CASE REPORT

Diagnosis and Treatment of Paratesticular Adenomatoid Tumors

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ABSTRACT

Paratesticular adenomatoid tumor (PAT) is the most common paratesticular tumor. It is still a concern for today's urologists because it cannot be distinguished from malignant testicular tumors by clinical symptoms, routine examination and imaging methods. Because of the predominant benign nature of paratesticular masses, testicular preservative treatments get to the foreground. However, the fact that virtually all of the solid scrotal masses are malignant and are treated with radicel ingainel orchiectomy (RIO) remains a cause of concern. In this study, we discuss the diagnosis and treatment of 12 paratesticular adenomatoid tumors treated between 2012 and 2017 in two centres. We suggest that a frozen section should be done with the help of an experienced pathologist; and a meticulous microscopic evaluation should be the gold standard in case of a benign tumor suspicion.

Key Words: *Paratesticular. Adenomatoid tumor. Frozen section. Testis-sparing surgery.*

INTRODUCTION

Paratesticular adenomatoid tumor (PAT) is the most common paratesticular tumor.[1] Even though the epididymis is the most common site of involvement, it can involve other testicular layers and spermatic cord. Heart, lymph nodes, adrenal glands, omentum, and retroperitoneal involvement can be seen extra-peritoneally. It is most commonly seen in 3rd and 4th decades.[2] There is usually painless, small, paratesticular mass on clinical examination. Its treatment is testis-preserving surgery (TPS). Since it is difficult to distinguish these tumors from malignant testicular tumors, PAT pathology may be encountered in orchiectomy materials.

CASE REPORT

We present data of 12 PAT patients with ages ranging from 27 to 61 years, who were treated between 2012 and 2017 in two centres. Four of these patients were admitted to our clinic with complaints of infertility, 3 had scrotal pain, 2 had scrotal pain and a hard mass, and 2 had scrotal swellings. One of the patients was injured with a firearm injury and testicular trauma was detected. Although this trauma patient was taken to an emergency operation, tumor markers were done in all the other patients, and alpha-fetoprotein elevation (10.52 ng/mL) was detected in 1 patient and lactate dehydrogenase elevation (224 U/L, 237 U/L, 567 U/L) was detected in 3 patients. Other tumor markers were in the normal range. Localisation was in the right in 7 patients, and in the left scrotum in 5 patients.

Scrotal doppler ultrasonography (SDU) was performed on all patients except trauma patient who was taken to emergency operation. Scrotal mass in 7 of the patients could not be distinguished from the testis. Radical inguinal orchiectomy (RIO) was done directly in 4 of these 7 patients. In the other 3 patients, frozen section was done with benign mass suspicion. Paratesticular mass was reported in SDU in 4 patients and; intra-operative frozen section was done in these patients. Five patients with benign paratesticular neoplazm in frozen section were treated with TPS. Two patients were diagnosed as epididymal adenocarcinoma and spermatic cord adenocarcinoma in frozen section and RIO was applied to these 2 patients; but final pathology was reported as PAT. Testicular parenchymal loss was observed in the patient who underwent scrotal exploration due to gunshot injury and scrotal orchiectomy was done (Table I).

Surgical pathology of all 12 patients was reported as PAT. Rete testis invasion, lymphovascular invasion, and intratubular germ cell neoplasia were not detected in orchiectomy materials and normal spermatogenesis findings were detected in testis (Figures 1 and 2). Clinical, biochemical and imaging relapse was not observed in the follow-up of these patients.

DISCUSSION

While it is known that the vast majority of scrotal masses are of testicular origin and are usually malignant; paratesticular masses constitute 2-3% of all scrotal masses and they are usually benign. PAT constitutes approximately 30% of all paratesticular-localised neoplasms and 60% of all benign neoplasms and is the

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Table I: Twelve paratesticular adenomatoid tumor cases treated between 2012 and 2017 in two centres.

Patient	Age	Side	Clinic presentation	Tumor Markers	Radiological localisation	Frozen section	Operation	Surgical pathology
Case 1	34	L	Mass	AFP +	Testicular mass	Ø	RIO	PAT
Case 2	28	L	Infertility	LDH +	Testicular mass	Ø	RIO	PAT
Case 3	41	R	Pain	LDH +	Testicular mass	Ø	RIO	PAT
Case 4	27	R	Infertility	-	Testicular mass	Ø	RIO	PAT
Case 5	37	R	Gunshot injury	Ø	Ø	Ø	SO	PAT
Case 6	43	L	Pain	-	Paratesticular mass	Epididymal adenocarcinoma	RIO	PAT
Case 7	30	R	Infertility	LDH+	Testicular mass	Spermatic cord adenocarcinoma	RIO	PAT
Case 8	61	L	Pain	-	Paratesticular mass	Benign paratesticular neoplasm	TPS	PAT
Case 9	57	L	Scrotal swelling	-	Paratesticular mass	Benign paratesticular neoplasm	TPS	PAT
Case 10	31	R	Infertility	-	Testicular mass	Benign paratesticular neoplasm	TPS	PAT
Case 11	44	R	Scrotal swelling	-	Paratesticular mass	Benign paratesticular neoplasm	TPS	PAT
Case 12	37	R	Mass	-	Testicular mass	Benign paratesticular neoplasm	TPS	PAT

R = right; L = left; (+) = Postive; (-) = Negative; AFP = Alfa fetoprotein; LDH = Laktat dehidrogenaz; Ø = Not taken; RIO = Radical inguinal orchiectomy; SO = Scrotal orchiectomy; TPS = Testicular preserving surgery; PAT = Paratesticular adenomatoid tumor.

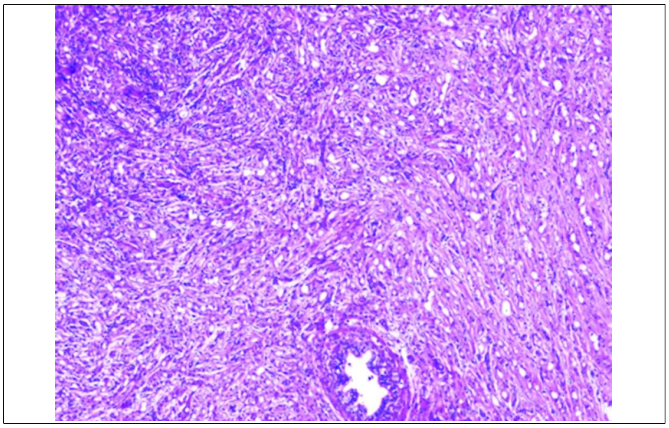

Figure 1: Adenomatoid tumor, which forms a solid pattern in the testis (H & E X40).

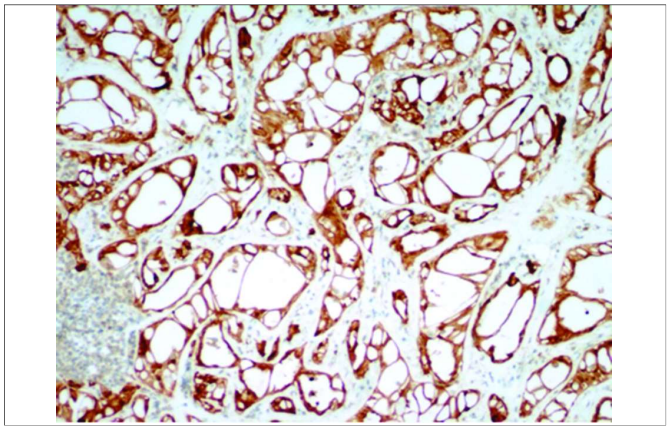

Figure 2: Diffuse positivity with Calretinin on immunohistochemical stain (Calretinin X40).

most common paratesticular neoplasm.[1] PATs are very rare benign tumors. Although they can be seen in all age groups, they are most common in 3rd and 4th decades.[2] PAT localisation is most commonly seen in the lower part of the epididymis. It can originate more rarely from tunica vaginalis, tunica albuginea, spermatic cord and rete testis.[3] It is clinically present as painless, hard, slow-growing mass smaller than 2 cm or may be detected incidentally.

The paratesticular area is an anatomic region with different structures; tumors are rarely seen in this area and include those originating from supportive tissues such as epididymis, spermatic cord, tunica vaginalis and vestigial embryological remnants. Therefore, the most important feature of the paratesticular region is that it is a source for many different types of tumors and lesions. In the differential diagnosis of PAT, other paratesticular lesions (cystadenomas, spermatoceles, hydroceles, varicoceles, hernias, scrotal calculi, poly-orchidism, solitary fibrous tumor, leiomyoma, neurofibroma, fibroma of the tunica, idiopathic fibromatosis and malignant tumors) and intratesticular lesions (tunica albuginea cysts, testicular simple cysts, epidermoid cysts, cystic ectasia of the rete testis, intratesticular varicocele, adrenal rest tumors, splenogonadal fusion and malignant tumors) should be considered.[4]

Ultrasonography is the first diagnostic method and helps to define solid, cystic features of the mass. On ultrasound, there is usually well-defined, homogeneous, less than 2 cm, hyper echoic paratesticular mass. However, there is no ultrasonographic finding that could clearly distinguish malignant testicular tumors from PAT. Nevertheless, computerised tomography and magnetic resonance imaging may be the guiding factor in defining characteristics of the mass and its relation to surrounding tissues.[5] In a very recent study, it has been shown that 18F- FDG PET / CT scan may be used in the diagnosis of testicular germ cell tumors.[6]

Although it is controversial, PAT is considered as mesothelial-derived. In microscopy, it consists of flattened and cubical single-row, epithelial cleft-like structures and tubules. In addition, storiform pattern, intraluminal mucin secretion, and stony ring-like cells can be seen. Immunohistochemically, these are WT1, calretinin, vimentin, and D2-40 positive.[7]

Malignant tumors can also be seen in the paratesticular area and compared to benign neoplasms, they present in older age and grow rapidly in a shorter period of time.[3] These are hard, generally solid, infiltrating into the

surrounding tissues with unclear margins, sometimes cystic and about 30% are sarcomas.[5] As paratesticular malignant neoplasms are very rare tumors, treatment options are based on a retrospective study of few cases.[8]

Although there is no consensus about the treatment of PAT, testis-preserving surgery seems to be adequate in the literature. Orchiectomy should be avoided if possible for these tumors, which do not show malignant transformation, do not affect spermatogenesis, and recurrence is not reported after enucleation.[9] As previously described, it may mimic clinically a malignant neoplasm and the vitality of the remaining parenchyma may cause concern and RIO can be applied in this case.[10] Therefore, preoperative diagnosis and intraoperative frozen sections are very important to prevent unnecessary orchiectomies in cases where differential diagnosis is not possible.

It is stated that tumor markers may be helpful in preoperative diagnosis to separate PAT from testicular tumors. However, it is known that tumor markers may be negative in 49% of malignant testicular tumors. Alpha-fetoprotein elevation (10.52 ng/mL) was detected in one of these patients with final PAT diagnosis and LDH (224 U/L, 237 U/L, 567 U/L) was detected in 3 patients with final PAT diagnosis.

In this series, scrotal doppler ultrasonography was reported as paratesticular mass in 4 patients. All of these patients underwent a frozen section during surgery. In addition, frozen section was also performed on 3 masses, which were determined to be radiologically indistinguishable from the testis, but were found to be paratesticular or epididymal during surgery. A total of 5 patients, who had benign paratesticular neoplasms in their frozen sections, were treated with testis TPS. In two patients, the frozen section was reported as spermatic cord and epididymal adenocarcinoma, so RIO was applied but final pathology was reported as PAT. However, scrotal mass was not distinguished from testis radiologically and surgically in 4 patients. RIO was done in these patients before the frozen section was performed, but the surgical pathology was reported as PAT. In three of these patients, the elevation of tumor markers and the radiological suspicion of malignant testicular tumors were distracting. However, the fact that the RIO has been applied before the frozen section can be regarded as an error.

Because of the predominant benign nature of paratesticular masses, testicular preservative treatment should be the preferred option. However, the fact that virtually all of the solid scrotal masses are considered malignant and are treated with RIO remains a cause of concern. We think that a frozen section should be done with the help of an experienced pathologist and a meticulous microscopic evaluation should be the gold standard in case of a benign tumor suspicion.

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