

CASE REPORT

Extra-gonadal mature teratoma: A case report of retroperitoneal location with a literature review

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Abstract

Extra-gonadal mature teratoma is a benign tumor occurring rarely in adults. The retroperitoneal localization constitutes less than 4%. Treatment consists of surgical resection. Histological examination is essential for definitive diagnosis. We reported an unusual case of mature retroperitoneal teratoma discovered in a young man with abdominal pain.

KEYWORDS

dermoid cyst, retroperitoneal space, teratoma

1 | INTRODUCTION

Teratomas are uncommon neoplasms that comprise a mixed dermal elements derived from the three germ cell layers.¹⁻² They can be mature benign without malignant cells or immature with malignant cells.³ They commonly occur in the sacrococcygeal region in children and in gonads in adults especially in women. The retroperitoneal localization is not common. It constitutes less than 4% of all extragonadal teratomas.⁴ In addition, those benign tumors are most often asymptomatic. In most cases, they are identified incidentally.

In this paper, we report a case of mature extra-gonadal retroperitoneal teratoma occurring in a young adult man. We focus on clinical, radiological, biological, and

histological presentations, and we realize a literature review of this rare location.

2 | SEARCH STRATEGY

2.1 | Publication search

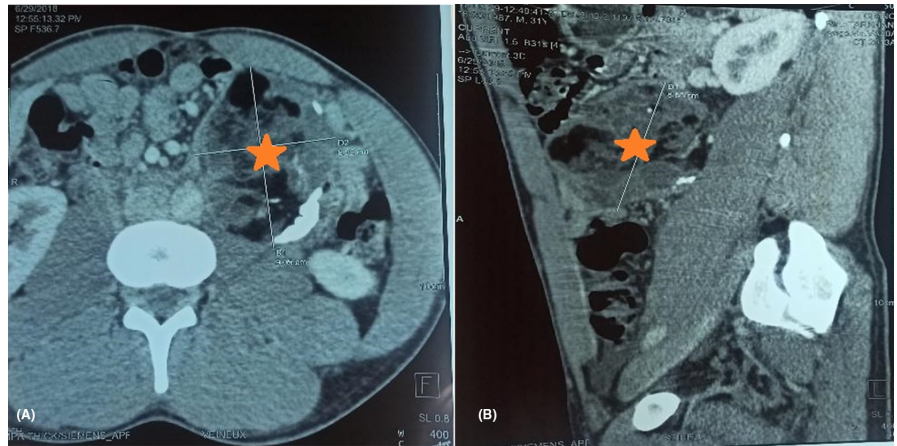
We performed a literature search in PUBMED for English language sources using the following keywords chosen from the Medical Subject Headings (MeSH) of MEDLINE: (“teratoma”[Mesh]) AND (“retroperitoneal space”[Mesh]), (“dermoid cyst”[Mesh]) AND (“retroperitoneal space”[Mesh]). Only cases occurring in adults were included.

TABLE 1 Cases of primary mature extra-gonadal retroperitoneal teratoma occurring in adult patients

Reference	Age/sex (Year)	symptoms	Clinical findings	Laboratory findings	Imaging investigations	Surgery procedure	Histological findings	Outcomes
Shakir et al. ³	31/female	Food indigestion, vomiting, abdominal pain	Palpable mass in left lumbar region	Normal	<i>Ultrasound</i> : cystic mass in left hypochondrium <i>CT scan</i> : mass in left lumbar region anteromedial to left kidney	Laparotomy with mass resection	Benign mature cystic teratoma	Simple
Lukanovic et al. ⁴	24/female	Pelvic pain and pressure	NS	NS	<i>CT scan</i> : mass in the left paravesical fossa altering the symmetry of the gluteal/perineal region	Laparotomy with mass resection	Benign mature cystic teratoma	Simple
Chen et al. ⁵	50/female	Low back pain, night sweats	Normal	Normal	<i>CT scan</i> : oval hypodense mass appearing arise from the left adrenal gland	Laparoscopic exploration: resection of an independent mass	Benign mature cystic teratoma	Simple
Sherer et al. ⁶	26/female	Pelvis pressure	Mild left lower abdominal tenderness, deviated uterus with a posterolateral mass	Normal	<i>Transvaginal ultrasound</i> : encapsulated heterogeneous mass with debris <i>CT scan</i> : displaced vaginal vault and rectum by a large pelvic mass	Laparotomy: the mass was opened, and the thick fluid contents of the cyst were removed by suction.	Benign mature cystic teratoma	Simple
Talwar et al. ⁷	23/32 weeks pregnant female	Fever, upper abdominal pain	Tender mass was palpable in right upper abdomen	NS	<i>Ultrasound</i> : hypoechoic mass suggestive of a large collection in subhepatic space <i>CT scan</i> : tumor with fluid, soft tissue, fat, and bone densities in the retroperitoneal space on right side	Laparotomy: resection of a capsulated mass	Benign mature cystic teratoma with abscess	Spontaneous labor simple
Kao et al. ⁸	24/female	Low back pain with sciatica	Mild throbbing pain in the right flank	NS	<i>Lumbar spine radiograph</i> : calcified tissues on the upper right quarter of the abdomen <i>CT scan</i> : delimited retroperitoneal mass, with extension into the intraspinal space at the L2-L4 level <i>MRI</i> : delimited intraspinal extradural cystic lesion	Right subcostal incision: a mass compressing the kidney and ureter to the left with extension to L2-L3 neuroforamen into the posterior lateral epidural space hemilaminectomy with liberation and resection of the mass	Benign mature cystic teratoma	Simple
Our case	30/male	Left sided abdominal pain	Palpable oblong mass of 10 cm long axis in the left hypochondrium and the left flank	Normal	<i>CT scan</i> : heterogeneous retroperitoneal mass of the left flank exerting a mass effect on the left ureter, and responsible for a slight dilatation of excretory cavities	Midline laparotomy: resection of a left retroperitoneal mass blowing the left mesocolon	Benign mature cystic teratoma	Simple

Abbreviations: CT scan, computerized tomography scan; MRI, magnetic resonance imaging; NS, not specified.

FIGURE 1 Abdominal computerizing tomography (CT) scan on axial (A) and sagittal (B) plane showing a heterogeneous retroperitoneal mass of the left flank measuring 9 cm in long axis, with mixed components of water, fat, and calcium, exerting a mass effect on the left ureter, and responsible for a slight dilatation of excretory cavities



The search included studies published from the database inception to April 2022. A total number of 6 articles were included in this review article.

2.2 | Collection data

The following data were collected from each case report: age, gender, symptoms, clinical examination, biologic findings, radiologic findings, intervention, histological findings, and outcomes.

The identified cases are shown in [Table 1](#).^{3–8}

3 | CASE PRESENTATION

3.1 | Case history and examination

Informed written consent was obtained from the patient before the publication of the case details and accompanying images.

A 30-years-old male patient, without previous comorbid, was referred to our surgery department for left sided abdominal pain evolving for 2 months. He had neither fever nor loss of weight and appetite. He had no digestive complaints or urinary symptoms.

The physical examination revealed an overweight, a normo-colored conjunctiva, a normal blood pressure, and a normal thermal profile. Cardiovascular, respiratory, and neurological examinations were normal. There was no testicular mass. On abdominal examination, there was a palpable oblong mass of 10 cm long axis in the left hypochondrium and in the left flank. It was deep, immobile on breathing, with firm consistency. Auscultation of bowel sounds was normal. Colonoscopy did not show any abnormality.

3.2 | Differential diagnosis, investigations, and treatment

Abdominal computerizing tomography (CT) scan showed a heterogeneous retroperitoneal mass of the left flank, with mixed components of water, fat, and calcium ([Figure 1](#)). It measured 9 cm in long axis, exerting a mass effect on the left ureter, and responsible for a slight dilatation of excretory cavities. The chest X-ray was normal. There was no other suspected location.

The operability assessment was normal. World Health Organization performance status grade was 0. Hemoglobin level was 14.2 g/dl. Renal function was normal, and urines were sterile on cyto-bacterial examination.

An exploratory midline laparotomy for mass resection was planned. Per-operative exploration showed a left retroperitoneal mass of 8 cm long axis, blowing the left mesocolon ([Figure 2](#)), and having a thick and inflammatory capsule. A careful left colic mobilization was performed after incision of the left mesocolon, in an avascular zone, in contact with the mass, from inside to outside. Gerota's fascia was intact. The mesocolic gap was sutured to avoid internal herniation later. The vascularity of the colon was not compromised. Enucleation of the mass was performed ([Figure 3](#)).

3.3 | Outcome and follow-up

Post-operative course was simple. The patient was discharged home on the third postoperative day.

Pathological examination showed a heterogeneous mass on section, with adipose lobules, cartilaginous hair and mucus, and covered with false membranes ([Figure 4](#)). On microcopy, there were 3 types of epithelium; a

keratinizing squamous epithelial, a respiratory type, and a secretory mucous gastric one with an extensive inflammatory remodeling (Figure 5). There was no immature component. The diagnosis of mature cystic teratoma was made. After 03 months of follow-up, the patient reported pain disappearance with a normal digestive transit.

4 | DISCUSSION

Our case is of interest because it provides an overview of a rare extra-gonadal mature teratoma occurring in a young adult man in the retroperitoneal region.

Teratomas develop from the three embryonic germ layers.¹ There are two variants: cystic teratomas which are composed of fully mature elements and are usually benign and solid teratomas which are more likely to be malignant.^{9,10} Primary retroperitoneal teratomas constitute 1 to 11% of retroperitoneal neoplasms and are most commonly found in neonates and in young adults.² They occur in female twice than male. Benign teratomas are usually asymptomatic.¹¹ Symptoms are related to the increase in volume causing compression of surrounding structures.^{5,12} As in our case, patient may present with back or abdominal pain, genito-urinary, or gastro-intestinal symptoms. Lower extremity or genital edema may be also noted.² Additionally, some complications may happen, such as abscess or rupture causing peritonitis.^{7,13}

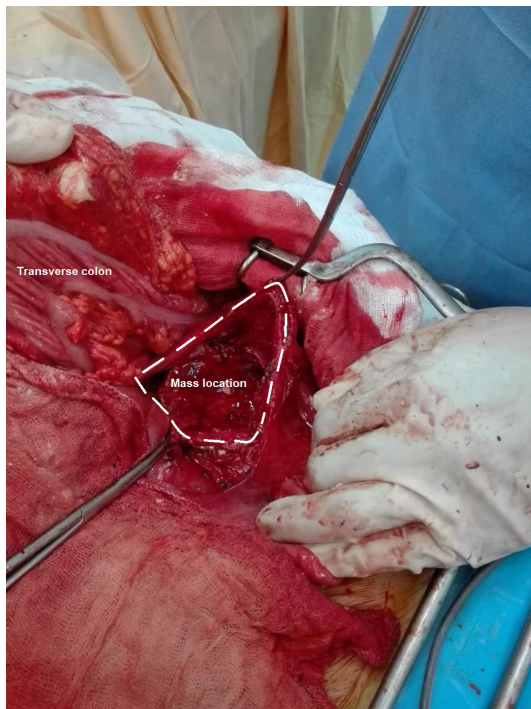


FIGURE 2 Per-operative exploration by midline laparotomy showing a left retroperitoneal mass blowing the left mesocolon

Physical examination may found a palpable abdominal mass, a tenderness, and distension.^{2,14} This mass may be confused with other tumors such as renal cysts, adrenal tumors, gonadic tumors, fibromas, hemangiomas, enlarged lymph nodes, and sarcomas. Imaging investigations can help guide the diagnosis. Abdominal ultrasound can reveal tumors with cystic and solid components and echogenic spots with acoustic shadows with some limitations in detecting fat and calcifications.¹⁴ Hence, CT scan offers a better identification of fat, sebum, and adipose tissue. Therefore, identification of sebum on CT is very suggestive for teratoma.² However, the diagnostic accuracy remains histological.^{8,15} Surgical resection is of greatest importance for both diagnosis and treatment. Midline or transverse incision is used for most patients. When possible, laparoscopic methods may be an excellent alternative but requires an adequate laparoscopic skills.¹⁶ Usually, primary retroperitoneal teratoma does not infiltrate adjacent structures and can be fully resected without significant complications. Prognosis is excellent in patients with complete resection of benign teratomas. Malignant transformation of mature teratoma is uncommon. Also, malignant teratomas with germ cell elements may have a grim prognosis, hence, the interest of a complete surgical resection with a histological confirmation.¹⁷

Through our literature review, we came across 6 cases of primary mature extra-gonadal retroperitoneal teratoma occurring in adult patients. They are presented in Table 1.³⁻⁸ The age of patients ranged from 23 to 50 years with an average of 29 years. They were 6 females. Abdominal pain was the most common first clinical presentation. Abdominal scan was performed for all patients. The mass was localized in the left retroperitoneal region for most of them. Complications were noted in two cases, one with abscess, and one with intra spinal extension.

Regarding therapy, laparotomy was performed in 5 cases and laparoscopy in one case. Simple outcome was noted in all cases.

5 | CONCLUSION

Primary retroperitoneal mature teratomas with extra-gonadal location are rare especially in young adult man. They are usually identified when they exert a mass effect on adjacent structures. Surgical resection remains the mainstay of therapy, and histological examination is essential for definitive diagnosis. Patients with complete resection of a benign teratoma have an excellent prognosis.

Additionally, we have highlighted teratoma as a differential diagnosis of retroperitoneal mass, and we emphasized the importance of complete resection to confirm

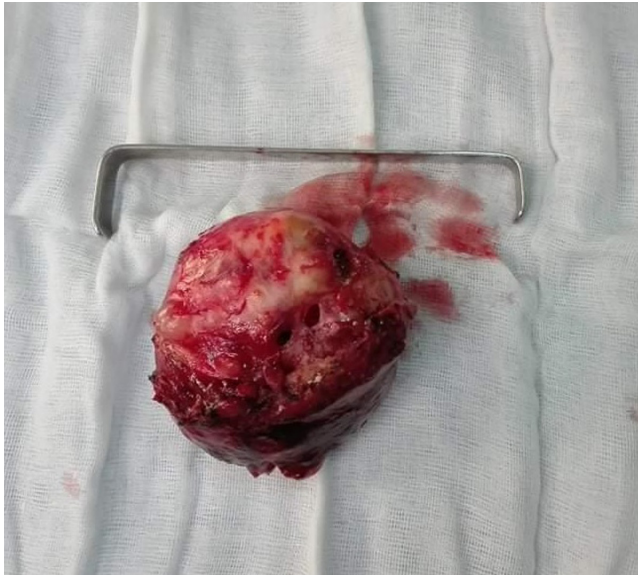


FIGURE 3 Enucleated retroperitoneal mass



FIGURE 4 Macroscopic examination showing a heterogeneous mass on section, with adipose lobules, cartilaginous hair and mucus, and covered with false membranes

the diagnosis and to avoid complications and malignancy transformation.

AUTHOR CONTRIBUTIONS

All the authors participated in the design of the study and wrote the manuscript. All authors read and approved the final manuscript. Mohamed Farès Mahjoubi has drafted the work. Ghassen Hamdi Kbir has substantively revised the work. Mohamed Maatouk, Sohaib Messaoudi, and Bochra Rezgui have made substantial contributions to the literature research. Mounir Ben Moussa has made substantial contributions to the conception of the work.

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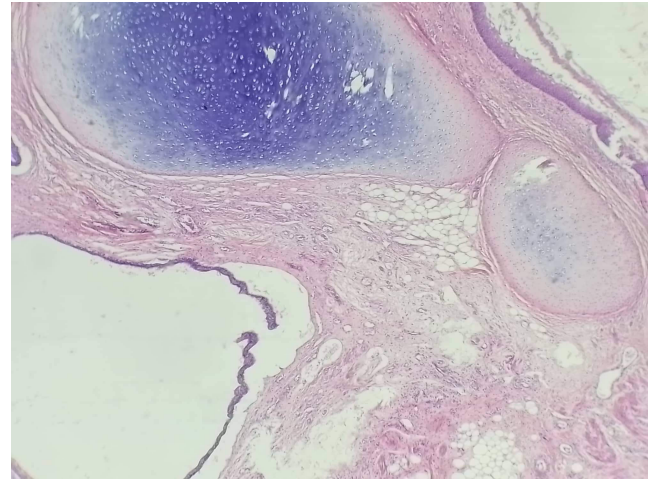


FIGURE 5 Histopathology examination of the mass showing 3 types of epithelium; a keratinizing squamous epithelial, a respiratory type, and a secretory mucous gastric one with an extensive inflammatory remodeling (HE $\times 100$)

CONFLICT OF INTEREST

The authors have no conflict of interests.

DATA AVAILABILITY STATEMENT

The data and supportive information are available within the article.

ETHICAL APPROVAL

Our locally appointed ethics committee “Charles Nicolle Hospital local committee” has approved the research protocol and informed consent has been obtained from the subject.

HUMAN AND ANIMAL RIGHTS

Our study complies with the Declaration of Helsinki.

CONSENT

Written informed consent was obtained from the patient.

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