Urology Case Reports 54 (2024) 102725

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Oncology

A unique case of large cell calcifying type sertoli tumor

Matthew DeSanto *, Samuel Deem

Charleston Area Medical Center, Charleston, WV, USA

ARTICLE INFO

Keywords:
Sertoli cell
Testicular cancer
Orchiectomy

ABSTRACT

Sertoli cells tumors are a rare testicular malignancy, with prevalence estimated to be less than 1% of testicular cancers. Furthermore, testicular cancer only accounts for approximately 0.5% of all diagnosed cancers. This is a unique case of a 19-year-old male where physical exam and ultrasonography findings revealed an intratesticular mass that, after pathologic analysis, demonstrated T1 large cell calcifying type Sertoli cell tumor which is infrequently documented in the literature.

1. Introduction

Testicular cancer is a somewhat rare genitourinary malignancy and makes up approximately 0.5% of all diagnosed cancers.[1] Once diagnosed, gold standard treatment is orchiectomy with ensuing treatment based on tumor staging and pathology review. Testicular malignancy treatment and outcomes vary greatly depending on if the mass is pathologically classified as germ cell tumors (GCT) or non germ cell tumors (NGCT). Germ cell tumors are more common and consist of seminomas, non-seminomas, and mixed germ cell tumors. On the other hand, non-germ cell tumors consist of sex cord stromal tumors, and other miscellaneous tumors such as leiomyomas or lymphomas. Sex cord stromal tumors includes tumors such as Leydig cell tumors, Sertoli cell tumors, granulosa cell tumors, thecomas, and gonadoblastomas.[2] NGCTs encompass approximately 5–10% of testicular malignancies.[3,4] Moreover, Sertoli cell tumors comprise only roughly 1 % of all.[5] Sertoli cell tumors can be classified into 4 main subgroups: large cell calcifying type, sclerosing, intratubular hyalinizing, and general sertoli cell tumor.[6] In this case report, we document a rare case of a 19-year-old male diagnosed with T1 large cell calcifying type Sertoli cell tumor that is infrequently documented in the literature.

2. Case report

This case reports details a 19-year-old otherwise healthy male who was diagnosed with T1 large cell calcifying type Sertoli cell tumor. The patient had no past medical history and presented to a local emergency department for persistent cough and fever. The patient was diagnosed with Covid-19 at this time. However, nearing the end of the visit, the patient also endorsed vague testicular pain localized to the right side of five days duration. Examination revealed a nodular, firm intratesticular mass. Exam demonstrated no sign of feminization including testicular atrophy or gynecomastia. Scrotal ultrasound revealed an irregular right intratesticular 1.8 × 1.5 × 1.6 cm calcified, hyperechoic mass with shadowing and a 1 cm branch extending from the main lesion (see Fig. 1). Physical examination and scrotal ultrasonography revealed no abnormalities of the L testicle or scrotal wall. Pre-operative levels serum tumor markers were drawn and within normal limits as such: AFP 1.1, B-HCG 0.6, and LDH 145. Computed tomography (CT) of the abdomen pelvis did not reveal any lymphadenopathy or metastatic lesions.

The patient was promptly scheduled for right radical orchiectomy. Pathological analysis revealed the right testicle and cord with a mass consistent with sertoli cell tumor, large cell calcifying type, limited to the testis. The decision was made within the multidisciplinary team that the patient would not need further treatment given favorable pathology and characteristics of the tumor invasion.

3. Discussion

This case is quite notable as the diagnosis of testicular cancer was incidentally found. Additionally, final pathological analyses revealed a rather uncommon form of testicular cancer being consistent with sertoli cell tumor, specifically pertaining to large cell calcifying type sertoli cell tumor. Several clinically relevant points can be ascertained from this case.

First, the presentation of this case alludes to the need for routine self-administered testicular examinations. As mentioned previously, young males remain the highest risk age group for development of testicular

* Corresponding author. 3401 Noyes Ave, Charleston, WV, 25304, USA.
E-mail address: mdesanto@neomed.edu (M. DeSanto).

https://doi.org/10.1016/j.eucr.2024.102725
Received 27 October 2023; Received in revised form 20 March 2024; Accepted 23 March 2024
Available online 27 March 2024

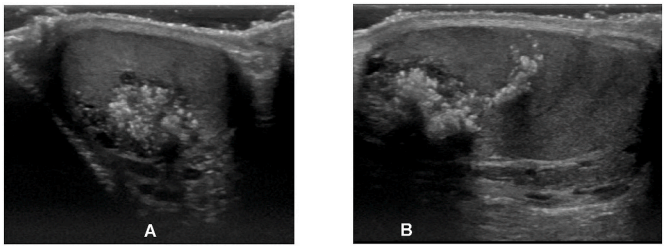

Fig. 1. Ultrasonography of the right testicle with intratesticular mass, (A) superior to inferior, (B) lateral to medial views.

cancer. It could be theorized that if the patient had not presented for care of other concurrent medical issues that diagnosis would have been delayed. It can be reasoned that there should be some sort of formal education for young males on how to properly do a self-assessment of the testicles, along with how often.

Second, large cell calcifying type sertoli cell tumor is not a commonly seen diagnosis in the clinical setting. There are only roughly 100 documented cases of large cell calcifying sertoli cell tumor in the literature.[4] The mean age of sertoli cell tumor diagnosis is 52 months of age; large cell calcifying type sertoli cell tumors are typically identified in prepubertal children and adolescents.[6] Malignancy rate for sertoli cell tumors is approximately 17%–25%, but certain smaller series have reported even lower rates.[7] Although typically benign, there can be clinical associations with Carney's syndrome as well as Peutz-Jeghers syndrome.[4,8] There have also been a few cases reported of adrenocortical hyperplasia as well as cardiac myxoma with sertoli cell tumors, however, these are rare occurrences.[9,10]

Further characterizing oncologic risk of sertoli cell tumors relates to high-risk unfavorable features. These include mass $>$ 5cm in size, vascular invasion, high mitotic activity, tumor necrosis and cellular atypia.[6] Pathology was favorable as the mass was contained to the testis and no high-risk tumor characteristics were identified. The mass was $<$5 cm, without vascular invasion, increased mitotic activity, or necrosis noted. In these scenarios, treatment via orchiectomy is typically deemed to be sufficient.[6] In cases of high risk features in children older than 5 years of age, full staging workup is suggested followed by RPLND if retroperitoneal metastatic disease is uncovered.[6] In the absence of these features, orchiectomy is substantial as a singular therapy. Furthermore, approximately 10% of these tumors can be hormonally active leading to virilization, feminization, or precocious puberty.[6] These features were not evident in this patient.

Discussion of treatment in a multidisciplinary setting is essential for cases such as this to provide the most appropriate care for the patient. There is a paucity of data on metastatic potential of sertoli cell tumors and minimal analysis of oncologic outcomes based on subtype. Often, sertoli cell tumors are characterized as benign entities.[11] Given this, guideline-driven treatment algorithms are lacking for large cell calcifying type sertoli cell tumor. A previous systematic review has demonstrated that 50 of 435 patients with any subtype of sertoli cell tumor developed metastatic disease, and 20 of these 50 were diagnosed at staging workup.[4] Yet, there are no concrete guidelines concerning sertoli cell tumor follow-up and treatment post-orchiectomy. Given our clinical review, the most appropriate action deemed was serial exams to the contralateral testis plus follow up imaging via CT abdomen pelvis for at least two years accompanied with genetic testing.

4. Conclusion

Sertoli cell tumors compose only about 1% of all testicular malignancies, and even fewer are classified as large cell calcifying type. Although not clinically common, it is imperative to have early multidisciplinary team involvement for prompt treatment, with the mainstay of orchiectomy, to attain the most successful patient outcomes.

CRediT authorship contribution statement

Matthew DeSanto: Conceptualization, Investigation, Project administration, Writing – original draft, Writing – review & editing. **Samuel Deem:** Conceptualization, Investigation, Methodology, Project administration, Supervision, Writing – review & editing.

Declaration of competing interest

None.

References

1. C National Cancer Institute. *Cancer Stat Facts: Testicular Cancer*. vol. 12. 2021. January https://seer.cancer.gov/statfacts/html/testis.html. Accessed February 15, 2021.
2. Brekelbaum CE, Abreo F, Fowler M, Kubricht WS, Eastham JA. Undifferentiated sex cord/stromal testis tumor. *Urology*. Mar 2000;55(3):436. https://doi.org/10.1016/s0090-4295(99)00482-3.
3. Dilworth JP, Farrow GM, Oesterling JE. Non-germ cell tumors of testis. *Urology*. May 1991;37(5):399–417. https://doi.org/10.1016/0090-4295(91)80100-l.
4. Grogg J, Schneider K, Bode PK, et al. Sertoli cell tumors of the Testes: systematic literature review and meta-analysis of outcomes in 435 patients. *Oncol*. Jul 2020;25(7):585–590. https://doi.org/10.1634/theoncologist.2019-0692.
5. Idrees MT, Ulbright TM, Oliva E, et al. The world health organization 2016 classification of testicular non-germ cell tumours: a review and update from the international society of urological pathology testis consultation panel. *Histopathology*. Mar 2017;70(4):513–521. https://doi.org/10.1111/his.13115.
6. Ferrer F. In: Wein A, Kavoussi L, Campbell M, Urology Campbell-Walsh, eds. *Pediatric Urology Oncology: Bladder and Testis*. twelfth ed. Elsevier Saunders; 2020: 5014–5025.
7. Kumar H, Tanveer N, Gupta N, Mishra K. Large cell calcifying sertoli cell tumour of testis-A rare case report. *J Clin Diagn Res*. Nov 2016;10(11):Ed03–ed05. https://doi.org/10.7860/jcdr/2016/22075.8842.
8. Alikasifoglu A, Gonc EN, Akcoren Z, et al. Feminizing Sertoli cell tumor associated with Peutz-Jeghers syndrome. *J Pediatr Endocrinol Metab*. Apr 2002;15(4):449–452. https://doi.org/10.1515/jpem.2002.15.4.449.
9. Coleman S, Barazani Y, Roma AA, Sabanegh E Jr. Testicular sclerosing sertoli cell tumor: a case report and review of the literature. *Rev Urol*. 2014;16(4):191–193.
10. Martinez-Aguayo A, Rocha A, Rojas N, et al. Testicular adrenal rest tumors and Leydig and Sertoli cell function in boys with classical congenital adrenal hyperplasia. *J Clin Endocrinol Metab*. Dec 2007;92(12):4583–4589. https://doi.org/10.1210/jc.2007-0383.
11. Gomha FS, Bhatti KH, Yousif AA, et al. The sclerosing Sertoli cell tumor of the testis is an extremely rare entity. *Ther Adv Urol. © The Author(s)*. 2020;2020, 1756287220960295.