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Bilateral intra-abdominal testicular tumor: Case report

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ABSTRACT

INTRODUCTION: Bilateral intra-abdominal testis is a very rare clinical entity. These testes may develop cancer in an adult patient with empty scrotum.
CASE PRESENTATION: A case of a huge intra-abdominal solid mass in a 32-year-old gentleman is presented. Physical examination revealed an empty scrotum. Laboratory investigations, imaging studies, laparotomy and histopathological examination showed that the solid mass was a mixed germ cell tumor of the left testis. The contra-lateral testis also had a tumor. Resection of the solid mass and contra-lateral orchiectomy was performed. Adjuvant chemotherapy was given. Six months after surgery, his follow-up parameters were all within normal limits.
DISCUSSION: Since bilateral intra-abdominal testis tumor is a very rare clinical entity, there are no patient management guidelines available. Management strategies differ significantly among groups and they are based mainly on the experience reflected in the context of anecdotal case reports.
CONCLUSION: Surgical exploration and adjuvant chemotherapy seems as a reasonable treatment option in the setting of bilateral intra-abdominal testis tumor in an adult patient.

1. Introduction

Testicular tumors constitute 1% of solid tumors encountered in males [1,2]. Its incidence is in the range of 3–10%. Germ cell testicular tumors constitute the most common type; 90–95% of testicular tumors have germ cell origin. These tumors can be seminomatous or non-seminomatous. Both genetical and epidemiological factors contribute to their pathogenesis [1]. History of cryptorchidism or presence of an undescended testis is considered as the most important epidemiological risk factor [3].

It is widely known that classical manifestation of testis tumor is painless testicular mass [1]. However – since undescended testis is associated with a long-term risk of developing testicular cancer-recognition of empty hemiscrotum or scrotum in an adult patient should also raise the suspicion of testicular tumor [4]. In this scenario testicular tumor may present with retroperitoneal mass.

In this work which is reported in line with SCARE criteria, we report an adult patient with empty scrotum who presented with the complaint of abdominal pain; imaging studies showed a retroperitoneal mass histopathological examination of which revealed a mixed germ cell testicular tumor [5].

2. Case presentation

A 32-year-old male patient presented to general surgery clinic with the complaint of abdominal pain. Past history of the patient was unremarkable except for an appendectomy which was performed 12 years ago. A mass was palpated in the right upper abdominal quadrant. Complete blood count and blood biochemistry results were within normal limits. Abdominal ultrasonography (USG) showed a 15-cm diameter solid mass adjacent to right kidney which had an ectopic pelvic location. Abdominopelvic magnetic resonance imaging (MRI) revealed a 14 × 9 cm solid mass with a cystic component in its centre (Figs. 1 and 2). Patient was referred to our clinic with the MRI images.

The mass was anterior to the pelvic right kidney but it did not originate from the kidney. We proceeded with scrotal examination; it revealed an empty scrotum with which he was not bothered. There were no palpable lumps in the groins. There was no supraclavicular adenopathy and gynecomastia. Genital examination showed a circumcized penis without hypospadias. Serum

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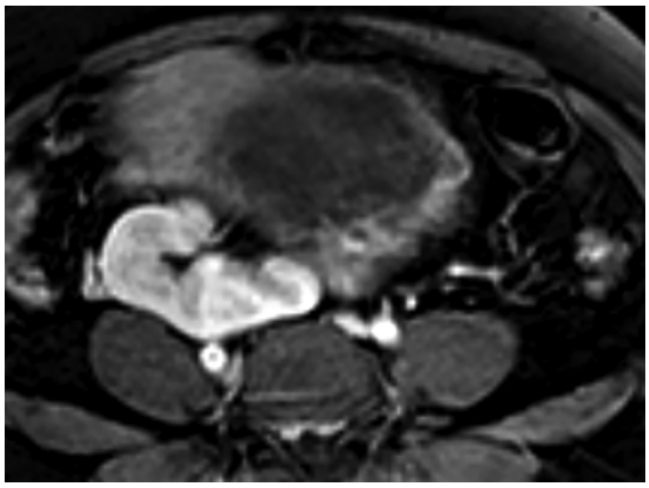

Fig. 1. Transverse magnetic resonance image of the solid mass anterior to the right ectopic pelvic kidney with a cystic component at the centre.

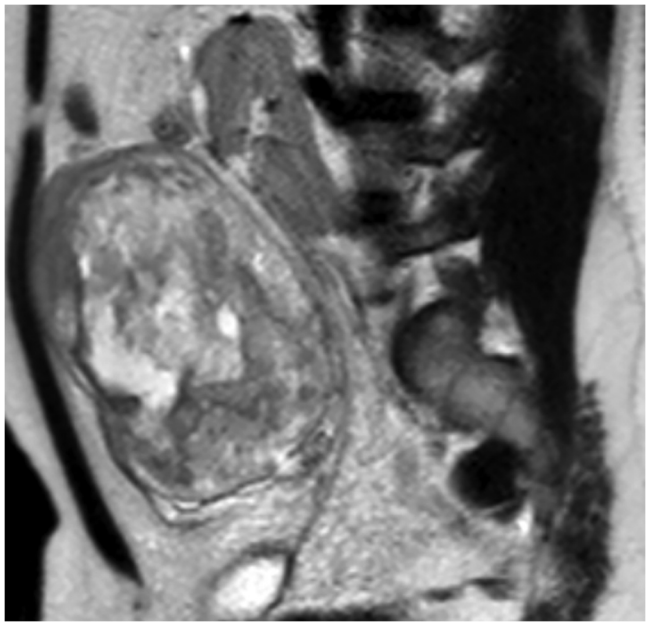

Fig. 2. Sagittal magnetic resonance image of the solid mass.

testicular tumor markers were analyzed; alpha fetoprotein (AFP) level was measured as 50,307 (normal: 0.6–6.0 ng/ ml), beta human chorionic gonadotropin (β-HCG) was measured as 3.8 (normal: 0–5 mU/ml) and lactate dehydrogenase (LDH) level was 412 (normal: 25–248 U/L). Serum total testosterone level was measured as 2.37 (normal: 3–10 ng/ dl) however the patient had a normal virile appearance. He was single and he did not want to have any children in the future. Therefore, he refused sperm banking.

Patient was admitted; abdominal exploration and excision of the retroperitoneal mass was planned. Surgical exploration revealed that the right-sided mass was posterior to the urinary bladder and invaded the descending colon. En bloc resection of the mass necessitated excision of a 10-cm segment of the colon and subsequent end-to-end anastomosis. Exploration of the left side led to identification of left testis at the level of iliac bifurcation. It was $4 \times 2 \times 2$ cm in length, width and height respectively with an extremely hard consistency and irregular borders. Left orchiectomy was performed. There were no intra-operative or post-operative complications.

Histopathological examination of the retroperitoneal mass was reported as mixed (90% non-seminomatous and 10% seminomatous) germ cell tumor with negative surgical margins (Fig. 3). Pathological assessment of the left testis also revealed a mixed tumor (atypical seminoma and mature teratoma) which constituted 5% of the testicular volume (Fig. 3). The non-tumoral tissue had diffuse intratubular germ cell neoplasia. Evaluation of colonic tissue confirmed colon invasion by the yolk sac tumor component of the retroperitoneal mass (Fig. 3). Immunohistochemical staining was positive for AFP, glipikon and panCK but negative for CD30 in non-seminomatous components while seminomatous tumor components were PLAP, OCT and CD117 positive.

Serum tumor marker analysis was repeated 7 days after surgery; AFP, β-HCG and LDH levels were measured as 3000 ng/ml, 0.5 mU/ml and 261 U/L respectively. Six weeks postoperatively, patient was referred to oncology clinic where 4 cycles of chemotherapy (BEP: Bleomycin 0.5 U/ kg IV, Etoposide 100 mg/m^2 IV, Cisplatin 20 mg/m^2) was administered at 21-day intervals. Tumor marker assessments performed 6 months after surgery revealed that all marker levels were within normal limits. Thoraco-abdominopelvic computerized tomography scan did not reveal any findings consistent with local recurrence or metastasis. Currently the patient is followed up every 2 months and is on testosterone replacement therapy with intramuscular testosterone propionate injections (250 mg, every 3 weeks).

3. Discussion

Testis cancer is most frequently diagnosed in men aged 15–44 [1,3]. It is usually found at an early clinical stage and cured by radical orchiectomy procedure. However, treatment of the patients diagnosed at a later stage can be more challenging [6,7]. In our patient who presented at a later stage, detection of a retroperitoneal mass and an empty scrotum led to the suspicion and subsequent diagnosis of testicular cancer. He had to undergo exploratory laparotomy including resection of the retroperitoneal mass and orchiectomy in addition to chemotherapy for complete treatment.

Although very rare, our case is not the first case of testis cancer presenting with intra-abdominal mass and an empty scrotum. Shrestha et al diagnosed testis cancer in a 30-year-old male patient who had a 13×12 cm retroperitoneal mass and an empty scrotum [7]. In line with our approach, they performed laparotomy first. The mass originated from right intra-abdominal testis and invaded the small bowel. Contra-lateral testis was also intra-abdominal; however it had a soft consistency. Therefore, these authors resected the mass and performed concurrent left orchiopexy. Pathological assessment revealed seminoma. This patient underwent adjuvant BEP chemotherapy which led to complete treatment.

Another similar report was published by Liss et al. [4]. Their patient was also in the 4th decade of life however–as a significant difference- this patient had persistent mullerian duct syndrome (PMDS) which is a rare disorder of sexual differentiation. Patients with PMDS are phenotypically male but they have undescended testes and mullerian structures [8]. Liss et al. performed biopsy of the abdominal mass as the initial step of their management [4]. Pathological examination revealed seminoma. They proceeded with BEP chemotherapy which led to significant reduction in tumor size. Since positron emission tomography did not demonstrate any viable tumor tissue they did not perform retroperitoneal exploration. They performed a laparoscopic bilateral orchiectomy procedure during which they diagnosed PMDS. Subsequently testosterone replacement therapy was started.

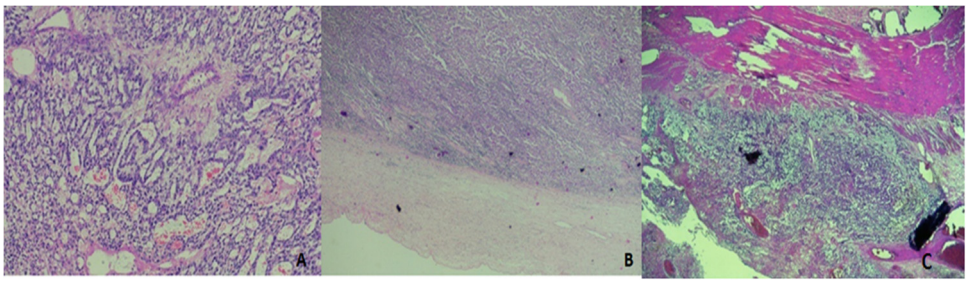

Fig. 3. (a) Non-seminomatous (yolk sac) component of the intra-abdominal right testicular mixed germ cell tumor (H&E staining, ×100 magnification) (b) Atypical seminoma cells between the septae invaded by lymphocytes in the left intra-abdominal testis (H&E staining, ×40 magnification) (c) Colon segment invaded through the serosal layer by the yolk sac component of the right testicular mixed germ cell tumor (H&E staining, ×20 magnification).

Al Harbi et al reported a case in which a 45-year-old patient who presented with an abdominal mass and empty scrotum was diagnosed with PMDS during abdominal exploration [9]. Interestingly, they started neoadjuvant BEP chemotherapy without a tissue diagnosis. The 20 cm diameter mass reduced remarkably in size and they performed a post-chemotherapy abdominal exploration which led to the tissue diagnosis of mixed germ cell tumor.

Velez et al presented a 32-year-old patient who had a 17 cm diameter abdominal mass and an empty scrotum [10]. In line with our case, this patient did not have PMDS. However, in contrast to our management strategy, these authors initially biopsied the abdominal mass and diagnosed seminoma. They proceeded with neoadjuvant BEP chemotherapy and subsequently resection of the residual mass with retroperitoneal lymph node dissection. This therapeutic approach led to complete treatment of their patient.

A case of a 23-year-old man who complained about abdominal pain and was later diagnosed with bilateral intra-abdominal seminoma was presented by Agrawal et al. [11]. After recognition of empty scrotum, these authors biopsied one of the intra-abdominal masses however histopathological examination findings were inconclusive. Subsequently laparotomy during which two intra-abdominal masses were resected was performed. This patient also had bowel invasion and the pathological examination revealed bilateral pure seminoma. Four cycles of BEP chemotherapy was given and patient was cured without any local recurrence or metastasis during a 5-year follow-up period.

Since the cases of bilateral intra-abdominal testis tumors associated with empty scrotum are extremely rare, there are no guidelines referring to the most appropriate management of these patients [7,10,11]. Probably due to this fact, there are differences between the therapeutic approaches of different groups [7,10,11]. Shrestha et al performed laparotomy as their initial management step and proceeded with BEP chemotherapy as per pathology report [7]. Our approach was similar. On the other hand, Velez et al initially biopsied the intra-abdominal mass and subsequently gave BEP chemotherapy [10]. Laparotomy for excision of the residual mass was their final management step. Despite the fact that management strategies differed between groups, all groups –including ours- reported successful results [7,10,11].

As per our opinion there are some lessons to take from the previous case reports as well as our report in terms of the management of adult patients who present with an intra-abdominal mass and empty scrotum. First, this clinical picture should raise the suspicion for an intra-abdominal testis tumor in the setting of long-term cryptorchidism. Second, surgical exploration should not focus solely on the resection of the intra-abdominal tumor; a search for the contra-lateral testis should also be performed. Contra-lateral testis -if found- should be examined and the surgeon should always consider the possibility of a bilateral intra-abdominal testis tumor. Third, patient should be consented and future fertility plans should be addressed pre-operatively since some intra-operative and post-operative decisions such as contra-lateral total orchiectomy and testosterone replacement therapy can be given more comfortably in that setting.

4. Conclusion

In conclusion, intra-abdominal testis tumor with or without associated PMDS must be considered as a potential diagnosis in an adult patient who presents with empty scrotum. Patients in the third or fourth decades of life seem to convey the highest risk. Since there are no guidelines, these patients should be evaluated individually with the guidance of anecdotal case reports. In addition, factors such as presence or absence of associated PMDS, pre-operative serum testosterone level, future fertility plans and status of the contra-lateral testis must all be considered during making these patients'individualized management plans.

Conflicts of interest

Authors of this manuscript do not have any conflicts of interest to declare.

Funding

Authors did not use any funding in this study.

Ethical approval

This study has been exempt from ethical approval in our institution.

Consent

Patient gave both verbal and written consent for the case report.

Author contribution

FS is the first author of this manuscript. SC revised it. SGC, BB, GG and AI performed the surgery. UH performed the histopathological examination. AK followed the patient postoperatively. GII gave the adjuvant chemotherapy.

Registration of research studies

This study has not been registered to any research registry.

Guarantor

Sertac Cimen.

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