Rare Metastatic Testicular Cancer in a Veteran

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Key Words

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Endometrioid variant yolk sac tumor

Abstract

Testicular yolk sac tumor (YST) is a nonseminomatous germ cell tumor that predominantly affects prepubescent boys. Pure endometrioid variant YST is rare, with only 1 report in the literature. We present the first reported case of endometrioid variant YST with mature teratoma in the retroperitoneal specimen.

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Introduction

Testicular neoplasms account for only 1–2% of cancers in men, commonly between the ages of 20 and 40 [1]. The incidence of nonseminomatous germ cell tumor (NSGCT) rose by 24% over 2 decades and currently represents approximately 40% of men with germ cell tumor (GCT) [2]. The first testicular yolk sac tumor (YST), a histologic subtype of NSGCT, was identified in 1910 in a 15-month-old boy [3]. However, it was not classified as such until the 1940's when Telium's detailed observations led to further categorization [4]. Pure YST rarely occurs

in adults, occurring more commonly as a component of mixed GCT [1]. Furthermore, it can demonstrate various histologic patterns in adult males, including microcystic, endodermal and sarcomatoid among others. We present the first reported case of pure endometrioid variant YST with mature teratoma in the retroperitoneal specimen.

Case Report

A 32-year-old man presented with a 2-day history of acute onset of left testicular pain. He had no lower urinary tract symptoms, hematuria, or other prior urologic history. He was subjectively healthy and was on no prescription medications. Examination revealed a tender mass at the superior pole of the left testicle with associated skin erythema. Urinalysis was negative for hematuria or pyuria. Scrotal ultrasonography revealed a 3.4 × 2.4 cm mass with heterogeneous internal echotexture, punctate calcifications and several cystic areas. Tumor markers revealed an alpha-fetoprotein (AFP) of 21.6 ng/ml (normal: < 9 ng/ml), lactic dehydrogenase of 210 IU/l (normal: 100–190 IU/l), and beta-human chorionic gonadotropin < 0.6 mIU/ml (normal: < 5 mIU/ml). Staging computed tomography (CT) revealed a heterogeneous partially calcified para-aortic mass, measuring 4.0 × 4.9 cm (fig. 1).

Pathology after a left radical inguinal orchiectomy revealed endometrioid variant of YST with extensive necrosis, hemorrhage, and granulomatous reaction (fig. 2). The tumor cells stained positive for AFP and SALL4 supporting the diagnosis of endometrioid variant of YST. The patient's tumor markers normalized post-operatively, resulting in American Joint Committee on Cancer stage pT1N2M0S0 with an International Germ Cell Consensus Classification stage IIB. He was referred to medical oncology and received 4 cycles of neoadjuvant cisplatin and etoposide without complication. Repeat imaging with CT revealed a stable retroperi-

Fig. 1. Well demarcated heterogenous soft tissue mass with scattered calcification, without evidence of local extra-lesional infiltration or fat disruption.

toneal mass. Pathology from his post-chemotherapy retroperitoneal lymph node dissection (RPLND) revealed no evidence of germ cell malignancy in 31 lymph nodes. However, the retroperitoneal mass revealed nodular proliferation with cartilage and foci of calcification in adipose tissue suggestive of mature teratoma. He had an uneventful recovery and is now on a surveillance protocol per the National Comprehensive Cancer Network guidelines.

Discussion

Whereas the majority of YST present as an asymptomatic mass affecting boys under age 16 months [6], it is rare after the age of 5 years, occurring at a rate of 6%. When seen in adults, it is almost always part of a mixed NSGCT [1]. As in our case, AFP is elevated in 40–60% of advanced NSGCT [5]. In adult pure YST, retroperitoneal lymph node metastasis occurs in 33% of patients [7].

Endometrioid variant ovarian YST was first described by Clement et al. [8] in 1987. It is typically well circumscribed with multiple cystic and hemorrhagic areas with tubulopapillary glands lined by columnar cells with prominent subnuclear vacuolization, atypical nuclei and abundant mitoses. This pattern must be distinguished from endometrioid adenocarcinoma, as the majority of the latter are malignant [9]. Similarly, the endometrioid

Fig. 2. Endometrioid variant of YST. Columnar cells with cytoplasmic vacuoles, atypical vesicular or hyperchromatic nuclei, with conspicuous nucleoli (Hematoxylin & Eosin stain, magnification 40 ×).

variant of testicular YST can be mistaken for glandular elements of a mature teratoma, which is always considered malignant [10]. As of 2015, there was one reported case of pure endometrioid variant testicular YST [9]. Two other cases have been described: one as part of a mixed GCT and the second case with retroperitoneal metastasis involving teratoma and embryonal carcinoma [10].

To our knowledge, our patient is the first in the literature to have an endometrioid variant of YST with residual mature teratoma in the RPLND specimen. Based on National Comprehensive Cancer Network guidelines, testicular cancer patients should be treated based on their stage and risk category. Patients with stage IIB NSGCT should be managed with either primary RPLND or 3 cycles of bleomycin, etoposide and cisplatin followed by post-chemotherapy RPLND. Follow-up includes quarterly tumor markers, chest imaging and CT semi-annually for the first 2 years and annually thereafter. Overall survival rates for stage IIB NSGCT are 75–80% [1]. Given the paucity of endometrioid variant YST, information is lacking regarding individualized treatment and prognosis.

References

- 1 Stephenson AJ, Gilligan TD: Neoplasm of the testis; in: Wein AJ, Kavoussi LR, Novick AC, Partin AW, Peters CA (eds): Campbell-Walsh urology, ed 11. Elsevier Saunders, Philadelphia, 2015, pp784–814.
- 2 McGlynn KA, Devesa SS, Graubard BI, Castle PE: Increasing incidence of testicular germ cell tumors among black men in the United States. *J Clin Oncol* 2005;23:5757–5761.
- 3 White CP: A case of carcinoma myxomatodes of the testis occurring in infancy. *J Pathol Bacteriol* 1910;14:522–544.
- 4 Young RH, Teilum D, Talerman A: History of gynecological pathology XXV. Dr. Gunnar Teilum. *Int J Gynecol Pathol* 2013;32:520–527.
- 5 Gilligan TD, Seidenfeld J, Basch EM, Einhorn LH, Fancher T, Smith DC, Stephenson AJ, Vaughn DJ, Cosby R, Hayes DF: American Society of Clinical Oncology Clinical Practice Guideline on uses of serum tumor markers in adult males with germ cell tumors. *J Clin Oncol* 2010;28:3388–3404.
- 6 Woodward PJ, Mostofi FK, Talerman A, Heidenreich A, Hailemariam SKaplan GW: Germ cell tumors; in: Eble JN, Sauter G, Epstein JI, Sesterhenn IA (eds): Pathology and Genetics of Tumours of the Urinary System and Male Genital Organs. WHO Classification of Tumours. Lyon, France, International Agency for Research on Cancer (IARC) Press, 2004, pp221–249.
- 7 Foster RS, Hermans B, Bihrlé R, Donohue JP: Clinical stage I pure yolk sac tumor of the testis in adults has different clinical behavior than juvenile yolk sac tumor. *J Urol* 2000;164:1943–1944.
- 8 Clement PB, Young RH, Scully RE: Endometrioid-like variant of ovarian yolk sac tumor. A clinicopathological analysis of eight cases. *Am J Surg Pathol* 1987;11:767–778.
- 9 Chander A, Otis CN, Cole LL: Pure glandular endometrioid-like yolk sac tumor of the testis. *Int J Surg Pathol* 2015;23:30–31.
- 10 Hazarika P: Endometrioid like yolk sac tumor of the testis with small teratomatous foci: a case report and review of the literature. *Indian J Pathol Microbiol* 2015;58:557–559.