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## Case Report

# A massive retroperitoneal mature teratoma from a “burned-out” testicular teratoma and seminoma<sup>☆</sup>

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## ABSTRACT

Germ cell tumors are the most common nonhematologic malignancy of young men which often present with metastasis to the retroperitoneum, however a primary retroperitoneal mass should also be considered. The case presented herein reports a 42-year-old male presenting with a massive heterogenous retroperitoneal mass determined to be a mature cystic teratoma. Further investigation revealed a multifocal right testicular mass containing both a viable pure seminoma and a fibrous scar demonstrating germ cell neoplasia in situ thus representing the rare phenomenon of a “burned-out” mixed germ cell testicular tumor. When the radiologist is faced with a large retroperitoneal fat-containing mass, the differential includes a renal angiomyolipoma, liposarcoma, or germ cell tumor (whether primary or secondary). If pathology reveals a germ cell tumor, it is imperative to perform a thorough evaluation of the gonads, as it is much more common for a retroperitoneal germ cell tumor to be a metastasis from the gonads, rather than primary in origin.

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## Introduction

Testicular tumors are subdivided into two major categories including germ cell and stromal tumors, the former accounts for 90–95% of tumors. Germ cell tumors (GCTs) are divided into

seminomatous and non-seminomatous GCTs (NSGCTs). NSGCTs are divided into four subtypes: choriocarcinoma, yolk sac, embryonal, and teratoma. Approximately 60% of GCTs contain multiple subtypes and even if a tumor predominately contains seminomatous elements, it is classified as a non-seminomatous GCT. The most common pure GCT is a semi-

Abbreviations: GCT, Germ Cell Tumor; NSGCT, Non-Seminomatous Germ Cell Tumor; RP, Retroperitoneum.

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noma (30–60%) followed by teratoma (5–10%), embryonal (3–4%), and choriocarcinoma (1%) [1].

GCTs are the most common nonhematologic malignancy in men between ages 15 and 49, however represent up to 1% of all male tumors and, fortunately, one of the most treatable and curable cancers with a survival rate over 95% if discovered early [2]. The lifetime risk of developing testicular cancer is one in 268 men, whereas the risk of death is one in 5000 [2]. Moreover, although its relatively uncommon, the incidence has more than doubled over the past 40 years [3].

The vast majority of GCTs arise from the gonads, [4] as such, the typical presentation includes a painless testicular mass although up to one-third of patients are initially misdiagnosed with epididymitis, orchitis, or hydrocele. At initial diagnosis, approximately 30% of patients will have metastatic disease which often presents with malaise, weight loss, and abdominal or back pain secondary to the lymphatic drainage to the retroperitoneum [5].

### Case report

A 42-year-old male with past medical history of liver cirrhosis currently on the transplant list who previously required weekly paracentesis presented with moderate, constant, dull, pressure-like, non-radiating left sided abdominal pain over the last few months which was associated with intermittent nausea and constipation. He reports decreased appetite and a weight loss of 10 pounds over the last two weeks. He denies fevers, chills, shortness of breath, chest pain, vomiting, and diarrhea.

Vitals signs on presentation were within normal limits. Laboratory evaluation was remarkable for a mild leukocytosis of 14,300 per cubic millimeter and a MELD score of 10 including a mild hyperbilirubinemia of 1.5 mg/dL and a hyponatremia of 134 mmol/L.

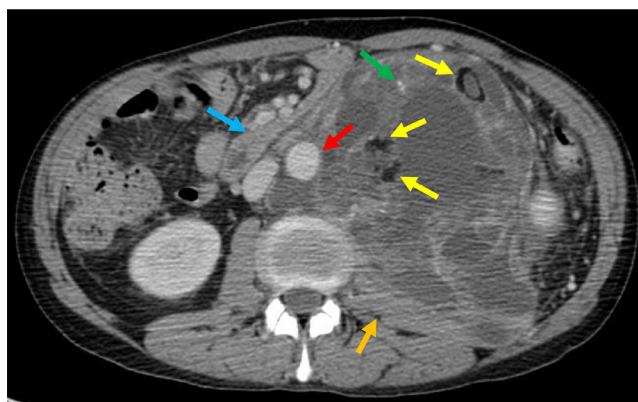
Computed tomography (CT) of the abdomen and pelvis with intravenous contrast revealed a large heterogenous predominantly cystic mass measuring 10.9 × 12.2 × 12.7 cm (Figs. 1 and 2) within the left retroperitoneum. Imaging characteristics favored this to be separate from the left kidney, psoas, and other retroperitoneal structures and may originate from the retroperitoneal lymph nodes. Based on the multiple regions of solid, enhancing components, as well as areas of fatty, cystic components, and scattered calcifications, the main differential diagnoses include a primary liposarcoma (particularly due to its size), metastatic primary testicular GCT, or a primary retroperitoneal teratoma.

Intraoperative findings revealed no nodularity of the peritoneum and a small amount of ascites (which was sent for pathologic evaluation). The left colonic mesentery was draped over and inseparable from the mass. The left renal vein, artery, and left ureter was also difficult to separate from the mass. Therefore, the left retroperitoneal mass was resected with en-bloc removal of the left kidney and left colon (Fig. 3), followed by an end-to-end colocolonic anastomosis.

Pathologic evaluation revealed a mass with endodermal, mesodermal, and ectodermal tissues including skin, gastric mucosa, enteric mucosa, pancreatic tissue, bronchial mucosa,



**Fig. 1** – Axial CT of the abdomen and pelvis with IV contrast at the level of the superior mesenteric artery demonstrates the superior-most aspect of the left retroperitoneal mass (yellow arrow). The left renal artery (red arrow) and partially visualized left renal vein (blue arrow) are displaced superiorly by the mass. There is also moderate right-sided hydronephrosis (green arrow).

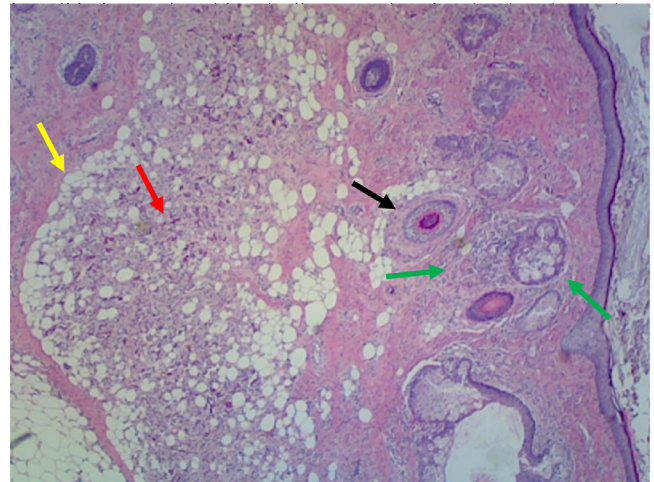


**Fig. 2** – Axial CT of the abdomen and pelvis at the level of the transverse duodenum, inferior to Fig. 1, demonstrates a large heterogenous predominantly cystic left retroperitoneal mass. The duodenum (blue arrow) is displaced anterolaterally. The left psoas muscle (orange arrow) is displaced posteriorly. The aorta (red arrow) is mildly displaced anteriorly by a small cystic portion of the mass. The mass contains a few small areas of fat (yellow arrows) and a small calcification (green arrow).

adipose tissue, smooth muscle, peripheral nerves, cartilage, and bone (Figs. 4 and 5). Focal atypia was present, however, no immature components or somatic-type malignancy was identified thus representing a mature teratoma. The tumor appeared completely excised with negative margins and no involvement of the left kidney or left colon. Sections of the kidney revealed mild to moderate chronic interstitial subcapsular inflammation with moderate arterio- and arteriosclerosis. Sections of the left colon demonstrated significant serosal adhesions but otherwise histologically unremarkable. The peritoneal fluid was also negative for malignant cells.



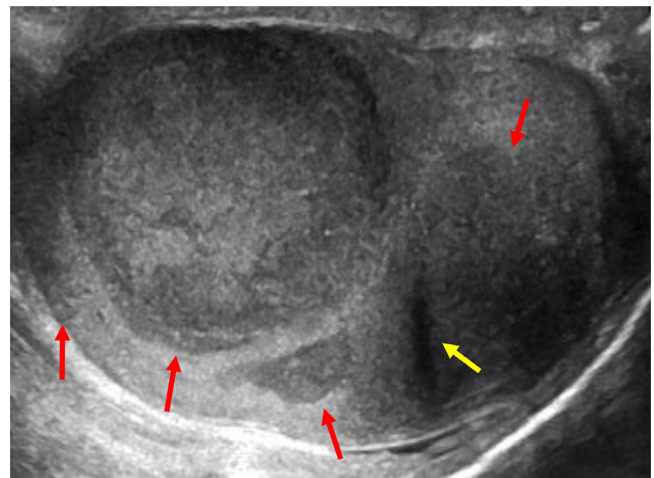
**Fig. 3** – Intraoperative photograph of the left retroperitoneal mass with en-bloc resection of the left kidney and left colon.



**Fig. 5** – Photomicrograph demonstrates cystic cavities lined by skin with hair follicles (black arrow) and adnexal structures (green arrows). Mature adipose tissue (yellow arrow) with a disordered collection of capillaries (red arrow) is also noted.



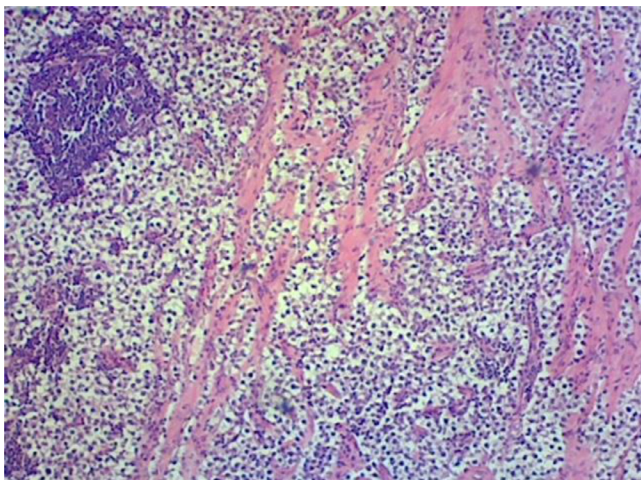
**Fig. 4** – Photomicrograph demonstrates a disorganized mixture of mature tissues including bone (black arrow), cartilage (yellow arrow), and gastric-type tissues (green arrow).



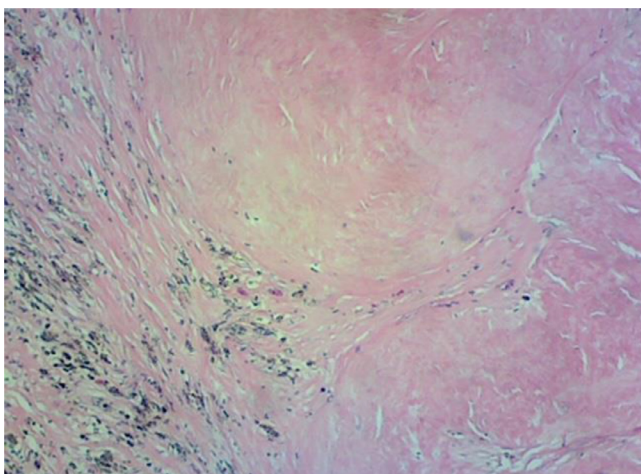
**Fig. 6** – Sagittal gray-scale sonograph of the right testis demonstrates four predominantly hypoechoic intratesticular masses (red arrows). The largest mass measures 2.4 x 1.8 x 2.5 cm while the smallest mass measures 0.3 x 0.3 x 0.3 cm. The most inferior mass demonstrates a small focus of posterior acoustic shadowing (yellow arrow) representing a macrocalcification.

Given the diagnosis of a teratoma, clinical attention was then turned to the testicles. The patient reported intermittent innocuous vague right testicular discomfort for which he did not feel required medical evaluation. Tumor markers AFP, hCG, and LDH levels were within normal limits. Sonographic evaluation of the right testicle revealed four separate predominantly hypoechoic masses (Fig. 6). The left testicle was unremarkable. The patient underwent an uncomplicated right orchiectomy. Gross pathology revealed a 2.5 cm tan-red fleshy mass in the superior pole and a firm 1 cm white fibrous scar

at the inferior pole. Immunohistochemical analysis revealed that the tumor was positive for OCT-4, D2-40, and SALL4 and negative for CD-30, AFP, and SOX2 consistent with a seminoma without lymphovascular invasion (Fig. 7). The white fibrous scar demonstrated germ cell neoplasia in situ (Figs. 8 and 9) which, along with the retroperitoneal mass, was diagnostic for a regressed GCT. Final AJCC staging was stage IIC (T1aN3M0S0).



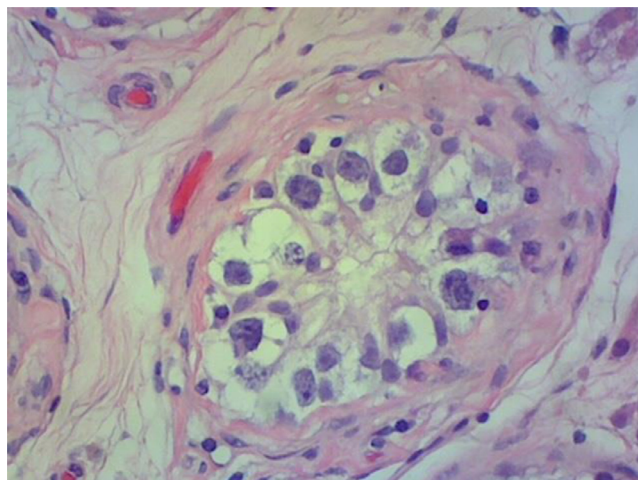
**Fig. 7** – Photomicrograph demonstrates sheets of cells with clear cytoplasm, fibrous septa, and a lymphoid aggregate (upper left corner) consistent with a seminoma.



**Fig. 8** – Photomicrograph demonstrates a region of scarring with hemosiderin collections at the bottom left corner which was adjacent to the seminoma (unable to capture portions of the seminoma in this low power field view).

## Discussion

When the radiologist is faced with a mass in the retroperitoneum (RP), the first step is to determine the location of the mass and where it is arising from. If the mass does not originate from the organs such as the kidney, adrenal glands, pancreas, bowel loops, or lymph nodes it can be classified as primary. Radiographic signs such as anterior displacement of these structures and lack of the beak sign, phantom organ sign, and embedded organ sign favors a mass to be primary in origin [6]. The most common primary RP mass is a liposarcoma representing approximately 30% which typically occur in the fifth and sixth decades of life. A teratoma can also be seen in the RP, however only 1-11% of primary teratomas arise in the RP [7]. Differentiation of liposarcoma and teratoma can be difficult. Imaging characteristics that favor liposarcoma



**Fig. 9** – High power photomicrograph within the scarring noted on Fig. 8 demonstrates a single seminiferous tubule lined with atypical cells, consistent with germ cell neoplasia in situ.

include a lesion greater than 10 cm, the presence of thick (greater than 0.2 cm) septa, and foci of nodular enhancement. RP teratomas are characterized by macroscopic fat, cystic areas, calcifications, fat-fluid levels, and heterogeneous enhancement. However, calcifications are also seen in 30% of dedifferentiated liposarcomas and are suggestive a worse prognosis [8]. If a teratoma (or any GCT) is suspected, it is essential to evaluate the gonads for a primary source, as only 10% are truly primary retroperitoneal GCTs [9].

Sonographic imaging of testes is the gold standard for the evaluation of the testes and has a near 100% sensitivity for the detection of testicular neoplasms. Most seminomas are homogenous while heterogeneity often corresponds to NSGCTs. A heterogeneous mass with cystic changes and calcifications suggests a mixed NSGCT with a teratomatous component [10]. Increased vascularity of a lesion is not specific to testicular tumors [2]. Meticulous sonographic evaluation of the testes is essential in the case of a suspected RP mass as findings may be subtle. The term “burned out” tumor is used to describe a regressed testicular tumor that presumably outstrips its blood supply and atrophies and was first documented in 1927 [11]. It has been shown that choriocarcinoma is the most likely to burn-out, followed by embryonal carcinoma; regression is uncommon for seminomas and probably does not occur in teratomas [12]. Sonographic characteristics include subtle ill-defined hypoechoic areas, a single 5 mm calcification, microcalcification with testicular atrophy, or even a curvilinear hyperechoic band [13,14]. Approximately 50% of “burned out” testicular tumors continue to harbor malignancy despite systemic chemotherapy and therefore orchiectomy is still performed [15].

## Conclusion

The case presented herein demonstrates the rare phenomenon of a “burned-out” GCT, defined as the presence of

a metastatic extragonadal GCT with histological regression of the primary testicular lesion.

Given the histologic diagnosis of a pure testicular seminoma and a mature retroperitoneal teratoma, it can be inferred that the primary tumor in this case was a mixed GCT (with both seminomatous and non-seminomatous components) of which the teratomatous portion underwent spontaneous regression while the seminomatous portion remained viable.

Not all fat-containing, aggressive-appearing masses in the retroperitoneum are liposarcoma. Although histologically benign, retroperitoneal teratomas (either primary or secondary) can radiographically appear alarming, especially when large. Every retroperitoneal mass must be considered secondary and thorough evaluation of the gonads is required, as it is much more common for a retroperitoneal germ cell tumor to be a metastasis from the gonads, rather than primary in origin.

### Patient consent

Informed written consent was obtained from the patient for publication of this Case Report and all imaging studies. Consent form on record.

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