

Retroperitoneal extension of massive ulcerated testicular seminoma through the inguinal canal: A case report

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Summary *Introduction: Testicular cancers represent about 5% of all urological malignancies and 1-1.5% of all male neoplasms. Most of the testicular cancers are localized (68%) at diagnosis. Bulky masses in the scrotum are rare. We present a rare case of bulky testicular cancer with retroperitoneal spread through the inguinal canal. Case report: A 44-year-old man came to the emergency department referring weakness and the presence of a scrotal mass. At physical examination, a voluminous mass was found, with necrotic phenomena within the scrotum. Abdomen was tense and sore. Abdominal CT scan revealed a bulky testicular mass spreading to the retroperitoneal space through the inguinal canal with node enlargement. Patient underwent orchiectomy with excision of infiltrated scrotum skin. Histologic diagnosis confirmed a typical form seminoma. The patient was then treated with a cisplatin-based chemotherapy, with a partial response. The patient recently relapsed and he is being treated with a new line of chemotherapy and subsequent surgery with or without radiotherapy. Conclusions: We described a rare presentation of testicular cancer. This case highlights the importance of a multidisciplinary approach to rare testis tumour presentation and early diagnosis for testicular cancers.*

KEY WORDS: Testicular cancer; Large seminoma; Retroperitoneal space; Inguinal lymph nodes.

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INTRODUCTION

Testicular tumours (TTs) represent about 5% of all urological malignancies and 1-1.5% of all male neoplasms (1). The incidence of testicular cancers is 3-6 new cases per 100.000 males in Western countries, with an increase observed in the past 30 years (2), probably as a consequence of pollution. These rare tumours are more frequent between 18 and 35 years and in Scandinavian countries (1, 3).

Risk factors include the presence of a tumour in the contralateral testicle, *Germ Cell Neoplasia in Situ* (GCNIS), Klinefelter's syndrome, cryptorchidism or undescended testicle, family history of testicular cancer (2).

World Health Organization (WHO) classification distinguishes testicular neoplasms into germ cell-derived (95%) and non-germ cell neoplasms (2). The most frequent *germ-cell tumours* (GCTs) are seminoma (40-50% of cases). In about 80% of the cases, seminoma presents in a typical form (4).

TTs are often localized (68%) and confined to the testis. Locally advanced tumours usually remain confined to the scrotum. Although rare, extension of the primary tumour to the inguinal canal can be observed, mostly among *non-germ cell TTs* (NGCTTs) (5). To the best of our knowledge, no previous case of large seminoma spreading in the retroperitoneum through inguinal canal has been described. In this study we report the first case of testicular cancer presenting as a voluminous ulcerated testicular mass.

CASE REPORT

A 44-year-old man self-referred to the emergency room of our hospital because of a voluminous scrotal mass associated to abdominal and pelvic pain. The patient had no fever, poor nutritional conditions and pale skin. Clinical history included smoke and thyroid goitre. Physical examination showed a voluminous scrotal mass likely with colliquative necrotic phenomena and abdominal extension (Figure 1A). The abdomen was tense and slightly painful on deep palpation.

Laboratory tests showed an anaemia with reduction in *red blood cell* (RBC) count ($3.1 \times 10^6 \text{ mm}^3$; normal range $4.5\text{-}5.3 \times 10^6 \text{ mm}^3$), *haemoglobin* (Hgb) of 6.9 g/dl (normal range 13-16 g/dl), Hct of 24 % (normal value 37-49%). Tumour markers were elevated, in particular β -HCG was 4873 mIU/ml (normal range between 0-5 mIU/ml), α -fetoprotein was 33.4 ng/ml (normal values less than 6 ng/ml were evaluated) and LDH was 9047 U/L (normal range 313-618 U/L). Complete blood tests are shown in the Table 1.

The patient underwent an abdominal CT scan, showing a voluminous scrotal sac (28 x 13 x 12 cm) with solid tissue sized 16 x 16 cm, occupying the scrotum with extension to the left inguinal canal and to the retroperi-

No conflict of interest declared.

Figure 1.

(A) Voluminous and necrotic scrotum at the diagnosis. (B) Intra-operative photograph. (C) Post-operative photograph of resected scrotal mass. (D-F) Computed tomography shows extensive abdominal diffusion of tumour.

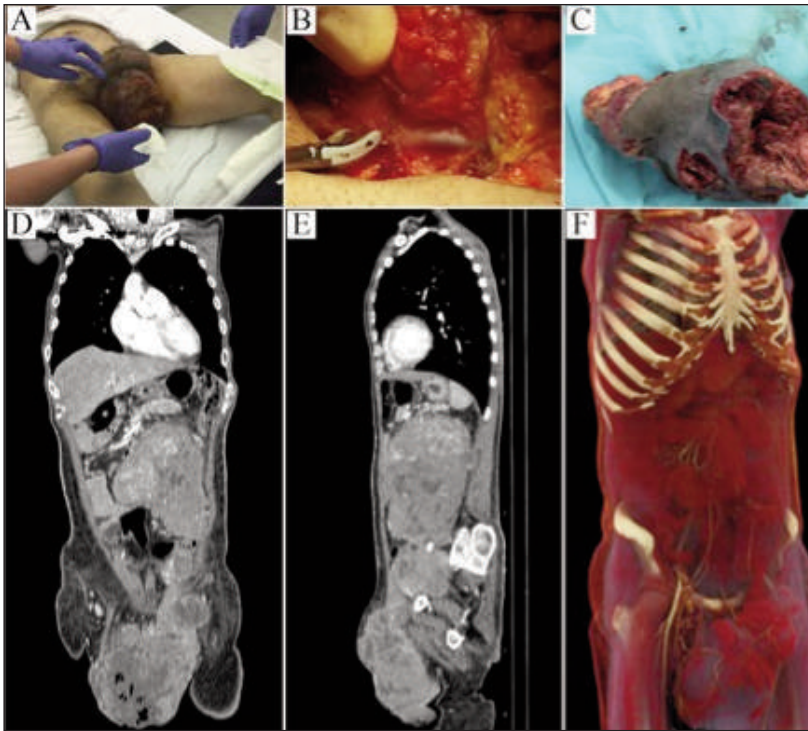


Table 1.
Blood tests.

	Result	Reference values
Haemoglobin (g/dl)	6.9	13.0-16.0
Red blood cells (mmc)	3.1	4.5-5.3 x 10 ⁶
Hematocrit (%)	24	37.0-49.0
MCH (pg)	21.8	25.0-35.0
MCHC (g/dl)	28	32.0-36.0
White blood cells (u/L)	8.97	4.00-10.0 x 10 ³
Platelets (mmc)	479	150-450 x 10 ⁴
Fibrinogen (mg/dl)	542	189-400
Glucose (mg/dl)	117	74-106
Creatinine (mg/dl)	0.90	0.66-1.25
Urea (mg/dl)	20.0	9.0-20.0
Albumin (g/dl)	2.6	2.5-5.2
Sodium (mmol/L)	138	136-146
Potassium (mmol/L)	5.40	3.50-5.10
Calcium (mmol/L)	2.3	2.1-2.55
AST (U/L)	35	15-46
ALT (U/L)	21	11-66
Amylase (U/L)	73	30-110
Lipase (U/L)	157	313-618
CPK (U/L)	21	55-170
Total Bilirubin (mg/dl)	0.60	0.20-1.30
LDH (U/L)	9047	313-618
Beta-HCG (mIU/ml)	4873	0-5
Alpha-fetoprotein (ng/ml)	33.4	< 6

MCH: Mean Cell Hemoglobin, MCHC: Mean Cell Hemoglobin Concentration, AST: Aspartate aminotransferase, ALT: Alanine aminotransferase, CPK: Creatin phosphokinase, LDH: Lactate Dehydrogenase, HCG: Human Chorionic Gonadotropin.

toneal space. Moreover, there was a pathological involvement of the left inguinal (11 x 7 cm) and iliac-obturator (10 x 6 cm) lymph nodes infiltrating the external iliac vein.

In addition, pathologic retroperitoneal lymphatic tissue was documented along the abdominal aorta for a longitudinal extension of about 20 cm, resulting in compression of the inferior vena cava and infiltration of the external iliac vein, left renal vein and left ureter with signs of post-renal obstructive uropathy. No distant lesions to parenchymal organs were detected (Figure 1D-F).

In the context of reduced Hgb, the patient underwent a transfusion and was hospitalized in the urology department. Unilateral orchiectomy with lymph node dissection was performed (Figure 1B). First, an inguinal incision was made and the enlarged nodes of left inguinal chain were identified. There was no clear distinction between metastatic lymph nodes and the testicular mass. After cautious isolation left inguinal nodes were dissected. Subsequently the inguinal portion of the tumour was also isolated and, after incision enlargement to the scrotum, was removed. Finally, scrotal

portion of the mass was resected alongside with the surfacing necrotic skin (Figure 1C). The right testis and penile shaft were preserved (Figure 2).

Histological examination showed a typical seminoma. The neoplasm infiltrated the skin up to ulcerating it and involved lymph nodes (pT4, pN3, pM1, S3). The presence of an intra-tumour phlogistic infiltrate was also revealed. Molecular morphology investigations with immunohistochemical characterization of the tumour showed positivity for *Octamer-binding transcription factor* (OCT) 3/4, *Placental alkaline phosphatase* (PLAP), β -HCG, CD117, *Leukocyte common antigen* (LCA, in the intra tumour inflammatory component) and CD30.

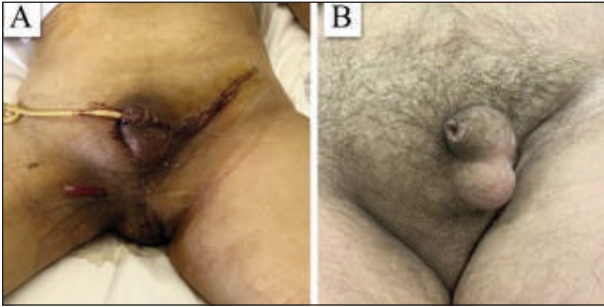
Following surgery, the patient received four three-weekly cycles of standard BEP (*Bleomycin* 30 UI IV weekly on days 1-8 and 15; *Etoposide* 100 mg/m² IV on days 1-5; *Cisplatin* 20 mg/m² IV on days 1-5 (6).

CT scan taken one month after the completion of chemotherapy showed a great deal of reduction in the retroperitoneal lymph node masses (6 x 4 cm current vs 17 x 12 cm prior). Serum level of tumour markers was also decreased (Table 2).

Subsequently, the patient underwent CT-scan at 3-month intervals. Abdominal and chest imaging showed a *stable disease* (SD) according to the *Response Evaluation Criteria in Solid Tumors* (RECIST) (7) with no parenchymal metastases for one year and half. A *progressive disease* (PD) was documented after 18 months. CT-scan showed a new dimensional increase in the left periaortic lymph node tissue (55 x 35 mm current vs 50 x 25 mm prior), along the left external iliac chains (57 x 44 cm current vs 47 x 37 cm prior) and the appearance of infiltration of the left iliac and

Figure 2.

(A) Post-operative photograph. An inguinal and scrotal incision was made and the voluminous necrotic scrotal mass was removed. (B) Photograph of inguino-scrotal region on follow-up at 12 months.

**Table 2.**

Tumour markers in the eighteen months following chemotherapy.

	November 2018	March 2019	June 2019	April 2020
Alpha-fetoprotein (ng/ml)	9	3.20	1.60	60.79
Beta-HCG (mIU/ml)	<1	<1	<1	<1
LDH (U/L)	422	179	216	2580

psoas muscles by the pathological lymph node tissue (7). Moreover, the patient underwent a PET-CT scan that showed an intense metabolic activity corresponding to a voluminous lymph node masses (10 x 13 cm) in the left iliac region, infiltrating the left ileo-psoas muscle.

After considering the disease progression, we had a multi-disciplinary meeting. As salvage chemotherapy the patient is being treated with four three-weekly cycles of standard TIP (Paclitaxel 175 mg/m² IV on day 1; Cisplatin 20 mg/m² IV on days 1-5; Ifosfamide 1000 mg/m² IV on days 1-5). In case of mass reduction, a combined surgical retroperitoneal lymph node dissection (RPLND) and radiotherapeutic approach will be evaluated.

DISCUSSION

In this report, we described a rare case of large seminoma extending to the inguinal canal with diffuse retroperitoneal spreading and skin ulceration. Presentation at advanced stage or even metastatic at diagnosis is more common for NGCTs (5).

Our case is paradigmatic for several reasons. First of all, the age of diagnosis. Our patient presented a primary testicular cancer in the absence of risk factors and at an age older than usual. This highlights the importance of genital examination at every age, even when the probability of a testicular tumour is low. Moreover, our patient had a very unusual presentation. Indeed, while most of testis cancers are diagnosed as localized tumours of few centimeters in diameter, in our case the patient turned to physicians only when symptomatic. When investigating the reasons why the patient delayed the primary intervention, a complex mix of personal, familiar and social causes emerged. Several studies have shown a detrimental effect of low socio-economic and familiar status on

cancer awareness and intervention timing (8, 9). In a recent analysis Maclooad *et al.* showed that socio-economic status was associated with poorer oncological outcomes and a more difficult access to primary treatment in patients with testicular cancer (10). Our case corroborates these evidences and suggests the importance of a social tissue that could lead to prompt access to primary care and early diagnosis. It is of note that the Italian one is a single payer healthcare system. In consequence, cure costs are not one of the major barriers to early diagnosis and treatment. However, even in this context weaker social strata still exist. Within this strata population could be more susceptible to experiment worse oncological outcomes. In fact, even after a wide surgical excision and associated chemotherapy, as recommended by international guidelines (11), we obtained only a partial response with a subsequent relapse of the disease.

Furthermore, in our case is evident how, even if cisplatin-based regimen is effective on testis cancer, a multidisciplinary approach should be warranted.

Early diagnosis, a multidisciplinary approach and a close follow-up remain mandatory to improve prognosis of testicular cancer (12, 13).

After relapse, our patient will undergo four cycles of TIP (14). Surgery and radiotherapy should be considered in the case of mass reduction and resectable masses with small residual tumour (15). Moreover, a close follow-up of all psychological aspects was planned in consideration of the high psychological burden of testis cancer.

CONCLUSIONS

In conclusion, we reported an extremely rare presentation of locally advanced testis cancer, resulting from the combination of cancer and patient related conditions. Early diagnosis is fundamental to guarantee a good oncological prognosis for testis cancer. Moreover, a multidisciplinary approach is important to guarantee a good oncological outcome.

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