Large teratoma at aortic bifurcation: an unusual metastasis of testicular cancer

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A 29-year-old man presented with a right testicular mass. Serum tumor markers were within normal limits. When compared to a previous computed tomography (CT) scan, a new 4 cm presacral mass was present. He underwent radical right inguinal orchiectomy that demonstrated a mature teratoma and seminomatous components. The patient received four cycles of chemotherapy. Over the course of chemotherapy, the mass grew in size and therefore he underwent retroperitoneal lymph node dissection. Pathology confirmed it to be a teratoma with negative retroperitoneal lymph nodes. The unusual presentation of an isolated metastasis to the presacral region raises the question of altered lymphatic drainage.

Key Words: retroperitoneal lymph node dissection, testicular cancer, teratoma

Introduction

We present the case of a Hispanic man who presented with right testicular mass and a separate, presacral lesion. We describe his presentation, clinical course and pathologic findings, and discuss the possible relations of the lesions.

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Case presentation

A 29-year-old Hispanic man presented with intermittent left testicular pain and epididymitis for several months. There was no history of cryptorchidism, orchidopexy or any prior inguinoscrotal surgery. Physical examination elicited left testicular tenderness localized to epididymis and a palpable nodule in a right atrophic testicle. Scrotal ultrasound study confirmed multiple right testicular lesions. Serum tumor markers were within normal limits. Indices of liver and renal function were normal.

The right testicular lesion prompted immediate intervention. The patient underwent a right radical

orchiectomy the following day that revealed extensive mature teratoma with small foci of seminoma(< 5% of total specimen). There was no lymphovascular invasion and all components were contained within the tunica vaginalis, and thus it was pathological stage T1. A staging computed tomography (CT) scan of thorax, abdomen and pelvis revealed a region of fluid attenuation with solid components inferior to the bifurcation of the abdominal aorta suggesting presacral teratoma, Figure 1. The lesion was not present in a prior CT scan of the abdomen for evaluation of appendicitis 15 months prior.

After consultation with medical oncology, it was determined that this clinical stage III testicular tumor would be treated with chemotherapy. With the large retroperitoneal mass and mixed germ cell testicular primary, a full course of chemotherapy was sought prior to any surgical therapy. A total of four courses of bleomycin, etoposide, and cisplatin (BEP) chemotherapy was completed 1 month after radical orchiectomy. At the termination of the chemotherapy, reimaging showed growth of the presacral lesion. Due to the large retroperitoneal mass and questionable occult lymphatic metastases, a bilateral full template retroperitoneal lymph node dissection (RPLND) with complete excision of the presacral mass, Figure 2, was then performed. The retroperitoneal lymph nodes showed no evidence of neoplasm and the presacral lesion showed mature teratoma. After an uneventful recovery from surgery, the patient has been disease free at 10 months and has been followed up monthly.

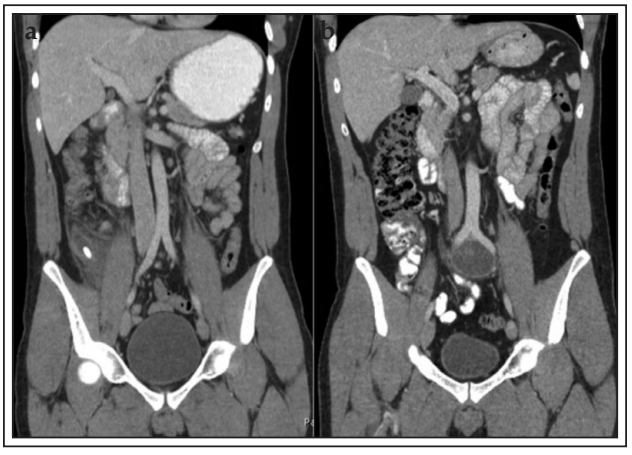

Figure 1. **a)** Coronal CT scan 15 months prior to presentation. **b)** Presacral tumor inferior to the bifurcation of the abdominal aorta measuring approximately 5.0 cm x 3.8 cm x 4.2 cm.

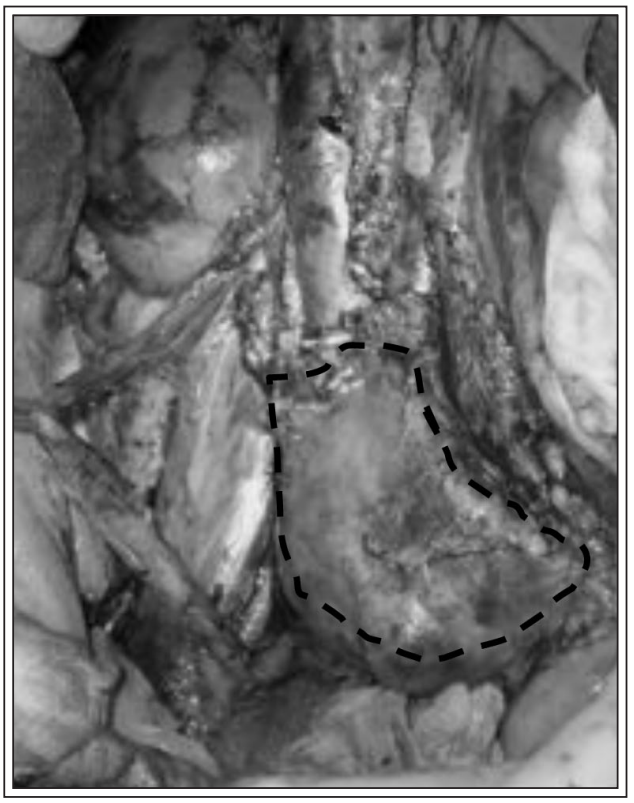

Figure 2. Retroperitoneal lymph node dissection with resection of presacral mass. Dash line outlines the presacral teratoma.

Discussion

To our knowledge, there are no reports of metastatic teratomas located at the aortic bifurcation as isolated metastasis from testicular cancer. Although teratoma is not an uncommon pathology during the post chemotherapy retroperitoneal lymph node dissection, this isolated location is rare. Non-seminomatous germ cell tumor (NGCT) may spread locally or via lymphovascular channels.[1] It has been demonstrated that the lymphatic drainage patterns of the testis are very predictable and stepwise. In the absence of previous inguinoscrotal surgery, the primary lymphatic drainage of the right testis is the interaortocaval nodes just below the renal vasculature, followed by precaval and paracaval lymph nodes. Left testis drains into the para-aortic and preaortic lymph nodes.[2] Retroperitoneal lymph nodes then drain into cisterna chyli. 85% of right-sided metastasis are confined to the ipsilateral lymph nodes, while 13% are bilateral and 1.6% are found in contralateral nodes only.[3] Caudal spread has been reported and is usually to the right

common iliac and right external iliac lymph nodes. This is frequently due to retrograde lymphatic spread and rarely, aberrant testicular lymph drainage. In men, the presacral nodes drain from the rectum while in women they drain the cervix, uterus and upper part of the vagina. The ascending pathway of the presacral lymph nodes extends into common iliac nodes, lumbar nodes or inferior mesenteric nodes. This case reflects the possibility of either retrograde testicular lymph drainage to the iliac nodes and then to the presacral area, or an anomalous connection from presacral area to the early testicular lymph pathway. The finding of all lymph nodes negative from RPLND renders the latter option more plausible.

Primary extragonadal germ cell tumors (EGCTs) account for only 1% to 5% of all germ cell malignancy.[4] A synchronous discovery of the presacral teratoma and TGCT with all negative lymph nodes raises the possibility of a primary EGCT with metachronous testicular cancer (MTC). In adults, primary EGCTs are more commonly found in the retroperitoneum or the mediastinum but occasionally found in the pineal gland, presacral area or liver.[5] The rate of MTC in men with EGCTs is largely unknown due to its low incidence of only 31 reported cases in literature.[6] However, the possibility of this patient having a primary EGCT in the presacral region is rather slim. In majority of sacrococcygeal germ cell tumors (SGCTs), the histology is mature teratoma which has a benign course. SGCTs are generally considered to have been present since birth and slow growing.[7] With a previous CT abdomen revealing no evidence of presacral mass, it is unlikely that a congenital teratoma suddenly advanced within a 15 months period.

With primary testicular NSGCT metastasizing to prescaral region being more likely, patient was managed as stage III NSGCT and received cisplatin-based induction chemotherapy following orchiectomy. We acknowledge that a teratoma would not respond well to chemotherapy, but we could not be sure the mass was all teratoma and with a large lesion felt that chemotherapy would the first option. In situations such as this, consideration can be given to reimaging mid-chemotherapy and performing surgery earlier if there is a poor response. Post chemotherapy RPLND and resection of chemo-resistant teratoma was performed subsequently. Comparably, if the presacral mass was a primary EGCT, the management is similar to advanced NSGCT of the testis, initial chemotherapy consisting of a cisplatin-based regimen followed by surgical removal of residual disease or teratoma is utilized.[8]

There are many templates for retroperitoneal lymph node dissection, which are reviewed well by Large et al.[9] Current full template RPLND and modified templates often include lymph nodes between the level of the renal vessels and the bifurcation of the common iliac artery.[10] However, it does not usually extend posteriorly to include lymph nodes of the internal iliac artery, which ultimately drain the presacral region. The rare location of extratemplate tumor reinforces the awareness of limitations of the mapping in underestimating the extent of retroperitoneal disease.

Conclusion

We believe this rare presacral metastasis of testicular cancer may represent an atypical lymphovascular pathway. The rare location of metastasis raises the concern for the possibility of a synchronous primary EGCT in adult and the limitation of the current retroperitoneal lymph node mapping. □

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