(Material)

A case of spermatocytic tumor in the testis

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ABSTRACT A male in his 50s presented to his urologist with a mass in his right testis. Contrast-enhanced magnetic resonance imaging revealed a round testicular mass approximately 23 mm in diameter with homogeneous contrast enhancement and diffusion restriction. Blood tests showed human chorionic gonadotropin < 2.3 mIU/mL, alpha-fetoprotein 3.5 ng/mL (normal range, 10-20 ng/mL), and lactate dehydrogenase 157 U/L (normal range, 120-220 U/L). A high orchiectomy was performed. A well-demarcated, white-colored mass was observed on gross examination of the testis. Histologically, the tumor exhibited three types of round cells of varying sizes, and the nuclear chromatin of intermediate and giant cells displayed granular or filamentous patterns. Mitotic figures were scattered, with minimal lymphocytic infiltrates. The presence of germ cell neoplasia in situ (GCNIS) remained uncertain. Immunohistochemistry revealed SALL4 positivity, partial weak positivity for c-KIT, but negativity for OCT-3/4 and D2-40 in tumor cells. Consequently, a diagnosis of spermatocytic tumor (ST) was made. ST is classified by the World Health Organization as one of the GCNIS-unrelated germ cell tumors and is rare, accounting for approximately 0.6-1.0% of testicular germ cell tumors. Although microscopically similar to seminomas, STs, and seminomas differ in clinicopathologic features. Accurate diagnosis relies on recognizing this rare entity, careful observation of hematoxylineosin staining and the adjunctive use of immunohistochemistry.

doi:10.11482/KMJ-E202551013 (Accepted on January 11, 2025)

Key words : Spermatocytic tumor, Testis, Pathology, Immunohistochemistry

INTRODUCTION

Spermatocytic tumor (ST) is classified as one of the germ cell tumors (GCTs) and is not related to germ cell neoplasia in situ (GCNIS) in the WHO classification¹⁾. STs are rare, accounting for approximately 0.6-1.0% of testicular germ cell tumors^{2. 3)}, and their incidence has been reported to be approximately 52-56 years of age⁴⁾. We present a case of testicular ST in which immunostaining was useful for diagnosis.

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Fig. 1. Contrast-enhanced MRI (T1) revealed a round testicular mass approximately 23 mm in diameter with homogeneous enhancement and diffusion restriction.

CASE REPORT

The patient was a 59-year-old man who presented with a stony, hard mass in his right testis. The patient noticed the lesion incidentally, and it was unclear how long the mass had been present. Blood tests showed human chorionic gonadotropin < 2.3 mIU/mL, alpha-fetoprotein 3.5 ng/mL (normal range, 10-20 ng/mL), and lactate dehydrogenase 157 U/L (normal range, 120-220 U/L). Contrastenhanced MRI (T1) revealed a round testicular mass approximately 23 mm in diameter with homogeneous enhancement and diffusion restriction (Fig. 1). A high orchiectomy was performed for this lesion.

On gross examination, a well-defined whitecolored mass was observed in the right testis (Fig. 2). Histologically, the tumor exhibited three types of round cells of varying sizes, with the nuclear chromatin of intermediate and giant cells displaying granular or filamentous patterns on hematoxylin-cosin staining (Fig. 3). Mitotic figures were scattered but not prominent, with minimal



Fig. 2. Well-defined white-colored mass in the right testis



Fig. 3. Tumor showing three types of round cells of different sizes (hematoxylin-eosin staining)

lymphocytic infiltrates. The presence of GCNIS remained uncertain. Atypical cells and necrosis were not observed. Immunohistochemistry revealed SALL4 positivity, partial weak positivity for c-KIT, but negativity for OCT-3/4 and D2-40 in tumor cells (Fig. 4). Based on these findings, a diagnosis of ST was made.

DISCUSSION

Testicular tumors are classified as GCTs and sexcord-stromal tumors. GCTs include seminoma



Fig. 4. (a-c) Immunohistochemistry showing SALL4 (a) positivity but negativity for OCT-3/4, (b) and D2-40, (c) in tumor cells

and non-seminoma histologies^{1, 4)}. The latter encompasses teratoma (postpubertal type), embryonal carcinoma, choriocarcinoma, yolk sac tumor, and mixed forms of these components. ST is a rare GCT derived from postpubertal type germ cells and was previously referred to as spermatocytic seminoma⁴⁾. ST is an exceptionally rare disease, accounting for approximately 0.6-1.0% of testicular cancers, and it exhibits distinct clinicopathologic features compared to other GCTs, such as seminoma $^{1-4)}$. Despite its rarity, the microscopic appearance of spermatocytic tumor can resemble that of a seminoma. The main histopathologic distinctions are as follows⁴): STs consist of three types of cells, including small cells (6-8 microns, with sparse cytoplasm), intermediate cells (15-20 microns, with round nuclei and chromatin ranging from granular to spireme-like), and giant cells (50-100 microns, with multiple nuclei). Unlike other GCTs, STs lack GCNIS and significant inflammatory infiltrate, including lymphocytes. In addition, immunohistochemical examination reveals negativity for several germ cell tumor markers, such as OCT-3/4, D2-40, and PLAP in STs.

The present case showed SALL4 positivity. SALL4 is a stem cell marker, which plays an essential role in maintaining the self-renewal and pluripotent properties of embryonic stem cells⁵⁾. The previous studies have demonstrated that SALL4 is expressed specifically in primitive germ cell tumors, including ST¹⁾. Furthermore, STs do not exhibit the expression of isochromosome 12p, which is commonly detected in seminomas, Although genetic analysis was not performed, the current patient displayed the aforementioned characteristic features of STs.

Most STs are diagnosed early, and orchiectomy alone is typically associated with a good prognosis $^{4)}$. However, two variants, anaplastic STs and STs with sarcomatous transformation, are resistant to systemic therapy and related to poor prognosis⁴⁾. The histopathologic features of anaplastic STs include increased mitotic count (> 30/10 HPF), prominent nucleoli, vesicular nuclei, bizarre giant cells, areas of necrosis, and abnormal mitotic figures⁴⁾. The sarcomatous component of STs with sarcomatous transformation is usually an undifferentiated spindle cell sarcoma⁴⁾. Our patient did not exhibit any of these features suggestive of aggressive behavior. The pathogenesis of sarcoma transformation in ST remains unclear. However, certain investigators have proposed the possibility of anaplastic transformation or dedifferentiation from a well-differentiated $ST^{4)}$.

CONCLUSION

We have described a patient with ST. Accurate diagnosis relies on recognizing this specific entity, meticulous observation of hematoxylineosin staining, and the adjunctive use of immunohistochemistry.

REFERENCES

- WHO Classification of Tumours Editorial Board: Urinary and Male Genital Tumours. Lyon (France), International Agency for Research on Cancer. 2022. (WHO classification of tumours series, 5th ed.; vol.8). https://publications.iarc.fr/610.
- 2) Carrière P, Baade P, Fritschi L: Population based incidence and age distribution of spermatocytic seminoma. J Urol. 2007; 178 (1): 125-128.
- 3) Ghazarian AA, Trabert B, Devesa SS, McGlynn KA:

Recent trends in the incidence of testicular germ cell tumors in the United States. Andrology. 2015; 3 (1): 13-18.

- 4) Secondino S, Viglio A, Neri G, Galli G, Faverio C, Mascaro F, Naspro R, Rosti G, Pedrazzoli P: Spermatocytic tumor: A review. Int J Mol Sci. 2023; 24 (11): 9529.
- 5) Ushiku T, Shinozaki A, Shibahara J, Iwasaki Y, Tateishi Y, Funata N, Fukayama M: SALL4 represents fetal gut differentiation of gastric cancer, and is diagnostically useful in distinguishing hepatoid gastric carcinoma from hepatocellular carcinoma. Am J Surg Pathol. 2010; 34; 533-540.