

Oncology

A case of contralateral superficial inguinal recurrence of testicular embryonal carcinoma

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ABSTRACT

Metastatic embryonal carcinoma to the subcutaneous tissues is rare. Prior cases have occurred in the setting of undiagnosed widely metastatic disease. Here we present the first case of metastatic embryonal cancer to the contralateral subcutaneous inguinal region in the absence of any other sites of metastatic disease.

Introduction

Testicular cancer is uncommon with 1 in 250 males developing this malignancy during their lifetime. Embryonal carcinoma is a type of non-seminomatous germ cell tumor (NSGT) that presents in the third decade of life. This type of testicular tumor is frequently metastatic at the time of diagnosis and is associated with early disease relapse. Subcutaneous metastasis of embryonal carcinoma is extremely rare and is thought to be concomitant with widespread disease and poor prognosis.¹ Below, we report a case of solitary left inguinal metastasis comprised of a mixed germ cell tumor presenting one-year after a right inguinal orchiectomy for a Stage 1 embryonal carcinoma.

Case presentation

A 27-year-old male with Bipolar disorder presented to his primary care provider (PCP) after noticing a “lump” in his right testicle. A scrotal ultrasound showed a 9.3 × 7 mm hypochoic mass in the right testicle with internal flow on Doppler. The left testicle was normal. Secondary to his Bipolar disorder the patient did not follow up with urology. He re-presented to his PCP six months later with a painful right hemiscrotum. Repeat ultrasound showed interval growth of the right testicular mass to 15 mm in the largest diameter. The left testicle was normal. He then underwent a right radical inguinal orchiectomy. Prior to surgery his LDH was 141, and both AFP and βhCG were within normal limits (WNL). Post-op tumor markers were WNL. Pathology showed a 1.4 × 1.1 × 1.0 cm embryonal cell carcinoma (pT1a) with no tunica vaginalis or lymphovascular invasion. Margins were negative. A staging computer

tomography (CT) scan of the chest/abdomen/pelvis was performed and was negative for metastatic disease. The patient elected for observation.

The patient re-presented one-year later complaining of a left groin mass. On exam, that patient was found to have a prominent, large, fixed non-tender mass in his left groin (Fig. 1). Tumor markers were again WNL. A CT chest/abdomen/pelvis showed a 4.5 × 6.7 × 5.2 cm heterogeneous mass with peripheral calcifications in the left inguinal region just deep to the skin and superficial to the femoral artery and vein (Fig. 2). No additional sites of metastasis were identified. A percutaneous biopsy of the mass was positive for teratoma. The patient underwent a wide local excision of the mass followed by a left inguinal lymph node dissection (Fig. 3). The mass was located just superficial to the femoral triangle and encased the saphenous vein. Pathology showed metastatic malignant mixed germ cell tumor (60% embryonal & 40% mature teratoma) associated with a deep inguinal lymph node (Fig. 4). The embryonal component was positive for CD30 with a focus of Glypican 3 immunoreactivity; AFP was negative. Margins and lymph nodes were negative. The patient underwent 3 cycles of Bleomycin, Etoposide, and Platinum (BEP). Six-months post-inguinal mass excision the patient is well with no evidence of disease recurrence.

Discussion

Nonseminomatous germ cell tumors (NGCTs) have a predictable metastatic pattern based on the lymphatic drainage of the testicles. Related to our case, the right testicle drains to the infrarenal inter-aortocaval lymph nodes and then to the paracaval and para-aortic nodes. As such, the retroperitoneum is the initial site of metastasis for

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Fig. 1. Prominent, large fixed non-tender mass in left groin one-year after a right inguinal orchiectomy for a pT1a embryonal cell carcinoma of the right testicle.



Fig. 2. CT C/A/P showing a 4.5 x 6.7 x 5.2 cm heterogenous mass with peripheral calcifications in the left inguinal region just deep to the skin and superficial to the femoral artery and vein.

80% of patients with testicular cancer.²

Interestingly, two enhanced CT scans (separated by one-year) did not show any retroperitoneal nodes of concern or other sites of distant metastasis. The only site of metastasis was the contralateral left subcutaneous inguinal region, which is an atypical location for metastasis for this type of malignancy. Although 30% of patients with testicular cancer are under-staged at their initial staging CT scan due to micro-metastatic disease,² it's surprising that no other lesions were identified in our patient's second scan performed one-year after right orchiectomy.

Embryonal carcinoma is a poorly differentiated NSGT that can transform into other NSGTs. This type of tumor is more aggressive and associated with a higher rate of early metastasis secondary to both lymphatic and hematogenous spread.² In fact, up to 37% of patients with embryonal carcinoma have distant metastasis at the time of diagnosis.³ However, subcutaneous metastatic embryonal carcinoma is exceedingly rare. Only three reports have described such an occurrence. In all cases, patients presented with an expanding subcutaneous mass (e.g. chest wall, breast) and were later found to have widely metastatic embryonal carcinoma or a mixed NSGT on further workup (e.g. visceral and pulmonary metastasis).^{1,4,5}

Unlike these cases, our patient presented with a solitary, large,



Fig. 3. Wide local resection of the 6.5 cm left inguinal mass followed by an inguinal lymph node dissection.

subcutaneous metastatic groin lesion one-year after a right inguinal orchiectomy for a pT1a embryonal carcinoma. The recurrent tumor was associated with a deep left inguinal lymph node that anatomically should not have drained either testicle, particularly the right testicle. It's unclear how this lymph node became involved. Repeat ultrasound of the left testicle showed it was free from masses or lesions. It's possible that the patient had secondary lymphatic channels draining to the common or external iliac nodes. Contralateral nodal involvement is more common in right-sided tumors, but such a distal contralateral recurrence is difficult to explain. In addition, unrecognized scrotal violation during the patient's inguinal orchiectomy could have disrupted normal lymphatic drainage. However, there was no evidence of this intra-operatively or on physical exam during groin mass excision. Regardless, the importance of meticulous surgical technique during inguinal orchiectomy cannot be understated.

Despite having metastatic embryonal carcinoma, our patient had normal tumor markers and no evidence of pulmonary or visceral metastasis. Thus, his 5-year relative survival likely approaches 80–90% after completing post-excision BEP chemotherapy. However, he will require intensive physical, biochemical, and radiographic surveillance to monitor for disease recurrence overtime given his atypical presentation of a solitary subcutaneous metastatic lesion.

Conclusion

Metastatic embryonal carcinoma to the subcutaneous tissues is rare. Prior cases have occurred in the setting of undiagnosed widely metastatic disease. Here we present a case of metastatic embryonal carcinoma to the contralateral subcutaneous inguinal region in the absence of any other metastatic sites with good response to post-excision systemic therapy. These types of patients will likely require intensive physical, biochemical and radiographic surveillance to monitor for disease relapse given their atypical re-presentation.

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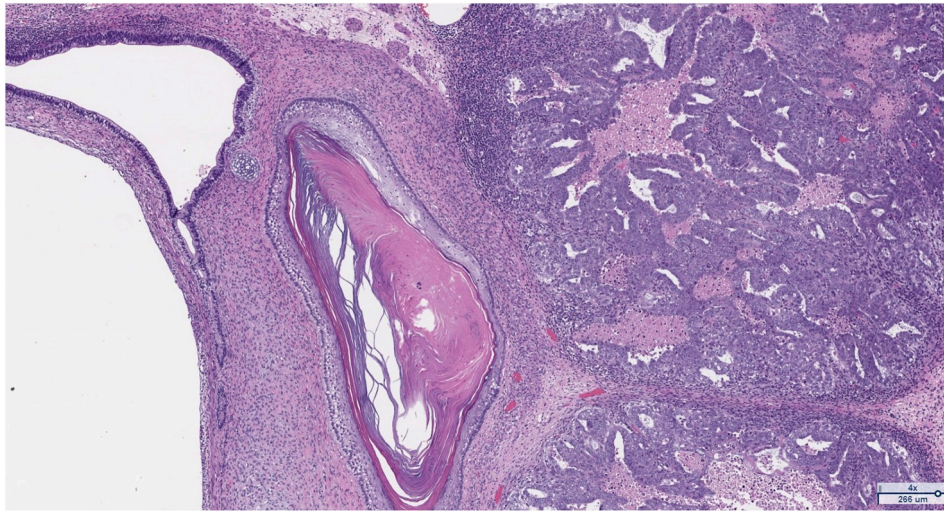


Fig. 4. Histology of the resected left inguinal mass shows a metastatic malignant mixed germ cell tumor comprised of embryonal carcinoma (right in the picture) admixed with mature teratoma (left in the picture, components including squamous epithelium with keratinization, columnar epithelium and a small focus of cartilage).

Conflict of interest and disclosure statement

The authors of this manuscript have no conflicts of interest or competing interests to report.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.eucr.2019.101012>.

References

1. Khan L, Verma S, Singh P, Agarwal A. Testicular embryonal carcinoma presenting as chest wall subcutaneous mass. *J Cytol.* 2009;26:39–40.
2. Stephenson AJ, Gilligan TD. *Neoplasms of the Testis.* Campbell-Walsh Urology. eleventh ed. Elsevier; 2011:784–815.
3. Vugrin D, Chen A, Feigl P, Laszlo J. Embryonal carcinoma of the testis. *Cancer.* 1988; 61:2348–2352.
4. Maubec E, Avril MF, Duvillard P, et al. Mixed nonseminomatous germ cell tumor presenting as a subcutaneous tissue mass. *Am J Dermatopathol.* 2006;28:523–525.
5. Nakahira M, Nakamura K, Koizumi Y. A case of subcutaneous metastasis of embryonal cell carcinoma of testicle. *Jpn J Urol.* 1961;52:682–686.