Bilateral synchronous plasmacytoma of the testis

Geetha Narayanan, MD, DM, Rona Joseph, MD, and Lali V. Soman, MBBS

Extramedullary plasmacytoma (EMP) is usually seen in the head and neck regions and in the upper respiratory, gastrointestinal, and central nervous systems. Testis is a rare site for EMP, and bilateral synchronous testicular plasmacytoma occurring as an isolated event at initial presentation has been reported only once previously. We present herein the second such report in a 70-year-old man who underwent bilateral orchidectomy.

Plasmacytoma is a discrete solitary mass of neoplastic monoclonal plasma cells occurring either in bone (solitary plasmacytoma of bone) or in soft tissues (extramedullary plasmacytoma, EMP). EMP represents 3% to 5% of all plasma cell neoplasms. The common sites of EMP are the head and neck, upper respiratory system, gastrointestinal system, and central nervous system (1). Testis is a rare site for EMP, and EMP accounts for only 0.03% to 0.15% of all testicular tumors (2–4). Testicular plasmacytoma usually occurs as part of multiple myeloma or as a site of recurrence. Rarely, isolated testicular plasmacytomas occur as a presenting feature, with a few cases reported (5–7). Even more rare is bilateral and synchronous testicular plasmacytoma, with only four cases reported in the literature, and in only one case it occurred as an isolated event at the initial presentation (8–11). We present the second report of isolated bilateral synchronous testicular plasmacytoma as the presenting feature in a 70-year-old man.

CASE REPORT

A 70-year-old man was evaluated for pain and swelling of the right testis of 6 months' duration. Right inguinal hernia and bilateral hydrocele were initially diagnosed. He underwent right hernioplasty and hydrocelectomy and eversion of the left tunica vaginalis sac. Swelling recurred after 4 months. Doppler ultrasound of the testis showed a grossly enlarged right testis (76 × 43 × 67 mm) with multiple hypoechoic lesions, lobulations, and increased vascularity. He underwent right orchidectomy, and the testis showed a circumscribed grayish-white lobulated lesion, 7.5 × 6 cm, with no normal testicular tissue seen. The section showed immature and atypical plasma cells in sheets infiltrating the interstitial tissue and surrounding the atrophic tubules (*Figure 1a*). On immunohistochemistry, the cells were positive for CD138 and kappa light chain staining (*Figure 1b*)

and negative for CD20 and CD5 and lambda, suggestive of plasmacytoma. Two months later, the patient noticed swelling of his left testis. Ultrasound showed similar hypoechoic lesions, and he underwent left orchidectomy.

The patient's hemoglobin level was 13.1 g/dL; total white blood cell count, 10,200/mm³; and platelet count, 2.88 lakhs/mm³; his total protein and renal function were normal. His serum IgG was 1838 mg/dL; IgA, 212 mg/dL; IgM, 86 mg/dL; free kappa light chain, 33.5 mg/dL; and free lambda, 36.7 mg/dL. Serum electrophoresis and serum immunofixation did not show any abnormal bands. His bone marrow aspiration was normal, and there were no lytic lesions on skeletal survey.

DISCUSSION

EMP occurs in 18% of patients with multiple myeloma (12). Testicular involvement by plasmacytoma is rare but well documented, and most patients have myeloma at the time of diagnosis or develop plasmacytoma during relapse (4, 13–16). EMP occurs rarely as a solitary plasmacytoma at initial presentation in the absence of systemic myeloma (2, 4–6, 14, 15, 17). Anghel et al reported 52 cases of testicular plasmacytoma, and most had prior or concurrent multiple myeloma (4).

Synchronous cancers are defined as malignant tumors that present simultaneously or within a 6-month period of identification of the original tumor. Bilateral involvement of testes by plasmacytoma is unusual, and most cases have occurred asynchronously at the time of relapse. Two cases of myeloma in remission relapsing later with bilateral testicular plasmacytoma were reported (9, 10). A 43-year-old man with myeloma was detected to have both testes involved by a positron emission tomography/computed tomography scan done prior to allogeneic transplantation (11). However, there has been only one previous report of bilateral synchronous testicular plasmacytoma presenting without evidence of myeloma occurring in a 48-year-old man (8). Our patient also presented initially with

From the Department of Medical Oncology, Regional Cancer Centre, Trivandrum, India.

Corresponding author: Geetha Narayanan, MD, DM, Professor and Head, Department of Medical Oncology, Regional Cancer Centre, Trivandrum 695011, Kerala, India (e-mail: geenarayanan@yahoo.com).

Figure 1. Orchidectomy section showing (a) sheets of plasma cells infiltrating interstitial tissue (hematoxylin and eosin, 100×) and (b) immunopositivity for CD138 (100×).

isolated bilateral testicular plasmacytoma and did not have any evidence of myeloma.

The mean age at diagnosis of testicular plasmacytoma is 55 to 60 years old (5). Patients usually present with a firm testicular mass which may be tender. Sonographic imaging has shown the masses to be either homogeneous or heterogeneous and typically hypoechoic. Hypervascularity on color flow and power Doppler imaging is a consistent finding in testicular plasmacytomas (6, 13). On microscopy, the tumor shows sheets of plasma cells with varying degrees of differentiation. The diagnosis is confirmed by immunostaining with CD138, CD79a, and light chain restriction (5). Our patient also showed positivity for CD138. Testicular plasmacytomas have a higher incidence of IgA than multiple myelomas (15, 18). The diagnosis of isolated testicular plasmacytoma requires studies to exclude systemic myeloma.

Testicular plasmacytoma, like other primary testicular tumors, requires radical inguinal orchidectomy. Local radiation may be considered in patients with incomplete resection or recurrent or refractory disease (5). The overall prognosis for patients with testicular plasmacytoma is poor, with a high rate of progression to multiple myeloma (14). Hence, these patients require close monitoring. The survival of patients with multiple myeloma has improved over the years as a result of better understanding of the disease biology and advancement in targeted treatment modalities.

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