

## CASE REPORT

# Retroperitoneal seminoma as a first manifestation of a partially regressed (burnt-out) testicular germ cell tumor

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### Abstract

Regressed (burnt-out) testicular germ cell tumors (TGCT) are rare clinical situations that are clinically difficult to recognize. This 43-year-old patient was admitted because of a suspicion of prostatic carcinoma, which eventually was followed by transrectal ultrasonography and a CT scan, both of which revealed a large retroperitoneal mass. Surgery showed extensive ureteral and vas deferens infiltration. Pathology was consistent with a classical seminoma. Eventually, testes were normal on palpation but ultrasonography only revealed areas of fibrosis and microcalcifications in the left testis, which was followed by a left orchidectomy. Microscopically, there were extensive areas of fibrosis and only a 2 mm area of seminoma was demonstrated. The few areas of uninvolved testicular tissue lacked lesions of intratubular germ cell neoplasia (IGCNU). Retroperitoneal germ cell tumors are rare in the male and consequently, an origin from an occult testicular tumor should always be discarded by image analysis and eventually a biopsy. Immunologic response may be responsible for tumor involution.

**Keywords:** retroperitoneal seminoma, burnt-out, ureter and vas deferens infiltration.

### Introduction

If usually TGCT are easily diagnosed after a testicular clinical evaluation, regressed tumors with retroperitoneal metastasis, as them first manifestation, might represent difficult cases prone to misdiagnosis.

The retroperitoneum is known as a source of various primary or metastatic tumors. Although primary germ cell tumors may exist in this location, the likelihood of a metastasis from a gonadal primary should always be borne in mind.

### Patient, Methods and Results

We present the case of a 43-year-old man with no previous medical records, presenting with progressive back pain and pollakiuria. On admission, he was diagnosed with a prostatic mass and elevated LDH serum level of 971 U/L and eventually admitted to the Urology Department. After a transrectal ultrasonogram and a normal PSA serum level, a prostatic tumor was excluded but a large retroperitoneal solid mass, in intimate contact but independent from the urinary bladder, was confirmed. For a complete evaluation of the tumor, a CT scan was performed demonstrating a multinodular mass with extensive areas of necrosis, imprinting the urinary bladder wall and infiltrating the left ureter. There were also multiple lymph nodes metastases.

Extensive retroperitoneal mass excision and partial resection of the ureter with a left cutaneous uretero-

stomy were performed. A small tumor fragment was sent for frozen section examination and a diagnosis of a pleomorphic tumor with extensive areas of necrosis was made.

Macroscopically, the 10×9 cm retroperitoneal tumor was partially necrotic with only a preserved peripheral rim, of white, solid tissue (Figure 1). There were also multiple, firm lymph nodes of 0.5 to 1.5 cm in diameter and a small segment of the left ureter and vas deferens, each measuring 1.5 cm in length.

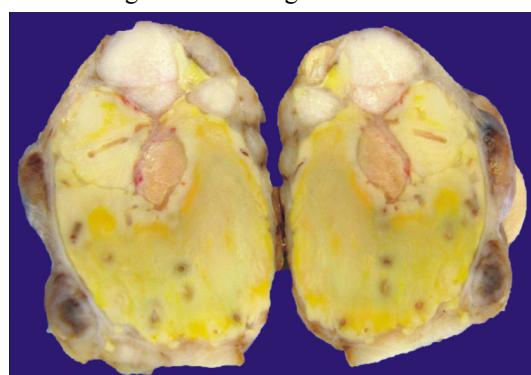
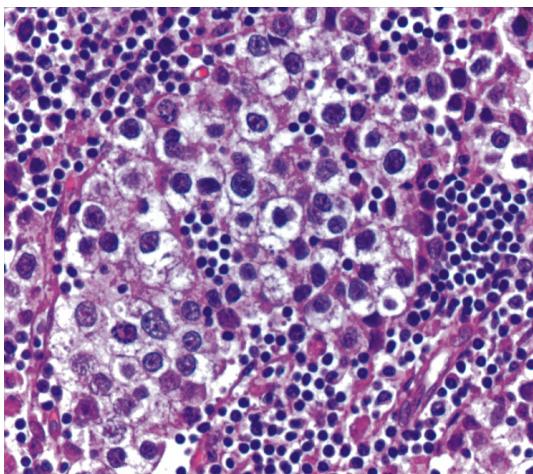


Figure 1 – Macroscopic aspect of the retroperitoneal tumor.

Microscopically, the tumor was solid and was constituted of large cells with clear to pale cytoplasm, evident boundaries, round vesicular nuclei with prominent nucleoli that were admixed with an abundant lymphocytic infiltrates (Figure 2) and extensive areas of necrosis.



**Figure 2 – Retroperitoneal seminoma with its characteristic microscopic features (HE stain,  $\times 400$ ).**

The ureter and the vas deferens were infiltrated and exhibited a thicker wall without compromising the lumen (Figures 3, A and B).

After the diagnosis of retroperitoneal seminoma infiltrating both the left ureter and vas deferens, associated with multiple lymph nodes metastases, the patient was re-evaluated even if a testicular mass was not evident on the previous clinical examination. When

the case was reconsidered, it was found that the left testis was smaller and firmer than the right one, revealing on ultrasound hyperechogenic areas compatible with fibrosis and microcalcification. AFP and hCG serum level were both normal but still associated with a high LDH serum level of 912 U/L. A subsequent left orchidectomy was decided.

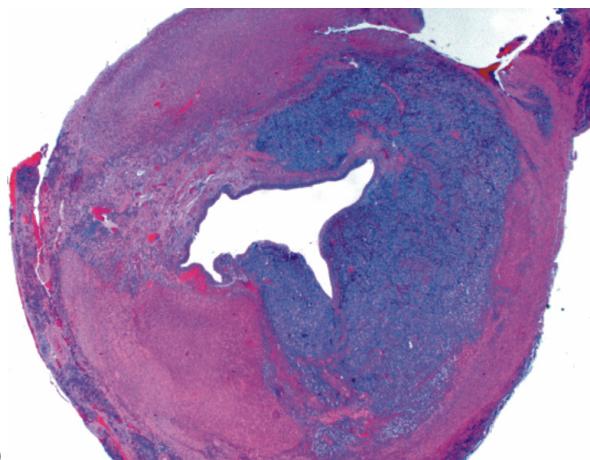
On cut section, the testicular parenchyma was almost entirely replaced by an ill defined, heterogeneous area of fibrosis (Figure 4).

Microscopically, there was an atrophic testicular parenchyma with fibrosis and numerous microcalcifications (Figure 5) and impaired spermatogenesis with a Johnsen 3 count.

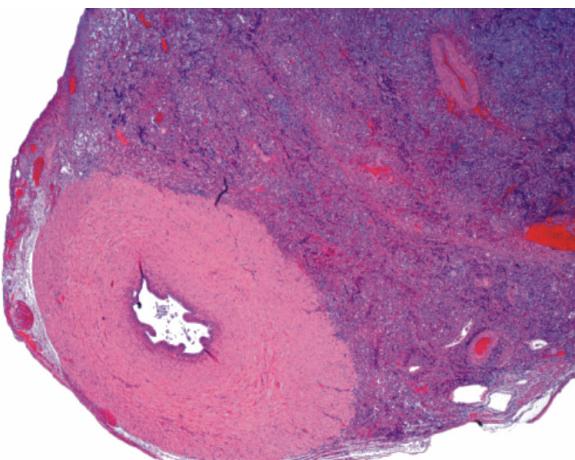
IGCNU was not evident at any level. However, only one block revealed a 2 mm diameter nodule corresponding to a testicular seminoma embedded in a fibrous matrix (Figure 6, A and B). Immunohistochemically the cells were diffusely positive for D2-40 and PLAP and negative for hCG, CD30 and AFP (Figure 6, C and D).

A final diagnosis of left partially regressed (burnt-out) TGCT with a residual seminoma nodule and seminomatous retroperitoneal metastasis with infiltration of the right ureter and vas deferens was done.

Five months after the surgery, the patient is well and receiving chemotherapy.

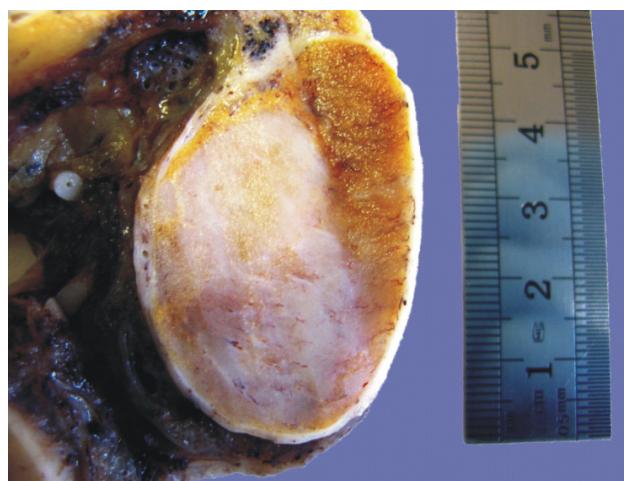


(A)

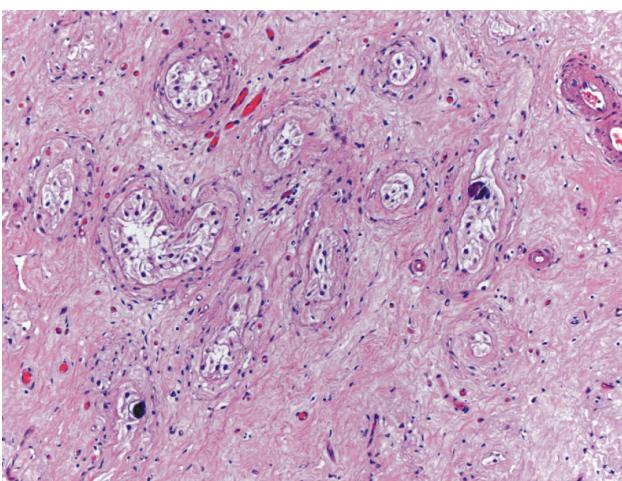


(B)

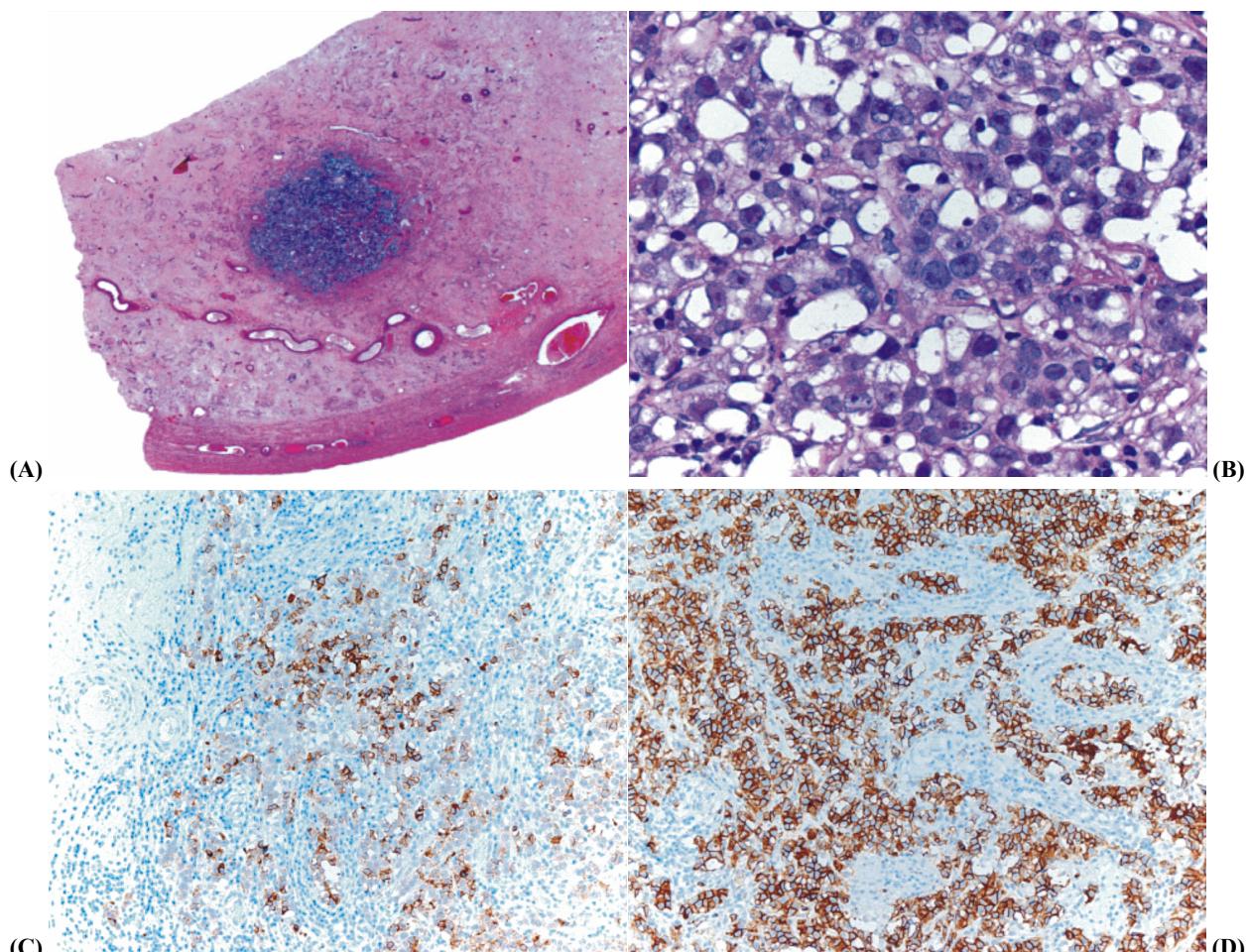
**Figure 3 – Infiltration of the left ureter (A) and vas deferens (B). HE stain,  $\times 25$ .**



**Figure 4 – Bivalve section of the left testis, demonstrating an almost entirely fibrous parenchyma.**



**Figure 5 – Testicular atrophy, with fibrosis and numerous microcalcifications (HE stain,  $\times 100$ ).**



**Figure 6 – Single nodule (A) of remaining seminoma (B), HE stain,  $\times 25$  respectively  $\times 400$ , with PLAP (C) and D2-40 (D) positive cells,  $\times 100$ .**

## Discussion

In the male, the testes are the most likely origin for GCT. Still, other midline locations, such as retroperitoneum, mediastinum and pineal region can represent locations where, albeit more infrequently, a primary GCT may occur. Isolated cases of GCT are reported in the sacro-coccygeal area in children, the retro-vesical space, lung, stomach, pancreas, esophagus, small bowel, the prostate, the seminal vesicle and the iliac fossa in males of all ages [1]. These ectopic tumors may develop from abnormally migrated primordial germ cells but dedifferentiation or neometaplasia of non-gonadal tissue have also been considered as a putative origin [2, 3].

Seminoma is the commonest TGCT associated with a spontaneous regression in the testis that can be associated with distant metastases [4]. Choriocarcinoma, due to its angioinvasive capacity, can also be associated with distant hematogenous metastases as first manifestation, sometimes without being identifiable in the testis [5–7]. Less frequently, embryonal carcinoma, an undifferentiated, pluripotent GCT, may do the same with intratubular coarse calcification as its only stigmata [4].

Generally, a testicular tumor can be demonstrated on ultrasound examination and confirmed after a microscopic study. Still, there are cases of testicular tumors that due to their small dimensions, central location or regression are clinically occult, as it was the present

case. It presented as a retroperitoneal mass with ureteral and bladder extrinsic compression, back pain. On physical examination, both testes seemed normal on palpation. Diagnosis was performed only retrospectively, after pathology diagnosis.

An exact incidence of burnt-out tumors cannot be specified, although it is documented in approximately 10% of the patients who died of metastatic TGCT [8]. Several mechanisms implicated in spontaneous regression are suggested, but the fact that cytotoxic T-lymphocytes present in seminoma can destroy tumor tissue explains the higher incidence of spontaneous regression in seminoma, a tumor invariably associated with an immunological response where T-cells predominate [9]. Even if in our case an inflammatory reaction was absent in the testicular tumor, an abundant lymphocytic infiltrate together with extensive areas of tumor necrosis was present in the retroperitoneal tumor. This would favor the immunological origin of burnt-out GCT as a source of retroperitoneal regressed tumors [10].

Characteristic for regressed TGCT, are the areas of ill-defined fibrosis, obvious even on macroscopic examination as a scar, which on microscopic evaluation are infiltrated by Hemosiderin-laden macrophages and lymphocytes. Sometimes, especially in cases of intratubular seminoma, round dense Hematoxylin bodies might be obvious. An intertubular growth is responsible for

fibrous, acellular, seminiferous tubules. Areas of dystrophic calcification and necrosis are generally reminiscent of a regressed embryonal carcinoma [4, 5, 7]. Sometimes, only remaining teromatous elements such as cartilage, intestinal or squamous epithelia can be recognized since they are unlikely to regress, as they are insensitive for chemotherapy [5].

In almost 50% of the cases, IGCNU can be the only microscopic evidence of a previous TGCT that might be helpful in confirming a testicular origin of an ectopic GCT [4]. Even if in our case a thoroughly sampling was performed, none of the slides did demonstrate the presence of IGCNU.

If a suspicion of a regressed testicular tumor exists, a systematical clinical evaluation, eventually followed by orchidectomy, should be performed, since a possible residual tumor might be a source of relapse. Also, the hidden primary might be resistant to chemotherapy due to fibrosis and an inadequate blood supply [4, 5, 11].

## Conclusions

In conclusion, our case presents an otherwise well known but rare case of a partially regressed testicular germ cell tumor with a residual seminoma nodule. As far as we know, the concomitant wide infiltration of left ureter and vas deferens is an otherwise unreported condition as first manifestation of a seminoma.

Regressed testicular germ cell tumors are clinically hard to recognize but they should be considered in the differential diagnosis of retroperitoneal masses, especially in young patients.

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