

EVALUATION OF INDOLENT EPIDIDYMAL MASS – ADENOMATOID TUMOR OF THE EPIDIDYMIS

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Abstract - Adenomatoid tumor of the epididymis is a rare benign neoplasm of mesothelial origin, mostly localized in the epididymis. It is usually presented as an indolent, painless scrotal enlargement. Immunohistochemically it is characterized by strong positivity for calretinin and epithelial markers, while tumor marker values are within the reference ranges. In this paper, two cases of 28- and 37-year-old men with adenomatoid tumors of the epididymis are described. In both, the tumor was presented as an indolent, slowly growing palpable mass in the epididymis. Tumor markers were in the normal range, digital examination found painless scrotal enlargement, while ultrasound showed a nonhomogeneous tumor. Immunohistochemically, they were characterized by strong positivity for calretinin and epithelial markers. If a localized epididymal mass is found, it is necessary to conduct an adequate diagnostic examination to differentiate inflammatory process from the neoplasm. An excision of the lesion is the most recommended option that also preserves testicular function and fertility. Histological and immunohistochemical evaluation are mandatory for a definitive diagnosis.

Key words: adenomatoid tumor; epididymis; immunohistochemical evaluation; preservation of testicular function.

INTRODUCTION

The adenomatoid tumor of the epididymis is a rare benign neoplasm of mesothelial origin (Hes, 2003). Mostly it arises from the epididymis, but other tumor localizations, such as the spermatic cord, testicular tunica and ejaculatory ducts have been observed (Resinav and Carranzab, 2010). Adenomatoid tumor is usually presented as a painless scrotal enlargement. It appears as a circumscribed, small, solid, firm, white, yellow or brown nodule (Maestro, 2009). Immunohistochemically, it is characterized by strong positivity for calretinin and epithelial markers, while tumor marker values are within the reference ranges (Kontos, 2008).

Preoperative diagnostic procedures need to distinguish between an adenomatoid tumor of the epididymis, infection or a testicular tumor, and when an accurate diagnosis is made, the recommended procedure that would also preserve testicular function is excision of the lesion (Amin and Parwani, 2009).

MATERIALS AND METHODS

We present the first case, a 28-year-old male, with no medical or surgical history of interest. A paratesticular enlargement was accidentally palpated during genito-scrotal examination performed for a penile rash. Both testes and penis were normal, but a

palpable indolent node was found in the right epididymis. Ultrasound imaging showed an inhomogeneous form 3 cm in diameter, mostly cystic on the upper pole of the epididymis.

Tumor markers, beta human chorionic gonadotropin (β -HCG), alpha-fetoprotein (AFP) and lactate dehydrogenase (LDH), were in referent ranges, as was the blood count. Microbiological analysis showed the absence of sexually transmitted disease (STD). We performed excision of the tumor with preservation of the epididymis. It was a firm, inhomogeneous well-circumscribed mass, 30 x 18 x 12 mm in size. Histopathological examination showed an adenomatoid tumor of the epididymis (Fig. 1A). Immunohistochemical examination found tumor cells diffusely and strongly positive for CK (AE1/AE3) and calretinin, and negative for CD31 (Fig. 1B-D). Twelve months after surgery the patient was asymptomatic, without local recurrence.

The other patient was a 37-year-old male, married, with two children. Several years before, he noticed a small painless node on the lower pole of the right testis, which enlarged a few months before visiting an urologist. Genito-scrotal examination found both testes to be normal with the small, hard, indolent node on the lower pole of the right testis. Ultrasound showed small, confined, inhomogeneous tumor, 18.8 x 18.8 mm in size, on the right epididymis (Fig. 2).

Tumor markers (AFP, β -HCG) were in the normal range. There were no other diseases, genito-scrotal infections or deviations in blood count. We performed excision of the tumor with epididymal preservation. Macroscopically it was a solid, whitish tumor. Histopathological examination revealed an adenomatoid tumor of the epididymis with similar characteristics to the previous case. After 6 months of follow-up, the patient was asymptomatic without any evidence of local recurrence.

RESULTS AND DISCUSSION

Paratesticular tumors are rare forms, presenting less

than 5-10% of intra-scrotal neoplasms, of which 70-80% are benign (Venyo, 2011). An adenomatoid tumor is a rare benign tumor of mesothelial origin that can be found in both female and male genital tracts. In females, the tumor is usually located in the myometrium, fallopian tubes or ovary hilus. More often it is found in males as an extratesticular mass, and it is one of the most common benign neoplasms. These tumors are more frequent in white males, and in most reported cases they were presented in patients in the third through fifth decades. Predominantly they are localized on the lower or upper pole of the epididymis, followed by the tunica vaginalis and spermatic cord (Evans, 2004).

Adenomatoid tumors are presented as either an incidental finding or a slow-growing scrotal mass. The usual form of presentation is a painless enlargement with normal scrotal skin and surrounding adnexa. At the time of diagnosis, according to ultrasound and pathological studies, the most common diameter of a tumor is 2 to 5 cm. They are presented as circumscribed, small, solid, firm, grayish white nodules, although cystic cases were also identified (Maestro, 2009; Evans, 2004; Williams, 2004).

Histologically, three patterns of tumor have been recognized based on the epithelial cell arrangement: tubular, angiomatoid and plexiform. The adenomatoid (tubular) type is mainly characterized by glandular structures, irregularly lined by cuboidal cells with vacuolated cytoplasm in certain cases, which resemble signet ring cells. The angiomatoid type consists of wide pseudovascular spaces, composed of flattened cells, scant cytoplasm, with an endothelial appearance. The solid (plexiform) type is formed of solid cords of cells with abundant, eosinophilic cytoplasm, with an epithelial-like appearance (Maestro, 2009; Yazaki, 1976; Tavora, 2011).

Immunohistochemically, an adenomatoid tumor exhibits positivity for calretinin and epithelial markers, such as AE1/AE3, epithelial membrane antigen (EMA), Cam5.2, CK5/6, and CK7, by which their mesothelial origin is proved. CK5/6 and CK7 may be only focally positive. Endothelial markers, such

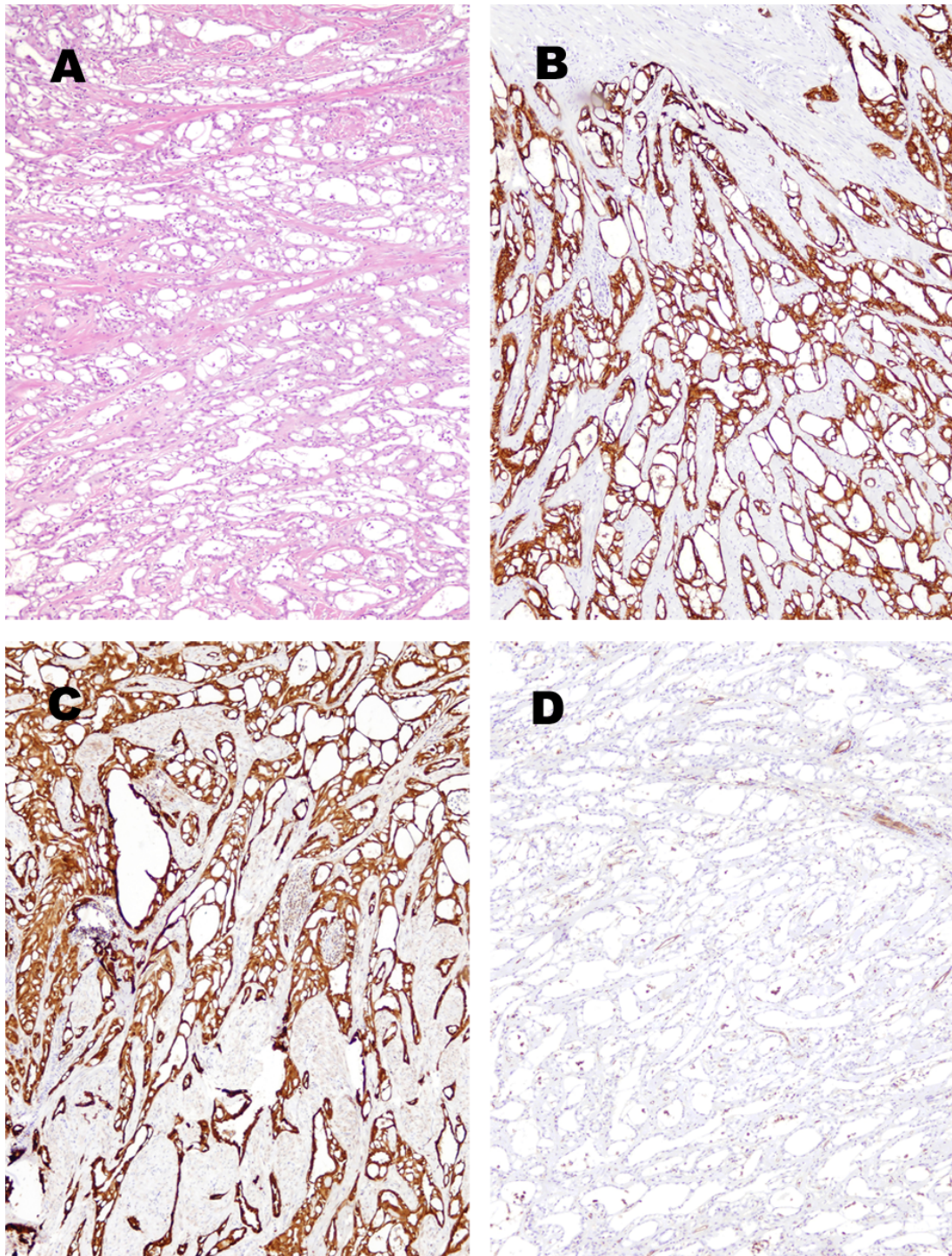


Fig. 1. Histopathological and immunohistochemical findings of the epididymal lesion showing: (A) an adenomatoid tumor of epididymis; tubular structures lined by attenuated cells infiltrate within fibrous and smooth muscle tissue. (B) Tumor cells positive for CK (AE1-AE3). (C) Tumor cells showing positive immunostaining with calretinin. (D) Tumor cells negative for endothelial marker CD31.

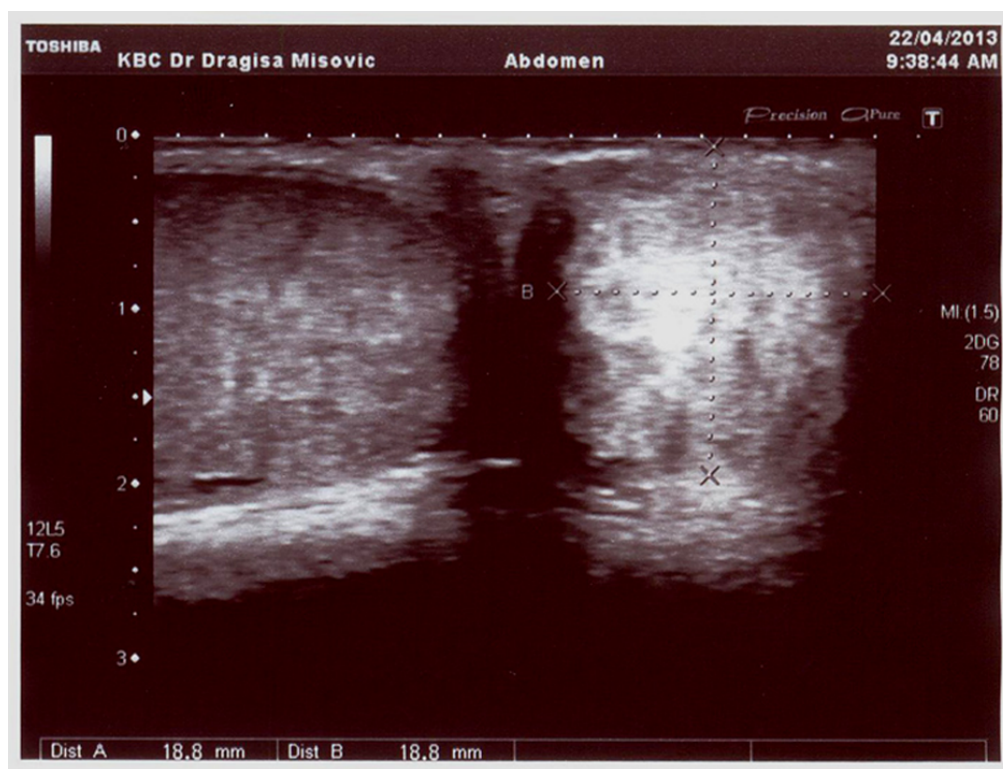


Fig. 2. Ultrasound scan showing a tumor on the right epididymis.

as factor VIII, CD31 and CD34, are negative (Hes, 2003). Referent values of tumor markers, such as β -HCG, AFP and LDH, help to exclude a malignant lesion (Delahunt, 2000; Sangoi, 2009).

Scrotal ultrasound is a useful imaging method to identify the lesion and differentiate it from testicular tumors. It may be hyper-echoic, iso-echoic, hypo-echoic or it may adopt any morphology and does not show significant vascularity with color Doppler examination (Amin and Parwani, 2009). In addition, specific computed tomographic (CT) and magnetic resonance imaging (MRI) can be performed, and these findings, according to tumor localization, morphology and tissue characteristics (attenuation on CT and signal on MRI), can narrow the differential diagnosis (Barry, 2005; Patel, 2004).

Fine-needle aspiration biopsy (FNAB) is not a recommended examination because of possibility of tumor dissemination in a malignant lesion. Some

authors performed extemporaneous biopsy in cases with intra-testicular tumor localization and doubt as to the malignant origin of the tumor (Mateu, 2010).

Local excision of a lesion is an adequate treatment, although some authors performed radical orchiectomy (tumor localization in testis) (Amin and Parwani, 2009; Alam and Maheshwari, 2011).

According to the recent literature data, few studies included analysis of numerous cases. In 2003, Chandrasekar et al. (2003) reported eight cases treated by excision of the tumor. Localizations of the tumors varied: 5 were from the epididymis, 2 from the tunica albuginea and 1 from the testis. Maestro et al. (2009) published a report of nine cases of adenomatoid tumor of which 7 were localized in the epididymis and 2 on the testis. They performed seven removals of the mass and two radical orchiectomies. Similar results were presented by Gonzalez Resina et al. in a study of nine cases with adenomatoid tumor (Gonzalez

Resina et al., 2010). All of them pointed out the importance of valid diagnosis for minimal surgery with preserving testicular function (Maestro, 2009).

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