CASE REPORT

Malignant transformation in abdominal cryptorchid testis 40 years after treatment of seminoma confined to other testis

Mustafa Sofikerim, MD,[1] Deniz Demirci, MD,[1] Figen Öztürk, MD,[2] Atila Tatlisen, MD[1]

[1]Department of Urology, Erciyes University School of Medicine, Kayseri,Turkey
[2]Department of Pathology, Erciyes University School of Medicine, Kayseri,Turkey

SOFIKERIM M, DEMIRCI D, OZTURK F, TATLISEN A. Malignant transformation in abdominal cryptorchid testis 40 years after treatment of seminoma confined to other testis. The Canadian Journal of Urology. 2007;14(2):3510-3513.

This report describes an exceptionally rare case of a 64-year-old man with seminoma in abdominal cryptorchidism, leading to intestinal obstruction 40 years after curative treatment for seminoma of the other (descended) testis.

Key Words: cryptorchidism, seminoma, testicular malignancy, intestinal obstruction

Introduction

Cryptorchidism is a known cause of testicular malignancy. The risk of developing testicular malignancy in a cryptorchid testis is 30 to 50 times higher than in a normal testis.[1] The position of the cryptorchid testis is related to the risk of malignant transformation, with the abdominal location having the highest risk of malignancy. Neoplastic transformation of a cryptorchid testis usually occurs at puberty, and seminoma is the most common type of malignancy.[2]

Accepted for publication January 2007

Address correspondence to Dr. Mustafa Sofikerim, Department of Urology, Erciyes University School of Medicine, 38039 Kayseri, Turkey

Case report

A 64-year-old man was admitted to the emergency unit of our university teaching hospital with abdominal pain, constipation, fever, and vomiting. These symptoms had started a few days earlier. A physical examination revealed a mass in the left side of his lower abdomen along with abdominal tenderness when palpated. The patient had acute abdominal pain. A digital rectal examination did not find any abnormal pathology. Palpation revealed that the right and left scrotum were empty. Bilateral inguinal incision scars were observed.

The patient's medical records revealed that 40 years earlier, he had had a high inguinal orchiectomy for a right testicular mass that was diagnosed as seminoma (based on pathology); he also had a left inguinal exploration for the absent left testis, but it was not found.

The patient underwent retroperitoneal irradiation for adjuvant treatment of right testicular seminoma. He was then followed for 14 years, during which time no relapse occurred. On the advice of his physician, he had not sought any further medical treatment.

After his medical history was obtained, blood samples were drawn to determine the levels of tumor markers. The laboratory test results showed normal levels of human chorionic gonadotropin (β-HCG) and α-fetoprotein (AFP) and an elevated lactate dehydrogenase (LDH) of 731 U/mL (normal range, 230 U/mL - 460 U/mL). A spiral computerized tomography (CT) scan of the patient's thorax revealed no evidence of disease, but a spiral CT scan of his whole abdomen revealed a 13 cm x 6 cm solid mass originating from the lower pole of his left kidney that spanned to the pubis vertically and across the midline horizontally; distension of the large bowel was evident above the mass, Figure 1.

An urgent explorative laparotomy was performed due to this intestinal obstruction. This revealed a large tumor with cystic and solid components; the tumor extended from the left side of the retropubic space across the midline, and it pushed the sigmoid colon to the right side. There was also marked adherence of the tumor to abdominal structures, likely due to previous pelvic radiation therapy. The tumor mass adhered tightly to the psoas muscle and surrounding abdominal organs. A sharp and blunt dissection released the left ureter from the tumor mass. A 6-cm segment of sigmoid colon was resected with the tumor, and then end-to-end anastomosis was done.

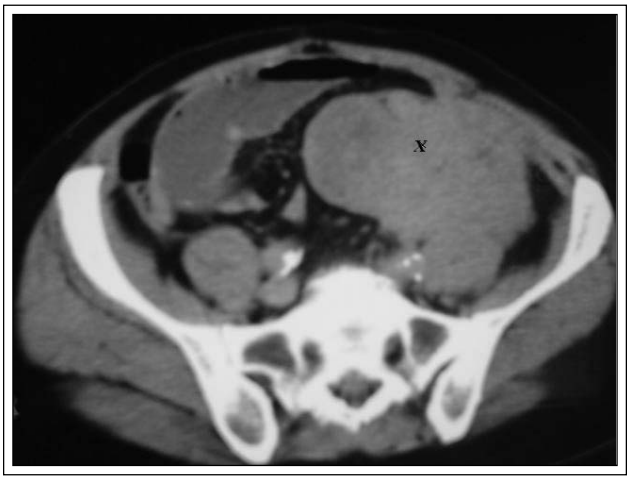

Figure 1. A contrast-enhanced computed tomography (CT) scan showing an irregular, solid pelvic mass (asterisk).

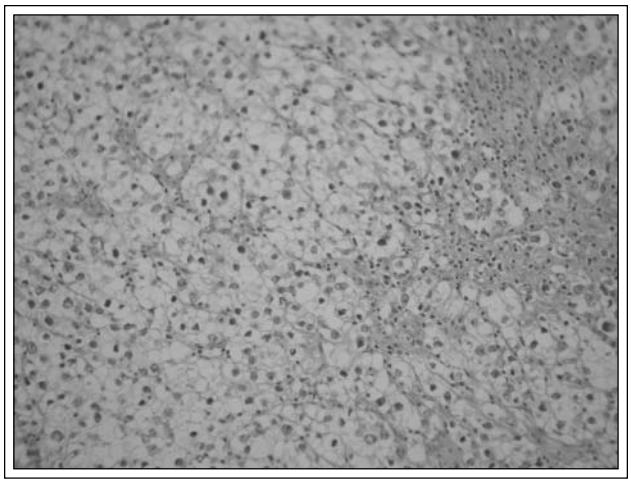

Figure 2. Tumor cells showing clear cytoplasm, distinct cell borders, and prominent nucleoli (HE stain, x20).

The tumor measured 13 cm x 8 cm x 7 cm, and markings from large blood vessels covered its surface. The whole lesion was sampled for histopathologic evaluation. This revealed neoplastic cellular proliferation composed of cells that were round to polygonal in shape with clear cytoplasm, distinct cell borders, and prominent nucleoli, Figure 2. Gross examination of the sigmoid colon resection material showed a 4 cm x 3 cm x 2 cm nodule in the serosal location. A histological microscopic examination of this nodule revealed seminomatose tumor cells, Figure 3. In one of the sections of the bulky tumor, tumoral infiltration of the ductus deferens was also noted, Figure 4.

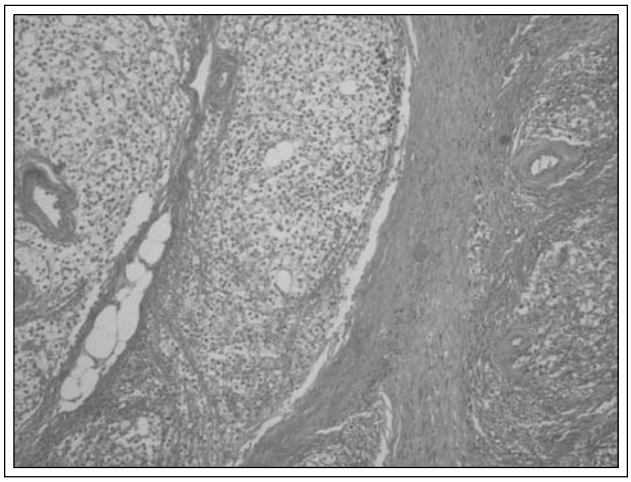

Figure 3. Tumor cells showing seminomatose histological findings in the serosal location of the sigmoid colon (HE stain, x20).

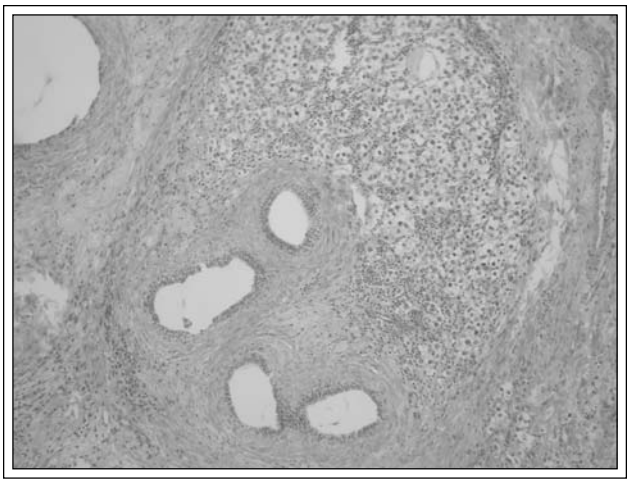

Figure 4. Tumor infiltration of the ductus deferens (HE stain, x20).

Case follow-up

In the present case, after the patient recovered from the operation, blood samples were drawn to determine levels of tumor markers. The laboratory test results were normal for β-HCG, AFP, and LDH, so radiologic evaluations were done. A spiral CT scan of the whole abdomen revealed a 3 cm x 1.5 cm residual solid mass in the para-aortic region. Multi-agent chemotherapy is highly recommended for abdominal cryptorchid testis complicated by seminoma.[3] Therefore, the patient received two courses of bleomycin, etoposide, and cisplatin (BEP) for consolidation chemotherapy. After the last course of chemotherapy, radiologic evaluations were done. A spiral CT scan of the whole abdomen revealed no evidence of disease. The patient was healthy at his last follow-up check-up 9 months after chemotherapy ended.

Discussion

This is a case of malignant transformation of an abdominal cryptorchid testis. The tumor infiltration of ductus deferens and the absence of a left testis suggested the presence of an abdominal cryptorchid testis. The bulky tumor was found to be entirely seminoma, which invaded the para-aortic region and a long segment of sigmoid colon. There was no evidence of another malignancy on final pathologic diagnosis. Because of the long time interval (40 years) from the initial curative treatment of seminoma confined to the right testis, as well as the history of infradiaphragmatic radiation and the absence of the left testis, the differential diagnosis of the tumor included primary gastrointestinal or retroperitoneal malignancy; late-onset secondary malignancies due to the past radiation history; malignancy due to intra-abdominal cryptorchid testis; or, less likely, late relapse of primary right-testicular seminoma.

Travis et al presented convincing data from a long-term follow-up of stage 1 seminoma patients treated with adjuvant radiation protocols that revealed that patients who survived beyond 5 years had an increased risk of developing secondary malignancies, and the risk increased with time from the initial diagnosis of cancer.[4] In the same report, the authors stated that significantly increased risk of colon cancer was limited to 20-year survivors, and that cancer of the small intestine occurred throughout a median follow-up of 11.5 years (range, 3-42.6 years).

On the other hand, Power et al reported that late-onset relapse of stage 1 seminoma usually occurs between the second and third year after an initial curative treatment.[5] Late relapse after as long as 40 years appears to be very rare; there is one report in the literature of a relapse that occurred 31 years after curative treatment.[6]

There are a few reports in the literature of malignant testicular tumors in a cryptorchid testis in the abdomen where presenting findings were abdominal pain, abdominal or inguinal masses, and adjacent visceral infiltration causing ascites.[7-9] Compared with an inguinal testis, an abdominal cryptorchid testis has a higher risk of developing malignancy, which happens in a gradual process over a long period of time.[1] The most common types of malignancies seen in a cryptorchid testis, in order of prevalence, are seminoma, embryonal carcinoma, and gonadoblastoma.[2] In the current case, the patient's history was unusual because of the very long (40-year) interval from his curative treatment for right testicular seminoma until the malignant transformation in the cryptorchid testis. Most cases of metachronal bilateral testicular tumors that have been 'previously reported occurred within 5 years of initial surgery of bilateral testicular tumors.[10]

Conclusion

In conclusion, when an abdominal cryptorchid testis is left untreated, it carries a high risk of malignant transformation and it can affect the surrounding viscera and present as acute abdominal pain. This case report reinforces the need for early diagnosis of a cryptorchid testis and proper medical management, which can result in a good long-term cure. □

References

1. Batata MA, Whitmore JR WF, Chu FCH et al. Cryptprchidism and testicular cancer. *J Urol* 1980;124:382-387.
2. Scully RE. Gonadoblastoma-a review of 74 cases. *Cancer* 1970;25:1340-1347.
3. Biswal BM, Madhavan M, Anas SR. Abdominal lump in an infertile man. *Postgrad Med J* 2000;76:719-729.
4. Travis LB, Curtis RE, Storm H et al. Risk of second malignant neoplasms among long-term survivors of testicular cancer. *J Natl Cancer Inst* 1997;89:1429-1439.
5. Power RE, Kennedy J, Crown J, Fraser I, Thornhill JA. Pelvic recurrence in stage I seminoma: a new phenomenon that questions modern protocols for radiotherapy and follow-up. *Int J Urol* 2005;12:378-382.
6. Strickland P. Seminoma testis with metastases after 31 years. *Br Med J* 1965;5431:356.
7. Raina V, Shukla NK, Gupta NP, Deo S, Rath GK. Germ cell tumors in uncorrected cryptorchid testis at Institute Rotary Cancer Hospital, New Delhi. *Br J Cancer* 1995;71:380-382.
8. Cheng C, Cheng PS. Cryptorchidism with a large intra-abdominal mass-a challenge. *Br J Urol* 1993;72:946-948.
9. Walker R, Rogers HS, Christmas TJ. Seminoma in an undescended testis presenting with frank hematuria. *Br J Urol* 1997;79:290-291.
10. Fukuhara Y, Shiga Y, Ken S. Bilateral testicular tumors recurring 31 years after the initial treatment: a case report and the literature review. *Nippon Hinyokika Gakkai Zasshi* 2005;96:17-20.