Case report

Malignancy in a Testis of a Bilateral Cryptorchid Man Revealed by a Large Intra-abdominal Mass – A case report

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. Aim of this article is 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 fourties presenting with a large intra-abdominal mass. Examination revealed bilateral cryptorchidism; historical enquiry 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. 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 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*

UNDER PEER REVIEW

1. INTRODUCTION

Men who are having a positive family history, cryptorchidism, gonadal dysgenesis or intra-tubular germ cell neoplasia carry a high risk of development of testicular neoplasia[1][2][3]. The incidence of testicular cancer is 3 to 30 times higher in men with cryptorchidism compared to general population[1][4]. Also, there is a direct correlation 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 and 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 tumor may go unnoticed. This is often true in case of cryptorchidism where testis in 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, that led to diagnosis of malignancy in the testis[6].

Oftentimes cryptorchidism is identified at pre-pubertal age by vigilant parents. Natural course of such presentation is surgical mobilization and orchidopexy within the scrotal sac to reduce the risk of malignant transformation and to facilitate early identification of such

transformation, if it were to occur[7]. Here, we report a case of an adult married male in his fourties who has been subfertile for 10 years without seeking treatment and had bilateral empty scrotum which he was unaware of, until presenting with primary tumor in one of the cryptorchid testes and retroperitoneal lymph nodal metastasis constitutive of a large intra-abdominal mass. This 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 sugical unit with the complaint of abdominal pain for 3 days. He has been evaluated at a regional tertiary care center where a large intra-abdominal mass in right lumbar and iliac fossa region has been found. Complete blood count 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 querying the suspicion of GIST or Lymphoma. Our evaluation revealed intra-abdominal mass corresponding to the ultrasound findings but also bilateral empty scrotum. Scrotal sac was under-developed. Right testis was not felt but left testis was felt in inguinal canal close to the superficial inguinal ring. On further questioning, patient revealed that he has been married for 10 years without children, for which he has not sought remedy. The absence of testes within the scrotum was not known by the patient.

Workup constituted Tumor markers panel 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). 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 after administration of intravenous contrast **(Fig.1)** reported the left testis to be visualized in the inguinal canal and a large heterogenously enhancing mass in right iliac fossa measuring 18.6 x 12.6 x 14.8 cm in size which show solid and cystic areas and high-density material within the mass. There was a vein draining into Inferior Vena Cava (IVC) below right renal vein which could be right testicular vein. Mass did not appear to invade 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.

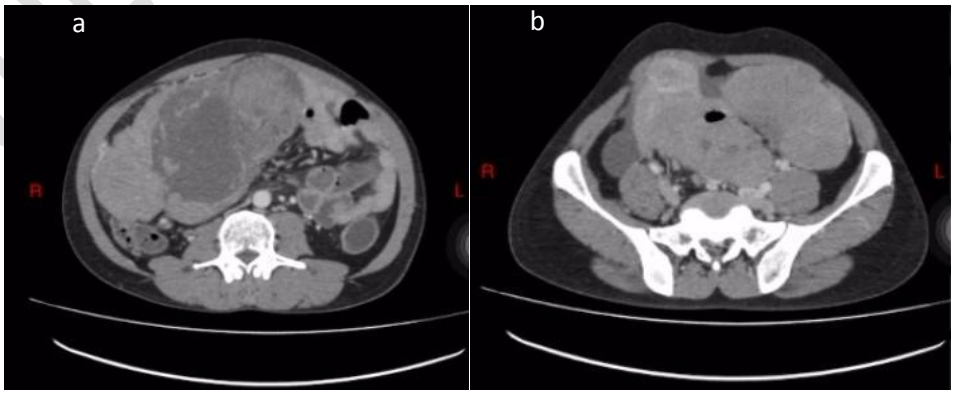

Fig. 1 Cross sectional CECT images showing **a.** Retroperitoneal lymph node mass, **b.** malignant testicular tumor

UNDER PEER REVIEW

Following multidisciplinary team (MDT) discussion, primary surgery for abdominal mass was decided to be done followed by adjuvant chemotherapy after final histo-pathology. Patient underwent exploratory laparotomy with midline incision. Large mass occupying the right iliac fossa and pelvis was identified **(Fig. 2)** pushing the small bowel to the left with dissectable adhesions to the bowel. However mid-segment of sigmoid colon was intimately involved by the tumor which required segmental resection of sigmoid colon with the specimen. Enlarged right testis sitting on the pelvis at retro-pubic position was attached to the main mass through a pedicle, and was removed en-bloc. Colo-colic anastomosis was done with single-layer interrupted suture technique.

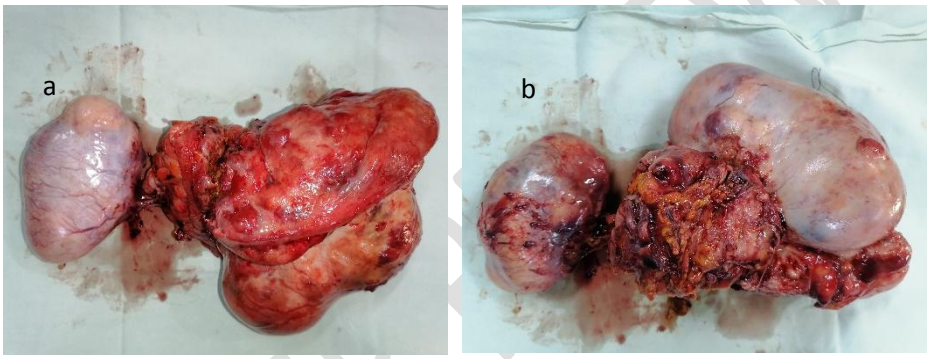

Fig. 2 Post-operative specimen of retroperitoneal lymph nodal mass and testicular tumor a**.** anterior view, b**.** posterior view with involved segment of sigmoid colon resected en-bloc

Final histo-pathology 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. Associated 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 male population[8]. GCTs can be either seminoma or Non-seminomatous Germ cell tumors (NSGCT), affecting males in 40s and 30s respectively. Seminomas have a slower progression and more favourable prognosis compared to NSGCTs. Tumor markers such as AFP, beta-hCG and LDH are helpful in determining the histological subtype, hence prognosis and staging of testicular tumors[9][10]. Although mainly secreted by choriocarcinoma, 5-10% of seminomas and 25% of yolk sac tumors can secrete beta-hCG. Although our patient who was in his fourties had a high beta-hCG level indicative of a

choriocarcinoma, final histology revealed it as a seminoma, which led to change in the predicted prognosis. This also implicates that correlation of tumor markers with 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. Higher the location of undescended testis, higher the risk of malignancy. Abdominal testis pose 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 majority of testicular tumors present with disease limited to 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.

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

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

Incidence of subfertility in bilateral cryptorchidism is 6 times higher compared to general population[13]. Azoospermia has been noted 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 small amount of semen and its unusual watery manner which has led himself to accept the state of subfertility without seeking treatment. That, and the fact that he could not produce enough sement for the test made us the decision not to order Seminal Fluid Analysis (SFA), which is rather an expensive investigation.

Pre-pubertal orchidopexy can reduce, although not completely eliminate the risk of malignancy as well as allow early identification of malignant transformation due to 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 unto his adulthood with bilateral cryptorchidism un-noticed, a married man with a long history of subfertility never to 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 evaluated as for a GIST or a lymphoma. Further evaluation revealed that patient has bilateral empty scrotum; Contrast enhanced CT revealed an intra-abdominal mass in right iliac fossa tomographically suspicious of malignancy-borne abdominal testis which in-fact 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 accuratelty pre-operatively. Intimately adhered segment of sigmoid colon, also identified intra-operatively was resected en-bloc followed by end-to-end colo-colic anastomosis. Contralateral inguinal cannalicular cryptorchid testis was not operatively handled simultaneously as per MDT decision, which needed orchidectomy in

UNDER PEER REVIEW

a staged fashion to avoid 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 routine evaluation of young-adults males presenting with abdominal masses, in order to avoid missing a malignant cryptorchid testis. Surveying seperately for the intra-abdominal primary tumor as well as for an associated retroperitoneal lymph node mass should be part of CT reporting when an intra-abdominal mass is evaluated in a cryptorchid man, to minimize intra-operative surprizes. 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 publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial Board members of this journal on request.

REFERENCES

1. Accardo G, Vallone G, Esposito D *et al.* Testicular parenchymal abnormalities in Klinefelter syndrome: A question of cancer? Examination of 40 consecutive patients. *Asian J Androl* 2015;**17**:154–8.

2. Rudzinski JK, Carlock HR, Beech BB *et al.* Empty hemiscrotum and a giant abdominal mass case report. *AME Med J* 2019;**4**:42–42.

3. Ferguson L, Agoulnik AI. Testicular cancer and cryptorchidism. *Front Endocrinol (Lausanne)* 2013;**4**:656–9.

4. Alamer. Right Testicular Seminoma in Bilateral Cryptorchidism: A Case Report. *World J Nephrol Urol* 2013;**2**:15–7.

5. Peng X, Zeng X, Peng S *et al.* The association risk of male subfertility and testicular cancer: A systematic review. *PLoS One* 2009;**4**:1–8.

6. Tazi MF, Riyach O, Ahsaini M *et al.* Tumor in undescended intrapelvic testis revealed by supraclavicular lymphadenopathy: A case report and literature review. *BMC Res Notes* 2013;**6**:2–4.

7. Banerji J, Chandra Singh J. Does early orchidopexy reduce risk of testicular cancer? *Indian J Urol* 2008;**24**:430–1.

8. Siegel RL, Miller KD, Jemal A. Cancer statistics, 2019. *CA Cancer J Clin* 2019;**69**:7–34.

9. Vasdev N, Moon A, Thorpe AC. Classification, epidemiology and therapies for testicular germ cell tumours. *Int J Dev Biol* 2013;**57**:133–9.

10. Milose JC, Filson CP, Weizer AZ *et al.* Role of biochemical markers in testicular cancer: Diagnosis, staging, and surveillance. *Open Access J Urol* 2011;**4**:1–8.

11. Woodward PJ, Sohaey R, O'Donoghue MJ *et al.* From the archives of the AFIP. Tumors and tumorlike lesions of the testis: Radiologic-pathologic correlation. *Radiographics* 2002;**22**:189–216.

UNDER PEER REVIEW

12. Hayes-Lattin B, Bleyer A. Testicular Cancer. *Pediatr Oncol* 2017:307–18.

13. Loebenstein M, Thorup J, Cortes D *et al.* Cryptorchidism, gonocyte development, and the risks of germ cell malignancy and infertility: A systematic review. *J Pediatr Surg* 2020;**55**:1201–10.

14. Rübben I. Hodenhochstand und Fertilität. *Urol* 2016;**55**:890–7.

15. Kolon TF, Herndon CDA, Baker LA *et al.* Evaluation and treatment of Cryptorchidism: AUA guideline. *J Urol* 2014;**192**:337–45.

UNDER PEER REVIEW