Olgu Sunumu
Case Report

Treatment Management of Leptomeningeal and Brain Metastases as a First Reccurence Site in a Case with Germ Cell Testicular Tumor

İLK NÜKSÜ LEPTOMENİNGEAL VE BEYİN METASTAZI OLAN GERM HÜCRELİ TESTİS TÜMÖRLÜ BİR OLGUDA TEDAVİ YÖNTEMİ

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ABSTRACT

Patients with testicular germ cell tumors (TGCT) usually presented with a testicular mass and retroperitoneal lymph nodes metastases, but brain and leptomeningeal metastases are rarely encountered in oncology practice. There are not any proven treatment options for relapsing TGCT with brain or leptomeningeal metastasis. We herein present a rare case of TGCT of brain and leptomeningeal metastasis as a first sign of recurrence with literature review.

Keywords: testicular germ cell tumor, brain metastasis, leptomeningeal metastasis

ÖZ

Testiküler germ hücreli tümör (TGCT) olan hastalar genellikle testis kitlesi ve retroperitoneal lenf nodu metastazları ile başvururlar ancak onkoloji pratiğinde beyin ve leptomeningeal metastazlara nadiren rastlanır. Beyin veya leptomeningeal metastazlı TGCT'nin tekrarlaması için kanıtlanmış herhangi bir tedavi seçeneği yoktur. Biz burada nadir görülen bir beyin ve leptomeningeal metastazlı TGCT olgusunu literatür taraması ile birlikte ilk rekürrens belirtisi olarak sunuyoruz.

Anahtar Kelimeler: testis germ hücreli tümör, beyin metastazı, leptomeningeal metastaz

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Central nervous system (CNS) metastases occur in approximately 1 percent of men with testicular cancer at the time of diagnosis, and between 0.4 and 4 percent subsequently develop brain metastases (1,2). Nevertheless, leptomeningeal metastasis of testicular germ cell tumors (TGCT) is very rare (3). Herein, we report an unusual case with leptomeningeal metastasis as a first sign of recurrence and a rare treatment schedule to contribute to the literature.

CASE

42-year-old men achieved complete response after front line treatment for metastatic TGCT. He presented with the complaint of hearing loss, imbalance and urinary incontinence. While the systemic disease was under control, tumor markers were elevated as follows: ß-HCG: 113.9 mU/l and AFP: 19.73 IU/ml. Brain MRI revealed newly developed parenchymal and perineural metastasis associated with the 7th cranial nerve and leptomeningeal metastasis was also found in the spinal MRI (Figure 1,2) and FDG-PET/CT scan.

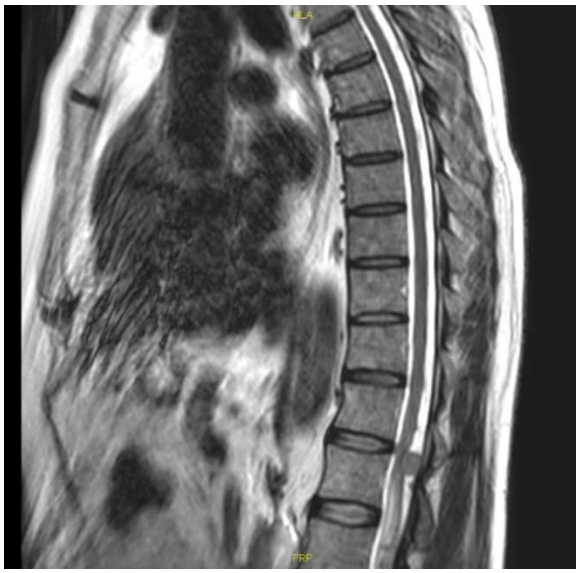

Figure 1: Contrast enhanced T1-weighted image shows right IAC (White arrow), left Vth nerve (White arrow) and right PCA (white small arrow) leptomeningeal metastasis.

Figure 2.

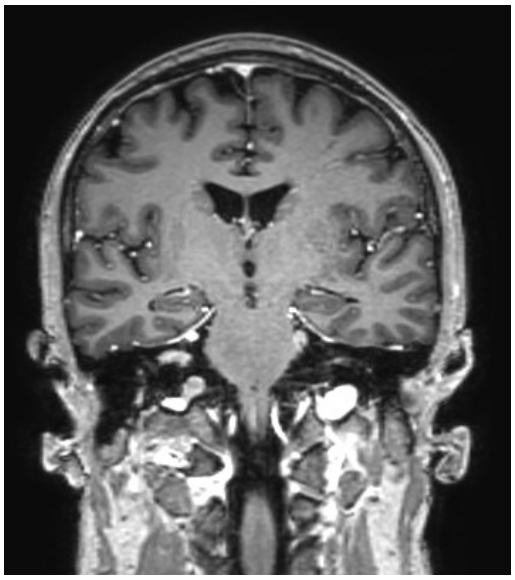

Figure 2: Coronal T2-weighted image shows one of the intradural extramedullary metastasis(arrow) displacing the spinal cord.

Figure 3, Coronal and sagittal FDG-PET/CT images shows leptomeningeal involvement with FDG uptake in spinal cord. Histopathological images of the biopsy taken from the brain lesion to confirm the diagnosis of germ cell tumor metastasis are shown in Figures 4,5 and 6.

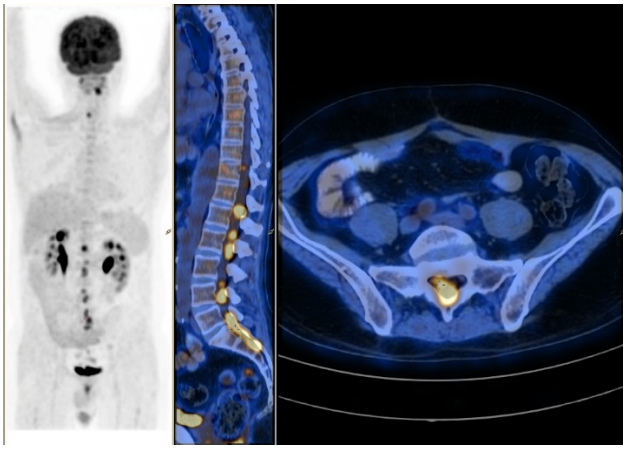

Figure 3: Coronal and sagittal PET/CT images showsleptomeningeal involvement with FDG uptake in spinal cord.

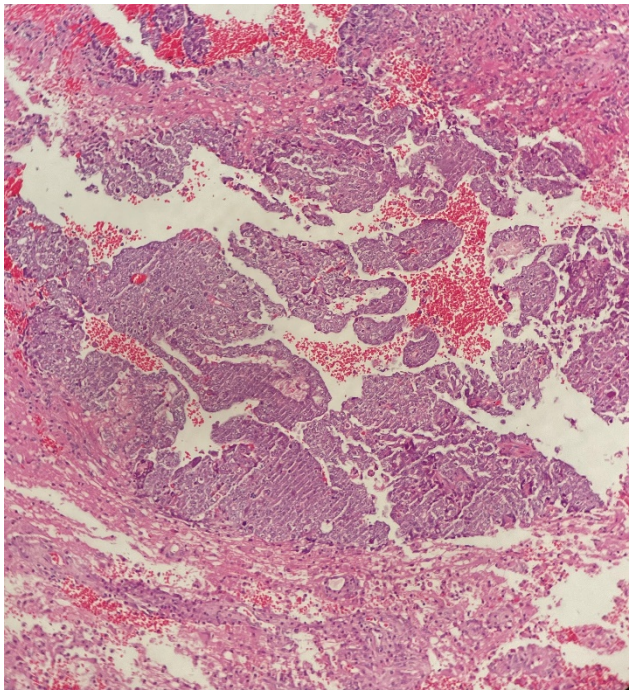

Figure 4: Large, epithelioid malignant tumor with prominent nucleoli showing in solid and papillary growth patterns; and adjacent glial tissue (HEx40)

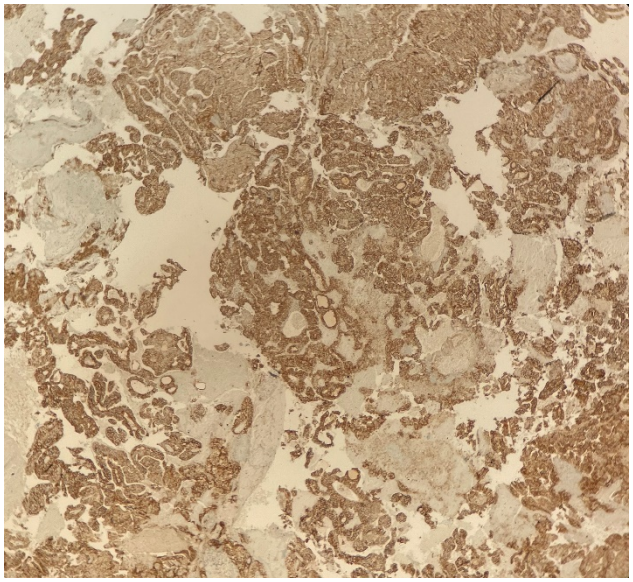

Figure 5: Most tumor cells express cd30, which indicates the embryonal carcinoma component (CD30 x100)

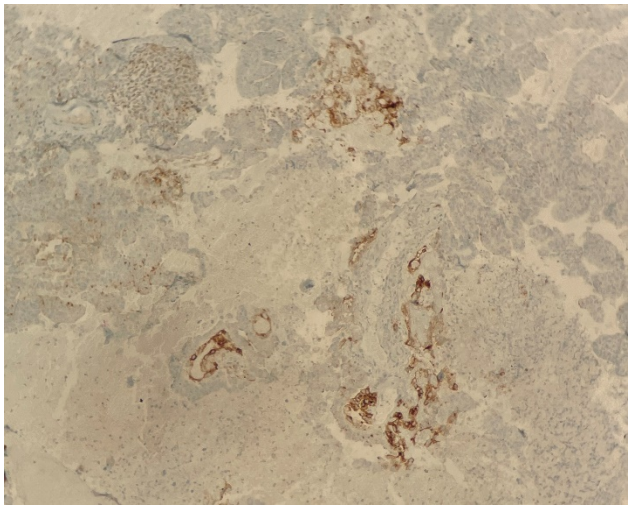

Figure 6: Glypican3 expression in yolc sac tumor component (Glypican X100)

Whole brain radiation therapy (WBRT) and whole spinal neuroaxis RT were administered. After three months systemic progression occurred and he treated with three courses of paclitaxel, ifosfamide and cisplatin (TIP) chemotherapy as salvage regimen, and then he underwent of autologous hematopoietic progenitor cells transplantation because of good response to RT and systemic chemotherapy. After transplantation complete radiological response was found. Also, tumor markers were within the normal limits. While the patient was routinely followed-up, he was hospitalized in February 2021 due to poor general condition and CMV infection. Fourteen months after brain and leptomeningeal metastasis, he died due to infection and disease progression.

DISCUSSION

Initial management of advanced TGCTs is based on the risk stratification (4). Metastatic seminomas are classified as either good or intermediate risk based upon whether metastases to organs other than the lungs or lymph nodes are present. On the other hand, non-seminomatous TGCTs are divided into good-, intermediate-, and poor-risk categories (3). For men with intermediate or poor risk, despite there is no data regarding the right number of chemotherapy cycle, four cycles of BEP regimen is usually the standard of care (4).

Brain and leptomeningeal metastasis are very rare but they are poor risk factor and their optimal treatment approach could not be clearly shown in patients with TGCTs. The treatment options include resection and/or RT and chemotherapy. In a report 228 of 523 patients with TGCT had brain metastasis at diagnosis, and 295 patients relapsed with brain metastasis and the primary tumor site was testis (83.1%) and most patients with brain metastasis died within first year (2). In our case, we initially preferred WBRT and whole spinal neuroaxis RT rather than chemotherapy because of systemic disease was under control and to gain quickly respond to neurological symptoms. Similarly to literature, patient's survival after brain metastasis occurred was 14 months.

High-dose chemotherapy with peripheral blood stem cell transplantation is an option for relapsed TGCTs. Kalra et al reported 19 relapsed TGCT patients with brain metastasis who treated with high-dose chemotherapy and/or local management, and 11 patients (44%) were alive at a median follow-up of 2 years (5). We used also stem cell transplantation after TIP regimen as salvage chemotherapy, in our patient.

Despite the brain metastasis, development of leptomeningeal metastasis is very rare, and there are just one pediatric case in the literature (6). A 9-year-old male patient was diagnosed with a germ cell tumor in the pineal gland and remission was achieved after chemoradiotherapy. One month after the completion of radiotherapy, a 1.3 cm mass was detected, and serum tumor markers also increased in follow up. Despite radiotherapy, high-dose chemotherapy and autologous stem cell transplantation the patient died after three months of treatment (6).

In the literature, there is no similar case of TGCT recurring with brain and leptomeningeal metastasis as a first site of recurrence. Patients with brain metastases are also very few. In patients with TGCT under follow-up who had neurological complaints, brain and/or leptomeningeal metastases should be kept in mind in in differential diagnosis of elevated tumors markers and neurological symptoms. Thus our case contributes to the literature with its treatment modality and imaging as a unusual relapsed TGCT.

Written informed consent was obtained from the patient. There is no conflict of interest.

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