Primary intratesticular leiomyosarcoma in a 78-year-old male

Wissem Hmida, Faouzi Mallat, Mouna Ben Othmen, Khaled Ben Ahmed, Sidiyaould Chavey, Sarra Mestiri, Amel Ben Abdallah, Faouzi Mosbah

ABSTRACT

Introduction: Primary leiomyosarcoma of the testis is a rare entity with only less than 20 cases were reported. The standard therapy is difficult to recommend because of the rarity of this tumor.

Case Report: We reported a case of primary intratesticular leiomyosarcoma in a 78-year-old male with unremarkable past medical history. The patient was presented within isolated testicular enlargement. Ultrasound revealed a large solid, homogenous and hyper echoic mass of the right testicle. Lactate dehydrogenase (LDH) was elevated, however, serum alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (β -hCG) were within normal limits. A right radical orchidectomy was performed. The diagnosis of primary leiomyosarcoma of testis was confirmed by histopathology.

Conclusion: The primary leiomyosarcoma of testis is a rare diagnosis which should be one of the differential diagnoses of testicular mass with normal tumor markers.

International Journal of Case Reports and Images (IJCRI)

International Journal of Case Reports and Images (IJCRI) is an international, peer reviewed, monthly, open access, online journal, publishing high-quality, articles in all areas of basic medical sciences and clinical specialties.

Aim of IJCRI is to encourage the publication of new information by providing a platform for reporting of unique, unusual and rare cases which enhance understanding of disease process, its diagnosis, management and clinico-pathologic correlations.

IJCRI publishes Review Articles, Case Series, Case Reports, Case in Images, Clinical Images and Letters to Editor.

Website: www.ijcasereportsandimages.com

CASE REPORT

OPEN ACCESS

Primary intratesticular leiomyosarcoma in a 78-year-old male

Wissem Hmida, Faouzi Mallat, Mouna Ben Othmen, Khaled Ben Ahmed, Sidiyaould Chavey, Sarra Mestiri, Amel Ben Abdallah, Faouzi Mosbah

ABSTRACT

Introduction: Primary leiomyosarcoma of the testis is a rare entity with only less than 20 cases were reported. The standard therapy is difficult to recommend because of the rarity of this tumor. **Case Report:** We reported a case of primary intratesticular leiomyosarcoma in a 78-year-old male with unremarkable past medical history. The patient was presented within isolated testicular enlargement. Ultrasound revealed a large solid, homogenous and hyper echoic mass of the right testicle. Lactate dehydrogenase (LDH) was elevated, however, serum alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (β -hCG) were within normal limits. A right radical orchidectomy was performed. The diagnosis of primary leiomyosarcoma of testis was confirmed by histopathology. **Conclusion:** The primary leiomyosarcoma of testis is a rare diagnosis which should be one of the differential diagnoses of testicular mass with normal tumor markers.

Keywords: Testis, Intratesticular tumor, Leiomyosarcoma, Radical orchidectomy

How to cite this article

Hmida W, Mallat F, Othmen MB, Ben Ahmed K, Chavey S, Mestiri S, Abdallah AB, Mosbah F. Primary intratesticular leiomyosarcoma in a 78-year-old male. Int J Case Rep Images 2014;5(10):717–722.

doi:10.5348/ijcri-2014128-CR-10439

INTRODUCTION

Leiomyosarcomas are malignant soft-tissue tumors arising from any tissues containing smooth muscles. However, intratesticular leiomyosarcoma are extremely rare compared with the paratesticular leiomyosarcoma [1].

We report a case of intratesticular leiomyosarcoma in a 78-year-old male who underwent right radical orchidectomy.

CASE REPORT

A 78-year-old male patient was presented with a two-year history of right scrotal mass. He had no significant past medical history. He did not receive any radiation therapy or anabolic corticosteroids in the past. No urologic or constitutional symptoms such as voiding complaints, weight loss, fatigue or fever were present. Physical examination revealed a hard, non-tender, right scrotal mass measuring 10x8x8 cm and did not reveal any superficial lymph node swelling. No ulceration of the overlying skin was evident. The digital rectal examination revealed a benign enlarged prostate.

Scrotal ultrasonographic examination revealed a solid homogenous hyperechoic mass of the right testicle measuring 9x8x8 cm. The left testicle and both epididymides were unremarkable and there was no evidence of hydrocele or paratesticular pathology.

Wissem Hmida¹, Faouzi Mallat¹, Mouna Ben Othmen¹, Khaled Ben Ahmed¹, Sidiyaould Chavey², Sarra Mestiri³, Amel Ben Abdallah², Faouzi Mosbah⁴

Affiliations: ¹MD, Department of Urology, Sahloul Hospital, Sousse, Tunisia; ²MD, Department of Radiology, Sahloul Hospital, Sousse, Tunisia; ³MD, Department of Pathology, Farhat Hached Hospital, Sousse, Tunisia; ⁴Professor, Department of Urology, Sahloul Hospital, Sousse, Tunisia.

Corresponding Author: Dr. Wissem Hmida, Department of Urology, Hospital of Sahloul, Sousse, Tunisia; Ph: +216 98452331; Email: wisshm@yahoo.fr

Received: 21 April 2014

Accepted: 03 June 2014

Published: 01 October 2014

Computed tomography (CT) scan of the chest, abdomen and pelvis revealed no evidence of metastatic disease or lymphadenopathy. Tumor markers serum beta-human chorionic gonadotropin (β -hCG) and alpha-fetoprotein (AFP) were within normal ranges. Serum lactate dehydrogenase (LDH) was raised (880 U/L, normal limit 100–190 U/L). The patient underwent a high ligation of the cord with right radical orchiectomy. Macroscopically, the tumor was a well-encapsulated grey white solid mass with hemorrhage and necrosis. There was no invasion to the spermatic cord or tunica vaginalis. The weight of the tumor was 450 grams and the size was approximately 11×10×9 cm.

Microscopic examination showed a high degree of cellular proliferation composed of spindle cells with round or oval-shaped nuclei implicating a storiform growth pattern (Figure 1). Necrosis was evident. Immunohistochemical examination revealed that the tumor cells were strongly positive for calponin, epithelial membrane antigen (EMA) smooth muscle actin and vimentin (Figures 2 and 3) but negative for S-100 and myogenic regulatory protein (MyoD1) proteins. The combined histologic and immunohistochemical findings were diagnostic of primary high-grade intratesticular leiomyosarcoma. With 24 months of follow-up, which included computed tomography scan of the abdomen, pelvis, bone, and chest, the patient remained free of disease.

DISCUSSION

Primary leiomyosarcoma of the testis is an extremely rare tumor. To our knowledge, only less than 20 cases have been reported (Table 1). Smooth muscle elements of the testicular parenchyma, especially blood vessels and the contractile cells of seminiferous tubules, appear to be the origin of intratesticular leiomyosarcoma [1, 18].

The real etiology of testicular leiomyosarcoma is already unknown, but hormonal stimulation has been suggested to have a role in the carcinogenesis of leiomyosarcoma [4]. In fact, it mostly occurs in young men in whom there is an associated history of high doses of anabolic steroids [4] or chronic inflammation [6]. Our patient does not have any risk factor. The clinical presentation and radiological finding of this tumor are non-specific. It seems to be not different from other malignant testicular tumors. Tumor markers (AFP, β -hCG and LDH) used in the previously reported cases were almost within the normal range, except two cases: one case with an elevated LDH [14] and the second with elevated β -hCG [16]. This case had an elevated LDH. Diagnosis is based only on histologic and immunohistochemical findings. Histologically, the nuclei of malignant smooth muscle cells are typically and selectively stain with antibodies against smooth muscle actin and desmin vimentin, but negative for S-100, CD-34, CD-68 and HMB-45. The high mitotic activity is considered an important criterion for malignancy [9].

Figure 2: Diffuse mesenchymal proliferation of malignant spindle shaped cells with eosinophilic, fibrillary cytoplasm and atypical nuclei displaying mitotic figures (H&E stain, x400).

Figure 3: Diffuse and intense immunostaining of tumoral cells with h-Caldesmon antibody (IHC, x100).

Figure 1: Diffuse mesenchymal proliferation of malignant spindle shaped cells with eosinophilic, fibrillary cytoplasm and atypical nuclei displaying mitotic figures (H&E stain, x100).

These tumors might spread via three routes: local invasion, lymphatic dissemination and hematogenous metastasis [8]. Metastasis was extremely rare, only one case developed pulmonary metastasis 14 months after surgery [5]. The treatment of leiomyosarcoma is not codified, it is difficult to recommend due to the few reported cases, and multi-therapy approach may be needed.

High inguinal orchiectomy (HIO) appears to be the treatment of choice in cases at low stage [18,19]. Although, adjuvant treatment chemotherapy and radiotherapy may be used for intratesticular leiomyosarcoma with high stage [14]. This case was treated with HIO without any adjuvant treatment and after 24 months of follow-up the patient was free of disease.

Table 1: The reported cases of intratesticular leiomyosarcoma.

Authors	Age	Side	Risk factor	Levels of tumor markers	Clinical stage	treatment	Follow-up (months)	Outcome
Yachia et al. [1]	55	R	-	N	I	HIO	24	Survived
Pellice et al. [2]	37	L	-	N	I	HIO	24	Survived
Washecka et al. [3]	47	R	-	N	I	HIO	49	Survived
Washecka et al. [3]	40	R	-	N	I	HIO	42	Survived
Frohner et al. [4]	32	R	Anabolicsteroid	Unknown	I	HIO + RPLND	79	Survived
Hachi et al. [5]	70	L	-	N	I	HIO	14	Died
Ali et al. [6]	65	R	Chronic inflammatio	N	I	HIO	12	Survived
Takizawa et al. [7]	76	L	-	N	I	HIO	12	Survived
Canales et al. [8]	30	R	Radiation	Unknown	I	HIO	6	Survived
Sattary et al. [9]	27		-	N		HIO	30	Survived
Wakhlu et al. [10]	8 months		-	N	I	HIO + CTx	12	Survived
Borges et al. [11]	19		-		I	HIO + CTx + RT	16	Survived
Labanaris et al. [12]	Not specified	R	-	N	I	HIO	28	Survived
Moona et al. [13]	45		-	N	II	HIO + CTx	Not specified	Survived
Bakhshi et al. [14]	60	R	-	LDH elevated	I	HIO + CTx + RT	12	Survived
Fadl-Elmula et al. [15]	20	R	-	N	Not specified	HIO + CTx + surgery for local recurrence + RT	More than 22	Not specified
Yoshimine et al. [16]	73	L	-	β-HCG Slightlyelevated	III	HIO + Chemotherapy	9	Survived
Kumar et al. [17]	65	R	-	N		HIO	6	Survived
Present case	78	R	-	LDH elevated	II	HIO	9	Survived

Abbreviation: HIO, high inguinal orchidectomy; CTx, node dissection; RT, radiotherapy. R, right; L, left; RPLND, retroperitoneal lymph node dissection.

CONCLUSION

Testicular leiomyosarcoma is a rare disease. Based on a review of literature, high radical orchidectomy and strict surveillance is the treatment of choice of intratesticular leiomyosarcoma at stage I. However, standard management of stage II or stage III disease is difficult to recommend.

Author Contributions

Wisseem Hmida – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Faouzi Mallat – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Mouna Ben Othmen – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Khaled Ben Ahmed – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Sidiya Ould Chavey – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Sarra Mestiri – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Amel Ben Abdallah – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Faouzi Mosbah – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2014 Wisseem Hmida et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Yachia D, Auslaender L. Primary leiomyosarcoma of the testis. *J Urol* 1989 Apr;141(4):955–6.
2. Pellice C, Sabate M, Combalia, Ribas E, Cosme M. Leiomyosarcoma of the testes. *J Urol (Paris)* 1994;100(1):46–8. [Article in French].
3. Washecka RM, Mariani AJ, Zuna RE, Honda SA, Chong CD. Primary testicular sarcoma: Immunohistochemicalultrastructural and DNA flow cytometric study of three cases with a review of literature. *Cancer* 1996 Apr 15;77(8):1524–8.
4. Froehner M, Fischer R, Leike S, Hakenberg OW, Noack B, Wirth MP. Intratesticularleiomyosarcoma in a young man after high dose doping with oral-Turinabol: A case report. *Cancer* 1999 Oct 15;86(8):1571–5.
5. Hachi H, Bougtab A, Amhajji R, et al. A case report of testicular leiomyosarcoma. *Med Trop (Mars)* 2002;62(5):531–3. [Article in French].
6. Ali Y, Kehinde EO, Makar R, Al-Awadi KA, Anim JT. Leiomyosarcoma complicating chronic inflammation of the testis. *Med Princ Pract* 2002 Jul-Sep;11(3):157–60.
7. Takizawa A, Miura T, Fujinami K, Kawakami S, Osada Y, Kameda Y. Primary testicular leiomyosarcoma. *Int J Urol* 2005 Jun;12(6):596–8.
8. Canales BK, Lukasewycz SJ, Manivel JC, Pryor JL. Post Radiotherapy intra testicular leiomyosarcoma. *Urology* 2005 Sep;66(3):657.
9. Labanaris AP, Zugor V, Smiszek R, Nutzel R, Kuhn R. Primary Leiomyosarcoma of the Testis. A Case Report. *Anticancer Res* 2010 May;30(5):1725–6.
10. Wakhlu A, Chaudhary A. Massive leiomyosarcoma of the testes in an infant. *J Pediatr Surg* 2004 Jul;39(7):e16–7.
11. Borges RP, Vila F, Cavdes V, et al. Primary testicular leiomyosarcoma: A case report. *Acta Urol* 2007;24:45–7.
12. Labanaris AP, Zugor V, Smiszek R, Nützel R, Kühn R. Primary leiomyosarcoma of the testes. A case report. *Anticancer Res* 2010 May;30(5):1725–6.
13. Moona MS, Fatima D, Turezbek A. Primary testicular leiomyosarcoma. *J Pak Med Assoc* 2011 Oct;61(10):1014–6.
14. Bakhshi GD, Wankhede KR, Tayade MB, Shenoy SS, Mundhe ST, Patel C. High grade leiomyosarcoma of the testes. *Clin Pract* 2011 Nov 30;1(4):e122.
15. Fadl-Elmula I, El Hassan LA, Elbadawi R, Arbab MAR, El Hassan AM. Massive primary leiomyosarcoma of the tes. *Sudan JMS* 2007 Sep;2(3).
16. Yoshimine S, Kono H, Nakagawa K, et al. Primary intratesticularleiomyosarcoma. *Can Urol Assoc J* 2009 Dec;3(6):E74–6.
17. Kumar M, Patne U, Kumar S, Shukla VK. Primary high-grade testicular leiomyosarcoma. *Indian J Pathol Microbiol* 2009 Jan-Mar;52(1):91–3.
18. Singh R, Chandra A, O'Brien TS. Primary intratesticularleiomyosarcoma in a mixed race man: A case report. *J Clin Pathol* 2004 Dec;57(12):1319–20.
19. Sattary M, Hazraty B, Saraii MB. Primary pure testicular low-grade leiomyosarcoma. *Iran J Med Sci* 2003;28:48–50.

ABOUT THE AUTHORS

Article citation: Hmida W, Mallat F, Othmen MB, Ben Ahmed K, Chavey S, Mestiri S, Abdallah AB, Mosbah F. Primary intratesticular leiomyosarcoma in a 78-year-old male. Int J Case Rep Images 2014;5(10):717–722.

Wissem Hmida is Assistant Professor at Department of Urology, Shloul Hospital, Sousse, Tunisia. His research interests include laparoscopy and kidney transplantation. Email: wisshm@yahoo.fr

Faouzi Mallat MD, Department of Urology, Sahloul Hospital, Sousse, Tunisia.

Mouna Ben Othmen MD, Department of Urology, Sahloul Hospital, Sousse, Tunisia.

Khaled Ben Ahmed MD, Department of Urology, Sahloul Hospital, Sousse, Tunisia.

Sidiyaould Chavey MD, Department of Radiology, Sahloul Hospital, Sousse, Tunisia.

Sarra Mestiri MD, Department of Pathology, Farhat Hached Hospital, Sousse, Tunisia.

Amel Ben Abdallah MD, Department of Radiology, Sahloul Hospital, Sousse, Tunisia.

Faouzi Mosbah is Professor, Department of Urology, Sahloul Hospital, Sousse, Tunisia.

Access full text article on
other devices

Access PDF of article on
other devices

Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals

Edorium Journals is a publisher of high-quality, open access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission

We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?

In less than 10 words - we give you what no one does.

Vision of being the best

We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services

We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review

All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review

All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version

Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status

From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks

You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.

Four weeks

After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.

Mentored Review Articles (MRA)

Our academic program "Mentored Review Article" (MRA) gives you a unique opportunity to publish papers under mentorship of international faculty. These articles are published free of charges.

Favored Author program

One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program

Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence

We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...

We request you to have a look at our website to know more about us and our services.

We welcome you to interact with us, share with us, join us and of course publish with us.

Edorium Journals: On Web

Browse Journals

CONNECT WITH US