hypertension and non-insulin dependent diabetes mellitus under medical control for one year. He suffered from right abdominal dull pain for three months and visited another hospital for help. After series of study, he was treated under the impression of hepatoma. Abdominal angiography with lipiodol embolization from right inferior hepatic artery was performed at that time.

Routine laboratory investigations showed mild anemia and chronic renal insufficiency. Computerized tomography showed a huge low attenuation mass with non-enhanced irregular thick wall, calcified spots or residual lipiodol and multiple enhancing mural nodules located just below the capsule. This mass occupies the space between the liver and right kidney, with suspicion of right portal vein invasion and indistinct fat plan between the liver, right kidney and adrenal gland (Fig 1, 2 and 3).

Exploratory laparotomy revealed a huge well-encapsulated mass, measuring about 20 cm in diameter, with cavitation or central necrosis over the right upper retroperitoneum. Severe adhesion to the kidney and liver was also identified. The right lobe of liver was markedly atrophic because of compression by the mass. Right radical nephrectomy and adrenalectomy were performed at laparotomy.

The specimen consists of a resected right kidney and a hemorrhagic adrenal gland. The cyst, measuring about $20 \times 17 \times 10$ cm, is composed of a dense fibrous capsule and remnants of yellow adrenal tissue. The
cyst contains yellow brown and bloody amorphous material.

Histologically, the cystic specimen is composed of dense fibrous connective tissue with focal calcification and adipose tissue metaplasia. However, no cellular lining is noted. The cyst contains fibrins and organizing hemorrhage. The diagnosis is adrenal pseudocyst (Fig 4).

Case 2
A 47-year-old woman suffered from epigastralgia and right flank dull pain for a period of time. She lived uneventfully before, apart from receiving total hysterectomy for uterine myoma 4 years ago. The systematic inquiry was unremarkable. There was no evidence of endocrinological disorder.

Figure 1. a. Precontrast CT shows a huge low attenuation mass with irregular thick wall and subcapsular calcified spots occupying right upper abdomen. b. Early arterial phase of contrast enhanced CT reveals enhanced subcapsular mural nodules and non-enhanced peripheral thick wall. c. Late venous phase shows most of the cyst wall is not enhanced.

Figure 2. Lower level than fig1. , postcontrast CT reveals non-enhanced soft tissue component and well-enhanced subcapsular wedge-shaped mural nodules.
Figure 3. a, b, c. Higher level than fig1. precontrast, early arterial phase and late venous phase of CT respectively, show that the lesion can not be well demarcated from liver.

Routine laboratory data were normal. Ultrasound showed a huge homogenous hypoechoic mass over right suprarenal region, about 10 cm in size. Right adrenal or retroperitoneal tumor was considered. Computerized tomography showed a slightly high attenuation cyst-like lesion over right upper retroperitoneum with indistinct fat plane between this lesion and the upper pole of the right kidney. Complicated right renal cyst was suspected on the basis of CT findings (Fig 5).

At laparotomy, a huge cyst, about 10 cm in diameter, derived from right adrenal gland was noted. The right kidney was separated from the adrenal cyst completely and about 400 ml of clear yellowish fluid was drained out before excision of this adrenal cyst and right adrenal gland.

Histologically, the cystic specimen is composed
of fibrous connective tissue and no cellular lining is noted. It contains blood clots without evidence of malignancy in the specimens examined. The diagnosis is adrenal pseudocyst.

**DISCUSSION**

Adrenal cysts are uncommon adrenal lesions [1]. They have multiple etiologies including cystic degeneration of cortical or medullary neoplasms, endothelial-lined cysts, and cysts secondary to infectious agents as well as pseudocysts [2]. Adrenal pseudocyst is defined as a non-neoplastic, nonparasitic cyst of the adrenal gland in which an epithelial or endothelial lining is not demonstrated. Non-neoplastic cysts of the adrenal gland have been divided into four categories: parasitic, epithelial, endothelial, and hemorrhagic pseudocyst [3]. True cysts have endothelial or epithelial linings, reflecting their origins. Endothelial cysts represent the predominant type, constituting of 45% of all adrenal cysts. They typically originate from lymphatics but rarely can derive from capillaries as well. Epithelial cysts and cystic adenomas are less common, constituting 9% of cysts. Pseudocysts represent the second most common type but are most frequently recognized clinically [3].

The adrenal pseudocysts are usually discovered incidentally at autopsy in earlier studies. Recent studies report that abdominal and/or flank dull pain may be the leading complaints, as that in our two patients, and their symptoms usually resolve following surgical removal [1,2,3,4,5,6]. The size of pseudocyst does not correlate well with the presence of symptoms [2]. Other reported clinical presentations include shock caused by massive hemorrhage [7], infection caused by infected hemorrhagic adrenal pseudocyst [8], and hypertension [9] etc.

On CT, the typical findings of adrenal pseudocysts in earlier studies are usually small, unilateral, and sharply demarcated lesions of homogenous water density. On ultrasonography, they are usually described as an anechoic cyst with well-defined wall and posterior acoustic enhancement [10,11]. In recent years, more and more so-called atypical imaging appearance of adrenal pseudocysts is reported, including: large size, thick wall, prominent septations, enhancing mural nodule, and irregular calcifications [10,11,12,13]. However, an adrenal pseudocyst with thick irregular wall and multiple enhancing mural nodules, as those in our first case, has never been reported. In our first case, it is very difficult to define the origin and nature before operation. The solid components of the irregular thick wall have been explained as resolving hematoma [10,14]. However, the cause of enhancing mural nodules has not been well explained. With the correlation of histopathology, we find that the subcapsular enhancing mural nodules seen on the CT are the nodular remnants of the adrenal gland.

According to initial presentation of our two cases, we have difficulty to define the origin of the lesion because of the large lesion sizes, the concomitant compression effect to both liver and kidney, and the limited scanning planes of CT scans. In lesions located between the liver and the right kidney, MR study may provide more information to define the tumor origin because of its multiplanar capability.

In general, asymptomatic adrenal pseudocysts need no treatment. Large cysts may be aspirated and

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*Figure 5. a. Precontrast CT reveals a huge, thin-walled low attenuation cyst-like lesion occupying the space between liver and right kidney. b. Contrast enhanced CT reveals slightly enhanced cyst wall.*
emptied by percutaneous fine needle aspiration. This, however, may result in bleeding, thus turning a non-symptomatic cyst into a symptomatic one. Indications for surgery of the adrenal pseudocyst are large, complicated cysts, parasitic cysts, functional, or malignant cysts [15].

In conclusion, we report two cases of giant right adrenal pseudocysts with atypical imaging appearance. Both of them are huge cystic masses with indistinct fat planes to the liver and right kidney, and one of them has irregular thick wall, calcified spots and multiple enhancing mural nodules that may be misinterpreted as a malignant liver tumor. It should be kept in mind that adrenal pseudocysts may have many atypical presentations.

**REFERENCES**

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腎上腺假性囊腫的非典型影像表現：病例報告

方瑞隆¹ 黃哲勳¹ 潘慧本¹ 盧怡吟² 周士程³

高雄榮民總醫院 放射診斷科¹ 一般外科² 病理檢驗科³

腎上腺假性囊腫是罕見的腎上腺疾病，而在歷史文獻曾經報導在影像診斷上有多種所謂典型以及非典型的表現，我們在此報告兩個發生在成人身上的個案，在這兩個個案均擁有報導過的所謂非典型影像表現，尤其第一個個案曾經在三家醫學中心被診斷為肝臟惡性腫瘤而分別被施以腫瘤血管栓塞以及手術切除的治療，其在影像診斷學上的特殊表現值得我們深入探討。

關鍵詞：腎上腺假性囊腫；肝臟惡性腫瘤