Golestan University of Medical Sciences

Jorjani Biomedicine Journal

Online ISSN: 2645-3509

Metachronous bilateral classical seminoma in a patient with bilateral cryptorchidism: A case report and review of the literature

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Article Type: Case Report

Article History

Received: 20 February 2026
Received in revised form: 20 March 2026
Accepted: 5 April 2026
Available online:
DOI: 10.29252/jorjanibiomedj.X.X.X

Keywords

Testicular Neoplasms
Metachronous Neoplasms
Seminoma
Testicular Cancer
Germ Cell Tumor
Cryptorchidism
Case Reports

Abstract

Background: Metachronous testicular cancer, defined as the development of a tumor in the remaining testis, is a rare phenomenon. It is exceedingly rare when both tumors share identical histology (Seminoma) and the patient has a history of bilateral cryptorchidism. The clinical significance of this case lies in determining the optimal duration of surveillance for such patients.

Case Presentation: A 54-year-old man with a history of bilateral orchiopexy in childhood presented with a left testicular mass at age 50. He underwent left radical orchiectomy, and pathological examination revealed classical seminoma. The left testis was located in the inguinal canal. The patient was regularly followed with ultrasound for four years, during which no abnormalities were detected. At age 54, a new hypoechoic mass measuring 11×9 mm was identified in the right testis, which was also located in the inguinal canal. The patient underwent right radical orchiectomy, and pathological examination again confirmed classical seminoma with the same immunohistochemical pattern. Tumor markers remained within normal limits (LDH=347, BHCG<5, AFP=2.8).

Conclusion: Metachronous seminoma with identical histology may occur despite regular follow-up. Therefore, long-term, meticulous surveillance is essential for patients with a history of cryptorchidism and testicular cancer.

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Highlights

What is current knowledge?

- Seminoma is the most common type of testicular cancer.
- Bilateral testicular cancer is rare, usually metachronous, and most second tumors occur within 5 years.
- Cryptorchidism markedly increases cancer risk, and orchiopexy does not eliminate it; guidelines generally recommend 5-10 years of surveillance.

What is new here?

- A patient with bilateral cryptorchidism developed two histologically identical classical seminomas, four years apart, in the inguinal testes.
- The second tumor occurred despite regular ultrasound surveillance, challenging the adequacy of the conventional 5-year follow-up window.
- The case supports a possible lifelong field defect and suggests that long-term or lifelong surveillance may be warranted in high-risk patients.

Introduction

Testicular cancer is a rare malignancy, accounting for approximately 1-5.5% of all cancers in men. Its incidence increased between 1992 and 2009 (1,2). The most common and aggressive testicular cancers are germ cell tumors (GCTs), which are classified as seminomas (The most common) and non-seminomas (Including embryonal carcinoma, choriocarcinoma, and teratoma) (3).

Men with a history of testicular GCTs are at increased risk of developing a second primary tumor in the contralateral testis. Synchronous seminomas typically occur within 5 years of the initial diagnosis (4). Cryptorchidism (Undescended testis) is a well-established risk factor for testicular cancer. The risk of malignancy is 11 times higher in testes located outside the scrotum but outside the abdomen and 50 times higher in intra-abdominal testes (5).

Bilateral testicular tumors are rare, with 80% being metachronous (Non-synchronous). Overall, metachronous bilateral testicular tumors are uncommon (6). The incidence of bilateral involvement is approximately 0.8 per 1,000,000 men aged 15-40 years (7). Although bilateral testicular cancer accounts for only 1-5% of all testicular cancers, its clinical significance has increased because of improved long-term survival rates resulting from advanced diagnostic methods and effective treatments (8). Intratubular germ cell neoplasia (ITGCN) is a precursor to nearly all testicular GCTs and represents one of the strongest risk factors for developing contralateral testicular cancer (9). Radical orchiectomy remains the standard treatment for testicular cancer; however, in selected cases, such as a solitary testis, bilateral cancer, or tumors occupying less than 30% of the testicular volume, testis-sparing surgery may be considered (9). A review of the literature identified only a few published reports of metachronous contralateral testicular cancer developing after orchiectomy, particularly with identical histology and bilateral cryptorchidism. The interval between tumors in these reports varies widely, with most tumors occurring within 5 years (4,10). However, cases with longer intervals have been documented, suggesting that the risk persists beyond the typical surveillance period. The uniqueness of this case lies in the presentation of metachronous classical seminoma with identical histology four years after the first tumor in a patient with bilateral cryptorchidism, highlighting the need for extended surveillance protocols. This case also emphasizes the importance of meticulous physical examination and imaging in patients with undescended testes, even after orchiopexy.

Case presentation

Patient information

A 54-year-old man presented with a painless right testicular mass. He had a history of a left testicular mass four years earlier, for which he underwent left radical orchiectomy. The previous pathological diagnosis was classical seminoma. During the four-year interval, he remained asymptomatic and underwent regular follow-up. The patient had a history of bilateral orchiopexy performed in childhood (Table 1). He denied any history of trauma to the right testis. He reported no family history of tumors or malignancies, had no history of smoking or alcohol consumption, and worked as an office clerk.

Clinical findings on initial presentation (Age 50)

On physical examination, the left testis was not palpable in the scrotum; it was located in the inguinal canal and contained an approximately 1.5 cm mass that was fixed, adherent to the testis, and non-tender. The right testis was also not palpable in the scrotum.

Diagnostic assessment

Ultrasound examination revealed a 1.5 cm calcified mass in the left inguinal testis. Laboratory evaluation showed normal tumor markers (LDH, BHCG, and AFP within reference ranges). A metastatic workup, including abdominal and pelvic CT scans, revealed no pathological findings. The patient underwent left radical orchiectomy, and histopathological examination confirmed classical seminoma with negative margins. The testicular capsule, vas deferens, seminal vesicles, and surgical margins were free of tumor cells. The pathological stage was pT1N0M0, Stage I.

Follow-up period (Years 1-4)

The patient underwent periodic ultrasound examinations every 6-12 months. No mass was observed in the right testis during this period, and tumor markers remained within normal limits.

Second presentation (Age 54)

During a routine ultrasound examination, a hypoechoic mass measuring 11×9 mm was observed in the right inguinal testis (Figure 1). The mass was well-defined and showed internal vascularity on Doppler imaging. CT examination revealed no evidence of retroperitoneal lymphadenopathy.

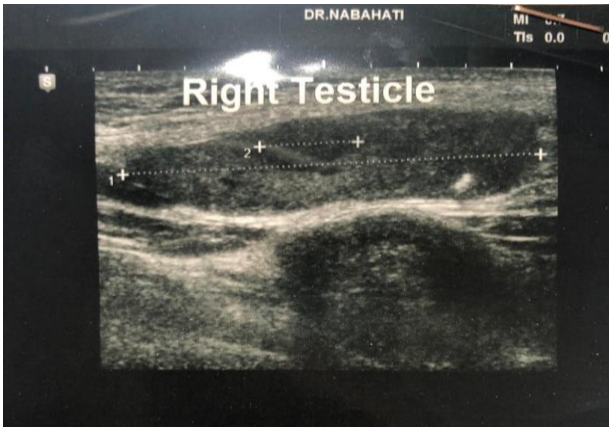

Figure 1. Ultrasound image of the right inguinal testis showing a hypoechoic mass (11×9 mm) with internal vascularity on Doppler imaging (Arrow). The mass was well-defined and located within a normal-sized testis in the inguinal canal.

Table 1. Timeline of clinical events

Age	Event
Childhood	Bilateral Orchiopexy
50 years	Left testicular mass detected; left radical orchiectomy; pathology: classical seminoma pT1N0M0
50-54 years	Regular surveillance with ultrasound and tumor markers; no abnormalities
54 years	Right testicular mass (11×9 mm) detected on ultrasound Right radical orchiectomy; pathology: classical seminoma pT1N0M0
54 + (18 months follow-up)	No recurrence; tumor markers normal; no complications

Laboratory findings

- LDH = 347 U/L (Normal: 140-280 U/L)
- BHCG < 5 mIU/mL (Normal: < 5 mIU/mL)
- AFP = 2.8 ng/mL (Normal: < 10 ng/mL)

Note: LDH was mildly elevated, possibly because of tissue necrosis or benign causes, and returned to normal postoperatively.

Therapeutic intervention

Right radical orchiectomy was performed. The decision to perform radical orchiectomy was based on the patient's history of contralateral malignancy, the presence of a testicular mass in a non-scrotal location, and the high suspicion of malignancy. The patient was counseled regarding the options of surveillance and adjuvant therapy. Given the Stage I disease and negative margins, surveillance was recommended in accordance with NCCN guidelines (11).

Pathology findings (Second tumor)

Gross examination revealed a well-circumscribed 11×9 mm tumor. Histopathological examination showed classical seminoma with a characteristic "fried egg" appearance (Figure 2). Immunohistochemistry was positive for OCT3/4, CD117, and PLAP and negative for CK and EMA. There was no lymphovascular invasion, and the tumor was confined to the testis (pT1). The surgical margins, vas deferens, and spermatic cord were free of tumor.

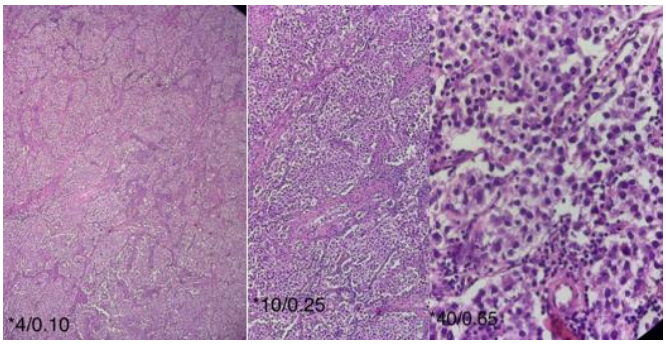

Figure 2. Seminoma tumor at different magnifications

Follow-up after second surgery

The patient has been followed for 18 months since the second orchiectomy. Serial imaging has shown no evidence of recurrence or metastasis. No complications (e.g., wound infection, hematoma, or psychological distress) have been reported. The patient expressed satisfaction with the diagnostic and therapeutic process.

Discussion

In this report, we present a rare case of metachronous bilateral classical seminoma with identical histology in a 54-year-old man with bilateral cryptorchidism. The interval between the two tumors was four years, consistent with the typical 5-year window reported in the literature for synchronous/metachronous tumors (4). However, intervals of up to 31 years have also been reported (10), suggesting that the risk of a contralateral tumor persists throughout the patient's lifetime.

Comparison with published cases

Our case is notable because of the patient's bilateral cryptorchidism, which significantly increases the baseline risk of malignancy. The occurrence of both tumors in inguinal testes, even after orchiopexy, underscores the importance of lifelong surveillance in this population.

Clinical implications

The primary takeaway from this case is that surveillance of patients with cryptorchidism and a history of testicular cancer should not be limited to 5 years. Current guidelines recommend follow-up for 5-10 years, but our case, together with others (10), suggests that the risk of metachronous tumors persists well beyond this period. Clinicians should maintain a high index of suspicion and continue regular physical examinations and ultrasound imaging indefinitely. Furthermore, this case highlights the importance of self-examination and patient education, particularly among patients with non-palpable or ectopic testes. The presence of a testis in the inguinal canal does not eliminate the risk of malignancy, and orchiopexy does not abolish this risk, although it may facilitate earlier detection.

Pathological considerations

The identical histology (Classical seminoma) of both tumors supports the hypothesis of a field defect or genetic predisposition rather than metastasis. The absence of lymphovascular invasion and the low stage suggest a favorable prognosis. The patient's elevated LDH level before the second surgery normalized postoperatively, consistent with a reduction in tumor burden.

Patient perspective

The patient reported that he was initially anxious about the possibility of a second tumor but was reassured by the regular follow-up and early detection. He expressed satisfaction with the care provided and appreciated the clear communication regarding treatment options. He has maintained a normal postoperative quality of life, with no functional or psychological complications.

Limitations

This study has several limitations. First, it is a single case report, and the findings may not be generalizable to all patients. Second, the follow-up period after the second surgery is relatively short (18 months), and long-term outcomes are not yet available. Third, genetic testing for hereditary syndromes (e.g., Klinefelter syndrome and familial testicular cancer) was not performed because of patient refusal. Fourth, the patient's occupational and environmental exposures were not thoroughly investigated. Finally, the patient's perspective was collected informally and may not fully capture the broader patient experience.

Supplementary material

CARE Checklist: A completed CARE (CAse REport) guidelines checklist is provided as a Supplementary File.

Conclusion

Metachronous bilateral classical seminoma with identical histology can occur in patients with bilateral cryptorchidism despite regular surveillance. This case demonstrates a 4-year interval between tumors and highlights the importance of extended lifelong follow-up for these patients. Radical orchiectomy remains the standard treatment, and surveillance is appropriate for Stage I disease. Clinicians should remain vigilant for contralateral tumors and educate patients about self-examination and the importance of regular imaging. We recommend that future guidelines consider extending surveillance beyond 5 years for patients with cryptorchidism and a history of testicular cancer, particularly those with bilateral involvement or ectopic testes.

Acknowledgement

Not applicable.

Funding Sources

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical Statement

This case report was conducted in accordance with the principles of the Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this case report and the accompanying images. A copy of the consent form is available for review by the Editor. Patient confidentiality was strictly maintained, and all identifying information was anonymized.

Conflicts of Interest

The authors declare no conflicts of interest.

Author Contributions

Dr. Omid Majidian: Conceptualization, Methodology, Supervision, Writing-Original draft. Dr. Farzin Valizade: Formal analysis, Validation, Writing-Review and Editing, and Visualization.

Data Availability Statement

Not applicable.

Use of Artificial Intelligence

We confirm that we have used AI-assisted tools for language polishing and formatting of this manuscript. All authors take full responsibility for the accuracy, originality, and integrity of the content, and have reviewed and approved the final version.

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Cite this article as:

Majidian O, Valizade F. Metachronous bilateral classical seminoma in a patient with bilateral cryptorchidism: A case report and review of the literature. *Jorjani Biomedicine Journal.* 2026;X(X):X. http://dx.doi.org/10.29252/jorjanibiomedj.X.X.X