



Teratoma of the adrenal gland: clinical experience and literature review

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Abstract: Teratoma originates from pluripotent cells of two or more than two germ cell layers, and most of them are benign. Teratomas are found in the ovaries and testes. Retroperitoneal teratoma is rare, especially adrenal teratoma. Here, we describe a rare case of a 17-year-old woman who was diagnosed with pulmonary tuberculosis and a right adrenal mass at the age of eight. So, she received anti-tuberculosis treatment. Nine years later, chest X-rays showed prior lesions in both lungs, and abdominal CT showed the mass in the right adrenal gland was larger than before, during this period she had no clinical symptoms. She underwent retroperitoneal laparoscopic adrenalectomy, and the pathological diagnosis was a mature teratoma of the right adrenal gland. During a one-year follow-up, the patients recovered well without any discomfort. Thirty-two cases were found in the literature review, among which no patients had a history of pulmonary tuberculosis. Adrenal teratoma is often seen in females and the left adrenal gland. The imaging features of adrenal teratoma can be cystic, solid, and cystic solids. Mature fat and calcification can be seen in most teratomas. Comprehensive analysis of clinical features and imaging characteristics can enhance the diagnostic confidence of radiologists in adrenal teratoma.

Keywords: Case report; adrenal teratoma; tuberculosis; imaging

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Introduction

Teratomas are most common from ovaries and testes, and patients usually have no overt clinical symptoms. Most teratomas are found in health examinations or gynecological examinations. Teratoma can also occur in the mediastinum, intracranial, sacrococcyx, and retroperitoneal space. Nevertheless, retroperitoneal teratoma is rare, especially adrenal teratoma, the incidence rate of which is 0.13% (1,2). The pathological classification of teratoma includes mature teratoma and immature teratoma. Mature teratoma means that there are no malignant and immature components in the tumor, and there are no similar lesions in other parts

of the body (3). About 1.5–2% of teratoma is malignant. Immature teratoma contains immature embryonic tissue, about 26% of which is malignant (4).

The imaging characteristics of adrenal teratoma are partly like those of adrenal angiomyolipoma, myelolipoma, and adenoma (5,6). It is difficult to diagnose before the operation (7,8). Here, we describe a rare case of a 17-year-old woman who was diagnosed with pulmonary tuberculosis and right adrenal mass at the age of eight. Nine years later, she underwent retroperitoneal laparoscopic adrenalectomy with a pathological diagnosis of a right adrenal teratoma. We summarized the clinical

manifestations and CT characteristics of adrenal teratoma and conducted a review of the relevant literature, and the information is summarized in *Table 1*.

We present the following article in accordance with the CARE reporting checklist (available at <http://dx.doi.org/10.21037/gs-20-648>).

Case presentation

The 17-year-old Chinese woman first appeared in our hospital in July 2010, when she was only eight years old. CT showed a spherical mass of about 3.2 cm × 3.2 cm × 4.0 cm in the right adrenal region, with a clear and smooth boundary and multiple calcifications in the mass. The radiologist diagnoses adrenal myelolipoma or neuroblastoma, and she also suffers from tuberculosis in the middle lobe of the right lung and Upper lobes of both lungs (*Figure 1*). One month after anti-tuberculosis treatment, chest CT showed the absorption of pulmonary lesions decreased, and there was no change in the right adrenal gland mass.

She reappeared in our hospital in February 2019, during this period she had no clinical symptoms. CT showed a mixed density mass of about 5.2 cm × 4.2 cm × 5.6 cm in the right adrenal region with a CT value of -133 to 2,169 HU. The mass has fat, soft tissue, and calcification. The three-dimensional reconstruction showed that there was a tooth-like high-density shadow in the mass, and the soft tissue components were enhanced on a contrast-enhanced scan. Chest X-rays showed multiple patchy and nodular high-density shadows in both lungs (*Figure 1*). Laboratory tests showed that norepinephrine: 1,174.0 pg/mL (normal range <600 pg/mL), and adrenaline, dopamine, supine position, aldosterone, and standing position aldosterone were all normal. Ferritin: 10.2 µg/L (normal range 11.0–306.8 µg/L), and carbohydrate antigen 19-9, carbohydrate antigen 153, carbohydrate antigen 125, alpha-fetoprotein, carcinoembryonic antigen were all normal. She then underwent retroperitoneal laparoscopic adrenalectomy under general anesthesia and the tumor was completely removed. The vital signs of the patients were stable during the operation, the patients received hemostasis and symptomatic support treatment after operation, and the incision healed in one stage. The postoperative gross pathology showed that the tumor was gray white cystic with a size of 7.0 cm × 2.5 cm × 2.0 cm, containing hair, teeth and bean curd residue like substances. Microscopically, skin, sweat glands, hair follicles, sebaceous glands and

adrenal tissues were seen. Pathological diagnosis was mature teratoma (*Figure 2*). Postoperative CT showed that the right adrenal teratoma was resected entirely, and no tumor recurrence or metastasis was found. During a one-year follow-up, the patients recovered well without any discomfort.

The ethics committee approved this study of The Affiliated Hospital of Zunyi Medical University, and informed consent was obtained from the patient for the anonymous use of the clinical, imaging, and histologic data for publication. All procedures performed in this study involving human participants were in accordance with the Declaration of Helsinki (as revised in 2013).

Literature review

PubMed, Medline, Web of Science, and Ovid databases were searched for English-language case reports and case series of adrenal teratoma published in the last 30 years from April 1990 to March 2020. The following keywords were used: (Adrenal Glands or Adrenal Gland or Gland Adrenal or Glands Adrenal) and (Teratoma or Teratomata). The flow chart of the literature screening process is set out in *Figure 3*. A total of 26 full-text articles involving 32 cases were included for analysis (*Table 1*).

According to the literature, before our case, only 32 cases, including 34 adrenal teratoma lesions, were reported in English-language. Adrenal teratoma patients are mainly adolescents with an average age of about 27 years. The adrenal-related hormones are normal and most of them had no clinical symptoms. Adrenal teratomas are unilateral, one case with bilateral adrenal involvement. Adrenal teratoma is often seen in females and the left adrenal gland. It should be noted that imaging features of adrenal teratoma can be cystic, solid, or cystic solid. Mature fat and calcification or bone tissue can be observed in most teratomas (*Figure 4*). Most adrenal teratomas are benign, mature teratomas, two with focal carcinoid foci. Fifteen of the 36 patients were followed up for 3 months to 4 years, with an average follow-up time of about 1 year. All patients had a good prognosis and no tumor recurrence or other discomfort.

Discussion

Teratoma is a type of germ cell tumor originating from peripheral pluripotent stem cells, which composed of tissues from the ectoderm, mesoderm, and endoderm tissues. The tumor may be solid, unicystic, multilocular, or cystic solid,

Table 1 Clinical and image features of the case of adrenal teratoma from the literature review

Case No.	Author/year/country	Sex/age	Size (cm)	Side	Presentation	Imaging findings			Histologic diagnosis	Follow-up (month)
						Morphological	Tissue composition	Contrast enhancement		
1	Lam/1999/China	F/18	11×8×7	L	GP	Solid	-	-	-	7
2	Lam/1999/China	M/17	7.5×6×3	R	GP	Cystic	-	-	MT	6
3	Lam/1999/China	F/37	10	L	GP	Cystic	Fat/fluid/calcification	-	MT	8
4	Bedri/2002/MA	F/57	8.0×7.0×5.0	L	AP	Cystic	Fat/fluid/calcification/	Enhancement	MT	-
5	Polo/2004/Spain	F/21	38×30×30	L	AP	Cystic	Fat/fluid	-	MT	-
6	Castillo/2006/Chile	M/8	8	R	GP	Solid	Fat/calcification	-	MT	3
7	Castillo/2006/Chile	F/61	8/54	L	IF	Solid	-	-	MT	-
8	Rais-Bahrami /2007/MA	M/33	5	L	IF	Cystic solid	Fat/soft tissue/calcification	-	MT	-
9	Sato/2010/Japan	M/37	20×14×13/1,530	L	IF	Cystic solid	Fluid/calcification	No enhancement	MT	-
10	Ersoz/2011/Turkey	M/8	10×8.5×6	R	IF	Cystic solid	Fat/calcification	-	MT	-
11	Giordano/2011/Italy	M/62	4.7×3	L	IF	Cystic solid	fat/calcification	-	MT	48
12	Li/2011/China	F/4	3×3	L	IF	Cystic	Fluid	No enhancement	MT	13
13	Bhatti/2013/Saudi Arabia	M/22	9×9.2×10.8	L	FP	Cystic	Fat/calcification	-	MT	6
14	Ciftci /2013/turkey	M/0.1	14×10×8	L	Vomit	Cystic	Fat/fluid	-	MT	-
15	Li/2015/China	M/49	6.7×7.0×11	R	IF	Cystic	Fat/calcification	No enhancement	MT	8
16	Nadeem/2015/Pakistan	M/19	8×6×4	R	FP	Cystic solid	Fat/calcification	-	MT	12
17	Bhatia/2016/Indira	F/24	7.6×6.5	L	FP	Solid	Fatty/fluid/calcification	-	MT	-
18	Kateoka/2016/Japan	F/32	5.1×3.6×3.4	R	IF	Cystic solid	Fat/calcification	-	MT	29
19	Naria/2016/Indian	F/2	6×5×3	R	AP	Cystic solid	-	No enhancement	MT with carcinoid tumor	8
20	Garg/2017/USA	F/0.3	10×10×8	R	AD	Cystic solid	Bony areas	-	MT	-
21	Kuo/2017/USA	M/26	8.5×5.5×4.8	R	FP	-	Fat/calcification	Enhancement	MT	-
22	Kuo/2018/USA	F/29	2.5×2.1×0.5	L	IF	-	Calcification	-	MT	-
23	Kuo/2017/USA	F/24	11.5×9.0×3.0	L	AP	Cystic	-	-	MT	-

Table 1 (continued)

Table 1 (continued)

Case No.	Author/year/country	Sex/age	Size (cm)	Side	Presentation	Imaging findings			Histologic diagnosis	Follow-up (month)
						Morphological	Tissue composition	Contrast enhancement		
24	Niu, M/2017/China	F/36	8×7×6.1	R	IF	Solid	Fat/soft tissue/calcification	Significant enhancement	MT	-
25	Pandit/2018/Nepal	F/16	12×10	L	AL	Cystic	Fluid/calcification	-	MT	-
26	Ramakant/2018/India	F/25	19×15	R	FP	Solid cystic	Fatty/calcification	-	MT	-
27	Zhou/2018/China	F/69	10×6×4	L	IF	Solid	Fat/soft tissue/calcification	-	MT	12
28	Zhou/2018/China	F/29	2.5×2.1×0.5	L	IF	-	Fluid/calcification	-	MT	12
29	Ban /2019/India	M/60	12×11×11	L	FP	Cystic solid	Fat/hyperdense internal contents/calcification	-	MT	-
			5.8×5×4.7	L	FP	Cystic solid	Fat/internal hyperdense content/calcification	-	MT	-
30	Sharma/2019/India	F/7M	9.8×9×8	L	AL	Cystic	Fat/soft tissue	No enhancement	MT	12
31	Wang/2019/China	F/22	10.4×10.1×13.2	L	IF	Cystic solid	Fat/soft tissue/bone	No enhancement	MT	-
32	Kiran/2020/India	M/2	12.3×10.5×12.5	R	AL	Cystic solid	Fat/soft tissue/bone	No enhancement	MT	10
			13×8×13	L	AL	Cystic solid	Fat/soft tissue/bone	No enhancement	MT with carcinoid tumor	10

CT, computed tomography; MRI, magnetic resonance imaging; AP, abdominal pain; IF, incidental finding; FP, flank pain; EP, epigastric pain; AL, abdominal lump; AD, abdominal distention; -, not recorded; MT, mature teratoma.

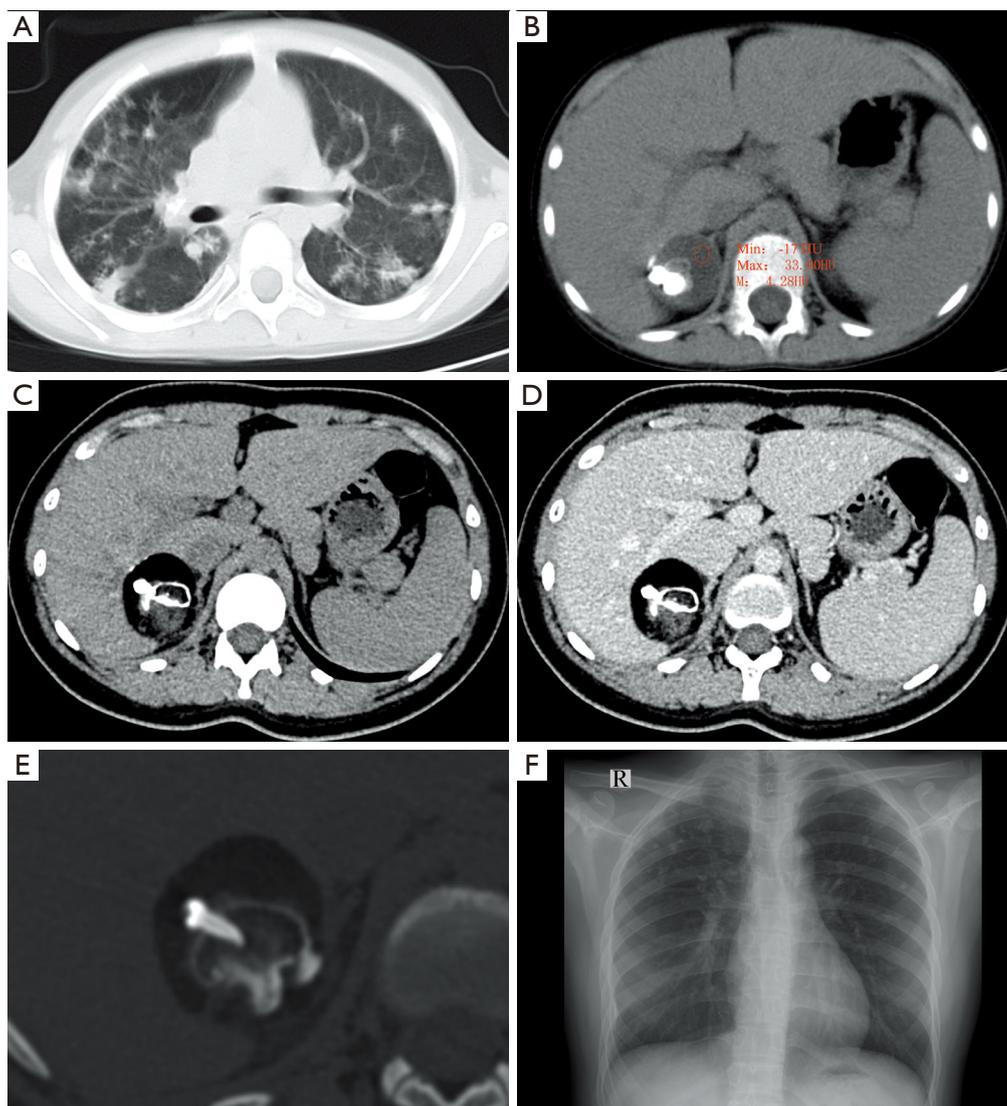


Figure 1 (A,B) A 17-year-old women at the age of 8. (A) Multiple patchy and nodular high-density shadows in the middle lobe of the right lung and upper lobes of both lungs, some of which were accompanied by punctate calcification [window width (WW) 1,382, window level (WL) -360]. (B) A spherical mass in the right adrenal region, with predominantly liquid density and multiple calcifications in the mass (WW 246, WL 65). (C,D,E,F) A 17-year-old women, CT image of upper abdomen. (C) Right adrenal mixed density mass with fat, soft tissue and calcification (WW 220, WL 40); (D) enhanced scan showed mild enhancement of soft tissue in the mass (WW 220, WL 40); (E) three-dimensional reconstruction showed there was tooth-like high-density in the mass (WW 2,612, WL 960); (F) chest X-ray, multiple patchy and nodular high-density shadows in both lungs.

containing hair, sebum, cystic fluid, calcification, or teeth (9). Teratomas are predominantly located in the gonads (testis or ovaries), 15% occurring in the extragonadal, 4% in the retroperitoneum, the adrenal gland is even rarer (10). According to the literature review, 32 cases of adrenal teratoma were reported in English-language literature

before this. Patients with adrenal teratoma usually have no clinical manifestations. Additionally, some patients presented with low back pain, abdominal mass, upper abdominal pain, among many others. Adrenal-related endocrine tests are usually regular.

The imaging characteristics of adrenal teratomas are like

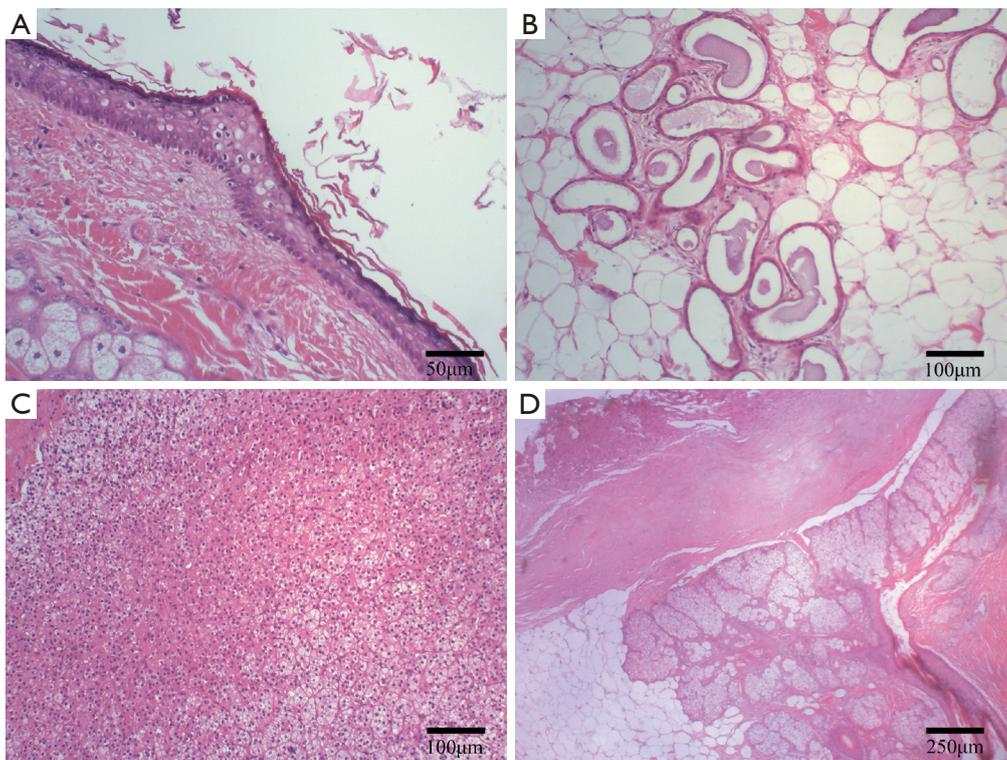


Figure 2 A 17-year-old women, microscopic examination of the mass. (A) Skin [hematoxylin and eosin (H&E) staining, magnification, $\times 200$]; (B) sweat gland (H&E staining, magnification, $\times 100$); (C) adrenal gland and keratin (H&E staining, magnification, $\times 100$); (D) adrenal and sebaceous gland (H&E staining, magnification, $\times 40$).

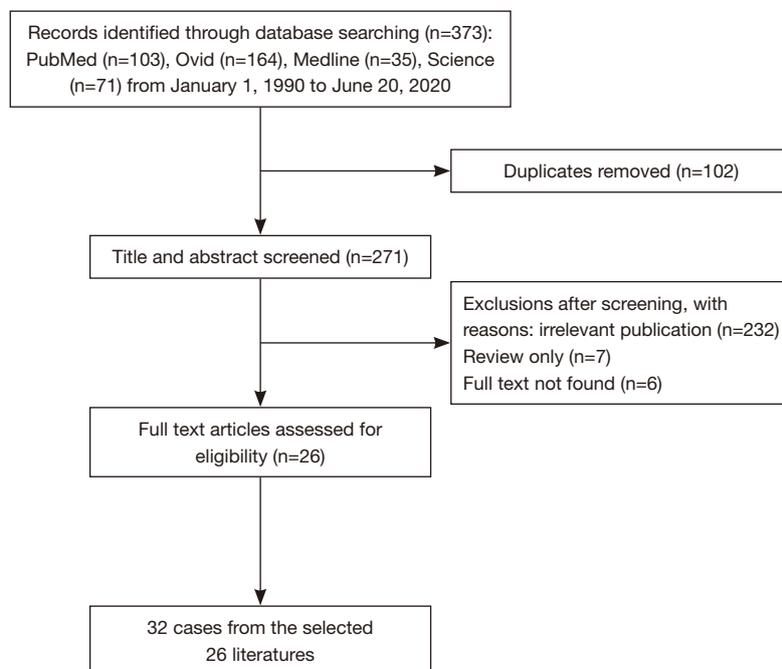


Figure 3 Flow chart of the literature screening process for adrenal teratoma.

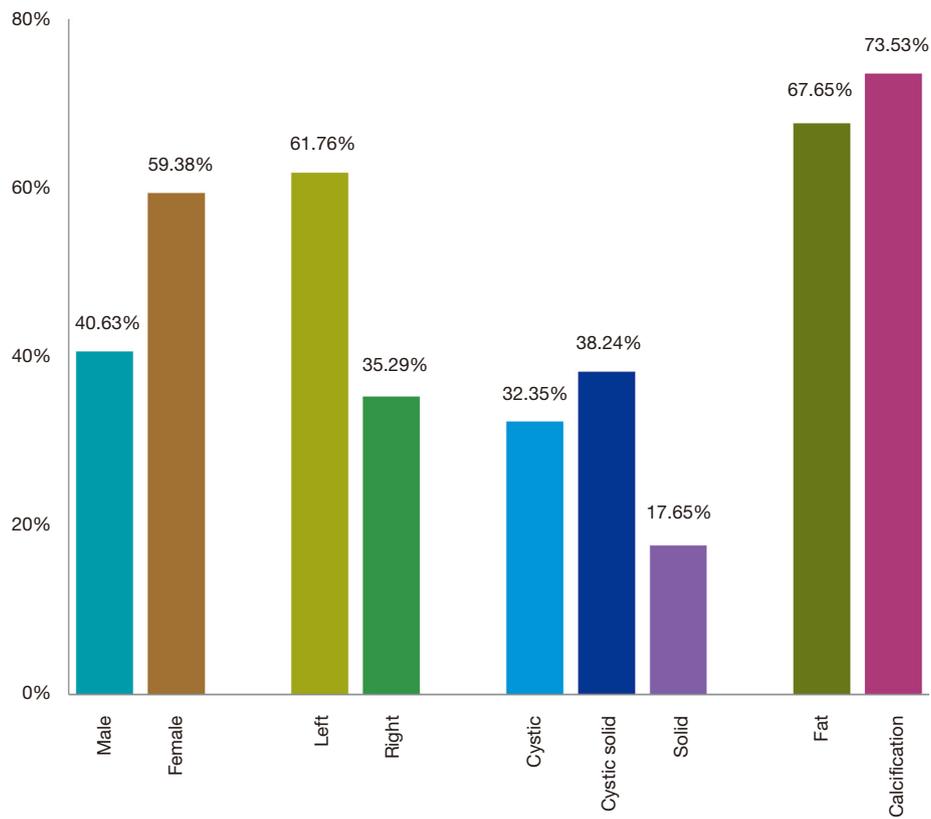


Figure 4 Clinical distribution and imaging characteristics of adrenal teratoma. Adrenal teratoma is often seen in females and the left adrenal gland. The imaging features of adrenal teratoma can be cystic, solid, or cystic solid. Mature fat and calcification can be seen in most teratomas.

those teratomas in other locations, most of which present as mixed density masses with predominant fat density/signal, and contain calcification, bone, teeth, soft tissue, and fluid components. Calcification can be punctate, radial, and linear. Soft tissue components can be accompanied by varying degrees of enhancement. The most critical preoperative diagnosis of adrenal teratoma is to differentiate it from other adrenal diseases with fat and calcification.

Differential diagnosis of adrenal teratoma and other fatty tissue containing lesions

Adrenal adenomas are the most common adrenal tumors, and about 70% of adenoma cells contain intracellular lipids (11,12). Adrenal adenomas are divided into functional and non-functional types. Functional adenomas are usually less than 3 cm with adrenal hormone abnormalities. The volume of non-functional larger adenoma, and the larger adenoma, the more likely there is a hemorrhage, necrosis,

and cystic degeneration. Most adrenal adenomas show uniform watery density, clear boundary on CT, and mild enhancement on the contrast-enhanced scan. The signal intensity of the adrenal adenoma on MR is like the liver, the signal on the out-phase image is significantly decreased, and the enhanced scan showed slight enhancement (13).

The most common adrenal lesion containing mature adipose tissue is myelolipoma, a benign tumor containing bone marrow components (14,15). Adrenal myelolipoma has no endocrine function and is often found incidentally during physical examination (16). Adrenal myelolipoma presents as a soft tissue mass, and with fat density on CT, 20% of which accompanied by calcification, and the soft tissue part is enhanced. The adipose component shows a high signal on both T1WI and T2WI on MR, while bone marrow shows a low signal on T1WI and isosignal on T2WI, the signal on fat suppression image decreases significantly. There is no signal decreasing on in-phase and out-phase images.

Adrenal angiomyolipoma is also rare. The content of these components determines the imaging findings of adrenal angiosarcoma. The larger ones (>4 cm) may be accompanied by scattered calcification, and the enhanced scan shows significant enhancement (17). The calcification of adrenal teratoma is lamellar, annular, or linear, and the teratoma has no enhancement or mild enhancement, which can supply the basis for the differential diagnosis of radiologists.

Differential diagnosis of adrenal teratoma and other calcified lesions

Metastases are the most common malignant tumors of the adrenal gland, and adrenal metastases from different sources have different imaging characteristics. Metastases from colon, breast, and small cell lung cancers can be accompanied by calcification and need to be differentiated from adrenal teratomas (18,19). Patients with adrenal metastases usually have a history of primary tumors, and most are multiple metastases with bilateral adrenal involvement. The most crucial distinguishing point between adrenal metastases and teratomas is whether the lesion has adipose tissue or not.

In the case we reported, the patient had a history of tuberculosis, so the possibility of adrenal tuberculosis must be considered. Adrenal tuberculosis is often caused by blood invasion of *Mycobacterium tuberculosis*. The clinical manifestations are low fever, fatigue, night sweats, adrenal dysfunction, among many others (20). The CT features of adrenal tuberculosis are bilateral adrenal enlargement, irregular shape, and high density (21). When there is caseous necrosis, the density of the mass is uneven and may be accompanied by an insufficient amount of calcification, complicated with large retroperitoneal lymph nodes, and circular enhancement. In the later stage, the whole adrenal gland was calcified, or part of the cheese was calcified. Contrast-enhanced scans often showed marginal enhancement, low-density casein necrosis in the center, or inhomogeneous enhancement (22).

Conclusions

In conclusion, primary adrenal teratomas are exceedingly rare. Imaging examination showed an adrenal mass with mature fat and irregular calcification or teeth, which had high value in the diagnosis of an adrenal teratoma. Comprehensive analysis of clinical features and imaging

characteristics can enhance the diagnostic confidence of radiologists in adrenal teratoma.

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Footnote

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <http://dx.doi.org/10.21037/gS-20-648>). The authors have no conflicts of interest to declare.

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