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Malignant Cryptorchid Testis Presenting as an Intra-Abdominal Mass: A Case Report

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Authors' contributions

This work was carried out in collaboration between both authors. Author PMPNM wrote this paper. Author SVY reviewed and proofread the content. Both the authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aims: An individual with testicular malignancy can present with a large abdominal mass. Such mass can be secondaries in retroperitoneal lymph nodes (RPLN), a malignant cryptorchid testis itself, or both. This article aims to highlight the importance of thorough genitourinary examination in males presenting with abdominal masses and of vigilance at image interpretation and explorative surgery for multiplicity of masses.

Presentation of Case: We report a case of one adult male in his forties presenting with a large intra-abdominal mass. Examination revealed bilateral cryptorchidism; historical inquiry revealed a state of subfertility for nearly 10 years. Case has neither sought treatment for subfertility, nor was aware of emptiness of his scrotum. Computed Tomography (CT) revealed one retroperitoneal mass suspicious of malignant intra-abdominal testis. At laparotomy, we encountered two separate intra-abdominal masses connected through a pedicle, a RPLN mass, and a malignant intra-abdominal testis.

Discussion: Cryptorchid abdominal testis poses increased risk of malignancy. Cases as such present in interesting ways; an intra-abdominal mass is the commonest. Such a mass can contain either or both primary testicular tumor and enlarged RPLN which, CT can often clearly define.

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However, there are times when surgeon face intra-operative surprises, encountering two masses instead of one, as reported by the imaging.
Conclusion: This case reinstates the necessity of a thorough genito-urinary examination as a part of routine evaluation of males presenting with abdominal masses. Radiologist and operating surgeon need to be vigilant to look for multiplicity of masses, corresponding to primary testicular tumor and RPLN mass.

Keywords: Testicular tumor; cryptorchidism; retroperitoneal lymph node mass; subfertility; empty scrotum; seminoma.

1. INTRODUCTION

Men who have a positive family history, cryptorchidism, gonadal dysgenesis, or intra-tubular germ cell neoplasia carry a high risk of developing testicular neoplasia [1,2,3]. The incidence of testicular cancer is 3 to 30 times higher in men with cryptorchidism compared to the general population [1,4]. Also, there is a direct link between subfertility and the incidence of testicular cancer [5].

Metastatic spread of testicular cancer follows lymphatic drainage of testes to retroperitoneal lymph nodes according to their embryological origin. Such lymph nodal disease can manifest as large intra-abdominal masses. This may be the primary presentation of certain patients as testicular cancers are often painless, hence primary tumors may go unnoticed. This is often true in the case of cryptorchidism where testis is not present within the scrotal sac, to manifest any symptoms in the first place. There are cases where patients have presented initially with supra-clavicular lymph node enlargement, which led to the diagnosis of malignancy in the testis [6].

Oftentimes cryptorchidism is identified at pre-pubertal age by concerned parents. The natural course of such presentation is surgical mobilization and orchidopexy within the scrotal sac to limit the likelihood of malignant transformation, let alone to aid in early identification of such transformation, if it were to occur [7]. Here, we report a case of an adult married male in his forties who had been subfertile for 10 years with bilateral empty scrotum about which he was unaware, until presenting with a primary tumor in one of the cryptorchid testes and retroperitoneal lymph nodal metastasis constituting a large intra-abdominal mass. This report also highlights the importance of thorough genito-urinary examination during clinical evaluation of males presenting with abdominal masses.

2. PRESENTATION OF CASE

A 44-year-old tea-estate worker presented to our tertiary care surgical unit with a three-day history of abdominal pain. He had already been evaluated at a regional tertiary care center where a large intra-abdominal mass in the right lumbar and iliac fossa region had been discovered. The results of the Complete blood count (CBC) and serum biochemistry panels were normal. Ultrasound scan (USS) has shown a large solid mass occupying the right flank and pelvis measuring 20cm x 13.3cm, moderately vascular with few necrotic areas raising the suspicion of GIST or Lymphoma. Subsequent evaluation under our care revealed the intra-abdominal mass consistent with the ultrasound findings but also the presence of a bilateral empty scrotum. The scrotal sac was underdeveloped. The right testis was not felt but the left testis was felt in the inguinal canal close to the superficial inguinal ring. Further questioning revealed that the patient was married for 10 years but had no children. The patient was unaware of the absence of testes within the scrotum.

Workup constituted a panel of Tumor markers which revealed Alfa fetoprotein (AFP) level 1.0ng/mL (<8.78), Total Beta hCG level 213.80IU/L (<5), and LDH level 3021 U/L (125-220). The hormone panel revealed Prolactin 218.50 mIU/L (73-412), FSH 22.80 IU/L (1.4-15.4), LH 14.68 IU/L (1.2-7.8). Computed Tomography (CT) performed with the administration of intravenous contrast (Fig.1) reported the left testis to be visualized in the inguinal canal and a large heterogeneously enhancing mass in the right iliac fossa measuring 18.6 x 12.6 x 14.8 cm in size, showing solid and cystic areas and high-density material within. The mass was draining into the Inferior Vena Cava (IVC) via a vein below the right renal vein, which was assumed to be the right testicular vein. Mass did not appear to have infiltrated the abdominal wall, bowel, or nearby vascular or soft tissue structures. There were no suspicious lesions in

the liver or nodules in the lungs. Bilateral kidneys were normal. The diagnosis of stage II germ cell tumor (GCT) with retroperitoneal lymph node metastasis was made.

Following a multidisciplinary team (MDT) discussion, a decision was made to operate on the abdominal mass followed by adjuvant chemotherapy based on final histopathology. The patient underwent exploratory laparotomy with a midline incision. A large mass occupying the right iliac fossa and pelvis was identified (Fig. 2) displacing the small bowel to the left with adhesions to the bowel. However, the mid-segment of the sigmoid colon was deeply involved by the tumor necessitating segmental resection of the sigmoid colon with the specimen. The enlarged right testis sitting on the pelvis at the retro-pubic position was attached to the main mass through a pedicle and was removed en bloc. Colo-colic anastomosis was done with a single-layer interrupted suture technique.

Final histopathology of the resected specimen showed a 105x70x38mm sized seminoma with infiltration of rete testis stroma, vascular invasion, lymphatic invasion and omental tissue, serosa, and muscular wall of sigmoid colon infiltration by the tumor. The accompanying lymph node mass was 140x85x80mm in size. Staging was considered as pT4 pN3 pM1b, S3.

3. DISCUSSION

Germ cell tumor (GCT) of the testis is the most common solid malignancy in males, affecting 1-2% of the male population [8]. GCTs can be either seminoma or Non-seminomatous Germ cell tumors (NSGCT), affecting males in their 40s and 30s respectively. Seminomas have a slow growth and more favorable prognosis compared to NSGCTs. Tumor markers like AFP, beta-hCG, and LDH help determine the histological subtype, hence the prognosis and staging of testicular tumors [9,10]. Although mainly secreted by choriocarcinoma, 5-10% of seminomas and 25% of yolk sac tumors can produce beta-hCG. Although our patient who was in his forties had a high beta-hCG level indicative of choriocarcinoma, final histology revealed a seminoma, resulting in a shift in the projected prognosis. This also implies that the correlation between tumor markers and histological type is weak.

Cryptorchidism or undescended testes (UDT) is the most common congenital genitourinary abnormality encountered, affecting 1% of males [6]. Location of cryptorchid testis can be high scrotal, inguinal canalicular or abdominal. The higher the location of undescended testis, the greater the risk of malignancy. Abdominal testis poses a risk as high as 30% [6]. Our patient had an abdominal testis on the malignancy-borne side and inguinal canalicular testis on the contralateral side. Cryptorchidism is bilateral in 10% of cases [4].

Although a majority of testicular tumors present with disease limited to the testis, 20% may present with retroperitoneal lymph node metastasis [11]. This is often the case with malignancy-borne cryptorchid testis as the primary tumor is less likely to be detected early.

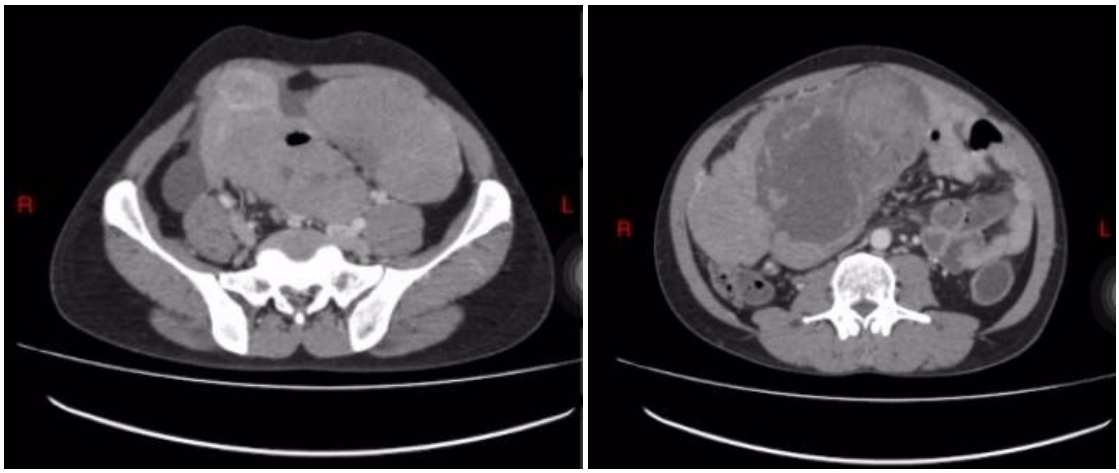

Fig. 1. Cross sectional CECT images showing a. retroperitoneal lymph node mass, b. malignant testicular tumor

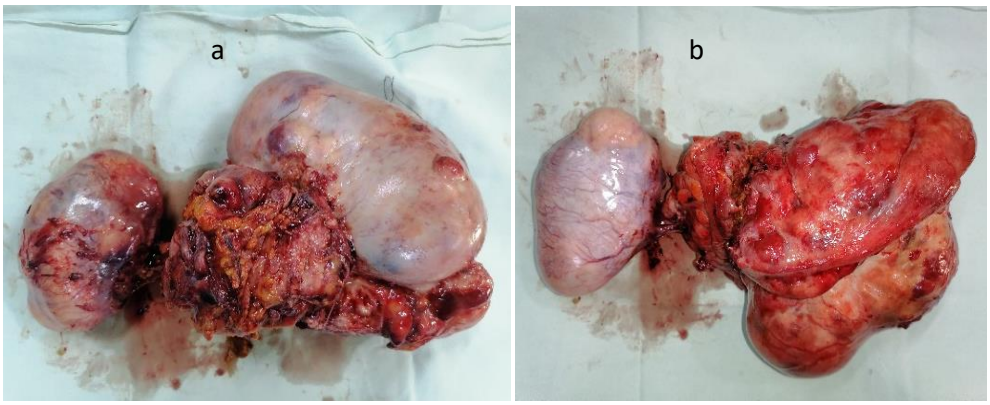

Fig. 2 Post-operative specimen of retroperitoneal lymph nodal mass and testicular tumor a. anterior view, b. posterior view with involved segment of sigmoid colon resected en-bloc

Treatment of GCTs is surgical excision of the primary testicular tumor followed by adjuvant chemotherapy for stage II, III, and IV diseases. Retroperitoneal lymph nodal disease (RPLND) of seminomas can alternatively be managed with radiotherapy for which it is far more sensitive, unlike NSGCTs. Persistent RPLND after systemic therapy, in either situation, is managed by consolidative surgical resection of the nodal mass. In our case, laparotomy was indicated to remove intra-abdominal primary testicular malignancy, and concurrent resection of associated RPLN mass, noted intra-operatively to cause compressive as well as infiltrative effect on neighboring organs, was done before chemotherapy. Interestingly, contrast-enhanced CT scan failed to distinguish separately the lymph node mass and the primary testicular tumor pre-operatively.

According to the prognostic staging of the International Germ Cell Cancer Collaborative Group (IGCCCG), due to extremely high LDH levels (more the 10 times the upper limit of normal), our patient fell into the 'high-risk' group. Therefore four cycles of adjuvant chemotherapy with BEP (Bleomycin, Etoposide, and Cis-platin) was recommended for this patient [12].

The incidence of subfertility in bilateral cryptorchidism is 6 times higher compared to the general population [13]. Azoospermia has been identified in 10% of men with unilateral and 32% of men with bilateral cryptorchidism [14]. Our patient has been vigilant enough to notice the production of a small amount of semen and its odd watery consistency which has led him to accept his state of subfertility without seeking therapy. That, and the fact that he could not produce enough semen for the test made us the decision not to order Seminal Fluid Analysis (SFA), which is rather an expensive investigation as well.

Pre-pubertal orchidopexy can lower, although not eliminate the risk of malignancy. It also enables early identification of malignant transformation due to its presence at a palpable location. Americal Urological Association (AUA) guidelines recommend close follow-up to those patients and encourage self-examination after puberty [15]. It is seldom, for a man to reach his adulthood with bilateral cryptorchidism un-noticed, a married man with a long history of subfertility to never seek treatment, but not infrequent for such an individual to develop malignancy in his testes and present at an advanced stage.

In summary, we present a case of large intra-abdominal mass which was initially misdiagnosed as for a GIST or a lymphoma. Further evaluation revealed that the patient has a bilateral empty scrotum; Contrast-enhanced CT revealed an intra-abdominal mass in the right iliac fossa tomographically indicative of malignancy-borne abdominal testis which corresponded to a lymph nodal mass encountered intra-operatively. The malignant right testis attached to the lymph nodal mass through a narrow pedicle was found in the retro-pubic region during operation which the CT failed to report accurately pre-operatively. The intimately adhered segment of the sigmoid colon, also noted intra-operatively was resected en-bloc followed by end-to-end colo-colic anastomosis. The contralateral inguinal canalicular cryptorchid testis was not operatively handled simultaneously as per the MDT decision, which needed orchidectomy in a staged fashion to avoid future malignant transformation. He was

subjected to adjuvant chemotherapy as per a high-risk case.

4. CONCLUSION

Thorough genito-urinary examination should be a part of the routine evaluation of young adult males presenting with abdominal masses, to avoid missing a malignant cryptorchid testis. Surveying separately for the intra-abdominal primary tumor as well as for an accompanying retroperitoneal lymph node mass should be part of CT reporting when an intra-abdominal mass is evaluated in a cryptorchid man, to avoid intra-operative surprises. The operating surgeon should also be vigilant to look for 2 retroperitoneal masses, corresponding to the primary testicular tumor and associated lymph nodal mass.

CONSENT

All authors declare that written informed consent was obtained from the patient for the publication of this case report and accompanying images.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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