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Adenocarcinoma of the Rete Testis

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Adenocarcinoma of the rete testis is very rare. It is highly malignant with potential of early metastasis. We are reporting such a case occurring in a 34-year-old man who remains tumor-free 7 years after orchiectomy. Another 20 reported cases of such a tumor from the English literature are reviewed.

KEY WORDS: **rare tumor of testis, rete testis cancer, adenocarcinoma of rete testis, unusual testicular malignancy**

INTRODUCTION

Adenocarcinoma of the rete testis is one of the rarest of testicular neoplasm. It is also highly lethal due to rapid metastases. We are reporting a case of rete testis carcinoma with a tumor-free course of 7 years. Another 20 reported cases from the English literature are reviewed.

CASE REPORT

A 34-year-old white man presented to the hospital in July, 1977 with a lump in the left scrotum that had been present for several years. The patient had noticed a recent enlargement of the lump and it had become tender and painful. Physical examination revealed an enlarged, tender, firm multinodular left testis with indurated left epididymis. The right testis was normal. The patient had undergone a left testicular exploration for spermatocele secondary to injury in 1965. The results of routine hematologic and chemical tests were normal. Serum acid and alkaline phosphatase levels were normal. Urinary human chorionic gonadotrophin (HCG) was absent. There was no increase of serum alpha-fetoprotein (AFP) level. Chest roetgenogram, total body scan, and liver-spleen scan were negative for any lesion.

The patient underwent a left orchiectomy. There were small white nodules on the tunica albuginea. Cut section revealed a 2 × 1.5 cm firm, white mass in the mediastinum testis invading the tunica albuginea. The testicular parenchyma was not invaded. Microscopic examination of the tumor showed a papillary adenocarcinoma (Figs. 1 and 2) with areas of necrosis.

One month later, the patient underwent an abdominal exploration and retroperioneal lymph node dissection. All the lymph nodes were microscopically free of metastatic tumor.

He did not receive any radiotherapy or chemotherapy. Almost 7 years later (in June 1984) the patient remains free of any recurrent or metastatic tumor.

DISCUSSION

The essential features of adenocarcinoma of the rete testis have been summarized in Table I.

Incidence

Carcinoma arising in rete testis is very rare. Only about 21 such cases have been described in the English literature. Among the reported cases there was a larger number of white patients.

Clinical Features

The rete testis carcinoma occurs in all ages. The oldest patient reported was 89 years of age [Goldstein et al, 1981]. The tumor has been noted in right or left testis with almost equal frequency. Only one case with bilateral involvement has been reported [Willis, 1967]. Though most patients had developed the tumor in a previously normal testis, there were a few cases with preexisting testicular abnormality, such as, undescended testis treated by orchiopexy [Dundon, 1952; Jacobellis et al, 1981], maldescended testis [Laird, 1954], chronic epididymitis [Desberg et al, 1964], history of trauma [Brown, 1964],

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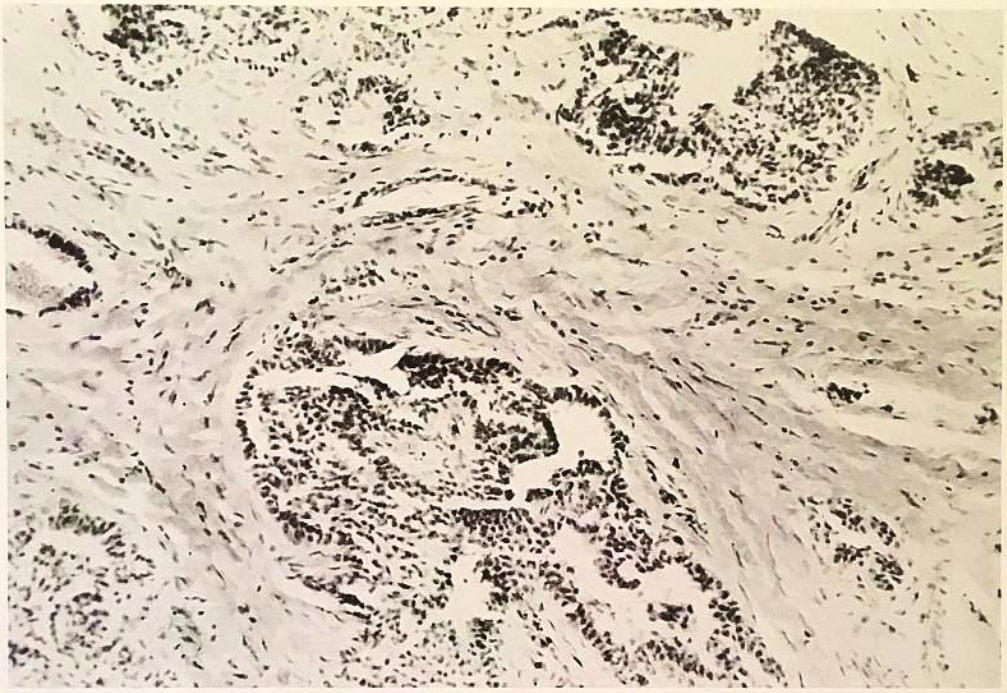

Fig. 1. Papillary adenocarcinoma of rete testis (H & E, ×60).

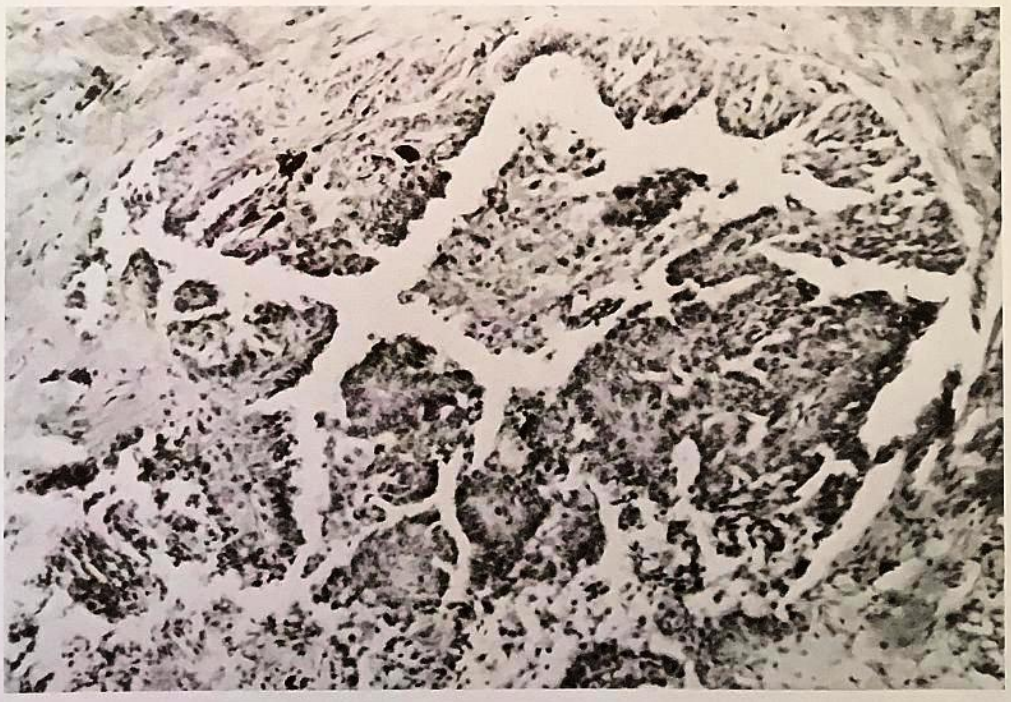

Fig. 2. Higher magnification of the tumor (H & E, ×120).

BLE I. Reported Cases of Adenocarcinoma of the Rete Testis

Authors/year	Age/race	Clinical features	Pathologic features	Treatment and course
Feek et al, 1945	59/Caucasian	Progressively increasing left scrotal—2 yr. Hemiplegia and urinary incontinence—2 mo	Upper pole mass, up to 2 cm thick with necrosis and hemorrhage; microscopically papillary carcinoma of rete testis	2 mo after orchiectomy, patient died of unknown cause. Brain metastasis was a possibility
Scully et al, 1948	48/Caucasian	Back pain radiating to right leg—5 mo. Rib metastasis showed adenocarcinoma on biopsy. Painful swollen right testis	Circumscribed, brown scirrhous mass, 3 × 1.7 × 2.2 cm in the hilus invading into the parenchyma	Bilateral orchiectomy followed by radiotherapy. Died 10 mo later with extensive systemic spread
Badenoch et al, 1951	30/?	Painful right scrotal swelling—2 wk	White, partly cystic lesion in the mediastinum testis invading into testis	Orchiectomy followed by radiotherapy. Disease-free 6 mo later
Shillitoe, 1952	44/?	Left scrotal swelling—3 wk	Upper 2/3 of testis occupied by a hemorrhagic tumor invading epididymis	One month after right orchiectomy lymph node metastases appeared followed by chest metastases. Good response to radiotherapy. Free of symptoms at 5 mo after presentation
Dundon, 1952	20/?	Pain and swelling of left scrotum—4 wk. History of undescended left testis and orchiopexy 10 yr earlier	5×3-cm lobulated, firm, white and yellow tumor occupying half of the testis. One lymph node contained tumor	Received radiotherapy. Free of metastases at 5-mo postoperatively
Laird, 1954	21/?	Right maldescended testis located lateral to the right external ring. Aching groin pain after exercise at presentation	Lobulated tumor, 6 × 6 × 3 cm surrounded by compressed testicular tissue. Areas of polycystic appearance	Received postoperative radiotherapy. No follow-up information
ıoen et al, 1959	80/Caucasian	5-yr history of left inguinoscrotal hernia. Recent enlargement of left scrotal mass	Left orchiectomy specimen showd friable, gray-white papillary excrescences on the outer surface of testis and white firm tissue on the cut surfaces of mediastinum testis	Tumor recurred at operative site and iliac lymph nodes were involved by metastasis, 8 mo after surgery. Chemotherapy (methotrexate) was administered. Patient died one month later
Desberg et al, 1964	71/?	12-yr history of chronic epididymitis. Painless enlargement of left testis—1 month	Irregular, nodular, firm, yellow-gray mass, 10 × 10 × 8 cm, completely replacing the testis	Received radiotherapy and chemotherapy. Died two months postoperatively with metastases to penile skin and inguinal lymph nodes
Brown, 1964	47/?	Painless selling of testis (side not mentioned)—1 yr. Past history of trauma	White-brown hemorrhagic masses mainly in the mediastinum testis with invasion of the body of the testis	Died 2 yr after orchiectomy; one metastatic nodule was found in the lung at autopsy
Willis, 1967	31/?	Painful right testicular enlargement—3 mo. Chest x-ray showed round shadows in both lung fields	At autopsy, right testis was replaced by a 8-cm firm white tumor	Patient died 4 mo after admission. There were extensive metastases in lymph nodes, lungs, liver and invasion of inferior vena cava
	31/?	Bilateral orchiectomy done for prostatic carcinoma	Both testes contained 2–7 mm round tumor nodules, some in the mediastinum	No follow-up information
Moghe et al, 1970	60/?	No clinical information	Capsulated, 8 × 6 cm tumor mass with invasion of spermatic cord. Capsular invasion and lymphatic invasion noted	Not described

Continued

Table I. *continued*

Authors/year	Age/race	Clinical features	Pathologic features	Treatment and course
Moghe et al, 1971	47/?	Painless left scrotal swelling—1 year. Abdominal swelling and loss of weight—6 mo. Enlarged inguinal lymph nodes. Chest x-ray showed metastatic tumors in both lungs	Gray-white solid nodules, 2–5 mm in size in the mediastinum testis, corpus testis, epididymis. Paraaortic and inguinal lymph nodes contained carcinoma	Orchiectomy and laparotomy were followed by radiotherapy and chemotherapy. No follow-up information given
Schapira et al, 1972 Gisser et al, 1977	73/Caucasian	Left scrotal mass—3 mo. Clinical hydrocele was aspirated, the testis then felt irregular, nodular, and hard	Orchiectomy specimen showed a white homogeneous mass in the upper pole invading the tunica albuginea	Scrotal skin recurrence within 18 mo. Received chemotherapy (5-fluorouracil). 4 yr later developed inguinal lymph node metastasis. Autopsy showed metastatic involvement of lungs, liver, and thoracic and abdominal lymph nodes
Whitehead et al, 1972	68/Caucasian	Enlarged right scrotum—several yr. Right groin mass and swelling of right leg—6 wk. Skin nodules over right groin on biopsy showed carcinoma	Orchiectomy specimen showed gray-white tumor in the mediastinum with invasion of testis parenchyma and tunica	received radiotherapy and chemotherapy. Patient died with metastases in skin, inguinal and retroperitoneal lymph nodes, and rib within several months after admission
Turner et al, 1973	75/Caucasian	Patient with bilateral inguinal hernia; examination under anesthesia revealed right testicular mass	Orchiectomy showed an irregular papillary mass, 6 × 4.5 cm in the hydrocele sac	Free of tumor recurrence or metastasis at 6 mo postoperatively. No radiation or chemotherapy
Mehan et al, 1977	35/white	Right hydrocelectomy, 5 yr previously. Enlarged indurated right testis since then	Orchiectomy specimen showed a granular lower pole	Received radiotherapy. Free of tumor 6 mo postoperatively
Roy et al, 1979	62/black	Came with draining scrotal sinus and urethral fistula. Swelling and pain of left scrotum—2 wk. Biopsy of bladder neck showed papillary adenocarcinoma	Autopsy revealed a 10 × 8 cm necrotic tumor replacing the left testis with direct invasion of prostatic urethra and trigone. Iliac lymph nodes contained metastatic carcinoma	Received radiotherapy. Expired 40 days after admission
Goldstein et al, 1981	89/Caucasian	Presented with symptoms of prostatism and right scrotal swelling due to hydrocele and a possible tumor	Orchiectomy specimen showed a gray-white focally nodular mass involving tunica vaginalis, epididymis, and spermatic cord	9 mo postoperatively the patient was clinically free of distant metastasis, but regional lymph nodes were thought to be involved
Jacobellis et al, 1981	34/Caucasian	Presented with lumbar pain radiating into scrotum and weight loss. Patient had undergone bilateral orchiopexy for cryptochidism 21 yr ago	Right orchiectomy specimen showed a 1-cm yellow-white irregular tumor in the mediastinum	Patient received chemotherapy. Metastatic tumor was noted in lung and paraaortic nodes by radiographic studies. He died 3 mo later
Sarma et al, present case	34/Caucasian	Left testicular mass—several years. Recent increase in size with pain and tenderness. Patient underwent left testicular exploration for spermatocele 12 yr ago	Orchiectomy specimen showed a 2 × 1.5 cm firm, gray-tan, nodular tumor involving testis, mediastinum, and tunica albuginea	All metastatic surveys yielded negative results. Retroperitoneal lymphadenectomy did not show any tumor. 7 yr later, patient remains free of tumor

previous hydrocelectomy [Mehan et al, 1977], and past testicular exploration for spermatocele [Sarma et al, present case]. Most commonly, the patients presented with scrotal or testicular mass with or without pain and tenderness of varying duration. Few presented with metastases. Other presenting features included hernia, hydrocele, and scrotal draining sinus.

Pathologic Features

Grossly, the tumor may be noted as a mediastinal nodule or multiple nodules in testis or as a mass completely replacing the normal testis. Size may vary from a few-millimeter nodule to 10-cm mass. Cut surfaces may be solid, necrotic, and cystic and papillary with gray-white, yellow-brown tumor tissue.

Microscopically, the tumors show a pattern of papillary adenocarcinoma, some showing solid sheets of undifferentiated epithelial cells. The intact rete testis ducts may show dysplasia and hyperplasia. There should be no seminoma or teratoma in the testis.

Course

The patients have been treated by orchiectomy alone or followed by radiotherapy with or without chemotherapy. Most of the patients die with metastatic disease within a year. Tumor may metastasize in the lymph nodes, bone, liver, lungs, and brain. Our patient has the longest survival (7 years) after undergoing orchiectomy and retroperitoneal lymphadenectomy. He remains free of any recurrent or metastatic tumor. Another patient survived 5.5 years after orchiectomy and chemotherapy; however, he died with extensive systemic metastases [Schapira et al, 1972; Gisser et al, 1977].

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