

**Case – “Burned-out” testicular tumor: A rare entity with diagnostic dilemma**Sunil Samnani<sup>1</sup>, Nimira Alimohamed<sup>2</sup><sup>1</sup>University of Calgary, Calgary, AB, Canada; <sup>2</sup>Tom Baker Cancer Centre, University of Calgary, Calgary, AB, Canada**Cite as:** Samnani S, Alimohamed N. Case – “Burned-out” testicular tumor: A rare entity with diagnostic dilemma. *Can Urol Assoc J* 2022 June 9; Epub ahead of print.<http://dx.doi.org/10.5489/cuaj.7879>

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**Introduction**

Most testicular cancers are germ cell tumours (GCTs) which occur in men aged 15-35 years, with germ cell histology accounting for 90% of testicular tumours. Risk factors for developing testicular cancer may include Klinefelter syndrome, history of cryptorchidism, positive family history of testicular cancer, and infertility.<sup>1-3</sup> The two most common histological patterns are non-seminomas, which account for approximately 40-45% of cases with a peak incidence at 25 years, and seminomas, which represent the other 55-60% of cases and have a peak incidence at 35 years of age. For patients with advanced GCTs, the 5-year overall survival probabilities are 98%, 96% and 66% for patients in good, intermediate, and poor-risk groups, respectively, based on the International Germ Cell Cancer Collaborative Group (IGCCCG) classification.<sup>3-6</sup> The treatment of advanced germ cell malignancies involves chemotherapy or radiation therapy, both of which can contribute to serious long-term complications.<sup>7-10</sup>

**Key Messages**

- Burned-out testicular tumour presents with an interesting therapeutic dilemma given spontaneous regression of retroperitoneal lymph nodes.
- The most histologic type of testicular tumour associated with a burned-out primary is seminoma followed by embryonal carcinoma.
- The most common histological features seen with burned-out testicular tumours are testicular atrophy, scar formation, lymphoplasmacytic infiltrates, and intratubular calcifications.
- Patient with normal tumour markers needs lymph node biopsy for diagnosis. Retroperitoneal lymph node dissection can be useful to remove all metastatic disease, and this avoid the short-term and long-term toxicity associated with cisplatin-based chemotherapy.

Spontaneous tumour regression in testicular tumour has been described as a “burned-out” testicular tumour, where there is a partial or complete regression of the tumour.<sup>2, 11</sup> Patients with burned-out testicular tumours can present without a clinically apparent tumour in the scrotum but usually have metastasis to other sites such as retroperitoneal and mediastinal lymph nodes.<sup>12, 13</sup> Patients may also present with elevated tumour markers and inconclusive findings on scrotal ultrasound. Most patients will proceed to radical orchidectomy and retroperitoneal lymph node biopsy (RPLND) to establish the diagnosis in the retroperitoneum. The most histologic type of testicular tumour associated with a burned-out primary is seminoma followed by embryonal carcinoma. The most common histological features seen with burned-out testicular tumours are testicular atrophy, scar formation, lymphoplasmacytic infiltrates, and intratubular calcifications.<sup>11</sup>

We present a case of a burned-out testicular GCTs with involvement of retroperitoneal lymph nodes which also spontaneously regressed. We discuss the treatment approach taken in this rare case of advanced GCTs with an unusual presentation.

### Case report

A 34-year-old male presented to the hospital after an ATV rollover accident. He underwent CT imaging of the chest, abdomen and pelvis and was incidentally found to have two periaortic lymph nodes (largest 2.1 cm as shown in Fig. 1) and a fracture of the transverse process of L1-L4. No other trauma-related injuries were noted. He denied any prior abdominal discomfort, distention, nausea, fever, or loss of appetite. The patient had no prior significant past medical history or risk factors for testicular cancer. He had not noted any previous testicular discomfort or swelling.

The levels of baseline tumour markers were as follows: Lactate dehydrogenase (LDH): 238 U/L (normal values: 100-225 U/L);  $\alpha$ -fetoprotein ( $\alpha$ -FP): 2 ug/L (normal values: <9 ug/L); and  $\beta$ -human chorionic gonadotropin ( $\beta$ HCG): <5 IU/L (normal values: < 5 IU/L). Scrotal examination was normal at baseline and abdominal examination was benign without any palpable masses. Ultrasound of the testes revealed a 1.2 x 0.7 cm region of subtle decreased echogenicity in the peripheral portion of left testicle, extending to the tunica with no overt mass-like features. The patient underwent an image-guided biopsy of an enlarged peri-aortic lymph node and pathology confirmed lymph node parenchyma involved with metastatic carcinoma with extensive coagulative-type tumour cell necrosis. This was reviewed by an expert genitourinary (GU) pathologist at a tertiary care centre and results confirmed a metastatic carcinoma, most likely an embryonal GCTs. A left radical orchidectomy was subsequently performed, showing testicular parenchymal scarring with no viable tumour or germ cell neoplasia in-situ. Repeat imaging was performed two months after the initial CT scan which revealed spontaneous regression in one of the periaortic lymph nodes from 19 mm to 15 mm (short axis) and stability in the other lymph node (11mm short axis) as shown in Fig. 2. The patient was reviewed at GU multi-disciplinary tumour board rounds with input from urologic oncology, medical oncology,

radiation oncology, pathology, and radiology. The consensus was to proceed to retroperitoneal lymph node (RPLND) dissection to confirm the pathological diagnosis. The patient went for an RPLND and recovered well. The final pathology revealed metastatic germ cell tumour in 3/15 lymph nodes: 2 of 8 para-aortic nodes and 1 of 4 left pelvic lymph nodes. On immunohistochemical staining, the paraaortic nodes stained positive for oct 3/4 cytokeratins, and CD117, suggesting this was non-seminomatous differentiation (predominantly embryonal with a small yolk-sac focus) in a seminomatous tumour. The pelvic lymph nodes were involved with metastatic seminoma.

Postoperatively, the patient's case was again reviewed by the multi-disciplinary tumour board and the consensus was to place the patient on a surveillance protocol. Chemotherapy was discussed but given this unique case, the group favoured surveillance with close monitoring. Patient was followed with repeat imaging in 3 months after his surgery which revealed no evidence of disease recurrence. The patient's tumour markers have remained normal throughout his treatment course. He will continue a surveillance protocol.

### Discussion

Burned-out testicular tumours with metastases to the retroperitoneal or mediastinal lymph nodes have unique characteristics.<sup>14</sup> A detailed physical examination of the abdomen and scrotum, along with a scrotal US is imperative as part of the workup in any young male presenting with a retroperitoneal mass.<sup>13</sup> Testicular atrophy and/or microcalcifications may be the only findings on ultrasound, yet a low threshold for radical orchiectomy should be held if the suspicion is high for testicular malignancy, given the high cure rates of this cancer and the need for accurate histologic diagnosis.

The management pathway for burned-out testicular tumours with metastatic disease to lymph nodes is typically the same as that for conventional testicular cancer with lymph node involvement. The standard of care, depending on extent of disease, involves chemotherapy or radiation therapy and/or RPLND. Seminomatous and non-seminomatous GCTs have different treatment pathways and thus it is important to identify the histological type to guide management and prognostication, particularly when tumor markers are normal or not clearly demarcating these two types. Patients with seminomatous GCTs with limited retroperitoneal metastasis can be managed by radiotherapy whereas patients with advanced non-seminomatous GCTs require chemotherapy.<sup>15</sup>

Serum tumor markers have low sensitivity, particularly in patients with seminomatous GCTs where the markers are likely to be normal compared to patients with non-seminomatous GCTs where 85% will have elevated tumor markers.<sup>16</sup> Isolated elevations in serum  $\beta$ HCG levels may be seen with seminoma and/or non-seminoma whereas elevations in both serum  $\beta$ HCG and AFP levels are likely indicative of non-seminomatous GCTs. Similarly, normal AFP levels are seen in both seminoma with non-seminoma GCTs.<sup>15</sup> Thus, if serum tumour markers are within normal range, it is essential to get a tissue biopsy to guide management. Conversely, if the tumor

markers are elevated ( $\beta$ HCG and AFP) in the presence of a normal testicular exam with retroperitoneal lymph nodes, this may be sufficient for the diagnosis of non-seminomatous GCTs and histological confirmation prior to chemotherapy may not be required.<sup>17</sup>

In this case, we report a 34-year male with incidental findings of retroperitoneal adenopathy and a burned-out testicular primary. This patient had spontaneous regression in the retroperitoneal lymph nodes. Subsequently, this patient has been able to avoid the short-term and long-term toxicity associated with cisplatin-based chemotherapy, which is the standard treatment for most advanced non-seminomatous GCTs.

### **Conclusions**

The burned-out testicular tumour presents with an interesting therapeutic dilemma given spontaneous regression of retroperitoneal lymph nodes. These cases require a multi-disciplinary approach to review all options for management.

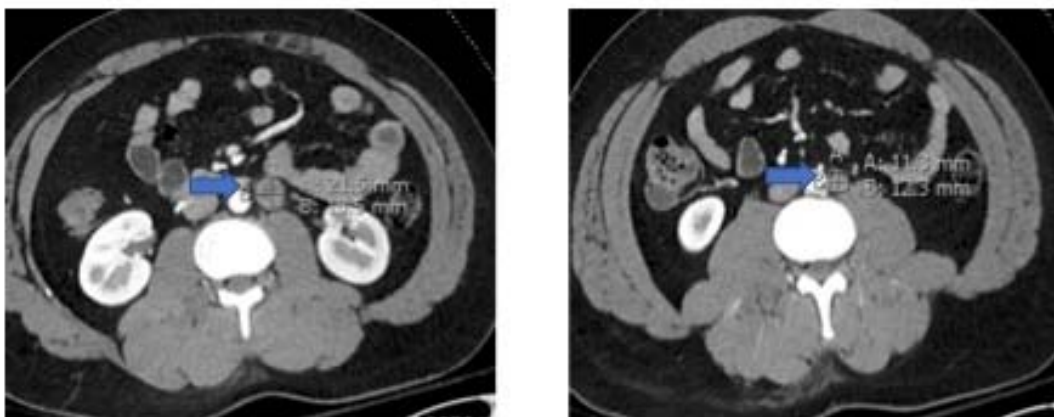
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Figures and Tables

**Figure 1.** Computed tomography scan of abdomen showing two para-aortic lymph nodes (2.1x1.8 cm and 1.1x1.2 cm).



**Figure 2.** Followup computed tomography scan showing regression in the size of the para-aortic lymph nodes (1.5 cm and 1.1 cm in short axis, respectively).

