

Case Report

Clinical characteristics and treatment of mature adrenal teratoma: a case series

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Abstract: Mature teratoma of the adrenal glands is rare, but more common in females and young people. We investigated the clinical characteristics and treatment efficacy of 10 patients. Six patients had a mass in the left adrenal gland and 4 patients had a mass in the right gland. Two patients presented with flank pain and 8 had adrenal masses incidentally discovered during physical examination. No patient had a history of hypertension. Plain CT scans revealed 3-9 cm round or dumbbell-shaped adrenal gland masses with mixed cystic and solid components. Plaque-like or arc-shaped calcifications and lesions with fat-like density were visible in the masses. Contrast CT showed enhanced tumor margins and soft tissue components inside the masses. All patients underwent adrenalectomy, and tumors were well-circumscribed and poorly vascularized. Microscopic examination revealed calcification, cartilage, fat, squamous epithelium, and sweat gland. Mature adrenal teratoma exhibits atypical clinical symptoms, and preoperative diagnosis primarily depends on imaging tests. Treatment with surgical resection provides a good prognosis.

Keywords: Teratoma, adrenal tumor, clinical features, surgical resection

Introduction

Teratomas are tumors derived from germ cells. They often contain multiple tissue components, including the ectoderm, mesoderm, and endoderm, and the cells are arranged disorderly. These cells can differentiate into tissues such as skin, nerves, fat, and teeth. Teratomas are mostly found in the testes and ovaries. Extragonadal teratomas, such as adrenal teratomas, are rare [1]. Due to insufficient understanding or atypical imaging findings, they are often misdiagnosed as adrenal lipomas, cysts, myelolipomas, and adrenal carcinomas. In this study, the clinical and follow-up data of 10 patients with mature adrenal teratomas were collected and analyzed and the efficacy of surgical treatment was evaluated.

Case presentation

Records were retrospectively reviewed for 10 patients with mature adrenal teratoma who were admitted to our hospital from January 2012 to December 2019. All 10 patients under-

went CT examinations and were routinely tested for complete blood work, renal and hepatic function, and electrolytes. Testing for catecholamines and aldosterone was performed. The diagnoses of all patients were confirmed by postoperative pathology. Patients were followed for a minimum of 10 months and data concerning demographics, presentation, surgical treatment, and pathology were tabulated and analyzed. This study was approved by the ethical review board of the hospital. Informed consent was not required due to the retrospective design of the trial.

The patients included 8 females and 2 males aged 21-51 years. Two patients presented with ipsilateral flank pain and 8 patients had incidental discovery of an adrenal mass during physical examination. None of the 10 patients had a history of hypertension. Clinical features of the patients are shown in **Table 1**. The results of routine laboratory testing, including complete blood work, hepatic and renal function tests, and electrolytes, were normal in all

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Table 1. Clinical features of the patients

| Case | Sex | Age (years) | Presentation | Location | Size (cm) | Surgery |
|------|-----|-------------|----------------------|----------|-----------|--------------|
| 1 | F | 21 | Incidental discovery | Right | 3.9 | Laparoscopic |
| 2 | F | 34 | Incidental discovery | Right | 4.7 | Laparoscopic |
| 3 | F | 43 | Incidental discovery | Left | 7.8 | Open |
| 4 | F | 29 | Flank pain | Left | 9.0 | Open |
| 5 | M | 30 | Incidental discovery | Right | 3.5 | Laparoscopic |
| 6 | F | 38 | Incidental discovery | Left | 5.6 | Laparoscopic |
| 7 | F | 47 | Flank pain | Right | 7.5 | Laparoscopic |
| 8 | M | 37 | Incidental discovery | Left | 6.2 | Laparoscopic |
| 9 | F | 51 | Incidental discovery | Left | 6.8 | Laparoscopic |
| 10 | F | 37 | Incidental discovery | Right | 4.5 | Laparoscopic |

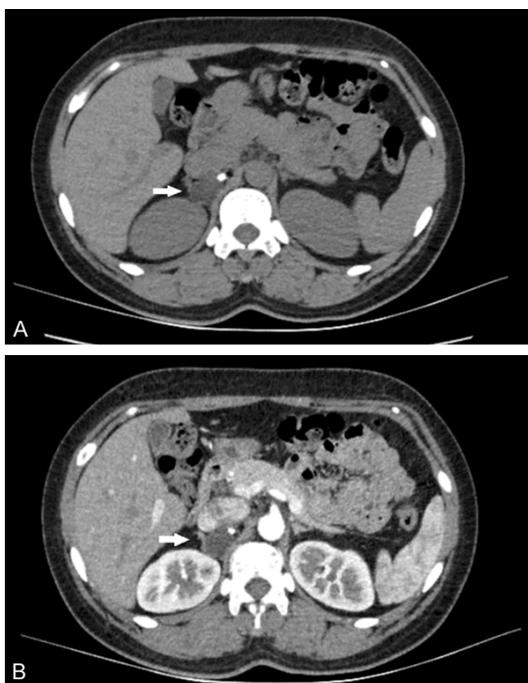


Figure 1. Representative computed tomography results. A. Plain CT scan shows irregular soft-tissue density in the right adrenal gland; the density is uneven, and calcified lesions, fat, and soft-tissue shadows can be seen. B. Contrast CT scan of the same patient shows enhancement of the mass margins in the right adrenal gland, and internal septations appear to be present.

patients. The results of adrenal function tests were also normal. CT scans showed that the masses were located in the adrenal gland region. They appeared round or dumbbell-shaped, with mixed cystic and solid components. The diameters ranged from approximately 3 to 9 cm. Calcified lesions were visible

in the masses, which were mostly plaque-like or arc-shaped. Internal septations could be seen in some masses. Lesions with fat-like density could also be observed in the masses (**Figure 1A**). Contrast CTs showed enhancement in tumor margins and soft tissue components inside the masses (**Figure 1B**).

Open surgical resection was conducted in 2 patients with large adrenal tumors, and laparoscopic resection was per-

formed in the remaining 8 patients. During the surgeries, all tumors were found to be encapsulated. The tumors were mainly solid, but some had cystic components. The tumors were poorly vascularized and were mostly well-demarcated, with clear boundaries and an absence of tight adhesions with surrounding tissues. The blood pressure of the patients did not fluctuate significantly when the tumors were palpated during the surgery.

Gross examination of the specimens showed that 6 tumors contained jelly-like or mucus-like substances as well as soft tissue masses, and 5 tumors had calcified or skeletal-like tissues (**Figure 2**). Smooth muscle tissue and sweat gland, as well as squamous epithelium, could be observed under the microscope (**Figure 3**).

The follow-up period was 10-80 months, with a mean of 43.2 months. All 10 patients had an uneventful postoperative course, and no tumor recurrence was observed.

Discussion

Teratomas are derived from pluripotent primordial germ cells. They are mostly benign, but their malignant tendency increases with age. Teratomas usually occur in sites anterior or paraxial to the midline of the body, and are commonly found in the sacrococcygeal region, mediastinum, and gonads. Retroperitoneal teratomas account for only 1% of all teratomas [2]. Patients with adrenal teratomas are usually asymptomatic in the early stages, and the masses are often discovered during physical examination. If tumors enlarge excessively and compress the surrounding organs, or in cases

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Figure 2. Representative surgical resection and gross examination. Photograph of gross specimen shows macroscopic appearance of the excised mass; a small amount of yellow adrenal tissue can be seen in the upper border of the solid component of the tumor.

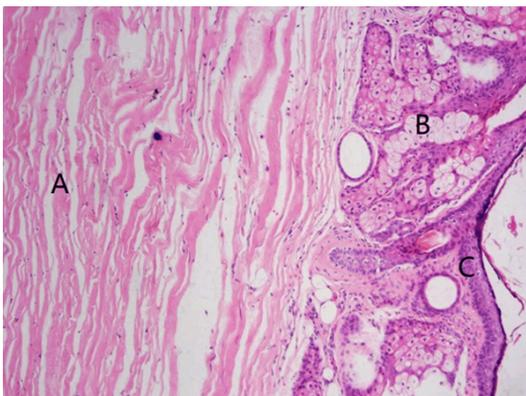


Figure 3. Representative histology. Postoperative pathology shows smooth muscle tissue (A) and sweat gland (B). Covered squamous epithelium (C) can be seen in some areas. (H&E staining $\times 100$).

of secondary infection, symptoms such as ipsilateral flank pain and paroxysmal abdominal pain may appear [3]. Adrenal teratoma is more common in females and young people. Primary adrenal teratoma is typically found in patients ≤ 35 years of age, and only a small proportion of patients are > 35 years of age [4]. To differentiate an adrenal teratoma from other tumors derived from adrenal tissue, endocrine-related hormone testing may be performed. The results of routine laboratory tests are mostly negative in adrenal teratomas [5].

Mature teratomas are pathologically benign. They are mostly parenchymal and are composed of mature, differentiated tissues. They

are encapsulated and can contain multiple cysts of different sizes. Tissues and organs derived from the ectoderm, mesoderm, and endoderm, such as hair, teeth, cartilage, sweat gland, smooth muscle, bronchi, or intestinal wall, can be found inside the teratomas. Some teratomas can undergo malignant transformation. Malignant teratomas consist of immature, undifferentiated tissues, and they manifest as abnormal proliferation of nerve tissue or epithelial tissue that exhibits invasive growth [6].

Calcification is a characteristic CT manifestation in adrenal teratoma, and mostly appears as spots, plaques, or arcs. Foci of fat density, which appear as a mixture of fat-fluid level, hair, and fatty fluid, are another typical CT finding. Contrast CT scans can also reveal that the parenchyma is slightly enhanced and the capsule and septa of the masses are significantly enhanced, while the cystic component is not enhanced. Calcification is present in 60%-80% of benign retroperitoneal teratomas, and is less common in malignant teratomas [7]. Calcification was shown by imaging and pathology in 80% (8/10) of the patients in the present study.

If a tumor in the adrenal gland demonstrates uneven density and contains fatty and calcified lesions, a diagnosis of adrenal teratoma should first be suspected. Mature adrenal teratoma must be differentiated from other nonfunctional adrenal tumors, such as adrenal myelolipoma, ganglioneuroma, and adrenal cysts. Adrenal myelolipoma is composed of adipocytes and bone marrow cells. Plain CT scan reveals fat density interspersed with linear septa. Contrast CT scan does not reveal additional features. The plain CT scan of an adrenal ganglioneuroma shows a mass with uniform soft-tissue density. Pathologically, the tumor is mainly composed of spindle-shaped nerve fiber bundles. The blood vessels are narrow and small in quantity. Therefore, the mass is not enhanced or only slightly enhanced with contrast scan. Pathologically, adrenal cysts are cystic lesions. As the cysts are fluid-filled, CT shows a uniform water-like density. The wall is thin and smooth, and there is no enhancement with contrast scan [8].

Although mature adrenal teratomas are benign tumors, tumors that grow to excessively large sizes may compress the surrounding tissues

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and organs. This may lead to corresponding symptoms and affect the normal functioning of the organs. Meanwhile, there is also a potential for malignant transformation. Therefore, adrenalectomy should be scheduled as soon as adrenal teratoma is diagnosed [9]. For small tumors or tumors without tight adhesions, laparoscopic adrenalectomy may be performed; for patients with large tumors or tight adhesions, open surgical resection may be performed [10]. As mature teratomas contain well-differentiated tissue, complete resection leads to an improved prognosis with little risk of recurrence and metastasis [11].

In summary, mature adrenal teratoma is a rare disease with atypical clinical symptoms and laboratory abnormalities. Imaging tests may facilitate the diagnosis of the disease, but pathological examination is required for a confirmed diagnosis. Complete resection of the tumor results in good prognosis. Additional study of these rare cases is necessary to assess longer-term outcomes and investigate potential signs that may facilitate earlier diagnosis.

Disclosure of conflict of interest

None.

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