

Original Research Article

Bilateral undescended testes with intraabdominal non seminomatous germ cell tumor – a rare clinical challenge

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ABSTRACT

Cryptorchidism is the most common congenital anomaly of the male genitalia, with nearly 72-77% of undescended testes located in the inguinal canal. Among malignancies arising in undescended testes, seminoma is the most frequent type, accounting for approximately 50-60% of cases. Tumors arising from intra-abdominal testes may grow to considerable sizes before detection, and delayed diagnosis often results in advanced-stage disease at presentation. We report a case of bilateral cryptorchidism in a young male who presented with vague lower abdominal pain, without any prior history of abdominal swelling or distension. The patient had noticed an empty scrotum since around 10 years of age but did not seek medical evaluation due to social stigma. Eventually, at the age of 21 years, he developed a large intra-abdominal non-seminomatous germ cell tumor arising from an undescended testis. The patient subsequently underwent an exploratory laparotomy with right radical orchidectomy and left orchidectomy, followed by advice to undergo adjuvant chemotherapy and regular follow-up.

Keywords: Cryptorchidism, Intraabdominal mass, Non seminomatous germ cell tumor, Testicular tumor, Undescended testes

INTRODUCTION

Descent of the testicles is a necessary factor for proper testicular development. Cryptorchidism is the absence of at least one testicle from the scrotum. Cryptorchidism (undescended testis) is a well-established risk factor for testicular germ cell tumours. Approximately 10% of testicular germ cell tumours occur in patients with a history of cryptorchidism, with seminoma being the most common histological subtype, although non-seminomatous germ cell tumours such as embryonal carcinoma may also occur. Undescended testicles (Cryptorchidism) are associated with decreased fertility (especially bilateral cases), increased testicular germ cell tumours (overall risk under 1%), testicular torsion, inguinal hernias, and psychological problems.^{1,2} 80% of undescended testicles palpated within inguinal canal or high scrotal area, 20% of undescended testicles not palpated in which 50% lie in abdomen, 50%

are atrophic.³ In our case, patient had bilateral testes located in the abdomen. About 3% of full-term and 30% of premature male infants are born with one or both testicles undescended. The testes normally descend by month 7 of gestation. Approximately 80% of cryptorchid testes descend by the third month of life after birth. This makes the true incidence around 1%. If the testis has not descended by six months of age, it is unlikely to do so spontaneously, and surgical correction should be considered.^{1,2} Seminomas are the most frequent histological type in cryptorchid testes (accounting for roughly 60% to 75% of tumors), non-seminomatous germ cell tumors (NSGCT) comprise ≈25 to 40% of cryptorchid-associated testicular malignancies. We encountered with case of bilateral cryptorchidism in 21-year unmarried male, with semen analysis suggestive of azoospermia who did not took treatment for undescended

testes. Due to which, our patient developed intraabdominal testicular non seminomatous germ cell tumor.

CASE REPORT

21-year unmarried male, presented with complaints of pain in lower abdomen since 1 month, intermittent, no history of lump in abdomen, no bowel or bladder complaints. Patient had history of bilateral cryptorchidism since childhood, but did not seek any treatment for the same. On examination, abdomen was scaphoid, no tenderness, no lump on palpation. Bilateral scrotal sac was empty. Hernial orifices were within normal limit, no cough impulse noted. Ultrasonography of abdomen and pelvis suggestive of a fairly well defined, heterogeneously hypoechoic mass lesion measuring approximately 8×6.2×8.1 cm, with interspersed hyperechoic areas within is noted in the pelvis more on the right-side superior to dome of urinary bladder and shows internal vascularity. Posteriorly it is abutting the right iliac vessels which however shows normal flow on color doppler. Bilateral scrotal sac appears empty. Suggestive of malignant neoplastic etiology likely arising from undescended testis. Beta human chorionic gonadotropin (beta hCG)-0.2 mIU/ml, serum LDH-609 u/l, alpha feto protein->1000 ng/ml. Computed tomography of abdomen and pelvis suggestive of empty scrotal sac. A large 8.1×8.2×8.1 cm (AP×ML×SI) sized well-defined hypodense lesion with few central hyperdense areas is noted in lower abdominal region in prevertebral region from inferior border of L4 to body of S2 vertebral level, in midline and to the left, superior to the overdistended bladder. On post contrast study, the lesion shows moderate and heterogeneous irregular predominantly peripheral progressive enhancement with central non enhancing areas within (suggestive of necrotic/cystic component). It shows arterial feeders from the right external iliac artery-suggestive of neovascularity. The lesion seen draining into the right gonadal vein. Posteriorly, the lesion is abutting left common iliac vessels, external iliac vessels and left internal iliac artery, angle of contact (<90°). The lesion is abutting L5-S1 intervertebral disc with focally effaced fat planes. It is seen compressing the left mid and distal ureter. It is also seen focally abutting left psoas muscle with effaced fat planes. Anteriorly, it abuts the anterior abdominal wall with effaced fat planes. Left laterally, it is abutting sigmoid colon with effaced fat planes. Right laterally, the lesion is seen abutting and displacing small bowel loops and sigmoid colon with effaced fat planes. Superiorly, it is seen abutting transverse colon and small bowel loops with effaced fat planes (Figures 1 and 2). The lesion is causing superior displacement of sigmoid colon which is long/redundant. Antero-inferiorly, it is compressing the dome of urinary bladder with effaced fat planes. Right testis not separately visualised from the lesion. A fairly well-defined soft tissue density enhancing oval structure measuring 3×2.1 cm with few peripheral foci of calcifications is noted in the rectovesical space on the left side, postero-inferior to the above-mentioned lesion, could represent left undescended testes. Above-

described features are suggestive of cryptorchidism with malignant neoplastic etiology of right testicular tumor like non-seminomatous germ cell tumor with possible tumour capsular concealed rupture. Semen analysis suggestive of azoospermia. Exploratory laparotomy with radical right orchidectomy with left orchidectomy was done after taking detailed informed consent. Gross appearance of specimen-a tumour is noted involving the entire testis not breaching the tunica, well encapsulated and having variegated appearance, having cystic areas, hemorrhagic areas as well as solid, whitish areas. The fibro fatty tissue has small whitish nodules on its surface (Figure 3). Microscopic findings - multiple section studied shows a tumour with predominant features of yolk sac tumour. Tumour show microcystic/reticular pattern, solid, endodermal sinus pattern. In few foci, sheets of pleomorphic cells with prominent nucleoli. Occasional foci show syncytiotrophoblast like giant cells. Lymphovascular invasion and extensive areas of necrosis and haemorrhage is seen. Spermatic cord margin is free of tumor. Tunica albuginea not breached. Impression: Germ cell tumor of testis-predominantly yolk sac tumor. Foci of embryonal carcinoma and choriocarcinoma to be confirmed by immunohistochemistry (IHC). Sections studied from left testis shows seminiferous tubules, few of them are calcified. No evidence of malignancy. The patient advised to follow up for further chemotherapy, but because of financial constraints, the patient did not follow up.



Figure 1: Computed tomography of abdomen and pelvis in axial section showing testicular tumor (black arrow).



Figure 2: Computed tomography of abdomen and pelvis in coronal section showing testicular tumor (black arrow).



Figure 3: Gross appearance of testicular tumor.

DISCUSSION

Cryptorchidism is a well-established risk factor for testicular cancer, increasing the likelihood of malignancy in the affected testis by approximately four-to sixfold. However, performing orchidopexy before puberty significantly reduces this risk, lowering the relative risk of testicular cancer to approximately two-to threefold compared with the general population.^{4,5} A meta-analysis evaluating patients with cryptorchidism demonstrated only a modest increase in the risk of testicular cancer in the contralateral normally descended testis, with a relative risk (RR) of 1.74 (95% confidence interval [CI]: 1.01-2.98).⁶ Serum tumor markers (AFP, HCG, and LDH) should be checked before any intervention, including orchidectomy, as these may be elevated in NSGCT. Serum alpha-fetoprotein (AFP) levels are elevated in approximately 50-70% of patients with NSGCT. Pure seminomas do not produce AFP. Therefore, an elevated AFP level in a patient with a histological diagnosis of pure seminoma suggests the presence of an occult NSGCT component, and such cases should be managed as mixed germ cell tumours. Human chorionic gonadotropin (β -hCG) is produced by syncytiotrophoblastic giant cells and may be elevated in some seminomas as well as in certain NSGCTs, particularly choriocarcinoma and embryonal carcinoma. Lactate dehydrogenase (LDH) is a relatively nonspecific tumor marker that is elevated in approximately 20-60% of patients with NSGCT. Its elevation is commonly associated with a high tumor burden and bulky disease.^{7,8} Radical inguinal orchidectomy is the standard procedure for obtaining a definitive tissue diagnosis and also serves as definitive treatment in patients with stage I disease. In contrast, testicular biopsy is generally avoided because it may result in tumor seeding and potential upstaging of the disease. Testicular cancer typically metastasizes through predictable lymphatic pathways. An important exception is choriocarcinoma, which has a marked propensity for early hematogenous dissemination.^{7,8} Contrast-enhanced computed tomography (CECT) of the abdomen and pelvis is the preferred imaging modality for assessing retroperitoneal lymph node metastases in patients with testicular cancer. In this case report, patient didn't have retroperitoneal lymph nodal metastasis. Radical inguinal orchidectomy remains the standard

primary surgical treatment for patients with testicular cancer who have a normal contralateral testis.⁹ The procedure provides a definitive histopathological diagnosis and enables accurate pathological T staging of the tumour. In patients with stage I disease, it is curative in approximately 75% of cases. Patients with stage I NSGCT who do not undergo retroperitoneal lymph node dissection (RPLND) require a more intensive surveillance protocol. In contrast, following RPLND, surveillance is generally less demanding, often limited to routine serum tumor marker assessment and chest radiography, with retroperitoneal relapse rates reduced to less than 1%.¹⁰ The majority (90%) of NSGCT patients who develop a recurrence will experience it within the first 2 years after definitive therapy. As the risk of recurrence is highest during the first two years, a more intensive surveillance protocol is recommended during this period. For most patients, follow-up includes a physical examination, chest radiography, contrast-enhanced computed tomography (CT) of the abdomen and pelvis, and serum tumour marker assessment every two months. Disease recurrence is often detected by multiple surveillance modalities, with abdominal and pelvic CT being the most sensitive and commonly positive investigation. Serum tumor markers alone are insufficient for the detection of recurrent disease. Therefore, periodic CT imaging remains an essential component of surveillance to identify radiologically occult recurrences.¹¹

Low-risk disease is associated with a favourable prognosis. It is defined by a primary testicular or retroperitoneal tumour, absence of non-pulmonary visceral metastases, and low serum tumour marker levels, as follows: AFP<1000 ng/ml, CG<5,000 IU/l (1,000 ng/ml), and LDH<1.5×upper limit of normal).¹⁰

Intermediate-risk malignancies account for approximately 28% of non-seminomatous germ cell tumours. These patients have an estimated 5-year progression-free survival of 75% and a 5-year overall survival of 80%. Intermediate-risk disease is defined by a primary testicular or retroperitoneal tumour in the absence of non-pulmonary visceral metastases, along with intermediate-range serum tumour marker levels, as follows: AFP $\geq$ 1,000 and $\leq$ 10,000 ng/mL or hCG $\geq$ 5,000 IU/l and $\leq$ 50,000 IU/l or LDH $\geq$ 1.5×N and $\leq$ 10×normal).

High-risk tumours carry a poor prognosis. They are defined by the presence of a primary mediastinal tumour, non-pulmonary visceral metastases, or markedly elevated serum tumour markers, as outlined below: AFP>10,000 ng/ml or hCG>50,000 IU/l (10,000 ng/ml) or LDH>10×upper limit of normal).

Close surveillance after orchiectomy as a standard treatment option in low-risk stage 1 NSGCT patients.¹² According to the EGCCCG and European Association of Urology (EAU) guidelines, a risk-adapted approach based on the presence of vascular invasion (VI), with administration of two cycles of PEB chemotherapy, is the

recommended standard of care. However, because vascular invasion is a relatively insensitive marker, this strategy results in an overtreatment rate of approximately 52%.¹³ In this case, the patient had lymphovascular invasion, but our patient lost to follow up even after counselling about the requirement of further adjuvant chemotherapy and risk of recurrence.

CONCLUSION

Intra-abdominal testicular tumors may attain remarkably large sizes before they are clinically detected. Therefore, a high index of suspicion is essential when evaluating a young male presenting with an intra-abdominal mass associated with an empty scrotum, as delayed diagnosis frequently results in advanced disease at presentation. Radiological evaluation, tumor marker assessment, and sound clinical judgment play crucial roles in guiding appropriate management. Documentation of such cases is important, as it adds to the limited existing literature and increases awareness of this uncommon yet clinically significant presentation.

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