Case Report

Bilateral synchronous seminoma with bilateral cryptorchidism of the testis

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ABSTRACT

Synchronous bilateral germ cell tumo (BGCT) of the testis is rare and its association with bilateral cryptorchidism is even rarer. We report one case of BGCT of testis with bilateral cryptorchidism who presented as blunt injury abdomen in emergencyand was not diagnosed preoperatively. Postoperatively after an appropriate diagnosis, he was managed with chemotherapy. In this report, we have reviewed the larger series of BGCT for the presentation and management of synchronous BGCT to derive some conclusions.

Key words: Bilateral germ cell tumor, synchronous, testicular tumor

CASE REPORT

A 23-year-old male presented with history of feeling of heaviness in the lower abdomen, indigestion, and constipation since 3 months, fever and low backache since 1 month. The patient also gave history of abdominal trauma a few months back. A clinical examination revealed distended, tender abdomen with ill-defined lump in the lower abdomen and his scrotal sacs were empty. An ultrasound of abdomen revealed a mass in the lower abdomen. A contrast enhanced CT scan of the abdomen showed bilateral abdominal mass with adherent small bowel loops, and there was no evidence of any lymphadenopathy [Figure 1]. FNAC from the mass was suggestive of acute infection. With a probable diagnosis of rupture of abdominal mass with abcess formation, the patient underwent exploratory laparotomy. Peroperative findings revealed two big masses in the lower abdomen encroaching midline. Right sided mass was bigger than the left and the small intestine was adherent to the mass. There was no lymphadenopathy, or any peritoneal seedling, and liver surface was normal. Both the lumps were excised and cut section of the mass was greyish white, hard with few foci of hemorrhage and necrosis [Figure 2]. Histopathological examination of the resected mass was suggestive of pure seminoma. An array of tumor markers like b-HCG, alpha feto protein, and serum LDH were normal. The patient was identified as Stage IA bilateral synchronous seminoma with bilateral cryptorchidism. Even though he had Stage IA disease, he was then given four cycles of BEP (bleomycin, etoposide, and cisplatinum) chemotherapy due to the presence of intestinal adhesions. Patient is presently on 5 years of follow-up,

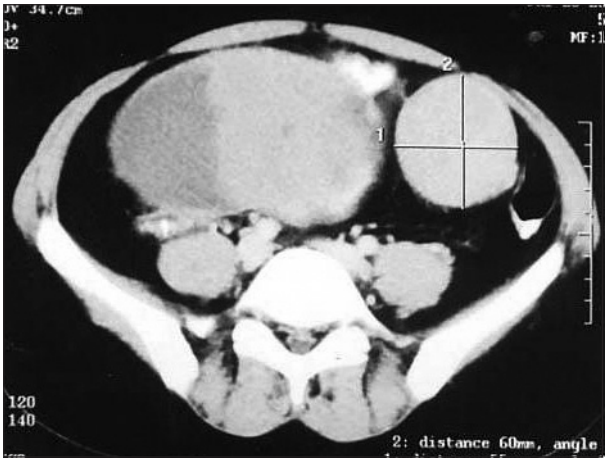

Figure 1: CT scan image of the patient with bilateral intra-abdominal testicular tumor

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Table 1: Patient characteristics, histopathology, treatment, and follow-up status of synchronous bilateral germ cell tumo reported by large series

Author	No. of patients	Med age	Histopathology (number of patients)	Stage (no of patients)	Postsurgery management (no of patients)	FU and status
Coogan[1]	5	30	Seminoma/seminoma (3) Teratoma/teratoma (1) Seminoma/embryonal (1)	A(2) B(1) C(1) B(1)	RT(2) BEP BEP BEP, RPLND	60 mo NED(5)
Holzbeierlein[3]	10	26	Seminoma/seminoma (3) Seminoma/NSCGT (7)	I(2) II(1) I(4) II(2) III(1)	RT(2) CT RPLND(3), surv(1) CT(1),RPLND(1) CT(1)	29.5 mo NED
Theodore[2]	14	26	Seminoma/seminoma (9) NSCGT/NSCGT (1) Discordant (4)	I (1) II(4) III(9)	RT(7) CT(7)	58 mo NED
Geczi[4]	19	NA	Seminoma/seminoma (13) Seminoma/NSCGT (3) NSGCT/NSGCT (3)	I (13) II (3) III(3)	CT(9) RT(4) RPLND+CT(2) CT+RT (1) Surv (1)	93 mo NED(16)
Hentrich[5]	14	30	Seminoma/seminoma (4) Seminoma/NSCGT (7) NSGCT/NSGCT (2) Unclassifiable (1)	I(2) II(2) I(3) II(3),III(1) III(2) III(1)	CT(1),RT(1) RT(2) RPLND(1),CT(2) CT(3) CT(2) CT(1)	34 mo NED (9) Dead (5)

RT, radiotherapy; CT, chemotherapy; RPLND, retroperitoneal lymphnode dissection; Surv, surveillance; BEP, bleomycin, etoposide, and cis-platin; NED, no evidence of disease, FU: follow-up status.

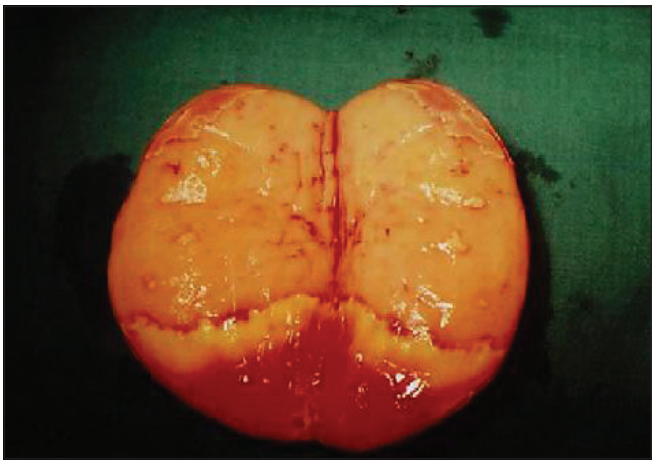

Figure 2: Opened-up tumor of right intra-abdominal testis

asymptomatic, follow-up CECT abdomen and pelvis are normal, tumor markers are normal, and is without evidence of any disease. His serum testosterone levels are nil and he has been put on testosterone supplementation which he is tolerating well.

DISCUSSION AND A REVIEW OF LITERATURE

The incidence of bilateral testicular germ cell tumors ranges from 1% to 5%. Bilateral germ cell tumors (BGCT) occur metachronously in 80–85% of cases and synchronously in 15–20% of cases.[1] Among patients presenting with seminoma, 1.8% develop BGCT as compared to 0.6% with nonseminomatous germ cell tumor (NSGCT). Synchronous testicular tumors commonly have concordant pathology in both testes. Several potential risk factors for developing a second testicular tumor are atrophy of the second testis, young age, infertility, and a family history of testicular cancer, atypical naevi, Down's syndrome, and testicular maldescent. Cryptorchidism is a known risk factor for the development of a testicular germ cell tumor. Bilateral synchronous seminoma with bilateral cryptorchidism is rare.[2] Incidence of intratubular germ cell neoplasia (IGCN) is high in patients with one or more risk factors (35–85%) and its detection allows curative tumor eradication with minimal morbidity or mortality along with the possibility of preserving testicular function.

Literature on the management of synchronous bilateral testicular tumors is insufficient probably because of the rarity of the condition. In a 50-year single institutional experience from Memorial Sloan Kettering Cancer Centre, reporting 58 bilateral testicular tumors,10 patients presented with synchronous tumors (the largest number reported for bilateral synchronous testicular tumors).[3] In this series, the predominant histology was a seminomatous with nonseminomatous tumor (7), which was followed by a seminoma with seminoma (3) and no patient had bilateral NSCGT. Most presented with Stage I and II disease.

Treatment was based on the histology and stage of the disease. In patients with a combination of seminoma and nonseminoma, Stage I patients were offered retroperitoneal lymphnode dissection, and rarely surveillance. Higher stage patients were managed with chemotherapy. Stage I bilateral seminoma were treated with postoperative radiotherapy, and more than Stage I patients with chemotherapy. Patients treated in the post-*cis*-platinum era had better overall survival than the patients treated in the pre-*cis*-platinum era.

There is a lot of heterogeneity in the reported series regarding the management of synchronous BGCT [Table 1] and only broad generalizations can be made from these. Post-orchiectomy management of these patients has been dictated by the higher stage of the tumor in either of the testis and the pathology with the higher malignant potential (NSGCT as compared to pure seminoma). Among the available options for Stage I patients with seminoma of surveillance, prophylactic para-aortic lymphnode irradiation, or one to two cycles of adjuvant chemotherapy, bilateral seminomas have a higher tumor burden and, therefore, these patients should not be kept on surveillance; rather they should be treated with prophylactic para-aortic lymphnode irradiation or one to two cycles of adjuvant chemotherapy. Patients in Stage II or higher should be treated with chemotherapy. For selected patients with tumors smaller than 25 mm confined to the testis and with normal preoperative testosterone, testis sparing surgery (TSS) to avoid lifelong androgen replacement and preservation of fertility should be offered to patients who are aware and accept the risk of a subsequent local relapse and who realize the importance of compliance during follow-up. Patients not suitable for TSS should be offered testosterone replacement therapy.

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