

CASE REPORT

Adenomatous hyperplasia of the rete testis: A rare intrascrotal lesion managed with limited testicular excision

Francesco Catanzariti¹, Lucilla Servi², Andrea Fabiani², Alessandra Filosa³, Gabriele Mammana²

¹ Surgery Department, Section of Urology, ASUR Marche Area Vasta 4, A. Murri Hospital, Fermo, Italy;

² Surgery Department, Section of Urology, ASUR Marche Area Vasta 3, Macerata Hospital, Macerata, Italy;

³ Section of Pathological Anatomy, Department of Clinical Pathology, ASUR Marche Area Vasta 3, Macerata Hospital, Macerata, Italy.

Summary

Introduction: Testicular cancer is one of the most frequent in young men and its incidence is increasing in recent years because of incidental finding during routine ultrasound exams. Adenomatous hyperplasia of the rete testis is one of the benign and rare pathological types incidentally detected and very few cases are described in the literature.

Case report: A 40 years old man come to our attention for a balanoposthitis without testicular pain. During andrological examination we performed palpation of the testes and we noticed a palpable nodule of hard consistency in the left testicle. We then performed an ultrasound exam of the testis which highlighted the presence of an intra-didymus neoformation with diameters of 1.2 x 1.6 cm and with the presence of cysts inside. We also performed blood tests to check tumor markers alpha fetoprotein, beta hCG and LDH which resulted inside the normal range. We then conducted a chest and abdomen CT scan that showed no pathological elements. Therefore, as we suspected that this tumor was benign, we performed an enucleation of the neoplasm. The definitive histological examination revealed the presence of dilated ducts lined with epithelial cubic-columnar cells with clear cytoplasm rich in glycogen and the pathologist so concluded that the tumor could be classified as adenomatous hyperplasia of the rete testis. At three months of follow up, the patient doesn't have any recurrent lesion to either testicles.

Discussion: Adenomatous hyperplasia of the rete testis is a very rare intrascrotal lesion. This histological type is the most frequent between benign lesion of the ovary, but few works in literature reported this histological type in the male gonad and, in most of these works, authors described these lesion at epididymis.

Conclusion: We believe that a conservative approach must be considered mandatory in case of testicular lesions 1.5 cm in diameter. A radical approach might have alterate fertility of the patient and also have caused psychological trauma more than an enucleation. However a longer follow up is needed to understand if this was the right decision for the oncological point of view.

KEY WORDS: Adenomatous hyperplasia of the rete testis; Testicular neoplasm enucleation; Benign lesion of the testis.

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INTRODUCTION

Testicular cancer is one of the most frequent tumours in young men and its incidence is increasing in recent years because of incidental finding during routine ultrasound exams (1). Even if the germ cell tumors are the majority of testicular neoplasms, there are some benign lesions those would require less aggressive treatment considering how the loss of a testicle may adversely affect those patients for the psychological point of view, moreover if we consider that these men come to a diagnosis of cancer

at a young age and in a phase of life fully sexually active. Adenomatous hyperplasia of the rete testis is one of the benign and rare pathological types incidentally detected and very few cases are described in the literature.

CASE REPORT

A 40 years old man came to our attention for a balanoposthitis without testicular pain. During andrological examination we performed palpation of the testes and we noticed a palpable nodule of hard consistency in the left testicle. We then performed an ultrasound exam (Figure 1) of the testis which highlighted the presence of an intra-didymus neoformation with diameters of 1.2 x 1.6 cm and presence of cysts inside. We then performed blood tests to check tumor markers alpha fetoprotein, beta hCG and LDH which resulted inside the normal range, respectively: 5.39 ng/mL, 0 mIU/mL and 151 U/L. We also conducted a chest and abdomen computed tomography (CT) scan that showed no pathological elements. We performed an enucleation of the neoplasm: after incision of skin, dartos and vaginal stratum and identification of the lesion we cut albuginea layer and we eas-

Figure 1.

Ultrasound exam of the left testis which show an intra-didymus neoformation with diameters of 12 x 16 mm and with the presence of cysts inside.

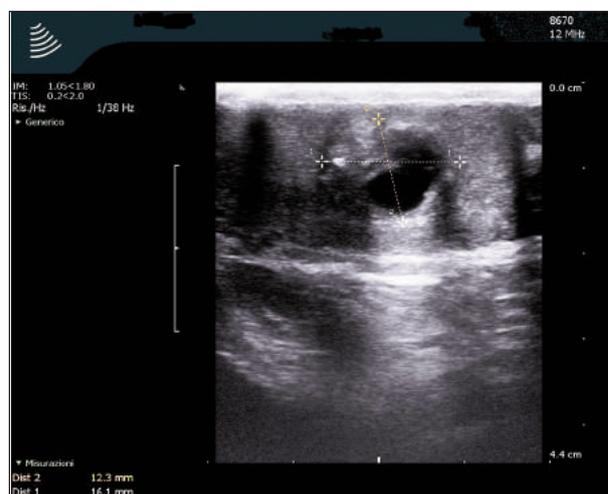
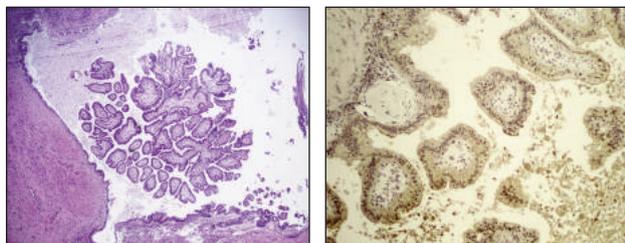


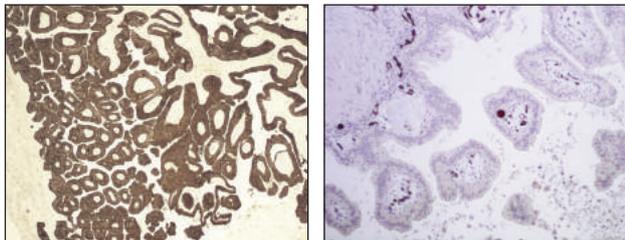
Figure 2.

Definitive histological examination: dilated ducts lined with epithelial cubic-columnar cells with clear cytoplasm rich in glycogen.



A) Hematoxylin and eosin.

B) Calretinine.



C) Cam 5.2.

D) wt1. **Diagnosis: adenomatous hyperplasia of the rete testis.**

ily removed the nodule, with low blood loss. Albuginea was then repaired with 3-0 absorbable suture. No clamp at spermatic cord was used. The definitive histological examination revealed the presence of dilated ducts lined with epithelial cubic-columnar cells with clear cytoplasm rich in glycogen (Figure 2). These cells were ck8 negative, ck18 positive, low weight ck positive, WT1 negative and Calretinine negative. The pathologist so concluded that the tumor could be classified as adenomatous hyperplasia of the rete testis. At three months of follow up, the patient doesn't have any recurrent lesion to either testicles.

DISCUSSION

Adenomatous hyperplasia of the rete testis is a very rare intrascrotal lesion. This histological type is the most frequent between benign lesion of the ovary, but few works in literature reported this histological type in the male gonad and, in most of these works, authors described these lesion at epididymis. The histogenesis of these ovarian-type epithelial tumours of the testis and paratestis remains unknown, but some Authors (2) explained that this lesion could be a Müllerian metaplasia within the testicular parenchyma due to inclusions of the mesothelium during the embryonic period.

One of the first studies about this rare tumor is by Kosmehl (3) in 1989. He described a 50 years old man with a unilocular cystic intratesticular tumour. He defined that lesion as the male analogue of the respective ovarian growth. After this, very few other studies have been published. The most important manuscripts in the last 5 years were those by by Elliot (4) in 2010 and by Olla in 2013 (5). Elliot and his colleagues described a 55-year-old man with painless right testicular swelling of 16 years duration that at ultrasound appeared as a hypovascular mass measuring 6.5 cm in diameter, containing an area of curvilinear calcification. Microscopically, the lesion had mucinous epithelium resembling endocervical type cells with focal intestinal type epithelium, qualifying the tumour as a mucinous

neoplasm of low malignant potential or “borderline” type. Two years ago a paper by Olla described a 58-year-old male patient with an occasional finding of two separate intratesticular nodules on the left testicle. At microscopic examination these lesions were classified as papillary serous cystadenoma with focal cytologic atypia for the presence of cysts with an epithelial lining almost entirely of ciliated columnar type with the presence of sporadic evidence of cytological atypia and no mitoses. In our case report pathologist described the lesion similar to these examples we cited, but the surgical approach we used was different from the others described in the literature. Therefore, as we suspected by ultrasound that this tumor was benign, we decided to perform an enucleation instead of an orchiectomy, also because of the young age of our patient. A radical approach might have affected its fertility and also have caused psychological trauma more than an enucleation. As previously reported, we believe that a conservative approach must be considered mandatory in case of testicular lesions 1,5 cm in diameter. The close collaboration with a dedicated uro-pathologist, also during frozen section examination, may be determinant (6, 7). However a longer follow up is needed to understand if this was the right decision for the oncological point of view.

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Correspondence

Francesco Catanzariti, MD (Corresponding Author)
 fracatanzariti@libero.it
 Surgery Dpt, Section of Urology, ASUR Marche Area Vasta 4, A.Murri Hospital, Fermo, Italy

Lucilla Servi, MD - Andrea Fabiani, MD - Gabriele Mammana, MD
 Surgery Dpt, Section of Urology, ASUR Marche Area Vasta 3, Macerata Hospital, Macerata, Italy

Alessandra Filosa, MD
 Section of Pathological Anatomy, Department of Clinical Pathology, ASUR Marche Area Vasta 3, Macerata, Italy