ADENOMATOID PARATESTICULAR TUMOR: REPORT OF 5 NEW CASES AND LITERATURE REVIEW


Summary.- OBJECTIVE: To report 5 cases of adenomatoid tumor of the epididymis that have been diagnosed in the last 11 years at our hospital. We performed a bibliographic review with discussion of diagnosis, differential diagnosis and treatment of this rare type of lesion.

METHOD: We have performed a retrospective analysis of epididymal adenomatoid tumors diagnosed during the last 11 years in our hospital, from January 2001 to June 2012.

RESULTS: The average age of the series was 44 years. The predominant reason for consultation was long duration painless scrotal mass, with palpable nodule, usually dependent of the epididymis. 60% of the patients have been studied with abdominal ultrasound, which showed a nodular paratesticular lesion, with an echogenicity compatible with solid tissue. Tumor markers associated with testicular tumors (alpha-fetoprotein [AFP], beta-human chorionic gonadotropin [b-HCG] and lactate dehydrogenase [LDH]) were analyzed in 60% of the cases, being normal in all patients. 3 lumpectomies, 1 epididymectomy and 1 radical orchiectomy have been performed.

CONCLUSIONS: The discovery of a solid epididymal mass is uncommon in clinical practice. Physical examination and imaging tests should confirm the paratesticular origin of the lesion, being then trans-scrotal epididymectomy the treatment of choice. If diagnosis is uncertain, inguinal approach and intraoperative biopsy are mandatory.

Keywords: Adenomatoid tumor. Epididymis. Diagnosis. Treatment.

Resumen.- OBJETIVO: Presentar los 5 casos de tumor adenomatoide epididimario, diagnosticados en los últimos 11 años en nuestro centro. Revisión bibliográfica y discusión del diagnóstico diferencial y tratamiento de este tipo de lesiones poco frecuentes en la práctica clínica.

MÉTODO: Se realiza un análisis retrospectivo de los casos de tumor adenomatoide epididimario diagnosticados durante los últimos 11 años en nuestro hospital, desde enero de 2001 a febrero de 2012.

RESULTADOS: La edad media de la serie fue de 44 años. El motivo de consulta predominante fue masa escrotal no dolorosa de larga evolución, con nódulo palpable, generalmente dependiente del epididimo. En el 60% de los casos se realizó ecografía abdominal, en la cual se observó lesión paratesticular nodular de ecogenicidad compatible con tejido sólido. En el 60% % de los casos se analizaron los marcadores tumorales relacionados con tumores testiculares: alfafetoproteína (AFP), beta-gonadotropina coriónica humana (b-HCG) y lactato deshidrogenasa (LDH), que resultaron normales. Se realizaron 3 tumorectomías, 1 epididimectomía y 1 orquiectomía radical.

CONCLUSIONES: El hallazgo de una masa sólida epididimaria es infrecuente en la práctica clínica. La exploración física y pruebas de imagen deben confirmar el origen paratesticular de la lesión, siendo entonces la epididimectomía transescrotal el tratamiento de elección. Ante duda diagnóstica, el abordaje quirúrgico por vía inguinal con biopsia intraoperatoria de la tumoración es mandatorio.

Keywords: Adenomatoid tumor. Epididymis. Diagnosis. Treatment.


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INTRODUCTION

The tumors located in the epididymis are rare, representing less than 5% of intrascrotal neoplasms, being the most frequent subtype the adenomatoid tumor, with 30% of the cases (3). Because of the fact that 75% of paratesticular tumors are benign, the correct diagnosis is essential in order to provide to the patient a conservative treatment.

We present 5 cases of adenomatoid tumor of epididymis that were treated surgically in our hospital, discussing aspects of their diagnosis, differential diagnosis and treatment.

CASES REPORT

In our series of 5 cases, the mean age was 44 years old, ranging from 33 to 60. None of our patients had urological history.

60% of the patients were referred to our center due to a painless scrotal mass of long duration, 1 of the patients for suspicion of epididymitis and another one of epididymal cyst.

On physical examination, we observed intrascrotal mass in epididymis in the 5 cases. Just the patient referred due to suspicion of epididymitis felt pain with the manipulation. Transillumination was negative in all the cases.

The determination of serum tumor markers (b-HCG, AFP and LDH) was performed in 3 patients, with values within normal limits in all cases.

60% of patients underwent scrotal ultrasound which showed a well-defined and intermediate echogenicity mass adjacent to tail of the epididymis. In only 1 of these cases it was impossible to exclude testicular involvement.

All patients underwent surgery to extirpate the lesion, but a more or less conservative attitude varied depending on the case: there were 3 lumpectomies (just one of these 3 was associated with an intraoperatively biopsy), 1 partial epididymectomy and 1 radical orchiectomy (in a 60-year old man whose ultrasound study could not rule out testicular involvement). We opted for a transesccrotal approach in 3 patients, the rest of them were treated using an inguinal approach (Table I).

Macroscopically, the lesions had a solid consistence, with an average diameter of 1.4 cm, well demarcated, brown,

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Reason for consultation</th>
<th>Location</th>
<th>Intraoperative biopsy</th>
<th>Treatment</th>
<th>Inguinal</th>
<th>Follow-up (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>37</td>
<td>Suspicion of epididymis cyst</td>
<td>Epididymis tail</td>
<td>No</td>
<td>Lumpectomy</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>60</td>
<td>Painful scrotal mass</td>
<td>Epididymis tail</td>
<td>No</td>
<td>Orchiectomy</td>
<td>E</td>
<td></td>
</tr>
<tr>
<td>46</td>
<td>Suspicion of epididymitis</td>
<td>Epididymis tail</td>
<td>No</td>
<td>Epididymectomy</td>
<td>79</td>
<td></td>
</tr>
<tr>
<td>33</td>
<td>Painless scrotal mass</td>
<td>Epididymis tail</td>
<td>No</td>
<td>Lumpectomy</td>
<td>70</td>
<td></td>
</tr>
<tr>
<td>45</td>
<td>Painless scrotal mass</td>
<td>Epididymis tail</td>
<td>No</td>
<td>Lumpectomy</td>
<td>13</td>
<td></td>
</tr>
</tbody>
</table>
greyish or, odder, whitish. Microscopic examination showed a proliferation of tubules composed of cuboidal cells, epithelioid, slightly vacuolated, forming solid cords (Figure 1) that alternate with areas of tubules lined by flat cells, simulating dilated vascular structures. In the 2 cases which were studied with immunohistochemistry, it showed positivity for pancytokeratine and epithelial membrane antigen (EMA) (Figure 2), with negative results for carcinoembryonic antigen (CEA) and factor VIII.

There were no perioperative complications in the series. In the case of the unique patient with pain on examination, there was no improvement after the intervention. With a mean follow-up of 36 months (range 5 to 79 months), there were no recurrences of the lesion in any patient.

DISCUSSION

The differential diagnosis of space-occupying lesions of the scrotal sac is very broad, with different therapeutic and prognostic implications depending on the characteristics of the lesion and its location (testicular or paratesticular). This differential diagnosis is based on 3 pillars: a thorough physical examination, determination of serum tumor markers for screening of testicular parenchymal tumors (beta-HCG, AFP and LDH) and imaging tests (usually ultrasound scrotal MR may be helpful in certain cases) (1). Using this diagnostic approach, we can observe the difference between a paratesticular tumor (which, given the generally benign nature, allows to preserve the gonad) and a testicular tumor, which would not allow such a conservative attitude (3).

In the epididymis, the majority of the space-occupying lesions are cystic and completely benign, highlighting the spermatoceles (or retention cysts) and serous cysts, typically of vestigial origin (4). However, the finding of an epididymal solid lesion is not unusual, and the 75% of them are benign. Therefore, there is a small percentage of epididymal malignant lesions, the most common, and in order of frequency: sarcomas (44%), metastases (27%) and primary epididymal carcinomas (24%). (5).

The diagnosis of epididymal adenomatoid tumors is usually performed incidentally between the 3 rd and 5 th decade of life (2, 6), because of its small size (usually less than 2.5 cm) and slow growth. The presence of pain is rare (1, 6), and it is associated with infarction and necrosis (2, 7).

Sonographically it is shown as a solid nodule with variable echogenicity with respect to adjacent tissues (4).

This tumor has a benign behavior, and metastases or recurrences have never been described (8). In fact, the appearance of cellular atypia and/or an infiltrative pattern in the excised lesion does not indicate malignancy (7, 9). The adenomatoid tumor of the epididymis may, in over 50% of the cases, show extension to rete testis and testicular parenchyma, and we cannot consider this a true invasion and it doesn’t add prognostic value (9). In practice, this occasional difficulty of demarcation between tumor growth and testicular or paratesticular structure causes that many paratesticular adenomatoid tumors are treated as a testicular tumor, especially if growth is rapid, it is associated with pain or another behavior more related to a malignant tumor or testis.

The treatment of choice is total or partial epididymectomy, radical orchietomy is not recommended as a first line
treatment. In most cases, with an adequate physical examination, determination of serum tumor markers, scrotal ultrasound, and if any doubt remains, with an intraoperative biopsy (1, 3, 9), we can rule out testicular origin of the lesion and its malignant nature, avoiding the removal of a healthy testis (2, 3, 9, 10). If there is any suspicion of malignancy, the inguinal approach is obligatory (2).

We emphasize that, although the conservative attitude was typical, 1 case, the oldest patient in the series, had been treated with radical inguinal orchiectomy. This was because the previous ultrasound did not detect a clear limit between the testis and the lesion.

However, in our opinion, a more appropriate attitude would have been to carry out an intraoperative biopsy of the mass, which could have prevented the loss of the testicle. Another diagnostic option, given the benign data provided by pre-surgical testing, could have been the fine-needle aspiration (FNA), although there is much controversy among authors, because of the risk of neoplastic dissemination in case of malignant tumor (1).

CONCLUSIONS

The discovery of solid epididymal mass is uncommon in clinical practice, and it is very usual to perform radical orchiectomy to prevent the risk of overlooking the clinical malignancy of the lesion. If you have the possibility of intraoperative biopsy of the lesion, and the pre-surgical and surgical data point toward a benign neoplasm, we must try to preserve the testicle.

On the contrary, if after a proper examination and the laboratory tests there are still doubts about the benign nature of the injury, it is mandatory inguinal surgical approach and intraoperative biopsy, in order to avoid an unnecessary orchiectomy.

REFERENCES AND RECOMMENDED READINGS

(*of special interest, **of outstanding interest)


