African Journal of Urology
1110-5704

Vol. 16, No. 3, 2010
88-92

Case Report

# Primary Plasmacytoma of The Testis with no Evidence of Multiple Myeloma: a New Case Report and Literature Review

**M. Bouassida[1], H. Ketata[1], A. Bahloul[1], S. Mseddi-Hdiji[2], S. Makni[3], J. Daoud[4], M. N. Mhiri[1]**

*Departments of Urology[1], Hematology[2], Histopathology[3] and Radiothorapy[4], Habib Bourguiba Hospital, Sfax, Tunisia*

## ABSTRACT

Plasmacytomas of the testis are extremely rare tumours, especially when occurring in the absence of a previous or concurrent diagnosis of multiple myeloma. We report a new case of solitary testicular plasmacytoma, with immunohistochemical studies showing monoclonal cytoplasmic production of IgG lambda light chains, in a 51-year-old man who had no evidence of multiple myeloma 3 years after the orchiectomy.

**Key Words:** Testis, plasmacytoma, multiple myeloma

**Corresponding Author:** Dr. Mehdi Bouassida, Department of Urology, Lapeyronie Hospital, 371, Av. Doyen Gaston Giraud. 34295 Montpellier cedex 5. France, Email: mehdi.bouassida@yahoo.fr

**Article Info :** Date received: 21/5/2010 Date accepted (after revision): 27/7/2010

## INTRODUCTION

The testis is an uncommon extramedullary site for plasmacytoma. Even more unusual are solitary testicular plasmacytomas, manifesting in the absence of a previous or concurrent diagnosis of multiple myeloma. To the best of our knowledge, 22 similar cases have been previously reported in the literature, of which 12 did not develop multiple myeloma during variable periods of follow-up (Table 1).

We report here the case of a solitary testicular plasmacytoma, with immunohistochemical studies showing monoclonal cytoplasmic production of IgG lambda light chains, in a 51-year-old man who had no evidence of multiple myeloma 3 years after the orchiectomy. This is the second case reported in the literature with IgG lambda in immunochemical studies. The case is discussed together with the data collected by a Medline-based review of the literature.

## CASE REPORT

A 51-year-old man presented with a 2-month history of painless right testicular enlargement. He had no past history of multiple myeloma or other lymphoproliferative disorder.

On examination, the right testis was hard and regularly enlarged. The tumor did not involve the epididymis or spermatic cord. Otherwise, the physical examination was unremarkable with no tenderness or inguinal lymphadenopathy.

**Table 1:** Immature plasma cells with eccentric nuclei and prominent nucleoli. H&E, reduced from x 100.

| Case | Author/Year | Side | Age | Serum M-protein | Treatment | Dissemination or progression to MM | Outcome |
|---|---|---|---|---|---|---|---|
| 1 | Anderson[12]/1949 | R | 72 | NA | Or + RT | NA | NA |
| 2 | Eckert and Smith[13]/1963 | R | 50 | NA | Or + RT | Yes / 1 y | DD / 1 y |
| 3 | Gowing[7]/1964 | R | 50 | NA | Or | Yes / 3 y | DD / 3 y |
| 4 | " | B | 50 | NA | Or | Yes / 1 y | DD / 1 y |
| 5 | Levin and Mostofi[14]/1970 | B | 46 | NA | Or | Yes | DD / 26 m |
| 6 | " | R | 48 | NA | Or | No | AND / 2 m |
| 7 | Oldham and Polmar[15]/1973 | L | 44 | Ig D λ | Or | No | AND / 3 y |
| 8 | Kaneshige et al[16]/1980 | L | 73 | Ig G κ | Or | No | AND / 1 y |
| 9 | Taylor et al[17]/1986 | L | 55 | IgA κ | Or | Yes / 6 y | DD / 7 y |
| 10 | Terzian et al[1]/1987 | R | 52 | IgA λ | Or | No | AND / 1 y |
| 11 | Oppenheim et al[5]/1991 | L | 71 | IgA λ | Or | No | AND / 9 m |
| 12 | Croft[8]/1992 | R | 74 | λ Chain | Or | No | AND / 1 y |
| 13 | White and Chan[2]/1995 | L | 72 | IgG κ | Or | No | AND / 41 m |
| 14 | Iizumi et al[9]/1995 | L | 83 | IgG λ | Or | No | AND / 14 m |
| 15 | Fischer et al [18]/1996 | L | 54 | IgA λ | Or | No | AND / 1 y |
| 16 | Ferry et al[19]/1997 | NA | 89 | NA | Or | Yes / 18 m | DD / 3 y |
| 17 | Reddi et al[20]/1998 | R | 62 | λ Chain | Or | No | AND / 16 m |
| 18 | Pham et al[21]/2000 | B | 48 | Λ Chain | Or | NA | NA |
| 19 | Suzuki et al[10]/2001 | R | 86 | IgG | Or | No | AND / 9 m |
| 20 | Hou et al[22]/2003 | R | 34 | K Chain | Or + CT | Yes / 3 y | AD / 7 y |
| 21 | Walker et al[23]/2005 | L | 67 | NA | Or | NA | NA |
| 22 | Chelly et al[24]/2007 | R | 65 | K Chain | OR + CT | No | AND / 6 m |
| 23 | Our case/2009 | R | 51 | IgG λ | Or + RT | No | AND / 30 m |

Ultrasonography showed a large, heterogeneous right testis with a hypoechoic and hypervascular lesion in the lower pole, suggestive of testicular tumor. Routine laboratory test results as well as serum testicular tumor markers including human chorionic gonadotropin (HCG) and α-foetoprotein (αFP) were normal.

Diagnosis was right testicular tumor and right radical orchiectomy was performed without complications. Macroscopically, the testis measured 8.5 x 5 x 4 cm. On cut section, there was a focally well-circumscribed grey-tan firm tumor measuring 5 x 4 x 4 cm in the lower pole of the testis. It extended to, but did not appear to penetrate the tunica albuginea. Histologic examination showed sheets of slightly atypical plasma cells, some with multiple nuclei (Fig. 1). Immunohistochemical study of the tumor specimens revealed cytoplasmic positivity for IgG and its lambda light chain. IgM, IgA and kappa chains were not identified, thus, confirming the monoclonal nature of this neoplasm (Fig .2).

Due to these findings, the patient underwent a diagnostic workup in search of systemic plasma cell disease. Serum protein electrophoresis performed 2

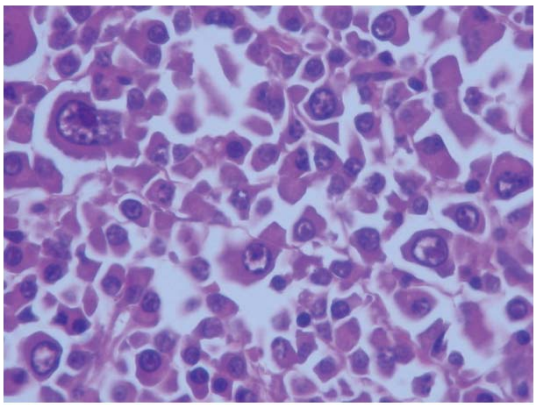

**Fig. 1:** Immature plasma cells with eccentric nuclei and prominent nucleoli. H&E, reduced from x 100.

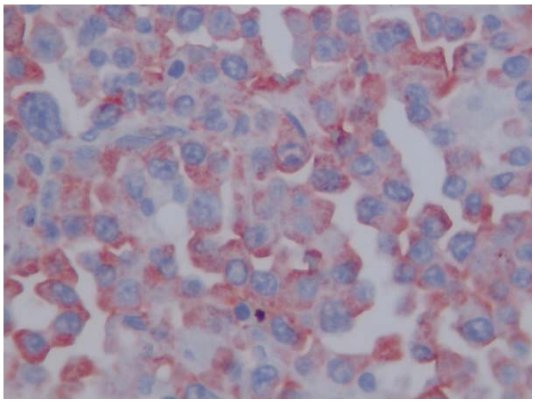

**Fig. 2:** Immunohistochemical stain demonstrates plasma cells strongly positive for lambda light chain. Reduced from x 100.

weeks postoperatively revealed a discreet gamma spike, which was characterized by immunoelectrophoresis as an IgG lambda protein. After one month, serum electrophoresis and immunoelectrophoresis were normal. Myelography and bone marrow biopsy showed no lesions suggestive of multiple myeloma. Computed tomography of the abdomen revealed no abnormal masses or lymphadenopathy. Serum and urinary proteins, creatinine and calcium were within the reference range.

The patient underwent external beam radiotherapy to the right scrotum. He tolerated this procedure without complications and remained well without any evidence of recurrence 3 years after the orchiectomy.

## DISCUSSION

Extramedullary plasmacytoma refers to plasma cell tumors arising in any organ outside the bone marrow. They most commonly involve the upper respiratory tract, lymph nodes, oral cavity and gastrointestinal tract[1,2]. Cases with proven testicular involvement are very rare. The majority of these lesions occur in the setting of multiple myeloma[3,4]. In 1991, Oppenheim et al[5] reported 37 cases of testicular plasmacytoma and only 6 developed in patients without previously or concurrently diagnosed multiple myeloma, testifying the extreme rarity of solitary plasmacytoma of the testis. In 1997, Castagna et al[6] compiled 57 cases, of which only 11 had no multiple myeloma during 2 months to 3 years of follow-up. In 2002, Anghel et al[3] compiled 51 cases, of which 17 were solitary plasmacytomas and only 7 of these did not develop multiple myeloma during follow-up.

With a Medline based review of the literature we collected 22 cases of solitary testicular plasmacytoma (Table 1). The mean patient age was 60 years (range 34-89 years). Ten were on the right side, 8 on the left one and 3 were bilateral (side not available in one case). Only 12 cases did not progress to multiple myeloma during 2 to 41 months of follow-up. Hence it cannot be over-emphasized that patients presenting with an apparently solitary testicular plasmacytoma either already have subclinical disseminated disease or will develop systemic myeloma if followed up for a sufficiently long time[2]. Full clinical investigation (serum protein electrophoresis, skeletal imaging and bone-marrow biopsy) and long-term follow-up are imperative to assess the risk of developing generalized multiple myeloma in patients with testicular solitary plasmacytoma. The better overall survival described for these patients needs to be confirmed by larger cohort studies[2-4]. Gowing[7] proposed that the assessment of the primary or secondary nature of an extramedullary plasmacytoma must be made on the basis of retrospective observation for about one year during which time no evidence of serum immunoglobulin or bone abnormalities is observed. Terzian et al[1] supported Gowing's criteria in their case report of testicular plasmacytoma. Our case fulfils these criteria and has no history

of other extramedullary plasmacytoma after 3 years of follow-up.

The clinical presentation of testicular plasmacytoma may be identical to that of any other primary or metastatic testicular neoplasms[4]. Ultrasonography of the scrotum is optimal for evaluating scrotal masses, and some testicular tumors have a distinctive echo pattern. Previous reports of testicular plasmacytoma described a relatively non-homogeneous or heterogeneous hypoechoic pattern, as in our case. However, this is not specific for plasmacytoma, and ultrasonography is unable to distinguish it from testicular germ cell tumors[8, 9]. There are only 2 reports where the preoperative diagnosis was made based on the hydrocele fluid cytology[10, 11].

The serum M-protein detected by immunoperoxidase staining in our patient, 2 weeks after the orchiectomy, was of IgG lambda isotype. After one month, serum electrophoresis and immunoelectrophoresis were normal, proving that there were no residual plasma cells in the circulating blood. Most of the reported patients analysed by immunoperoxidase staining had IgA, only one case was IgG lambda, like our patient[9].

The rarity of this condition does not permit delineation of a precise therapeutic approach. Management of the 22 reported cases consisted of radical orchiectomy with subsequent surveillance in 16, radiotherapy in 4 and chemotherapy in 2 cases (Table 1).

The prognosis of testicular plasmacytoma associated with multiple myeloma is poor, with postoperative survival ranging from 5 weeks to 48 months. The prognosis in the absence of previous or concurrent multiple myeloma is better, but requires strict long-term observation[4].

**Conclusion:** Although rare, plasmacytoma of the testis should be considered in the differential diagnosis of a testicular mass, particularly in a patient with known myeloma. Given the strong association between testicular plasmacytoma and multiple myeloma or plasmacytomas elsewhere, any patient who presents with testicular plasmacytoma should be investigated and monitored for the potential development of these entities.

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