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증례

고환의 백색막에 발생한 샘모양종양의 압착도말 세포소견

- 1예 보고-

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Touch Imprint Cytology of Adenomatoid Tumor of the Tunica Albuginea
- A Case Report -

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Adenomatoid tumor is a benign neoplasm of a mesothelial origin, and it usually occurs in the reproductive organs, especially in the epididymis. The author experienced a case of adenomatoid tumor involving the tunica albuginea and testicular parenchyme without any evidence of epididymis involvement. The patient was a 36-year-old man with a painless scrotal mass that he had experienced for 2 months, and this mimicked testicular neoplasia, including metastatic carcinoma, or other benign lesions. The imprint cytology of the tumor showed a hypocellular smear with mainly arranged cells in cohesive monolayered clusters along with occasional singly dispersed cells and naked nuclei in a clean background. The cellular clusters formed vague glandular and cord-like structures. The tumor cells were large polygonal to columnar cells with a relatively monomorphic appearance. The nuclei were oval to round shape and they showed vesicular, fine chromatin and inconspicuous nucleoli. The cytoplasm was moderate to abundant, and it contained fine vacuoles in some tumor cells. Mitoses and cellular pleomorphism were not present. Awareness of the cytologic finding of this lesion is necessary to screen or differentiate a testicular or paratesticular mass before and/or during surgery because the cytology may be useful as a diagnostic tool. Pathologists should be aware of the cytologic features of common lesions in this anatomic region so as to avoid performing aggressive and unnecessary surgical procedures.

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Key Words : Adenomatoid tumor, Tunica albuginea, Cytology, Imprint

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INTRODUCTION

Adenomatoid tumor (AT) is a benign neoplasm of a mesothelial origin that usually occurs in the male and female reproductive systems.[1] The epididymis is the most commonly involved site. AT is usually asymptomatic and it presents as small, solid, intrascrotal and extratesticular mass. Most ATs are usually diagnosed based on their anatomic locations, the gross appearance and the characteristic histologic features. However, some ATs with histologic alterations[2] or that have developed in unusual locations[3,4] may cause diag-

nostic difficulty. We report here on a case of an unusual location of adenomatoid tumor that occurred in the tunica albuginea and testicular parenchyme, and this caused confusion for making the differential diagnosis from primary testicular neoplasia, metastatic tumor and other benign lesions before surgery and/or during surgery. Awareness of the cytologic finding of this tumor is very helpful for making the diagnosis, especially together with the touch imprint cytology and the frozen sections. We describe here the imprint cytologic features of this lesion to emphasize the important role of preoperative or intraoperative cytology examination for differentiating this AT tumor from testicular and paratesticular lesions.

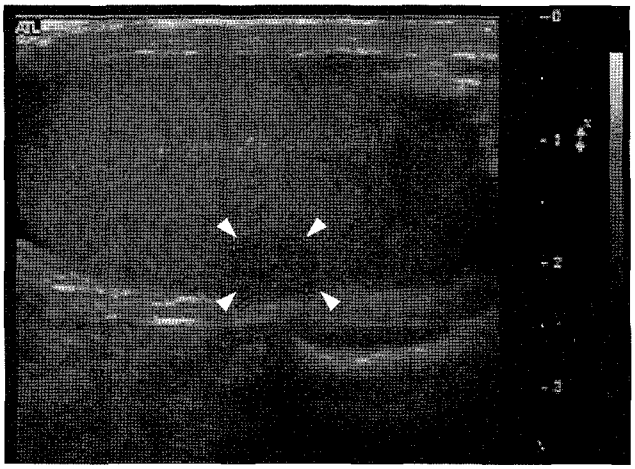

Fig. 1. Scrotal ultrasonography. A. 0.7 cm sized, well-defined homogeneously hypoechoic lesion (arrow head) is noted in the subcapsular portion of the right testis.

CASE

Clinical Presentation

A 36-year-old man visited to the urology department with a painless right scrotal mass that he' d had for two months. There was no history of recent trauma. He denied any urinary and constitutional symptoms. The past medical history was unremarkable, except for a bout of pulmonary tuberculosis 10 years ago. On physical examination, a small, hard, nontender, testicular nodule was palpable in the right scrotum. The serum tumor markers, including beta-human chorionic gonadotrophin (β-HCG), alpha-fetoprotein (α-FP) and lactate dehydrogenase (LDH), were all within normal limits. The clinical features strongly suggested the possibility of seminoma and the less likely possibility of tuberculosis. Chest X-ray revealed no demonstrable active lesion in the both lungs. Scrotal ultrasonography disclosed a 0.7 cm sized, ovoid, well-defined, hypoechoic, homogeneous solid lesion in the subcapsular portion of the right testis and an abnormal fluid collection within the scrotal sac (Fig. 1). The patient underwent exploration and a portion of the lesion was sent for a frozen section diagnosis during the operation. The frozen section revealed an ill-defined lesion that consisted of haphazardly arranged, dilated or collapsed tubular structures and randomly scattered clear spaces which were partially lined by cuboidal or flat endothelial-like cells. Tubular structures and solid epithelial cords were separated by interlacing stromal bundles with varying degree of hyalinization. Primary germ cell tumor was easily ruled out. The differential diagnosis on the frozen section included metastatic adenocarcinoma, sclerosing Sertoli cell tumor, vascular tumor and other benign lesions. The frozen section diagnosis was benign lesion with an inconclusive diagnosis. A right radical orchiectomy was performed. The testis revealed an ill-defined 0.8×0.7 cm sized, triangular shape, hard, solid nodule with a broad base on the tunica albuginea and the apex toward the testicular parenchyme. The nodule had a no relation to the epididymis or the other paratesticular structures.

Cytologic Findings

The imprint cytology showed a less cellular smear. The cellular elements were predominantly scattered with cohesive monolayered clusters together with occasional singly dispersed cells and naked nuclei in a clean background. The tumor cells mainly formed a vague or

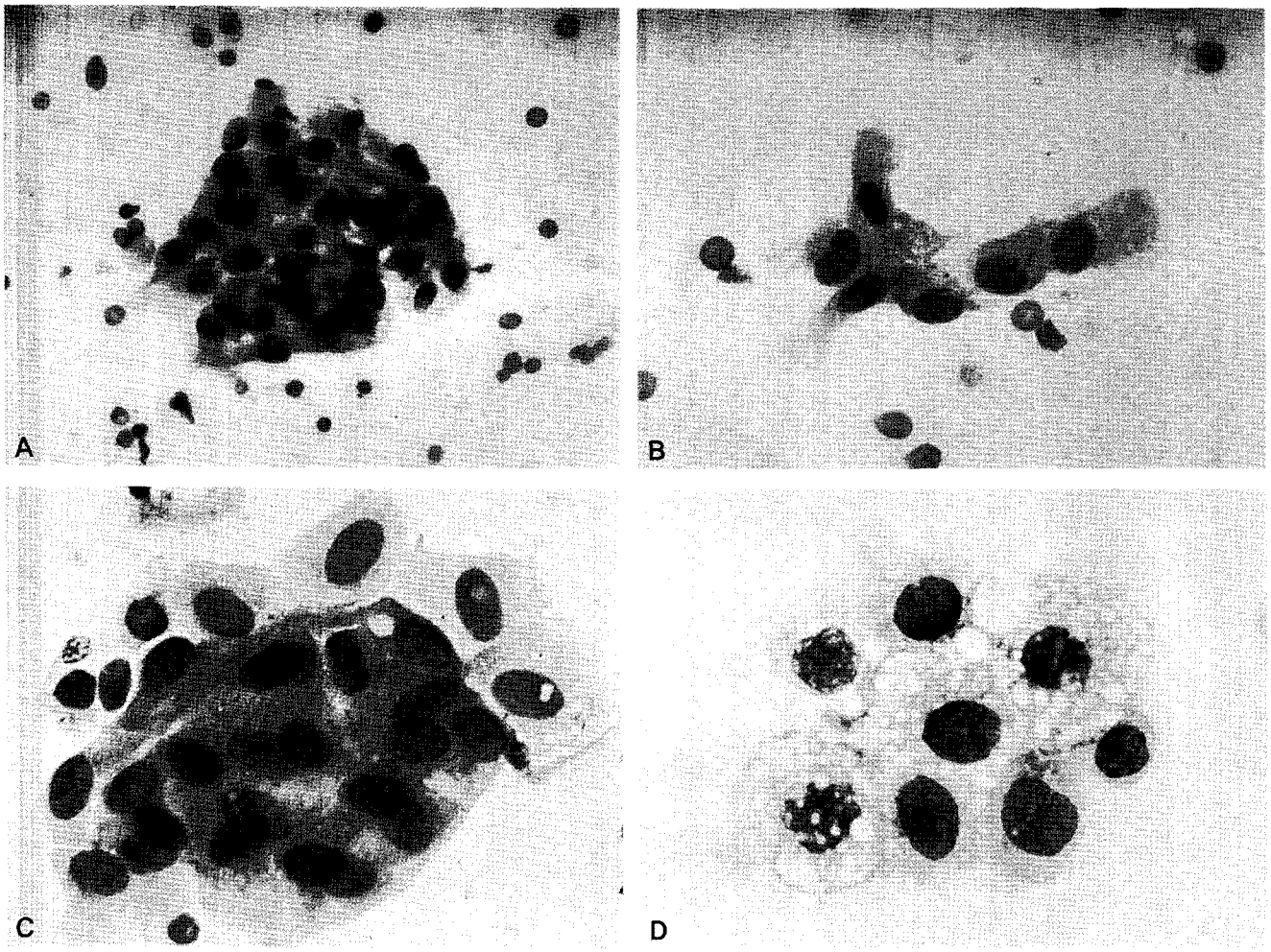

Fig. 2. Cytologic features on touch imprint smears. (A, B) The cohesive mono-layered cellular clusters shows vaguely glandular and cord-like arrangements. (C) The large polygonal to columnar tumor cells with relatively monomorphic appearance exhibit centrally or eccentrically located, oval vesicular nuclei with fine chromatin and inconspicuous nucleoli. The tumor cells have a plump cytoplasm with thin, wispy appearance. (D) Some tumor cells reveal numerous fine vacuolated cytoplasm. (H&E).

abortive glandular pattern and a linear or branched cord-like arrangement (Fig. 2A, 2B). Pattern-less loose cellular aggregates were occasionally present. The tumor cells were large polygonal to columnar with a relatively monomorphic appearance (Fig. 2C). The nuclear/cytoplasmic ratio was low. The nuclei were usually oval and vesicular, and a small number of elongated nuclei were also seen. The nuclei were centrally or eccentrically located and they showed fine and evenly distributed chromatin and inconspicuous nucleoli. Occasionally, a small number of tumor cells contained small or prominent nucleoli. The cytoplasm was moderate to abundant with a thin, wispy appearance. Some tumor cells were finely vacuolated without any nuclear indentation (Fig. 2D). Mitoses and cellular pleomorphism were not present.

Histologic Findings

The tunica albuginea was moderately thickened, and the mass showed as an ill-defined lesion with dilated tubules and microcyst-like clear spaces in a fibrotic background (Fig. 3). Abortive tubular structures and solid cords of epithelial cells of a cuboidal shape were also present. The tubules and cystic spaces were lined by cuboidal to flat epithelial cells that were similar to

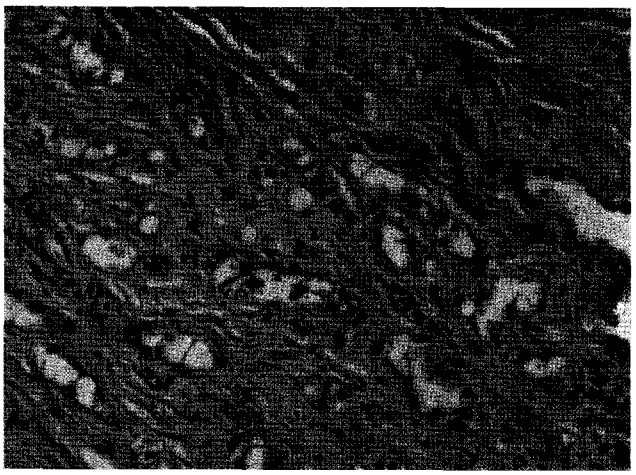

Fig. 3. Histologic findings. Dilated tubular structures with vacuolated cells are scattered. Vacuolatd cells resemble endothelial cells (H&E).

endothelial cells. The tumor cells exhibited prominent cytoplasmic vacuolization. The intraparenchymal portion consisted of smaller capillary-like tubules and clear spaces that were intermingled with lymphoid aggregates and hyalinized stroma. Immunohistochemically, the tumor cells were positive for cytokeratin, but negative for CD34, factor VIII, CEA and EMA. The final diagnosis of adenomatoid tumor was made.

DISCUSSION

Adenomatoid tumor (AT) is the most common benign neoplasm of the testicular adnexa, and it accounts for 32% of all the tumors in this location.[1] In spite of its prevalence, the cytologic reports concerning this entity are limited because the majority of ATs can be diagnosed by their typical anatomic location upon physical examination and the imaging findings. Pathologists rarely encounter the cytologic specimen of this tumor.

In this case, the patient showed a solid testicular nodule that clinically and radiologically mimicked an intratesticular neoplasm, particularly seminoma. Intraoperative frozen section excluded primary germ cell tumors and vascular tumors. However, the infiltrative growth of tubular and cord-like structures of cuboidal cells in the fibrohyaline background called for differentiating metastatic adenocarcinoma and sclerosing Sertoli cell tumor (SSCT). Additionally, the mixed cellular components, including the tubular epithelial nests, stromal cells and lymphoid aggregates suggested reactive lesions, including mesothelial hyperplasia. The post-operative imprint cytologic smears revealed vague or abortive glandular structures, linear or branched cord-like arrangements and simple, loose cellular aggregates. Metastatic adenocarcinoma was excluded because the cellular elements were arranged in predominantly monolayered cohesive clusters in a clean background and the tumor cells had a relatively monomorphic appearance that was characterized by the low nuclear/cytoplasmic ratio, the eccentrically placed vesicular nuclei with fine chromatin and inconspicuous nucleoli and the moderate to abundant cytoplasm. There were no tridimensional ball-like or papillary structures.

SSCT is a rare and biologically indolent neoplasm of the testis that usually presents as a small well demarcated intraparenchymal nodule in adults. Microscopically, SSCT are characterized by irregular cellular aggregates, anastomosing trabeculae, tubular structures and thin cords of Sertoli cells in a prominent collagenous background.[5,6] Although rare, we also considered the possibility of this entity on the frozen sections. Generally, benign Sertoli cell tumors, not otherwise specified (NOS), consist of less cohesive cellular aggregates and singly scattered cells.[7,8] Naked nuclei are prominent in some case.[8] Neoplastic Sertoli cells are polygonal and they show abundant finely granular eosinophilic cytoplasm and eccentrically located round to oval nuclei with fine chromatin. Terayama et al.[8] emphasize the presence of coffee bean nuclei and nuclear indentations in some tumor cells of benign SCT. In this case, the presence of cohesive monolayered clusters and the lack of coffee bean nuclei and nuclear indentations favored AT over SCT. However, the cytological differentiation between AT and SSCT may be difficult

because the two neoplasms share histologic features. Reactive mesothelial hyperplasia was also considered for the diagnosis. Most of reactive mesothelial hyperplasia usually show hypercellular smears that contain large tissue fragments, single cells with abundant cytoplasm, and large vesicular nuclei. Reactive mesothelial cells exhibit a mild to moderate degree of anisocytosis and pleomorphism. Nuclear grooves, lobulation and mitosis may be observed. In the present case, the cytology findings were different from reactive mesothelial lesion according to the monolayered, relatively monomorphic cellular clusters.

The cytologic features of this case were similar to those of the previous reports[3,9,10] but the overall cellularity was low and any stromal fragments were not identified. This limitation may be related to the different methods to obtain cytologic specimen such as FNAC, direct scraping and just touch imprint.

The imprint cytology is helpful to make the diagnosis when frozen sectioning is performed. This combination method may easily differentiate the testicular and paratesticular lesions including metastatic adenocarcinoma and reactive mesothelial hyperplasia. The pre- or intraoperative cytology may be used as an important diagnostic tool to evaluate the testicular and paratesticular masses. Pathologist should be aware of the cytologic features of common lesions to avoid aggressive or unnecessary surgical procedures.

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