

Adenomatoid tumor epididymis with infarction: Report of a rare case with Review of Literature.

Avni Bhatnagar, Karsing Patiri, Usha Singh.

Department of Pathology, Dr. Baba Saheb Ambedkar Hospital and Medical College, New Delhi, India.

1. Dr. Avni Bhatnagar, M.D. (Pathology), Senior Resident, Dept. of Pathology, Dr. B.S.A Hospital Rohini, New Delhi-110085 (India)
2. Dr. Karsing Patiri, M.D. (Pathology), Associate Professor, Dept. of Pathology, Dr. B.S.A College & Hospital, Rohini, New Delhi-110085.
e mail- drkarsingbsamch@gmail.com
3. Dr. Usha Singh, M.D. (Pathology), Consultant Pathology, Dept. of Pathology, Dr. B.S.A College & Hospital, Rohini, New Delhi-110085 (India). e mail- ushaucms@gmail.com
Corresponding Author*: Dr. Avni Bhatnagar, M.D. (Pathology)
Senior Resident, Dept. of Pathology, Dr. B.S.A Hospital,
Rohini, Sector-6, New Delhi-110085

Abstract:-

Adenomatoid tumor is a rare benign mesothelial neoplasm of para testicular region and mainly involves the epididymis. It is important to make a definite diagnosis because of difficulty in differentiating it clinically and radiologically from other intra scrotal lesions. We report a case of 52-year male presenting with swelling in right scrotum. High inguinal orchidectomy was performed and the histopathological examination confirmed diagnosis of adenomatoid tumor of epididymis. Based on its rarity we hereby, review the literature and discuss the clinical and histopathological aspects.

Keywords: Adenomatoid tumor, para testicular mass, epididymis, infarction, orchidectomy.

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I. Introduction: -

Para testicular neoplasms are rare and accounts for less than 10% of all intra scrotal tumors and most of them are benign in nature.¹ Adenomatoid tumor is a benign mesothelial neoplasm of the para testicular tissue, accounting for approximately 30%.² Most common sites of adenomatoid tumor are the male and female genital tract.³ Majority of the patients are asymptomatic. Most of the tumors are small, with most measuring less than 2cm. Becca et al found that adenomatoid tumor accounted for 55% of all epididymal tumors, followed by leiomyomas (11%) and by papillary cystadenomas in (9%).⁴ Surgical excision is required for histopathological diagnosis and to rule out other malignant tumors such as malignant mesothelioma, carcinoma rete testis and para testicular yolk sac tumor. Biopsies are contraindicated due to scrotal skin metastasis.⁵

II. Case Report: -

A 52-year male patient presented to our hospital with complaints of swelling in right scrotal region for the past 14-15 years. His only complaint was pain on and off in the scrotum. There was no history of trauma, hematuria, or difficulty in micturition. He was a tobacco chewer. Rest of the personal and family history was non-contributory. His serum LDH levels were mildly increased- 450U/L.

USG scrotum showed enlargement of right testis (6.5x4) cm with mixed echogenicity and altered echotexture. Large cystic areas were seen within. Few foci of calcification were also seen. No evidence of internal vascularity noted. Other side testis and epididymis were unremarkable (Fig-4). CECT whole abdomen showed loculated hypodense cystic lesion measuring (48x52x50) mm at upper pole of right testis. The tumor appeared to be involving the head of right epididymis. Few foci of calcification were seen in the cyst wall. Impression of benign epididymal cyst was suggested (Fig-5). Right high inguinal orchidectomy under spinal anesthesia was performed.

Gross examination revealed a cut open cystic mass (7x7x3) cm along with attached spermatic cord measuring 8cm in length. Serial sectioning showed a single cystic mass measuring (6x5) cm filled with necrotic and friable material. Normal testis was pushed towards the periphery (Fig-1). Microscopic examination revealed normal testis along with adenomatoid tumor of epididymal origin. The tumor cells were arranged in tubules

lined by cuboidal to flattened epithelium with large cytoplasmic vacuoles along with large areas of infarction (Fig-2 & Fig-3).

III. Discussion: -

Para testicular tumors are rare and benign in nature with adenomatoid tumor being the commonest one.⁶ At our institute we received 83 orchidectomy specimen over a period of five years (January -2018 to August 2022) and out of them only one case was of Adenomatoid tumor, rest came out to be granulomatous orchitis, chronic epididymo-orchitis or testicular neoplasm. Adenomatoid tumors were first reported in 1945 by Golden and Ash.⁷ Adenomatoid tumors have mostly been reported in the head of epididymis. The spermatic cord, prostate and ejaculatory ducts are also reported as affected sites.^{3,8,9} Adenomatoid tumors rarely involve the testicular parenchyma and present as intratesticular masses.^{8,10} In females, the uterus, fallopian tube, and ovary can also be affected.¹¹ Other than the genital organs mentioned above, adenomatoid tumors have also been found in the adrenal gland.¹²

Adenomatoid tumor may be seen in all ages, but most cases are seen in the third to fifth decades of life.³ Age group of patients reported in the literature ranged from 18-80 years.¹³ The origin of the adenomatoid tumor is uncertain. In the literature, variety of theories indicates that adenomatoid tumor is of mesothelial nature, as originally proposed by Masson et al.¹⁴ The structural and immunohistochemical studies support the mesothelial origin.

Clinically, patient presents as small, solid intra-scrotal mass which are found incidentally. Rarely adenomatoid tumor of epididymis is associated with pain and sometimes confused with torsion of the testis. Testicular tumors are also sometimes associated with pain so should be kept as differential diagnosis. Though infarction is rare and is associated with trauma as reported by Gupta et al¹⁵, in our case the patient though did not give any history of trauma, but the swelling gradually presented for 14-15 years. Histology showed infarction with calcification. Predominantly adenomatoid tumor is located on the left side than the right side and occurs in or near the lower pole or upper pole of the epididymis. A slightly higher incidence in the lower pole has been reported.¹⁶ Mostly adenomatoid tumor present as slow growing and smaller in size ranging from 1-5cm but few cases also reported up to 12 cm.¹⁴ In our case right upper pole of testis with head of epididymis was involved, pain was on and off and size of the tumor was large measuring (6x5) cm.

Microscopically, it shows cells arranged in cords, tubules and microcystic spaces with cuboidal to flattened epithelium along with vacuolated cytoplasm. The stroma is usually fibrous containing smooth muscle. Differential diagnosis includes chronic epididymo-orchitis, tuberculosis, leiomyoma, hemangioma, papillary cystadenoma, testicular neoplasm, metastatic carcinoma, malignant melanoma, and carcinoma of rete testis. Histomorphology and immunohistochemical features (calretinin and HMBE1) of these tumors support their mesothelial origin. Intermittent testicular torsion can lead to testicular ischemia and possibly infarction. It is characterized by repeated attacks of pain in scrotum and asymptomatic periods in between the episodes.¹⁷ It is less common in adults so often missed or misdiagnosed.¹⁸ Recurrent attacks of intermittent testicular torsion can lead to ischemic damage to testis.¹⁹ It can cause venous congestion with or without decreased arterial flow and leads to testicular damage. On histology, we can see atrophic seminiferous tubules, peritubular fibrosis, or lack of spermatogenesis.^{17,20,21,22} In our case adenomatoid tumor was accompanied by infarction and calcification. Possibility of torsion due to adenomatoid tumor cannot be excluded. Torsion would also explain the pain, infarction, and calcification. Infarction with calcification can be seen without a definite history of trauma.

No case of recurrence or metastasis have been reported after surgical excision.²³ Proper management includes orchidectomy with histopathological and radiological correlation.

Our case is unique because of: -

- 1) Long history of 14-15 years.
- 2) Large size.
- 3) Pain off and on.
- 4) Large area of infarction and foci of calcification.

Legends: - Adenomatoid tumor, Epididymis



Fig 1: - Gross specimen of Adenomatoid tumor, epididymis showing cystic area filled with necrotic tissue with normal testicular tissue (marked by black arrow) pushed to periphery.

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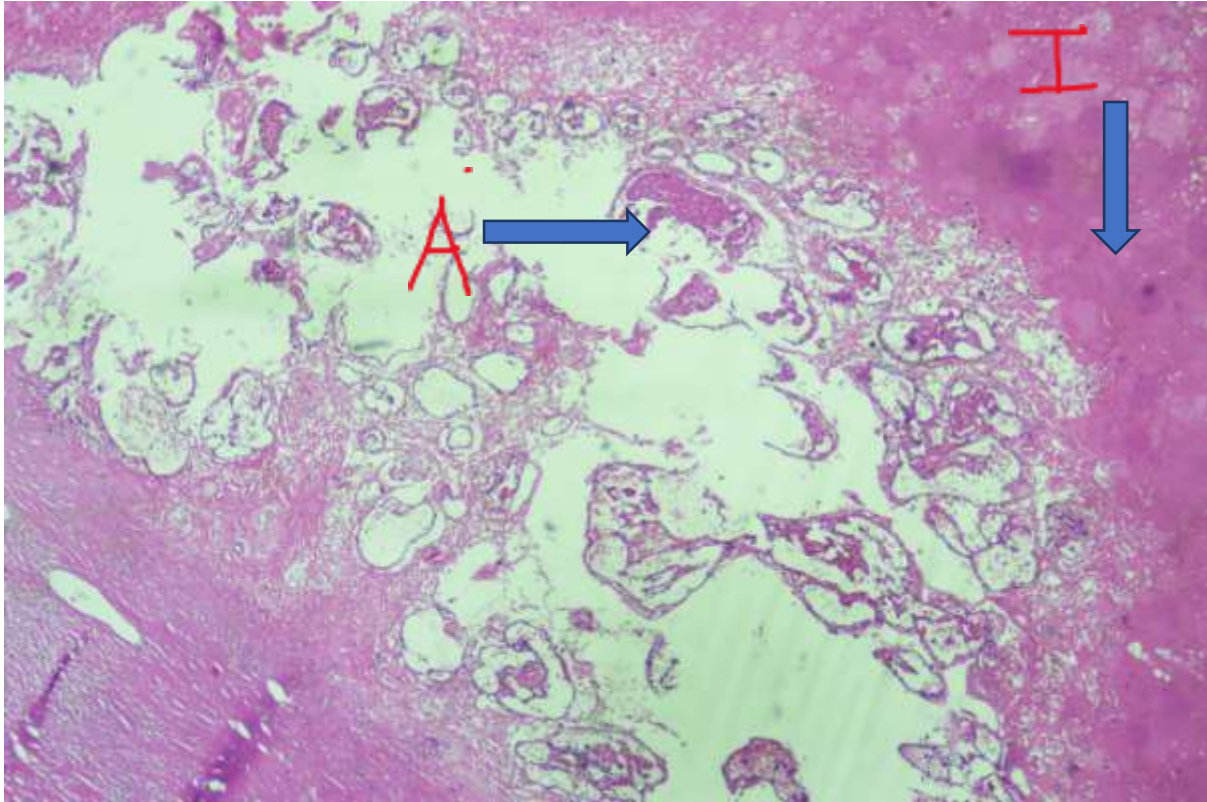


Fig 2: - Histological pattern of Adenomatoid tumor 4X10 (H&E), tumor cells showing tubular or nesting pattern along with large areas of infarction necrosis.

- I = Infarction
- A = Adenomatoid tumor

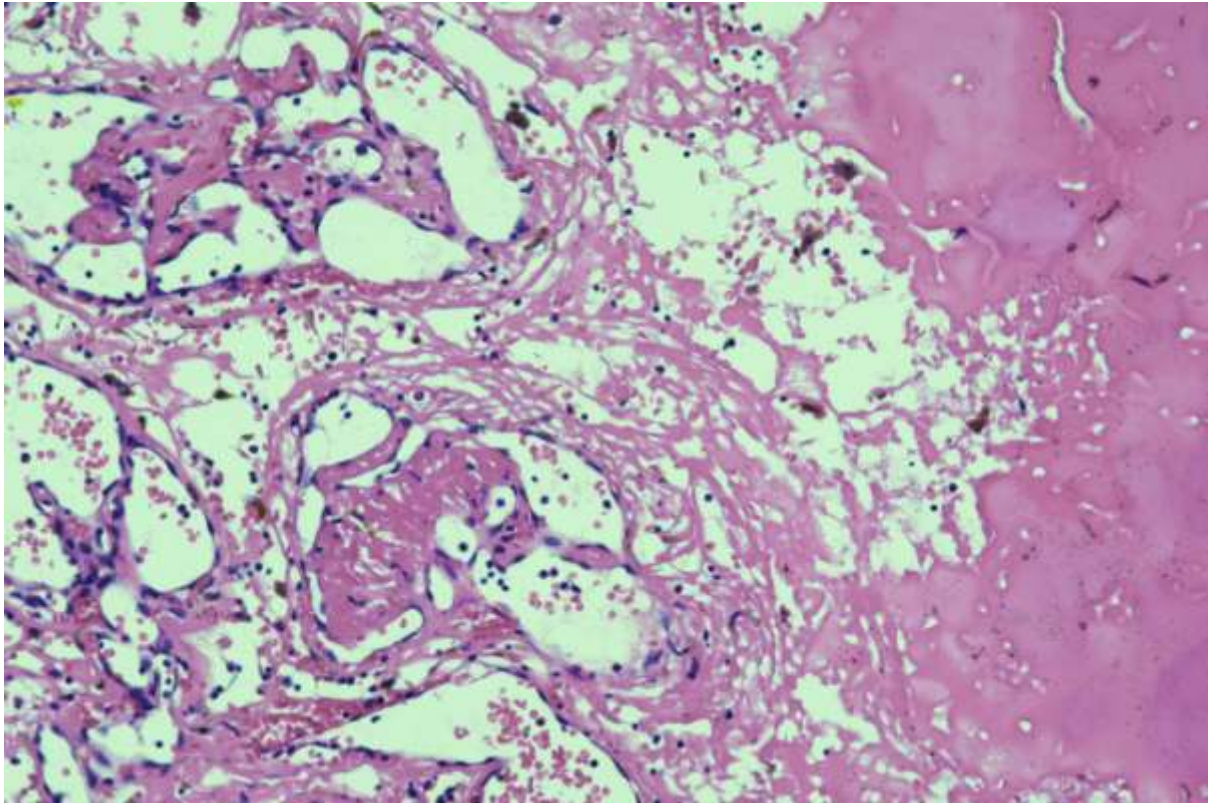


Fig 3: - Histological pattern of Adenomatoid tumor 10x10 (H&E), the tumor is lined by cuboidal cells with vacuolated cytoplasm along with areas of infarction necrosis.



Fig 4: - Ultrasonography of scrotum showing testicular degeneration with enlargement of size measuring 6.5x4.1 cm with large cystic areas and few foci of calcification. There is no evidence of internal vascularity.



Fig 5: - Computerized tomography scan showing loculated hypodense cystic lesion at the upper pole of right testicle, appears to be involving the head of right epididymis showing few foci of wall calcification.

Abbreviations: -

LDH- lactate dehydrogenase, USG- ultrasonography, CECT- high dose contrast-enhanced computed tomography.

Declaration of patient consent: -

The authors certify that they have obtained all appropriate consent forms. In the form the patient(s) has/have given his/her consent for his/her images and other clinical information to be reported in the journal. The patients understand that their name and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest: -

There are no conflicts of interest.

References: -

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