

Bilateral synchronous testicular seminoma: A rare presentation of a rare disease

Pedro Simões de Oliveira, Tiago Ribeiro de Oliveira, Sérgio Pereira, David Martinho, Tomé Lopes

Hospital de Santa Maria, Urology Department, Lisbon, Portugal.

Summary *Objective: To present a case of a bilateral synchronous testicular seminoma in a young male clinical stage IIB.*

Material and method: A 37 years old man presented a bilateral testicular mass with elevated tumoral markers. Histology of frozen section revealed bilateral seminoma and bilateral radical orchiectomy was performed.

Result: Enhanced chest and abdominopelvic staging CT scan revealed a lymphadenopathy of 30 mm within the inter-aorto-cava nodal chain (stage IIB). Patient received three cycles of BEP. Three months later 18F-FDG PET showed no evidence of hypermetabolic activity and serum tumoral markers were normal.

Conclusion: Bilateral testicular germ cell tumors are a rare disease. Management of this tumors is controversial. Bilateral radical orchiectomy is the standard of care, nevertheless, in order to preserve fertility and androgen production, an organ-sparing surgery can be attempted in selected cases. Although prognosis is good, with overall survival rates similar to patients with unilateral disease, life-long close follow-up may be advocated due to relapse risk.

terone levels (1). In order to preserve fertility and androgen production, organ-sparing surgery can be attempted when tumor volume is less than 30% of the testicular volume and surgical rules are respected (1). In those cases, the rate of associated germ cell neoplasia in situ (CIS) is high (up to 82%). Follow-up schedule is the same for patients with unilateral testis cancer (1).

Life-long follow-up may be advocated because of a small risk for late relapse and close clinical control, with ultrasound of the testis, may be recommended for patients undergoing testis-preserving surgery (1).

We present a rare case of a young male with bilateral voluminous testicular masses representing synchronous seminoma clinical stage IIB.

CASE REPORT

A 37 years old man, without any known risk factor, presented as an outpatient with chief complaints of bilater-

KEY WORDS: Seminoma; Bilateral; Synchronous.

Submitted 28 October 2017; 19 November 2017

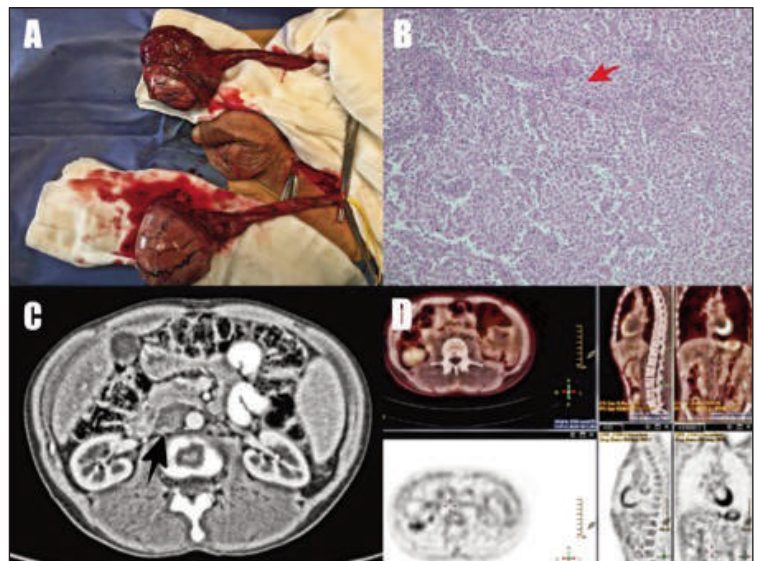
INTRODUCTION

Testicular germ cell tumors (TGCTs) are the most common malignancy diagnosed in males aged 20 to 40, nevertheless they are a rare disease representing only 5% of all urologic tumors (1). Risk factors in the development of testicular tumors include history of cryptorchidism or undescended testis, Klinefelter syndrome, testicular cancer in the first-degree relatives, presence of tumor or intratubular germ cell neoplasia (TIN) in the contralateral testis, and infertility (1).

Bilateral TGCTs (BTT) are even more rare, representing 1-2% of all cases at diagnosis. Approximately 35% of these bilateral tumors are synchronous and the majority are seminomas, being histologically identical almost always (1). Management of this tumors is controversial. Bilateral radical orchiectomy via an inguinal incision is considered the standard of care in cases of synchronous testis cancer with impaired pre-operative testos-

Figure 1.

A) Intra-operative image showing bilateral radical orchiectomy via inguinal incision; B) Pathological examination: nests of seminoma surrounded by bands of fibrous tissue infiltrated with lymphocytes (arrow); C) Enhanced chest and abdominopelvic CT scan revealing a lymphadenopathy of 30 mm within the inter-aorto-cava nodal chain (arrow); D) 18F-FDG PET showing no evidence of hypermetabolic activity.



No conflict of interest declared.

al scrotal swelling and heaviness, since last three months. On local examination swelling was present over bilateral hemiscrotum, hard in consistency, surface was irregular and it was associated with restricted mobility.

Transillumination was negative. There were no palpable inguinal neither supraclavicular lymph nodes. Testis ultrasound revealed multiple bilateral solid nodules occupying about 50% of each testicle. Serum tumor markers were LDH 322 U/L, AFP 1.3 ng/ml and hCG 29 U/L. Testosterone was 453 ng/dL.

Patient underwent inguinal exploration and bilateral testicular biopsy for frozen section histological examination which revealed bilateral seminoma (Figure 1A). A bilateral radical orchiectomy was performed with insertion of bilateral testicular prosthesis.

Pathological examination confirmed bilateral seminoma with invasion of tunica albuginea and vascular and lymphatic invasion (pT2) (Figure 1B).

Postoperatively, serum tumoral markers were normal (LDH 166 U/L, AFP 1.3 ng/ml and hCG 0.3 U/L).

Patient initiated testosterone replacement therapy.

Enhanced chest and abdominopelvic staging CT scan revealed a lymphadenopathy of 30 mm within the interaorto-cava nodal chain (stage IIB) (Figure 1C).

Patient was referred to Oncology and received three cycles of BEP (bleomycin, etoposide and cisplatin).

Three months later 18F-FDG PET showed no evidence of hypermetabolic activity (Figure 1D) and serum tumoral markers were normal.

CONCLUSIONS

BTT are rare and therefore literature is scarce, mainly case reports or small series, restricting management strategies for these patients.

Patients with BTT, present with different problems requiring careful management to permit a good quality of life. Bilateral radical orchiectomy is the standard of care treatment, nevertheless this approach has a vast impact on fertility and can render patients dependent on life-long testosterone supplementation (1).

In 1997 Heidenreich *et al.* (2) presented a series of 13 patients with BTT who underwent testis-sparing surgery. Six of the 13 patients underwent testicular radiation for CIS and five patients had adjuvant local therapy.

A testicular biopsy was taken 6 months post-operatively and revealed Sertoli cells only in all patients who had received radiation therapy. Only one patient had local recurrence 9 months after tumor enucleation.

Testis-sparing surgery must be performed whenever possible, when tumor volume is less than 30% of the testicular volume and surgical margins are respected (1), although multiple testicular biopsies are advocated in this setting in order to identify the presence of CIS and tumor multifocality (1).

The occurrence of CIS is a factor suggesting the need for irradiation therapy which can result in loss of fertility and hormonal function of the remaining part of the testicle, the important factors affecting patients' quality of life (1).

Holzbeierlein *et al.* (3) reported a series of 58 patients with bilateral testicular tumors treated at the Memorial Sloan Kettering Cancer Center between 1950 and 2001, ten had synchronous tumors while 48 had metachronous tumors, being seminoma the most frequent histology. The authors suggested that these patients had favorable outcomes when compared with patients with unilateral tumors and the prognosis was mainly determined by tumor histology and metastatic disease.

Although these patients usually present with a higher stage disease, they have an excellent prognosis with overall survival rates comparable with those with unilateral disease.

Follow-up is the same as unilateral disease, nevertheless life-long close follow-up is advisable due to the small risk of late relapse (1).

REFERENCES

1. Campobasso D, Ferretti S, Frattini A. Synchronous bilateral testis cancer: clinical and oncological management. *Contemp Oncol (Pozn)*. 2017; 21:70-76.
2. Heidenreich A, Hörtl W, Albrecht W, *et al.* Testis-preserving surgery in bilateral testicular germ cell tumours. *Br J Urol*. 1997; 79:253-7.
3. Holzbeierlein JM, Sogani PC, Sheinfeld J. Histology and clinical outcomes in patients with bilateral testicular germ cell tumors: The Memorial Sloan Kettering Cancer Center experience 1950 to 2001. *J Urol*. 2003; 169:2122-5.

Correspondence

Pedro Simões de Oliveira, MD (Corresponding Author)
pedrosimoesdeoliveira@gmail.com

Tiago Ribeiro de Oliveira, MD
tiagoribeirooliveira@sapo.pt

Sergio Pereira, MD
sergioahpereira@gmail.com

David Martinho, MD
martinho_david@hotmail.com

Tomè Lopes, MD
tomematoslopes@gmail.com

Avenida Professor Egas Moniz 1649-035 Lisbon, Portugal