

A Diagnostically Challenging Case of an Infarcted Adenomatoid Tumor of the Epididymis

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We describe a case of an infarcted adenomatoid tumor of the epididymis that was challenging to diagnose. A 20-year-old man presented with acute left scrotal pain. He was found to have a 2 × 1.5 × 1 cm tumor that was relatively well circumscribed on gross examination. There was a central necrotic area that exhibited gaping spaces and ghost outlines of epithelial cells. The periphery of the necrotic lesion showed focally viable adenomatoid tumor. The majority of the tissue adjacent to the necrosis consisted of granulation tissue, fibroblastic and myofibroblastic proliferation, and neutrophils. The fibroblasts and myofibroblasts showed plump nuclei, often with small nucleoli. No mitotic activity was present. The differential diagnosis for an infarcted adenomatoid tumor includes malignant mesothelioma, inflammatory myofibroblastic tumor, and inflammatory conditions. The key to diagnosing an infarcted adenomatoid tumor is to consider it in the differential diagnosis of any spindle cell tumor with necrosis occurring in the genital tract.

Key Words : Adenomatoid tumor; Infarction; Epididymis

Adenomatoid tumors are relatively rare benign genital tract neoplasms of mesothelial origin.¹⁻⁴ Based on their characteristic gross and microscopic appearance, they are easily diagnosed in most cases. However, infarction of an adenomatoid tumor may obscure the nature of the underlying lesion and cause diagnostic difficulty. We describe a case of an infarcted adenomatoid tumor of the epididymis that proved to be a diagnostic challenge.

CASE REPORT

A 20-year-old man presented with acute left scrotal pain. He had had a palpable left scrotal mass for the past 4 years. Physical examination of the scrotum revealed a well-defined, solid, firm nodular mass in the head of the epididymis, measuring 2 × 2 cm. Ultrasonography of the scrotum revealed an echogenic mass, suggestive of epididymitis with abscess formation. The patient underwent an epididymectomy.

The mass was relatively well-circumscribed, measured 2 × 1.5 × 1 cm, and showed central necrosis. Histologically, the necrotic area consisted of central nondescript necrosis with no recognizable underlying pattern, along with peripheral, pale-staining, mummified adenomatoid tumor cells (Fig. 1). Areas adjacent to the necrosis showed fibroblastic and myofibroblastic proliferation admixed with viable adenomatoid tumor components (Fig. 2). The fibroblasts and myofibroblasts had plump nuclei, often with small nucleoli. The viable tumor was composed of columns of polygonal cells with eosinophilic cytoplasm, prominent nucleoli, and occasional vacuoles, in a predominantly solid growth pattern. No mitotic figures were identified. Immunohistochemical staining for cytokeratin showed positive reactions in both viable and necrotic areas (Fig. 3). Immunohistochemical staining for calretinin was positive in the viable area and negative in the necrotic area. The soft tissue surrounding the mass contained marked fibrin deposition and a patchy infiltrate of acute and chronic inflammatory cells.

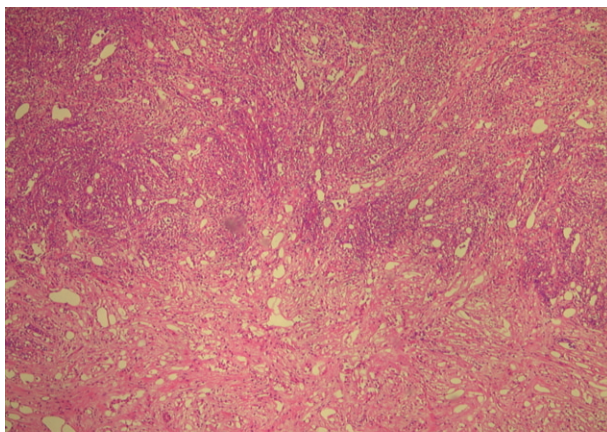


Fig. 1. Necrotic area with no recognizable underlying pattern (top) is surrounded by pale-staining area with ghost outlines of typical pattern of adenomatoid tumor (bottom).

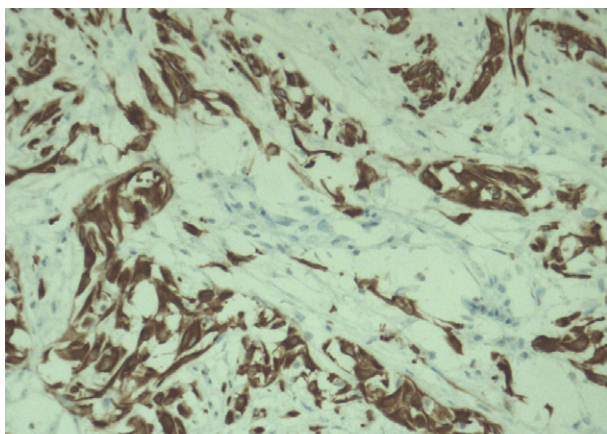


Fig. 3. The tumor cells show positive immunohistochemical reaction for cytokeratin.

DISCUSSION

Adenomatoid tumors are usually asymptomatic and well-demarcated, and they exhibit a typical microscopic pattern of tubules, cords, and nests.^{5,6} Adenomatoid tumor infarction was first described by De Klerk *et al.*, and the clinicopathologic features of five cases were described by Skinnider and Young.^{2,7} The clinicopathologic features of infarcted adenomatoid tumors-including our case-differ from otherwise typical adenomatoid tumor. Our patient, along with two patients described by Skinnider and Young, presented with acute scrotal pain simulating epididymitis, which is an unusual presentation for an adenomatoid tumor.⁷ Although infarcted adenomatoid tumors are usually well-circumscribed, reactive fibroblastic and myofibroblastic proliferation may blur the boundaries of the mass. One case in the Skinnider and Young series showed pseudoinfiltration of fat together

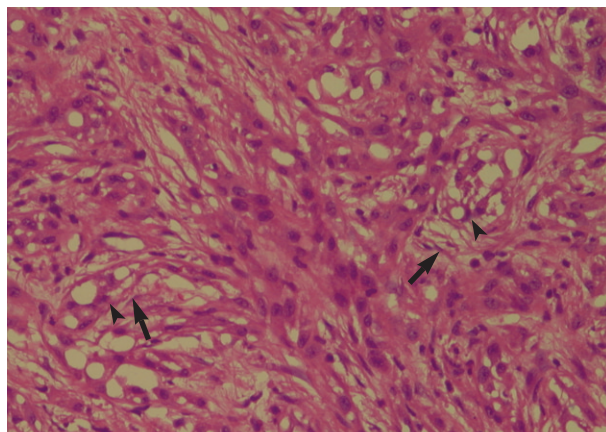


Fig. 2. Fibroblastic proliferation (arrows) merges imperceptibly into the viable adenomatoid tumor (arrowheads).

er with reactive tissue.⁷ Immunohistochemistry was helpful in accentuating the well-circumscribed border of the tumor.

Solid-like pattern is common in infarcted adenomatoid tumors. This pattern may increase the concern for the possibility of malignancy. However, it is not a true solid or diffuse pattern, but represents closely packed tubules, cords, and nests, probably as a result of the tissue reaction.

The differential diagnosis for an infarcted adenomatoid tumor includes malignant mesothelioma, inflammatory myofibroblastic tumor, and inflammatory conditions. Malignant mesothelioma should be considered due to spindle cell proliferation with atypia and necrosis. Careful assessment of the gross appearance is an important first step in sorting out this differential diagnosis. Malignant mesothelioma typically presents as multiple irregular masses that commonly have a grossly papillary appearance coating the tunica albuginea.⁵ Adenomatoid tumors do not have the typical well-defined papillary pattern of malignant mesotheliomas. Both tumors commonly have a tubular growth pattern, but adenomatoid tumors have tubules that are more elongated, with more flattened cells than are seen in malignant mesotheliomas. The tubules of malignant mesothelioma are round and hollow. The absence of mitosis also supports the diagnosis of adenomatoid tumor. Some cases of malignant mesothelioma have bland cytologic features with rare mitoses, but these are typically in areas with a striking papillary pattern.⁵

Because of the presence of granulation tissue and fibroblastic and myofibroblastic proliferation admixed with inflammatory infiltrate in a loose myxoid stroma, an infarcted adenomatoid tumor may be misdiagnosed as an inflammatory myofibroblastic tumor.^{4,8} However, careful examination of the tumor will allow for identification of solid areas of viable adenomatoid tumor. Immuno-

histochemical stains for calretinin and cytokeratin are useful for distinguishing between adenomatoid tumors and inflammatory myofibroblastic tumors.⁹ It should be noted that necrosis is uncommon in inflammatory myofibroblastic tumors.

Finally, an adenomatoid tumor with almost total infarction and a heavy neutrophilic infiltrate may be misdiagnosed as epididymitis. On histological sections, gaping spaces with no evident lining are major clues for the diagnosis.^{6,7} However, diagnosis may be difficult to make based on fine needle aspiration cytology (FNAC). Singh *et al.* described a case of chronic epididymitis misdiagnosed as an adenomatoid tumor on FNAC.¹⁰ Clusters of elongated to round epithelial cells were aspirated from hyperplastic epithelium lining the epididymal tubules, which led to the suggestion of an adenomatoid tumor. Epididymectomy may be curative, as well as diagnostic, in selected cases.

The cause of infarction remains unknown. Skinnider and Young suggested trauma because the majority of tumors were from the epididymis and testis, which are more susceptible to trauma. However, no documented history of trauma has been present in any of the reported cases, including ours.

In summary, we have described a case of an infarcted adenomatoid tumor. The key to diagnosing this entity is to consider this condition in the differential diagnosis of any spindle cell tumor with necrosis occurring in the genital tract.

REFERENCES

1. Broth G, Bullock WK, Morrow J. Epididymal tumors. 1. Report of 15 new cases including review of literature. 2. Histochemical study of the so-called adenomatoid tumor. *J Urol* 1968; 100: 530-6.
2. de Klerk DP, Nime F. Adenomatoid tumors (mesothelioma) of testicular and paratesticular tissue. *Urology* 1975; 6: 635-41.
3. Goddard MJ, Grant JW. Adenomatoid tumours: a mucin histochemical and immunohistochemical study. *Histopathology* 1992; 20: 57-61.
4. Hollowood K, Fletcher CD. Pseudosarcomatous myofibroblastic proliferations of the spermatic cord ("proliferative funiculitis"): histologic and immunohistochemical analysis of a distinctive entity. *Am J Surg Pathol* 1992; 16: 448-54.
5. Perez-Ordenez B, Srigley JR. Mesothelial lesions of the paratesticular region. *Semin Diagn Pathol* 2000; 17: 294-306.
6. Amin MB. Selected other problematic testicular and paratesticular lesions: Rete testis neoplasms and pseudotumors, mesothelial lesions and secondary tumors. *Mod Pathol* 2005; 18 (Suppl 2): S131-45.
7. Skinnider BF, Young RH. Infarcted adenomatoid tumor: a report of five cases of a facet of a benign neoplasm that may cause diagnostic difficulty. *Am J Surg Pathol* 2004; 28: 77-83.
8. Lam KY, Chan KW, Ho MH. Inflammatory pseudotumour of the epididymis. *Br J Urol* 1995; 75: 255-7.
9. Delahunt B, Eble JN, Nacey JN, Thornton A. Immunohistochemical evidence for mesothelial origin of paratesticular adenomatoid tumour. *Histopathology* 2001; 38: 479.
10. Singh I, Dev G, Singh N. Chronic epididymitis (epididymal nodule) mimicking an adenomatoid tumor—case report with review of literature. *Int Urol Nephrol* 2002; 34: 219-22.