

Oncology

Extramedullary plasmacytoma of the testis: A case report

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1. Introduction

Testicular cancer accounts for nearly 1% of all malignant disease in male patients. The annual incidence of testicular cancer in Japan ranges from approximately 0.7 to 1.8/100,000 patients. A total of 90%–95% of testicular cancers are categorized as germ cell tumors, including seminoma and non-seminoma. Other non-germ cell tumors account for <10%. Extramedullary plasmacytoma (EMP) is reported to account for only 0.03%–0.15% of all cases of testicular cancer.¹

EMP is a solitary mass of neoplastic plasma cells occurring in soft tissue without bone marrow. EMP represents only 5% of all plasma cell neoplasms,² and most commonly occurs in the head and neck region. We herein report a rare case of EMP of the testis.

2. Case presentation

A 79-year-old Asian (Japanese) patient was referred to our

hospital to undergo further examination for a right testicular tumor. The patient had glaucoma, hyperlipidemia, and benign prostate hyperplasia. The only remarkable physical examination finding was a right testicular mass that was elastic and hard, measuring 5 cm in diameter. The laboratory findings, including several tumor markers (HCG, <0.1 mIU/mL; CEA, 3.3 ng/mL; AFP, 3 ng/mL; CA19-9, 11 U/mL; CA125, 17 U/mL; SCC, 0.8 U/mL; NSE, 11.0 U/mL; sIL-2R, 448.4 U/ml), were normal. CT revealed right testicular mass of 5.4 cm in diameter, without invasion or metastasis [Fig. 1] (see Fig. 2).

Right radical orchiectomy was performed based on the clinical diagnosis of testicular cancer. The resected elastic firm specimen was 6.8 × 4.3 × 5.3 cm. The resected specimen showed an enlarged testis occupied by a white solid tumor. No normal testis was observed. Histologically, the tumor obliterated the underlying testicular parenchyma and there was intertubular growth of tumor cells in peripherally. Neoplastic cells showed plasmacytoid appearance of eosinophilic cytoplasm and vesicular nuclei with central nucleoli (Fig. 3a). Immunohistochemically, tumor cells were positive for λ light chain (Fig. 3b) and negative for κ light chain (Fig. 3c). Amyloid sedimented on vascular walls and basement membrane of testicular tubules. Amyloid deposits were Congo red-positive (Fig. 3d). Based on these findings, monoclonal plasma cells grew in neoplastic and immunoglobulin light chain had deviation. The tumor was therefore diagnosed as testicular plasmacytoma.

Based on the diagnosis of testicular plasmacytoma, blood and urinary protein analyses, PET-CT, and bone marrow aspiration were performed. M protein was not detected and PET-CT revealed no irregular findings. Thus, the final diagnosis was plasmacytoma arising from the testis. The patient is currently free from recurrence at four months after undergoing radical orchiectomy, without any additional systemic chemotherapy. PET-CT showed no metastasis. The serum immunoglobulin findings were as follows: IgG, 1048 mg/dL; IgA, 165 mg/dL; IgM, 33 mg/dL; IgD, <0.6 mg/dL; and IgE, 258 mg/dL. A serum protein sub-fraction analysis revealed the following findings: Alb, 63.2%; α1-glob, 3.7%; α2-glob, 8.1%; β1-glob, 5.6%; β2-glob, 4.5%; and γ-glob, 14.9%. M protein was not detected in the patient's urine or serum.

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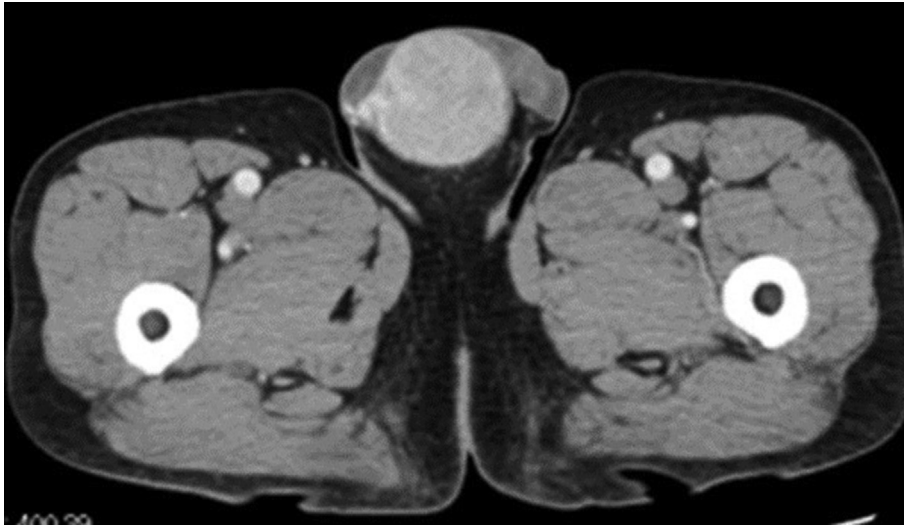


Fig. 1. Contrast-enhanced computed tomography revealed a right testicular tumor of 5.4 cm diameter with heterogeneous enhancement. No lymph-node or distant metastasis was observed.

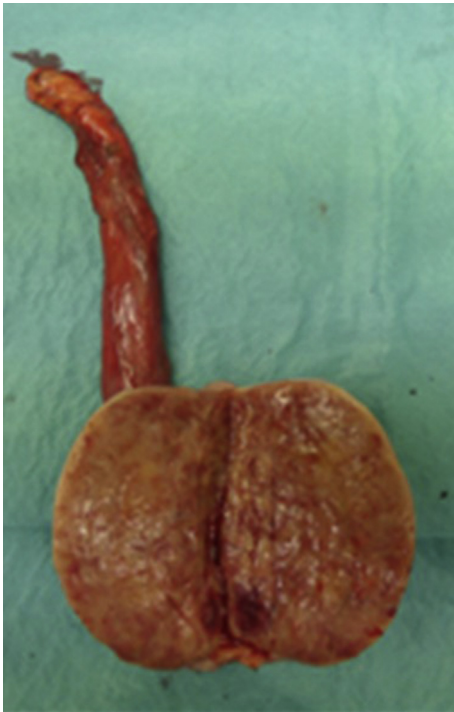


Fig. 2. The resected specimen was 6.8 × 4.3 × 5.3 cm in size, with a yellowish-white color; no normal testis was observed.

3. Discussion

Plasmacytoma is a monoclonal proliferation of plasma cells. Intra-bone marrow plasmacytoma is referred to as multiple myeloma, while bone or extra-bone marrow plasmacytoma is referred to as extramedullary plasmacytoma; these account for <5% of all plasma cell neoplasms.² The incidence in males is slightly

higher than that in females. Most cases involve individuals of 40–60 years of age.

A total of 80% of extra-bone marrow plasmacytoid tumors are located in the upper respiratory tract, followed by the lymphoid organs, gastric organs, colon and liver. Cases involving the skin and central nervous system are rare. Plasmacytoma of the testis is extremely rare, and accounts for only 0.03%–0.10% of all testicular tumors.³

Plasmacytoma of the testis is asymptomatic. There are no typical findings of extramedullary plasmacytoma on CT or MRI. US shows a heterogeneous low-echoic lesion with flow; however, the flow is reported to be low in comparison to other testicular tumors.⁴

The diagnosis of extramedullary plasmacytoma was based on the following criteria: 1) the presence of extramedullary monoclonal plasma cells, 2) the absence of serum or urinary M protein, 3) normal bone marrow, 4) no bone or organ invasion, 5) typical hematoxylin-eosin staining, and 6) monoclonal differentiation of immunoglobulin.

In cases without metastasis, extramedullary plasmacytoma shows a relatively favorable outcome, with a mean survival time of 8.3 years.⁵ However, plasmacytoma of the testis is extremely rare and the prognosis is still unknown. Some bone plasmacytoma cases are reported to progress to multiple myeloma; such cases show a poorer prognosis, the same as multiple myeloma.

Plasmacytoma of the testis is usually treated by radical orchiectomy; in cases involving advanced-stage plasmacytoma or recurrence, radiation therapy is usually added.³ In cases involving multiple tumor lesions, systemic chemotherapy is usually selected. Extramedullary plasmacytoma has higher sensitivity and is sometimes cured by radiation monotherapy.²

Our case was diagnosed based on the pathological findings after higher orchiectomy. No additional therapy was added because M protein was not detected in the bone marrow and because there were no findings in the other organs. A previous study reported on progression to multiple myeloma and recurrence; thus, continuous follow-up is required. The patient is currently free from recurrence at four months after surgery.

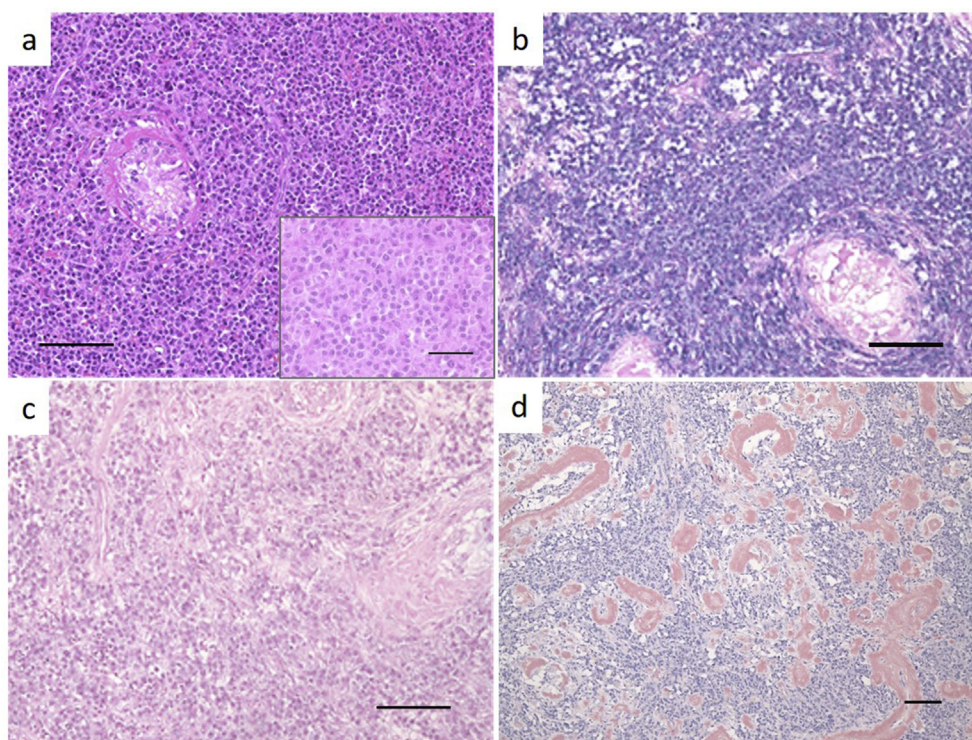


Fig. 3. a) Histological feature of the tumor (Hematoxylin and eosin staining) (bar = 100 μ m). Inset: Tumor cells showed plasmacytoid feature (bar = 50 μ m). b, c) Immunohistochemically, almost all tumor cells were positive for λ light chain (b), and negative for κ light chain (c). (bar = 100 μ m). d) Amyloid deposits were Congo red-positive (bar = 100 μ m).

4. Conclusion

We herein reported a rare case of plasmacytoma of the testis.

Abbreviations

Not applicable.

Declarations

Ethics approval and consent to participate & consent for publication.

The present study was approved by the IRB of Yokohama City University Medical Center.

Consent for publication

Written informed consent was obtained from the patient's legal guardian for the publication of this case report and any accompanying images. A copy of the written consent form is available for review by the Editor-in-Chief of this journal.

Availability of data and material

Due to ethical restrictions, the raw data underlying this paper is available upon request from the corresponding author.

Conflicts of interest

The authors declare no conflicts of interest in association with the present study.

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Author contributions

Conceived and designed the experiments: KS, TK, DT. Analyzed the data: KS, TK, SC, MY, HU. Performed the experiments: KS, TK, SC. Wrote the paper: KS, TK.

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References

1. Kojima T, Kawai K, Tsuchiya K, et al. Identification of a subgroup with worse prognosis among patients with poor-risk testicular germ cell tumor. *Int J Urol*. 2015;22:923.
2. Galièni P, Cavo M, Pulsoni A, et al. Clinical outcome of extramedullary plasmacytoma. *Haematologica*. 2000;85:47.
3. Khan M, Rajarubendra N, Azer S, et al. Plasmacytoma of the testis in a patient with relapsed and refractory multiple myeloma: case report and review of the literature. *Urol Ann*. 2015;7:530.
4. Bortolotto C, Ori-Belometti G, Rodolico G, et al. Plasmacytoma of the testis in a patient with previous multiple myeloma: sonographic appearance. *J Ultrasound*. 2016;19:153.
5. Fernandez LA, Couban S, Sy R, et al. An unusual presentation of extramedullary plasmacytoma occurring sequentially in the testis, subcutaneous tissue, and heart. *Am J Hematol*. 2001;67:194.