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Case Report

Teratoma involving adrenal gland - A case report and review of literature

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Abstract

Teratomas are germ cell tumors which are mainly gonadal in origin. Other common extra-gonadal sites are mediastinal, sacro-coccygeal and pineal regions. Adrenal teratomas are extremely rare and primary adrenal teratomas are even rarer. We reported a case of primary adrenal teratoma in a 60-year-old male. We reviewed literature from 2000 to till date, and found 29 adult cases and 6 paediatric cases of adrenal teratoma. Usually, they are asymptomatic and identified as an incidental finding. Imaging modality such as USG, CT and MRI are useful in diagnosis. Though these tumors are mostly benign, malignant transformation may occur. Treatment includes surgical removal.

Key words: Abdominal imaging; adrenal teratoma; computed tomography; germ cell tumor; teratoma; X-ray

Introduction

With the increasing use of cross-sectional imaging, adrenal lesions are frequently identified in routine practice and are seen in up to 5% of abdominal CTs.[1] Extra-gonadal teratomas are uncommon tumors and are less common in adults than children. These are mostly retroperitoneal in location. Moreover, primary adrenal teratomas are even rarer. Diagnosing adrenal teratomas is a challenge as these mimic myelolipomas, angiomyolipomas or liposarcomas. These tumors are mostly benign, whereas malignant transformation may occur more often in adults than children, thus becoming an important entity that requires appropriate management protocol. Here, we discuss such a case of primary adrenal teratoma identified in an elderly male. Also, a literature review regarding adrenal teratomas in adult and paediatric population published from 2000 till date, which were available on the internet, was performed.

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Case Report

A 60-year-old male presented to hospital with left flank pain and burning micturition since 2 months. Pain was colicky in nature and was relieved by medication. The patient had no history of weight loss or fever. There was no significant past or family history.

Physical examination and routine investigations were done including blood pressure and complete blood count all were within normal limits except for urine examination which revealed trace of blood.

Contrast-enhanced computed tomography (CT) scan of abdomen was performed which revealed two fat density

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lesions of size (12 × 11 × 11) cm and (5.8 × 5 × 4.7) cm in left adrenal gland. Larger one showed few heterogeneously hyperdense internal contents and bone like calcification with cortical and medullary differentiation. Smaller one also had internal hyperdense content and showed internal as well as peripheral rim like calcification [Figures 1 and 2]. Lesions showed acute angle with left kidney. Left adrenal gland was not distinctly visualised on imaging. There were few stones in left renal pelvis and left pelvi-ureteric junction [Figure 3] with left moderate hydronephrosis which explained colicky left flank pain and burning micturition. X-ray abdomen was performed, which showed peripheral and bone like calcification [Figure 4]. On imaging possibility of adrenal teratoma with second possibility of retroperitoneal teratoma was given.

Surgical resection of tumor was done along with left adrenalectomy without any intraoperative or postoperative complications. The specimen was sent for histopathological examination. On gross examination cystic mass with tooth and bone like areas and hair tuft was seen. On microscopic examination cells from all three germ layers were seen, residual compressed adrenal cortical tissue was evident in

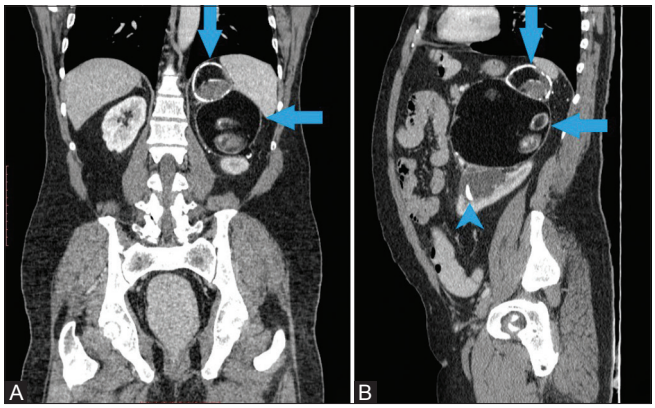

Figure 1 (A and B): Contrast-enhanced computed tomography (CECT) scan of abdomen (A) coronal view and (B) sagittal view showing two fat density lesions in left adrenal gland. Larger one (horizontal arrow) showing heterogeneously hyperdense internal contents. Smaller one (vertical arrow) showing internal hyperdense content and rim like peripheral calcification. Arrow head showing stone in lower calyx of left kidney

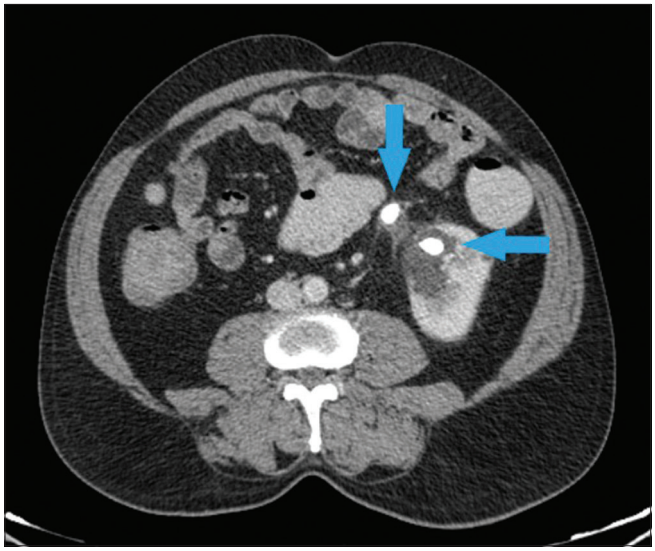

Figure 3: Contrast-enhanced computed tomography (CECT) scan of abdomen axial view showing stones in lower calyx of left kidney (horizontal arrow) and left pelvi-ureteric junction (vertical arrow)

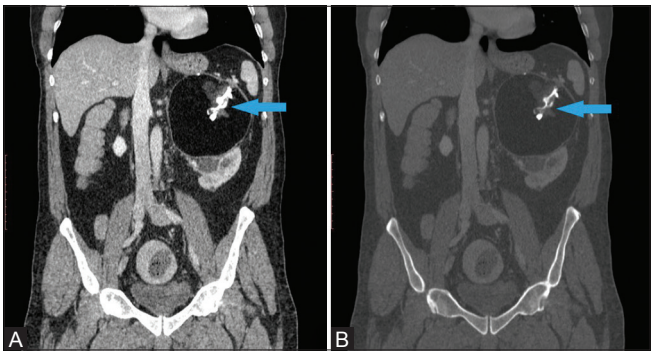

Figure 2 (A and B): Contrast-enhanced computed tomography (CECT) scan of abdomen coronal view (A) abdomen window and (B) bone window showing bone like calcification with cortical and medullary differentiation within the lesion

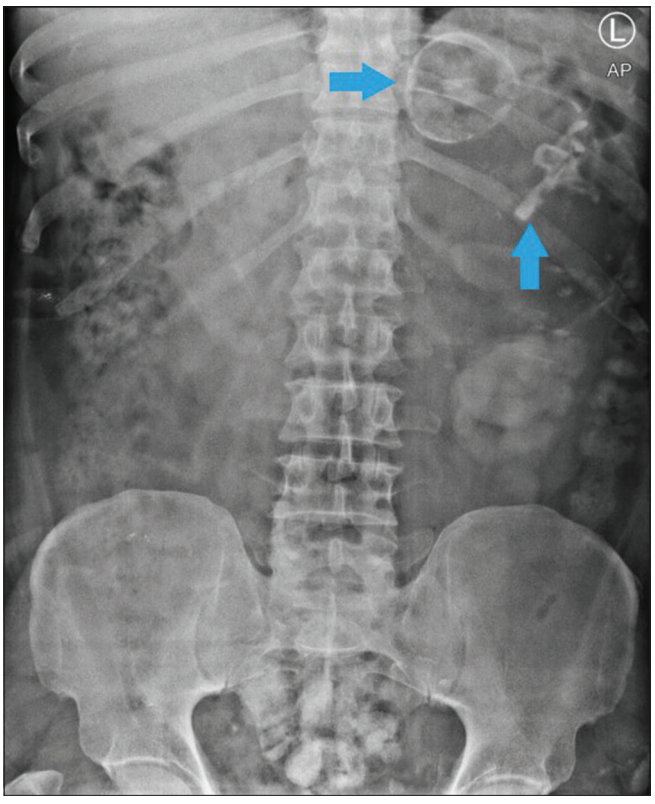

Figure 4: X ray abdomen antero-posterior view showing rim like calcification (horizontal arrow) and bone like calcification (vertical arrow)

the wall of cyst. Final diagnosis of mature cystic teratoma of adrenal gland was made.

Review of literature

We did an extensive search for recent literature—from 2000 to 2018—on the internet about adrenal teratomas in adults and children.

Cases of primary adrenal lesions were included where histopathological lesion origin was proven to be of adrenal gland. Primary retroperitoneal teratomas which involved the adrenal gland were also included. Cases where adrenal gland was found only to be compressed by the lesion or was completely uninvolved were not included in this review.

Findings of review of literature

Adrenal teratomas were more common in adults than in children (26 vs 6).

(1) Adult patients: 26 cases of adrenal teratomas were found. Majority of them were female accounting to 82.7% cases. Only 5 of them were male. 15 cases involved the left adrenal gland whereas 14 involved the right adrenal gland. All cases were surgically resected. Only 3 cases were of size less than 5 cm. Rest all were more than 5 cm Modalities used were CT, USG and MRI. CT was used in majority of cases. Typical imaging features of adrenal teratoma which include heterogeneous lesion, internal fat component and calcifications were evaluated in this review. All three features were present in 19 out of 26 cases.

Histopathological confirmation was mentioned in all cases except one [Table 1; case no - 18]. 7 cases were diagnosed as primary retroperitoneal adrenal teratomas, where their origin was not definitely identified to be from adrenal gland but adrenal gland involvement was affirmative. Among all these cases, one case of malignant transformation of teratoma and one case of rupture of adrenal teratoma in the hemi-thorax was noted.

(2) Paediatric patients: 6 cases of adrenal teratomas were found. 4 cases were female and 2 male. 5 cases were noted involving right adrenal gland. Modalities used were USG, CT and MRI. Typical imaging features of teratoma were noted in 3 out of 6 cases

All of them were surgically resected and histological proven cases. Follow-up of minimum of 6 months was noted in 3 cases, with no recurrence in any case [Table 2].

Table 1: Adult population with adrenal teratomas

Year	Case no	Age [yrs]	Sex	Site	Final Diagnosis
2018[2]	1	69	F	Left	Primary adrenal teratoma
	2	29	F	Left	Primary adrenal teratoma
2018[3]	3	25	F	Right	Primary adrenal teratoma
2017[4]	4	26	M	Right	Primary adrenal teratoma
	5	29	F	Left	Primary adrenal teratoma
	6	24	F	Left	Primary adrenal teratoma
2017[5]	7	36	F	Right	Primary adrenal teratoma with malignant transformation
2016[6]	8	14	F	Right	Primary adrenal teratoma
2016[7]	9	24	F	Left	Retroperitoneal adrenal teratoma
2015[8]	10	49	M	Right	Primary adrenal teratoma
2015[9]	11	21	F	Right	Primary adrenal teratoma
	12	16	F	Right	Primary adrenal teratoma
	13	43	F	Left	Primary adrenal teratoma
	14	49	F	Left	Primary adrenal teratoma
	15	51	F	Right	Primary adrenal teratoma
2015[10]	16	19	M	Right	Primary adrenal teratoma
2015[11]	17	29	F	Right	Retroperitoneal adrenal teratoma
2014[12]	18	39	F	Right	Retroperitoneal adrenal teratoma
2014[13]	19	21	F	Right	Primary adrenal teratoma
	20	35	F	Right	Primary adrenal teratoma
2014[14]	21	45	F	Left	Retroperitoneal adrenal teratoma
2013[15]	22	22	M	Left	Retroperitoneal adrenal teratoma
2013[16]	23	64	F	Left	Primary adrenal teratoma
2006[17]	24	61	F	Left	Primary adrenal teratoma
2004[18]	25	21	F	Left	Retroperitoneal adrenal teratoma
2002[19]	26	57	F	Left	Retroperitoneal adrenal teratoma

Table 2: Paediatric population with adrenal teratomas

Year	Case no	Age	Sex	Site	Final Diagnosis
2017[20]	1	3 months	F	Right	Primary adrenal teratoma
2016[21]	2	17 months	F	Right	Primary adrenal teratoma
2016[22]	3	2 yrs	F	Right	Retroperitoneal adrenal teratoma
2013[23]	4	3 months	M	Left	Primary adrenal teratoma
2011[24]	5	4 yrs	F	Left	Primary adrenal teratoma
2006[17]	6	8 yrs	M	Right	Primary adrenal teratoma

Discussion

Teratoma is an uncommon neoplasm with an incidence of 0.9/100,000 population.[25] Common sites for teratomas in infancy and children are extra-gonadal like mediastinal, sacro-coccygeal and pineal regions.[26-28] In adults, they are mainly gonadal. Retroperitoneal teratoma is rare and comprises of about 1% of all teratomas.[29] Adrenal teratomas are extremely rare. Adrenal teratomas form about 0.13% of all adrenal tumours.[30]

Commonly, these patients are asymptomatic with incidentally detected adrenal masses. However, sometimes they may present with vague symptoms like abdominal pain. Imaging plays an important role in diagnosis of these adrenal masses.

CT imaging usually shows a large heterogeneous lesion mainly comprising fatty components with few calcifications and absence of normal adrenal gland tissue. The differential diagnosis of adrenal teratoma includes other lipomatous masses arising primarily from the adrenal gland such as myelolipoma, lipoma, liposarcoma, angiomyolipoma, pheochromocytoma.[9,31] Also included in the differentials list are the retroperitoneal lipomatous lesions like liposarcoma

and teratoma, where adrenal gland is normally visualised or may be compressed and/partly involved. As retroperitoneal lesions can extend to involve adrenal region, it becomes difficult to determine the organ of origin in some cases.

In teratoma, calcifications can be punctate, shard-like or linear-strand with high density.[32] Attenuation of calcification higher than cortical bone is highly suggestive of teeth within the lesion.[32] These features distinguishes mature teratoma from other lipomatous tumors of the adrenal gland.[31] Calcifications can be demonstrated on 62% of plain radiographs.[32] This was evident in our case as well.

Typical imaging features include:

i. Heterogeneously echogenic mass with hyper-echoic fat components, hypoechoic cystic areas, calcifications with well-defined margins on ultrasonography[33]
ii. Mixed density lesion with fat, bone and other soft tissue densities along with calcifications on CT. It has been reported that 93% of lesions contain fat components and 56% contain calcifications[34,35]
iii. Iso-intensity to muscle on T1weighted imaging (T1WI) and iso to hyper-intensity on T2WI with well-defined boundary.[36]

Histopathology is required for confirmation of diagnosis where elements derived from more than one germ cell layer i.e., endoderm, mesoderm and ectoderm and different tissues such as fat, hair, skin and teeth can be seen within the specimen[31,32]

Benignity of teratomas is based on following features: (1) no immature elements present, (2) no indicators of malignant transformation, (3) no similar lesions elsewhere, (4) no recurrence on long-term follow-up.[37]

Indicators for malignant transformation are as follows:[5]

- Significant enhancement of the cyst wall and septations as well as mural nodules,
- Abnormal levels of hormones including cortisol, ACTH, aldosterone, and VMA.

However, malignant transformation is extremely rare.

Surgery is the method of choice for treatment of mature teratoma.[8] Surgery may be open or laparoscopic. Today, laparoscopic surgery is the gold standard for adrenal lesion removal.[38] The overall prognosis is excellent with a 5-year survival rate of nearly 100%.[8,15] A close follow-up after surgery is recommended in mature as well as immature teratoma.[9]

Conclusion

The purpose of this review article is identifying characteristic imaging features of teratoma and differentiating between the various lipomatous lesions involving adrenal region. Although adrenal teratoma is a rare tumor, it is essential to understand and acknowledge its characteristic features and means to make an appropriate diagnosis. Its characteristic imaging features help in identifying and distinguishing them from similar characteristic adrenal lesions. This eventually helps in appropriate line of management and better prognosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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