

## A CASE OF METACHRONOUS BILATERAL TESTICULAR SEMINOMAS

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We report a case of metachronous seminoma in the surgically corrected right undescended testis of a 21-year-old man who had 7 years previously developed a seminoma in the left testis along with hypospadias.

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**Key words:** Bilateral testicular tumors, Cryptorchidism, Hypospadias

### INTRODUCTION

Bilateral lesions have been found in 1.6 to 5.8% of the patients with testicular cancer<sup>1-7)</sup> which constitutes a relatively high risk of subsequent contralateral tumor development<sup>1,8)</sup>. The undescended testes also present a high risk for malignant changes<sup>8-10)</sup> although the relationship between testicular cancer and urogenital anomalies including hypospadias remains unknown<sup>9)</sup>. To date about 150 patients with bilateral testicular cancer have been reported in Japan. We herein report a case of metachronous seminoma in a surgically corrected undescended testis with a history of contralateral seminoma and hypospadias.

### CASE REPORT

A 21-year-old male patient presented at our hospital in October, 1991 complaining of a right testicular swelling persisting for about 11 months. He had a past history of hypospadias and right undescended testis, which had been treated with chordectomy and orchiorchidoplasty at 6 years of age and with urethroplasty at 9 years of age. He had subsequently undergone left orchietomy through a high inguinal incision for a

seminoma at 14 years of age. Physical examination at the time of presentation revealed no abnormal findings except the right enlarged testis. Laboratory data were normal, including values for lactate dehydrogenase (LDH), the  $\beta$  subunit of human chorionic gonadotropin ( $\beta$ -HCG), and  $\alpha$ -fetoprotein. Systemic work-ups for staging revealed no evidence of distant metastasis and right orchietomy was performed through a high inguinal incision. Since he had received a previous 39.6 Gy dose radiation therapy to the retroperitoneal and ipsilateral pelvic regions for the first seminoma, no further adjuvant therapy was performed. The patient has received testosterone replacement therapy and continues to have no evidence of disease 14 months after the second orchietomy without additional treatment.

**Pathological findings.** The resected specimen weighed 46 g. A solid tumor, sized 31 × 24 × 22 mm with expansive growth, was localized within the testicular capsule (Fig. 1). Cut surfaces showed bulging, grayish white tissue. Histologically the tumor was composed of uniformly distributed cells and delicate connective tissue stromal elements with a few scattered lymphocytes. Tumor cells were large and

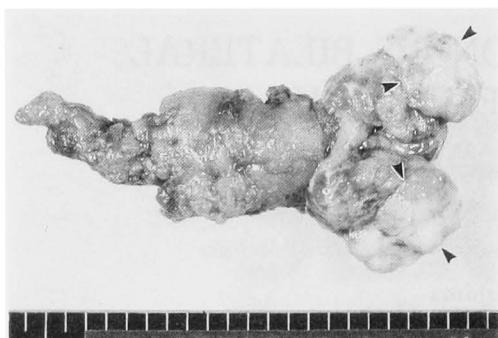


Fig. 1. Macroscopic appearance of the tumor in the right testis (arrowheads).

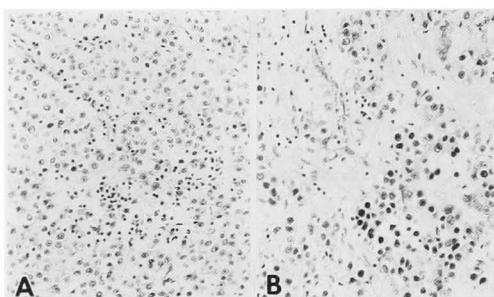


Fig. 2. Essentially similar histological appearance of the tumor developing in the right (A) and left (B) testes (HE staining,  $\times 85$ ).

polyhedral or round with large centrally located, hyperchromatic nuclei. Mitoses were infrequently observed (Fig. 2A). These findings corresponded to those of typical seminomas. Figure 2B illustrates the essentially similar histological appearance of the seminoma in the left testis which was resected 7 years previously.

#### DISCUSSION

Bilateral testicular germ cell tumors have been diagnosed in 1.6 to 5.8% of patients suffering from such lesions<sup>1-7</sup>, most of these cases being metachronous rather than synchronous, the ratio varying from 3:1 to 19:1<sup>2-5</sup>. In addition, several investigators have suggested a recent increase in incidence of bilateral tumors which could be related to the improved survival of patients due to advances in chemotherapy during the last decade<sup>4,7</sup>. The risk of a second tumor developing thus appears very much higher in patients with a first

testicular malignancy than in the normal healthy male population. Hamilton and Gilbert<sup>1</sup>, reviewing 7,000 cases of testicular tumor, reported that the likelihood of cancer in the second testis is from several hundred to several thousand times greater than expected by chance association. This finding indicates that all testicular tumor patients should be carefully followed for changes in the contralateral testis in the long term. Scheiber et al.<sup>4</sup> recommended periodic self-examination by the individuals concerned as well as periodic sonographic evaluation of the remaining testis.

An undescended testis is also a significant risk factor for testicular tumor development. The probability of a tumor arising in an undescended testis is in fact 10 to 48 times greater than in a normally descended testis<sup>8-10</sup>. Sokal et al.<sup>3</sup> reported that 7 to 15 (46%) patients with bilateral germ cell tumors for whom an adequate history had been obtained had suffered from maldescent. Four factors may contribute to the increased incidence of testicular tumors in cryptorchid cases: abnormal germ cells, interference with blood supply, endocrine disturbance, and gonadal dysgenesis<sup>9</sup>. Kratzik et al.<sup>5</sup> revealed the histocompatibility antigen (HLA)-B14 to be significantly increased in sequential bilateral testicular tumors and suggested that genetic factors might therefore be important in their development. The present patient had a hypospadias. The relationship between urogenital anomalies and testicular tumors clearly deserves further attention.

Regarding the interval between the first and second tumors, previous reports demonstrated 54 to 68% of all subsequent tumors to develop within 5 years of the first tumor diagnosis<sup>2-4</sup>. For example, Sokal et al.<sup>3</sup> reported 19 patients with second tumors after intervals of 4 months to 15 years with a median of 4 years. Scheiber et al.<sup>4</sup> reported that the interval between initial and secondary tumors varied from 2 months to 32 years with a median of 6 years in 19 patients with metachronous bilateral tumors. Therefore the in-

terval of 7 years in the present case was not atypical.

Regarding histological type of tumor Aristizabal et al.<sup>2)</sup> revealed seminomas to most commonly involve bilateral testes. Sokal et al.<sup>3)</sup> reported that of 9 of 13 (69 %) patients with seminoma as a first tumor had the same histological type as the second tumor, the same as in the present case. However, opinions are divided regarding the question of whether the histological type of the first tumor affects the risk of a secondary tumor. Sokal et al.<sup>3)</sup> considered the likelihood of a second tumor developing is slightly higher in seminoma patients. By contrast, Kratzik et al.<sup>5)</sup> argued that the histological type of the first tumor is not a useful parameter for determining the risk of the occurrence of a secondary testicular malignancy.

Management of a second tumor is determined generally in accordance with its histology and stage, but modifications may be required due to the prior therapy presented for the first tumor<sup>6)</sup>. Thus the present patient is under careful follow-up without any postoperative adjuvant treatments because the first tumor was diagnosed as a stage I seminoma necessitating adjuvant radiotherapy. Consideration of the risk benefit of additional therapy is clearly necessary in such cases.

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## 和文抄録

## 異時性両側性セミノーマの1例

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異時性両側性精巣セミノーマの1症例を経験したのでここに報告する。症例は21歳男子で尿道下裂および右停留精巣の既往歴を有していた。7年前に左精巣の

セミノーマ（stage I）を生じ、今回は固定術後の右精巣にセミノーマ（stage I）の発生を認めた。

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