A case of cervical spinal mass with cord compression and rib bone metastasis from presumably burned-out seminomatous testicular germ cell tumor

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Most germ cell tumors are located in the gonads however there are instances where these tumors are located elsewhere in which are termed extragonadal germ cell tumors. When primary lesion of the testicular tumor has regressed, the term "burned-out testicular tumor" has been proposed. We herein report the first case of burned-out seminoma of the testis presenting as a cervical spinal mass causing cord compression with bone metastasis.

Key Words: testis, germ cell tumor

Case

A 38-year-old Caucasian male with no past medical history presented with chief complaint of neck pain for 3 weeks. The pain was sudden in onset, sharp in nature, severe, radiating to right arm associated with numbness and not getting relieved with over the counter nonsteroidal anti-inflammatory drugs. There was no preceding history of trauma, infection or sick contacts. On examination, there was no visible spinal deformity, edema or erythema. Range of motion of the neck was limited due to pain. Tenderness of the spine was elicited at C7 level. The muscle strength of right arm was preserved but sensation to pin prick was decreased in the fingers.

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Magnetic resonance imaging (MRI) of the entire spine with contrast, Figure 1, showed destructive mass lesion at the posterior aspect of C7 vertebral body that extended to the spinous process, left lamina and the epidural space. There was displacement and mild compression on the lower cervical cord. Patient was immediately started on IV dexamethasone and underwent emergent posterior decompression of cervical spine. Biopsy, Figure 2, showed classic type seminoma; malignant germ cell tumor involving bone and soft tissue, with focal extensive necrosis. Tumor was noted to be composed entirely of seminoma component with interspersed scattered lymphocytic infiltrates. Immunostains show tumor cells to be positive for CD 117, focally positive for PLAP, a few scattered cells positive for AE1/AE3 (paranuclear dot- like pattern). The tumor cells were negative for AFP, glypican-3 and CD30.

On further work up, MRI of the abdomen with contrast showed a mass measuring 3 cm x 6.6 cm in length in the posterior right lung base that started in the pleural space extending to the ribs, subcutaneous space and the surrounding muscles. We performed

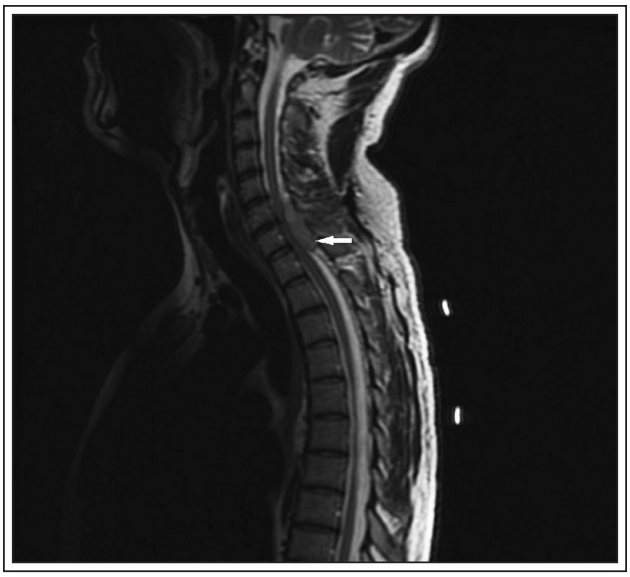

Figure 1. MRI of the entire spine showing a destructive cervical spinal mass (denoted by the white arrow) at the posterior aspect of the C7 vertebral body extended to the spinous process and the epidural space with mild compression of the lower cervical cord.

nuclear bone scan that showed 6 cm mass in the right eleventh rib. On scrotal ultrasound, there was mild inhomogeneous echotexture of the right testicle which was likely due to prior vasectomy no evidence of testicular mass, torsion or epididymitis was seen. MRI scrotum was also done that did not show any mass. MRI brain was negative for any metastasis.

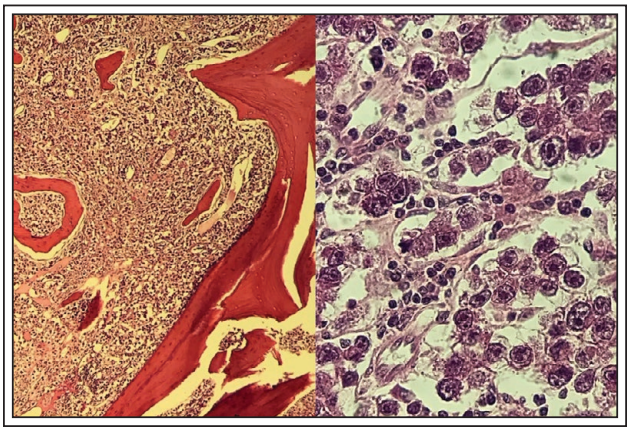

Figure 2. Microscopic finding of cervical spinal mass at C7 shows malignant germ cell tumor involving bone and soft tissue, with focal extensive necrosis. Tumor was noted to be composed entirely of seminoma component with interspersed scattered lymphocytic infiltrates.

His lab studies showed normal Complete blood count, liver function tests, LDH was normal (170U/l), normal AFP (4.7 ng/mL) , normal B HCG (< 0.1 MIU/ML). Final diagnosis was seminoma stage IIIC (T0 N0M1b S0). Patient was treated with chemotherapy with curative intent. He received four cycles of bleomycin, etoposide and cisplatin. The CT scan chest, spine and nuclear bone scan post treatment showed complete response to therapy.

Discussion

Testicular cancer is derived from mainly germ cell tumors (95%) and sex cord/gonadal stromal tumors (5%) such as Leydig and Sertoli cell tumors.[1] The incidence of testicular cancer in men is 5.9 per 100,000 per year.[2] In 2016, according to the National Cancer Institute of Surveillance, Epidemiology, and End Results (SEER) program, it is estimated about 263,137 men in the United States live with testicular cancer. In addition, analysis models from 2006 to 2015 show an up-trending rise of newly diagnosed testicular cancer cases at an average rate of 0.8% per year.[2] According to the World Health Organization (WHO) classification of testis tumors, germ cell tumors are further classified into: precursor lesions (intratubular germ cell neoplasia); pure forms (seminoma, spermatocytic seminoma, embryonal carcinoma, yolk sac tumor, trophoblastic tumors and teratoma); and mixed forms.[3] Germ cell tumors usually affect men in the late twenties and thirties.[3]

Most germ cell tumors are located in the gonads, however, there are rare cases in which these tumors are located elsewhere and are termed extragonadal germ cell tumors (EGCT).[4] Primary EGCT are uncommon and make up about 1%-2.5% of all germ cell tumors.[4] The most common site for primary EGCT are located in the mediastinum and the retroperitoneum.[4] Germ cell tumors located in the retroperitoneum, however, are usually resulting from metastasis from a primary gonadal germ cell tumor.[4] Infrequently there are cases where these tumors can also metastasize to the brain, tonsils, lungs, gastrointestinal tract, liver, spleen, kidneys, adrenal glands, peritoneum and bones.[5] According to Choyke et al, differentiating primary EGCT from secondary EGCT metastasis from a primary testicular tumor can be difficult; careful investigation of the testis with ultrasound is important and warranted as gonadal germ cell tumors can delay in development or could even possibly regress.[4]

When the primary lesion of the testicular tumor has regressed, the term "burned-out testicular tumor" has been postulated.[6] The first case of burned-out tumor of

the testis was reported by Prym in 1927.[6] In many cases, macroscopically, these tumors regress to fibrous scars and occasionally to precursor lesions of intratubular germ cell neoplasia; atrophy of the testes have been reported.[4,6] Microscopic evaluation of these burned-out tumors histologically include: hemosiderin-laden macrophages and lymphocytes deposits, fibrosis, Hematoxylin bodies, psammoma bodies, and calcium phosphate depositions.[4-6] Radiologically, findings from ultrasound in burned-out tumors include changes in the echogenicity of a foci possibly attributed to calcium deposits, fibrosis and even from the Hematoxylin or psammoma bodies.[4,5,7,8]

In our case, the histological findings of the cervical epidural mass revealed seminoma (classic type) with immunohistochemistry staining positive for CD117 and Podoplanin, focally positive for PLAP and CAM5.2; and few cells positive for AE1/AE3. MRI of the abdomen and bone scan revealed metastatic mass involving the right eleventh rib but testicular exam and imaging (including ultrasound and MRI) did not reveal any primary mass.

Based on the fact, no tumors were visualized on imaging, we believed orchiectomy was not appropriate at this time. In addition, given that an in-homogenous echotexture of the right testis was detected with a slightly smaller size right testis, we suspect these findings from ultrasound are consistent with what is frequently described in the literature as "burned-out" seminoma.

Conclusion

Spontaneous regression of testicular tumor with metastatic spread referred to as "burned–out" germ cell tumor has been described in case reports. On review of the literature, we were able to find one case report from 2017 describing solitary bone metastasis with burned-out seminoma of the testes;[9] to our knowledge, the case we describe appears to be the first case report of burned-out seminoma of the testis presenting as a cervical spinal mass causing cord compression with bone metastasis. □

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