Case Report

Mature cystic teratoma involving the left adrenal gland with complete colonic wall formation in a 24-year-old female: A rare case report

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ABSTRACT

Teratoma arising from the adrenal gland is a rare retroperitoneal tumor. Only a few cases, mostly in young patients have been reported. Mature cystic teratomas of the adrenal gland frequently contain intestinal type epithelium, but they are rarely associated with complete intestinal wall formation. We herein describe an exceptionally rare case of left adrenal teratoma with complete colonic wall development in a 24-year-old female.

Key words: Adrenal gland, colonic wall, mature cystic teratoma

INTRODUCTION

Though mature cystic teratomas of extragonadal sites are unusual,[1] those arising in the adrenals are exceptionally rare.[2,3] They are more common in childhood and rarely occur in adults.[4] Histologically, they are composed of variable proportions of tissue originating from the ectoderm, mesoderm, and endoderm. Although gastrointestinal epithelium is occasionally seen in these tumors, the presence of a complete intestinal wall is rare.[5] We report a mature cystic teratoma of the left adrenal gland with complete colonic wall formation.

CASE REPORT

A 24-year-old female patient presented with pain in the left hypochondrium since 2 years. Computed tomography (CT) scan with contrast showed a mass (19 × 16 × 9 cm) arising from the left adrenal gland. The mass was multiloculated, containing fat and calcification. A radiological diagnosis of an adrenal teratoma was made [Figure 1]. Patient underwent laparoscopic transperitoneal left adrenalectomy, and the resected mass was sent to us for histopathological examination. On gross examination, the adrenal mass was composed of multiloculated cysts with a tan smooth mucosal-like inner surface, and with a tan smooth and shining outer surface [Figure 2]. On histopathological examination, there were cystic spaces lined by keratinizing squamous epithelium along with sebaceous gland, hair, pancreatic tissue and multiple cystic spaces lined by mucous and respiratory epithelium. In addition, there was the formation of complete intestinal wall-like structure. The intestinal wall-like structure resembled a complete colonic wall, including mucosa, muscularis mucosa, submucosa with loose connective tissue, and two layers (circumferential and longitudinal) of muscularis propria [Figure 3].

DISCUSSION

Teratoma is a germ cell tumor derived from totipotential cells, which comprise tissues originating from more than one germ cell layer, usually all three, and giving rise to different tissues such as skin, muscle, nerve, fat, and tooth structures. Most teratomas are found in gonads. Nevertheless, many extragonadal sites have been reported, including the mediastinum, retroperitoneum, cranium, sacrococcygeal region, the large bowel and even the tongue.[1] Primary

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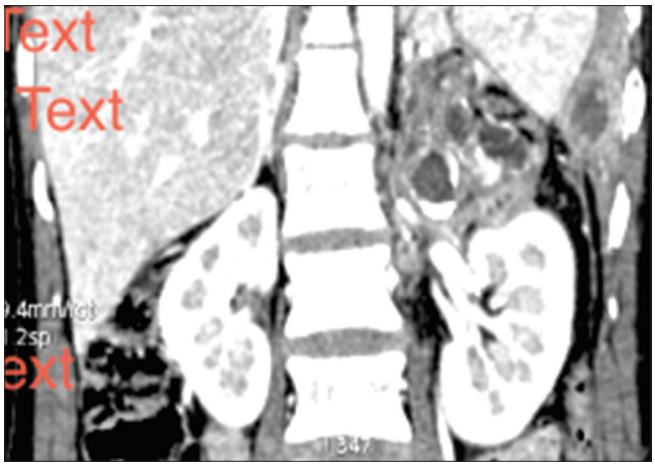

Figure 1: Coronal contrast-enhanced computed tomography showing left adrenal mass containg fat, calcification and peripharally enhancing cystic areas

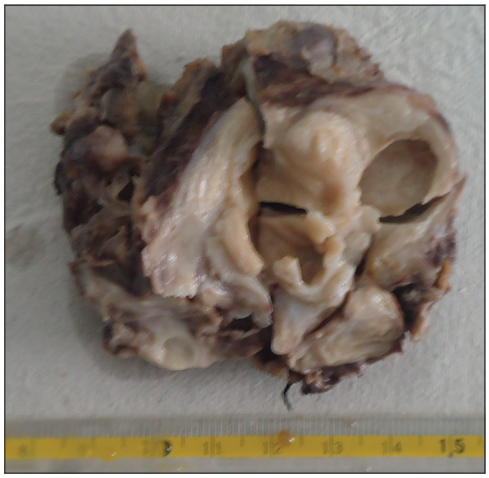

Figure 2: Gross specimen showing multilocular cut surface

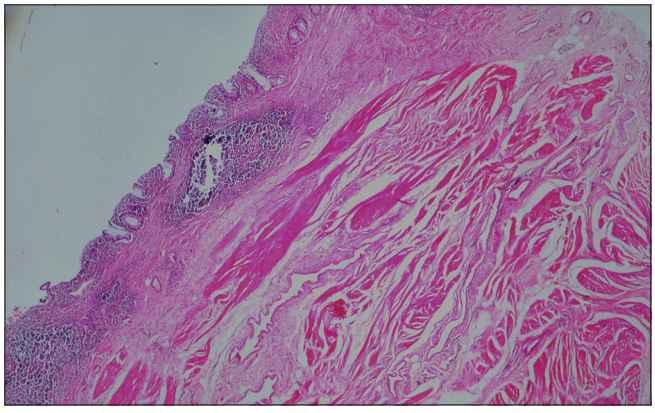

Figure 3: Photomicrograph showing complete colonic wall formation in the mature cystic teratoma

retroperitoneal teratomas arising from adrenal glands are exceedingly rare, accounting for only 4% of all primary teratomas.[2,3] Most teratomas in this region are secondary to germ cell tumors of the testes or ovaries. According to Lam and Lo,[6] adrenal teratomas represent only 3% of the surgically excised adrenal masses.

Adrenal teratomas are more common in childhood and are rarely seen in adults.[4] They are more frequently encountered in the left adrenal gland.[7] The majority of these cases are asymptomatic, present with nonspecific complaints, or are identified incidentally on routine investigations.[8] With respect to high index of clinical suspicion, retroperitoneal teratomas involving adrenal glands may present congenitally, or later in life when they grow to massive sizes.[9] Clinical presentations are variable and include nonspecific, abdominal/flank/back pain, obstructive gastrointestinal and genitourinary symptoms, as well as lower limb/genital swelling due to lymphatic obstruction.[10] They can rarely present with complications such as secondary infections (abscess formation),[11] traumatic rupture leading to acute peritonitis,[12] or malignant transformations.[13] On CT scans, teratoma is frequently shown as a heterogeneous fat dense mass with calcifications. Mature teratoma in the adrenal region can mimic other types of lipomatous adrenal tumor. Conventional imaging techniques cannot exactly distinguish the various types of lipomatous tumor.

Woodfield *et al*.[14] described the entire gastrointestinal tract from esophagus to the colon in a benign cystic teratoma. Complete intestinal wall formation is a rare finding in mature cystic teratoma. Fujiwara *et al*.[5] reported two cases of mature cystic teratoma containing complete intestinal wall, but both cases were in the ovary. The present case is the first case of adrenal teratoma showing complete formation of the colonic wall.

Early diagnosis and surgical resection are important in the treatment of these tumors. Laparoscopic transperitoneal adrenalectomy is a feasible, effective technique that enables excellent results.[15] Our patient underwent a laparoscopic transperitoneal adrenalectomy and is free of recurrence for longer than 18 months now.

CONCLUSION

Though adrenal teratomas have been reported extremely rarely in adults, it should be considered in the differential diagnosis of hormonally silent adrenal tumors. In particular, teratoma should be considered in the differential diagnosis of adrenal lipomatous tumors, not only in children and young adults, but also in elderly patients. The final diagnosis depends on the findings of the pathological examination.

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