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A Case Report of Giant Adrenal Cyst with Spontaneous Rupture and Hemorrhage

Qi Xu[1], Wanghong Sun[1], Lide Song[1],*

[1]Urology Surgery Department, Zhuji People's Hospital of Zhejiang Province, Zhuji, 311800, China
**Correspondence: lyt108529@163.com*

Abstract: *A case of huge adrenal cyst with spontaneous rupture and hemorrhage was reported. A 27-year-old female patient came to our hospital in emergency department due to "right upper abdomen and right low back pain and discomfort for 2 days". Whole abdominal enhanced CT revealed a right adrenal cyst with rupture and hemorrhage. An emergency exploratory laparotomy revealed a huge right retroperitoneal hematoma with a size of 25.0cmx20.0cmx18.0cm. Combined with pathology, adrenal endothelial cyst with bleeding was considered. To investigate the follow-up, diagnosis and treatment of giant adrenal cyst based on the literature.*

Keywords: *huge adrenal cyst, spontaneous rupture, hemorrhagic shock*

Adrenal cysts are relatively rare in clinic, and often have no specific clinical manifestations in the early stage. They are occasionally found in physical examination. The late clinical manifestations are mostly caused by cyst enlargement and compression of surrounding organs. Abdominal pain is the most common. Other symptoms such as gastrointestinal symptoms, hypertension and hormone abnormalities are rare. Hemorrhagic shock caused by spontaneous rupture and hemorrhage is even rarer[1]. In June 2020, Urology Surgery Department, Zhuji People's Hospital of Zhejiang Province, admitted a patient with a huge adrenal cyst with spontaneous rupture and hemorrhage. The report is as follows:

1. Case report

The patient, a 27-year-old female, came to the emergency hospital on June 13, 2020 because of “pain and discomfort in the right upper abdomen and right back for 2 days”. The patient suffered from pain and discomfort in the right upper abdomen and right waist and back without obvious inducement 2 days before admission, which was not severe, accompanied by nausea, vomiting and low fever (T37.4 ℃). One day ago, the patient’s pain was aggravated, affecting the whole abdomen, accompanied by dizziness and fatigue. Physical examination on admission: T37.4 ℃, pulse 120 times/minute, blood pressure 90/55mmHg, clear mind, irritability, anaemic appearance, abdominal bulge, a lump palpable under the right costal region, medium texture, tenderness of the whole abdomen, acute right upper abdomen, no rebound pain, positive percussion pain in the right renal region, negative percussion pain in the left renal region, unfilled bladder, negative mobility dullness. In the past three months, abdominal color ultrasound showed that the liver was normal in shape, normal in size, smooth in surface, and homogeneous in liver parenchyma. The size of the right lower lobe of the liver was about 11.6 cm x 8.7 cm x 10.0 cm, with no echo, and high in echo, with a range of 2.0 cm x 0.9 cm. The gallbladder was normal in size, smooth, and without obvious abnormal signals. The pancreas and spleen were normal in size, intact in envelope, smooth in surface, and clear in renal cortex and medulla. Therefore, the patient may have a liver cyst with hyperechogenicity (hemorrhage). The blood routine examination at the emergency admission showed that the red blood cell count was 2.24x1012/L, and the hemoglobin was 65.0g/L. Full-abdominal contrast-enhanced CT showed that the surface of the liver was smooth, and the proportion of each lobe was uniform. A mass of mixed density shadow with the size of about 13.9 cm x 14.0 cm was seen in the right adrenal area (Figure 1). The internal density was uneven. After the enhancement scan, the focus was not significantly enhanced, the intrahepatic blood vessels were not clearly displayed, the intrahepatic bile duct was not dilated, the gallbladder wall was not thickened, and no stone shadow was found; Spleen is not swollen, with uniform density. Admission diagnosis: right adrenal cyst with rupture and hemorrhage, hemorrhagic shock. Emergency exploratory laparotomy showed a small amount of bloody fluid in the abdominal cavity, and the shape and color of the liver and spleen were good. The right retroperitoneal giant hematoma is about 25.0cm x 20.0cm x 18.0cm in size, with high tension, which compresses the lower edge of the liver upward and adheres to

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the lower edge of the liver. During the operation, first free the liver curvature of the colon and the lower edge of the right liver, push the liver upward, and then free the inferior vena cava, expose the central vein and disconnect it after triple ligation. Open the retroperitoneum transversely at the upper edge of the right kidney, and see about 2000ml dark red blood clot behind the peritoneum. After clearing the blood clot, see the rupture and hemorrhage of the right adrenal giant cyst, and the right kidney is compressed and moved downward, and the shape and color of the kidney are good. Retain part of the gland of the right adrenal gland, separate the adhesion around the cyst, ligate the surrounding blood vessels, and remove the right adrenal cyst (Figure 2). It is estimated that the cumulative blood loss of the patient is about 3000ml, and a total of 8.5 units of suspended red blood cells and 1000ml of fresh frozen plasma were infused during the perioperative period. After the operation, the patient was sent to the intensive care unit for anti-inflammatory fluid replacement and anti-shock treatment. The vital signs were stable and the condition gradually improved. Postoperative pathological report: the fibrous capsule wall tissue is lined with flat cells, and immunohistochemical staining shows F8 positive. Irregular lamellar hemorrhage can be seen in the capsule wall, and pink liquid and blood can be seen in the capsule. Adrenal endothelial cyst is considered (Figure 3). The patient came to the hospital for follow-up one month after the operation. The patient had no pain and discomfort, and no obvious abnormality was found on the CT plain scan of the whole abdomen.

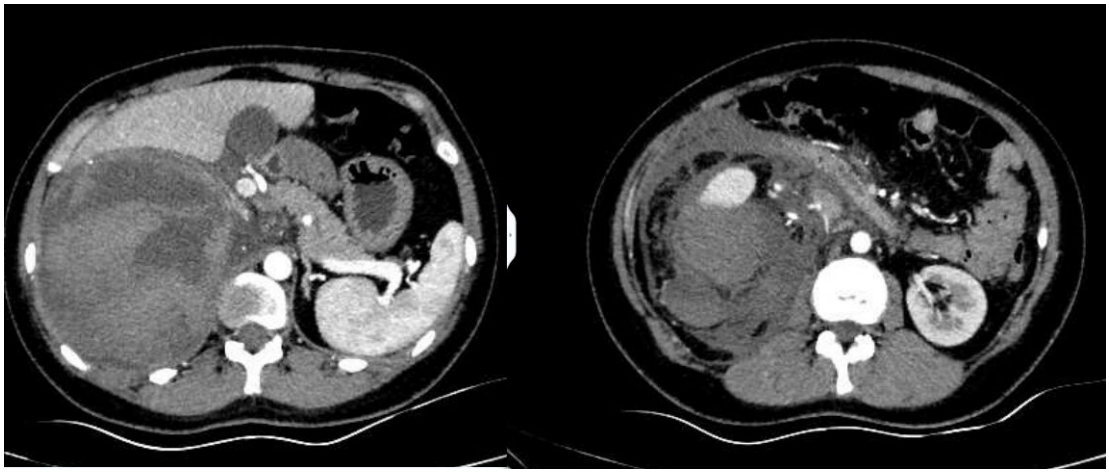

Figure 1: Full-abdominal contrast-enhanced CT: The right adrenal region has a huge mass with hemorrhage, adjacent to the liver and kidney.

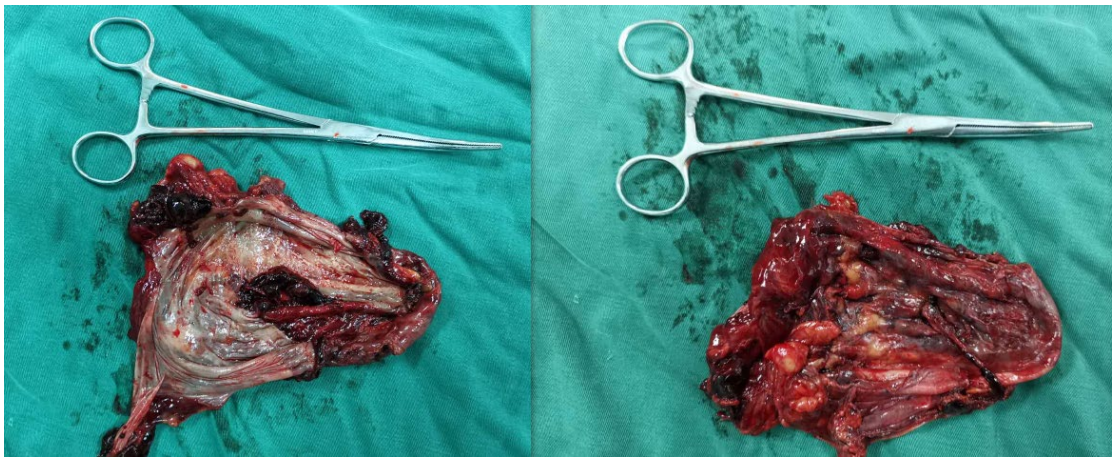

Figure 2: Adrenal cyst specimen: resected adrenal cyst, medial (left) and lateral (right)

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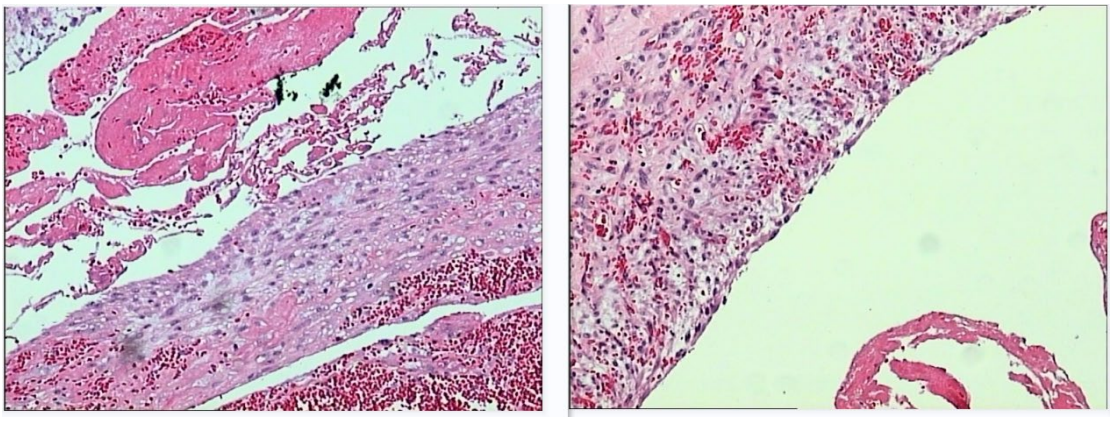

Figure 3: Pathology (HE × 100): The fibrous capsule wall tissue is lined with flat cells, irregular lamellar hemorrhage can be seen in the capsule wall, and pink liquid and blood can be seen in the capsule.

2. Discussion

Adrenal cysts are relatively rare in clinic. The age of onset is mainly 30 to 50 years old. The proportion of female patients is high, and the incidence of autopsy is even less than 0.2%[2]. In 1966, Foster [3] classified it into four types according to histological type and incidence rate: endothelial cyst (45%), pseudocyst (39%), epithelial cyst (9%) and parasitic cyst (7%), which is currently recognized as a classification method. Most patients have no specific clinical manifestations [4]. It is difficult to diagnose through medical history, physical examination and blood biochemical hormone examination, and often depends on imaging examination[5]. With the popularization of color Doppler ultrasound, CT, MRI and other auxiliary examinations, the diagnostic rate of adrenal cyst has gradually increased. Large adrenal cysts are prone to misdiagnosis. Before diagnosis, it is not only necessary to differentiate them from the surrounding cysts (liver, spleen, kidney, pancreas), but also from the cystic lesions of adrenal tumors. In clinical practice, multiple imaging methods are often used in combination with adrenal function examination to locate and determine the nature[6]. In this case, liver cyst was considered by color Doppler ultrasound in the past, which interfered with the follow-up diagnosis and treatment. The causes of misdiagnosis are as follows: ① The adrenal cyst of the patient is large, and it is closely adhered to the liver by pressing the lower edge of the liver. A single imaging examination (such as color Doppler ultrasound) cannot determine the origin of the cyst; ② Before the cyst rupture and hemorrhage, the patient did not show relevant clinical manifestations and laboratory abnormalities; ③ Giant adrenal cyst is rare, and clinicians have little experience, which has not attracted attention and further differentiation. The diagnosis of adrenal cyst with hemorrhage is not easy. The etiology is mostly related to pelvic and abdominal trauma, but there are also reports of spontaneous hemorrhage caused by stress, tumor and coagulation dysfunction [7]. This patient is a young woman with no definite history of pelvic and abdominal trauma before the onset of the disease. It is considered as adrenal cyst with spontaneous rupture and hemorrhage.

Adrenal cyst is a benign disease. At present, scholars have not reached a consensus on the treatment of adrenal cyst. Most scholars believe that the treatment principle is to avoid potential complications and reduce the risk of malignancy, so comprehensive evaluation is very important. The evaluation indexes include cyst volume, nature, clinical manifestation and complications. For asymptomatic patients with diameter less than 5cm, regular follow-up is often used; for cysts with diameter greater than 5cm, severe compression symptoms, endocrine dysfunction and possible malignant change, surgical treatment should be taken [8][9]. With the development of minimally invasive technology, laparoscopic adrenal cyst decapitation has become the preferred surgical method because of its small surgical trauma, rapid postoperative recovery, and preservation of normal adrenal tissue. Open surgery can also be used for patients with large volume[10].

With the popularization of physical examination, the detection rate of occult adrenal cyst is higher and higher. Its differential diagnosis and follow-up evaluation play an important role in diagnosis and treatment. Simple ultrasound examination has the risk of misdiagnosis. It needs to be combined with CT, MRI and other imaging examinations, and the final diagnosis needs pathological examination. Because the incidence rate of the disease is low and there are few cases reported in the literature, the

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optimal treatment is still controversial [11], but for cases with compression symptoms and asymptomatic cases with large volume or abnormal adrenal hormone, surgery is still the preferred treatment.

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