Adenomatoid Tumor of Epididymis: A Case Report with Correlation Between Histology and Cytology

Mega Lahori1*, Sakul2, K C Goswami3 and Arvind Khajuria3

1Dept of Pathology, Acharya Shri Chander College of Medical Sciences Jammu (J&K), INDIA
2Dept of Medicine, Acharya Shri Chander College of Medical Sciences Jammu (J&K) INDIA
3Dept of Pathology, Acharya Shri Chander College of Medical Sciences Jammu (J&K)

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ABSTRACT

Adenomatoid tumor is a rare and distinctive, benign mesothelial neoplasm of the paratesticular region in males, most commonly occurring at the tail of epididymis and constitutes 30% of paratesticular neoplasms. We present a case of adenomatoid tumor in a 35 year old male, who presented with a mass in right epididymis and was diagnosed by fine needle aspiration cytology and later confirmed on histopathology. The clinical, histopathological and cytological aspects of this rare case are discussed. FNAC plays a pivotal role in the preoperative diagnosis of these tumors as it is a simple, inexpensive, minimally invasive and reliable diagnostic modality. Due to its benign nature, the treatment of choice is local excision. We believe it is important for the physician to be aware of this interesting entity in order to make an accurate diagnosis and prevent unnecessary orchiectomy.

*Corresponding author:
Dr Mega Lahori, PG Resident, Deptt of Pathology, Acharya Shri Chander College of Medical Sciences Jammu (J&K) 180017, INDIA
Phone: +91 9419177133
E-mail: iammegha00@gmail.com
Introduction

Adenomatoid tumor is a rare and benign neoplasm of mesothelial origin seen in male as well as female genital tract along with extragenital sites, but is more common in male adnexa. It is seen usually in the third & fourth decades (mean age is 36 yrs).[^1] Adenomatoid tumor was first described in 1945 by Golden and Ash as a small, firm asymptomatic intrascrotal mass, with no pain or tenderness, occurring in third to fifth decades of life. These tumors represent 30% of the paratesticular tumors. Beccia et al studied 314 epididymal tumors, of which 75% were benign and 73% of those were diagnosed as adenomatoid tumors, followed by leiomyomas (11%), and papillary cystadenomas (9%). These tumors are benign in nature (even when they infiltrate into the adjacent sites), with no reported cases of malignant change, metastasis, or relapse after removal, and tumor excision is therefore the treatment of choice.[^1] Accurate diagnosis is the key to prevent unnecessary orchiectomy.[^3]

Case Report

A 35 year old male presented at the Urology OPD of our hospital with one year history of a slowly enlarging and painless right sided scrotal mass (felt at the lower pole of epididymis) which was nontender, non fluctuant and non transilluminant on local examination. Personal and family history was unremarkable. There was no history of epididymitis, torsion or trauma. Aspiration from the mass was done using a 10 ml syringe and 24G needle to obtain a scanty, whitish aspirate. FNAC from the mass revealed moderately cellular smears composed of monomorphic, round to oval tumor cells arranged in sheets and multilayered clusters. The cells were round to oval with pale vacuolated cytoplasm, eccentric nucleus having fine chromatin and small, central nucleolus within a background of naked nuclei and stromal bits (Fig 1). A cytological diagnosis of adenomatoid tumor was made; following which the patient underwent a conservative testis sparing surgery with the excision of epididymal nodule which was received in the Histopathology section of our Deptt of Pathology. Grossly, the nodule was globular, measured 1.5x1.3cm in diameter and was grayish-white on cut section (Fig 2a). Histopathological examination revealed a well circumscribed, non encapsulated tumor with characteristic features of adenomatoid tumor composed of tubules and cords of cuboidal cells having large intracytoplasmic vacuoles and gaping spaces (Fig 2b). The cellular vacuoles had a signet ring-like appearance in some fields. There was no mitotic activity or nuclear atypia. Prominent lymphoid collections were seen in a fibroblastic stroma, which is another important clue to the diagnosis. Resection margins were free of tumor cells. This tumor was diagnosed as adenomatoid tumor and the patient underwent no additional treatment.
Discussion
Most Adenomatoid tumors are asymptomatic masses reported by the patient and, generally remain unchanged in size for years. In males they occur in the epididymis, spermatic cord, prostate and ejaculatory duct. Mostly they arise at the lower pole of the epididymis. In females they occur in uterus, fallopian tubes and ovarian hilus.[1] They are also seen in extragenital sites like adrenal gland, lymph nodes, mediastinum, pleura, pancreas and heart in either sex.

They usually present as small (~2cm), solid, firm masses and on cut section, they appear grayish-white & homogeneous (the excised nodule in the present case also had solid, homogeneous cut surface). Occasionally, small cysts may be seen. These tumors are usually well circumscribed but non-encapsulated. They have a plethora of microscopic appearances, represented by four basic patterns: adenomatoid (tubular), angiomatoid (canalicular), plexiform (solid nests) and cystic (mixed) [the tumor in the present case had an adenomatoid (tubular) histological pattern]. Cells are cuboidal to flattened, with weakly eosinophilic & markedly vacuolated cytoplasm. Nucleus is eccentric with fine chromatin and small, central nucleolus and little to no mitotic activity. Cells may form solid cords alternating with channels having dilated lumina simulating vascular structures. Cystically dilated gaping spaces with no evident lining, representing a necrotic tubular component, and smaller gland-like spaces (giving the appearance of vascular spaces) are major clues to the diagnosis. Stroma is prominent and fibrous, with abundant smooth muscle and elastic fibers. Additional stromal features may be prominent lymphoid follicles, extensive necrosis, abundant desmoplasia and signet-ring like cells (in clusters or individually scattered).

The first cytological description of adenomatoid tumors was given by Perez-Guillermo et al in 1989. Cytology reveals sheets, cords, glandular patterns or multilayered clusters of monotonous, round to oval cells with pale, vacuolated cytoplasm. Nucleus is spherical & eccentric with fine chromatin and small central nucleolus.[4] They are considered to be of mesothelial origin (as originally proposed by Masson et al), which is supported by immunohistochemical studies (positivity for HMBE 1 & Calretinin) and genetic analysis of WT-1 gene expression.[1] These tumors show positive immunoreactivity for Calretinin, Epithelial markers (AE1AE3, EMA, Cam5.2, CK5/6, CK7), Vimentin, WT1, HMBE 1, D2-40 and are negative for Carcinoma markers (CEA, CD15, B72.3, MOC-31, Ber-ep4), Endothelial markers (FVIIIRA, CD31, CD34), Germ cell tumor markers (OCT 3/4, Nanog, Sox-2, AFP, PLAP) and Inhibin. Diagnosis is based mainly on histopathology, aspiration cytology & ultrasonography, which is supported by immunohistochemistry.[5,6] The ultrasound features of adenomatoid tumors may vary but usually include a well-defined, homogeneous, round, hyperechoic nodule. The differential diagnosis includes epithelioid hemangiomia, malignant mesothelioma, metastatic adenocarcinoma, papillary cystadenoma of epididymis, large cell calcifying Sertoli cell tumor, Epididymidal carcinoma, testicular rhabdomyosarcoma, and carcinoma of rete testis. Malignant mesotheliomas usually show abundant necrosis and mitoses while papillary cystadenomas can be differentiated on the basis of numerous papillary infoldings which project into cystic spaces. The absence of staining of epithelial markers is of utility in excluding carcinomas (CEA, PSA, MOC 31/BerEP4) and germ cell tumors (OCT 3/4, Sox2, AFP, PLAP) from the differential diagnosis, while negativity of vascular markers (CD34, factor VIII) excludes a diagnosis of epithelioid hemangiomia.

Conclusion
Adenomatoid tumor should be suspected in any intrascrotal mass lesion. Its separation from testicular tumors is important as paratesticular tumors have good prognosis compared to testicular tumors. We are highlighting the strong correlation between cytology and histology features. Both diagnostic modalities reveal a monotonous proliferation of tumor cells with similar cellular features. FNAC, as a preoperative diagnostic tool, can help to plan surgery as complete local excision of this benign tumor is both diagnostic as well as therapeutic and has had no reported case of recurrence or metastasis after excision. The main clinical consideration is accurate diagnosis in order to prevent unnecessary orchiectomy and preserve endogenous testicular function.

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