Adenoid Cystic Carcinoma of the Prostate Gland
--Pathological Review with a Case Report--

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A case of adenoid cystic carcinoma of the prostate gland in a 38-year-old Korean man is described. Microscopically, variable patterns, that is, glandular, trabecular, cribriform and solid areas, were seen. The unusual location of this tumor in our patient highlights the ubiquitous distribution of this malignant neoplasm.

Key Words: Adenoid cystic carcinoma, prostate gland

Adenoid cystic carcinoma is an extremely uncommon but distinctive tumor in the prostate gland. It is frequently found in the major and minor salivary glands (Thackray et al. 1974) but has been infrequently described in the breast (Cavanzo et al. 1969), uterine cervix (Gallager et al. 1971), Cowper's gland (Carpenter et al. 1971), lacrimal gland (Adam et al. 1971), external ear (Althaus et al. 1970), upper respiratory tract (Ash et al. 1974), esophagus (Melms et al. 1972), and skin (Headdington 1978). It is rarely found in the prostate gland and there have only been seven well documented reports written in English (Frankel et al. 1974; Tannenbaum 1975; Kramer et al. 1978; Kuhajda et al. 1984; Shong-Sanc et al. 1984; Gilmour et al. 1986; Young et al. 1988). When adenoid cystic carcinoma occurs in the prostate gland, it is not distinguishable clinically from ordinary adenocarcinoma other than that it tends to occur at a relatively younger age and serum acid phosphatase is within the normal range. The authors recently experienced a case of adenoid cystic carcinoma in the prostate gland of a 38-year-old male, and herein report it.

CASE REPORT

A 38-year-old man was admitted with nocturia and dysuria of 5 year's duration. Three to four months ago, urinary frequency and nocturia increased to 2-3 times per night. His past and family histories were unremarkable.

Rectal examination revealed an enlarged, stony hard, irregular prostate. Peripheral blood revealed normal hemoglobin 14.7g/dl, WBC 9,000/mm³, and platelet count 273,000/mm³. Serum acid phosphatase (1.3 u/l), BUN (14.0mg/dl), creatinine (0.9g/dl) and urinalysis were also within normal ranges. Computerized tomography revealed an enlarged prostate gland displacing the urinary bladder to an anterior superior position with signs of invasion into the seminal vesicle. This indicated that the tumor in the prostate gland was malignant. A transurethral prostatectomy resulted in a diagnosis of adenoid cystic carcinoma. A prostatectomy was then performed.

PATHOLOGIC FINDINGS

Gross findings

The prostatectomy specimen, 8x6x4cm in size and weighing 100 grams, consisted of the prostate gland and the seminal vesicle; it was relatively well demarcated from the surrounding tissue, irregularly oval in shape, light yellowish brown in color and
Fig. 1. The cut surface shows a well-defined pinkish gray firm mass. The mass is lobulated by thin fibrous septae with loci of hemorrhagic necrosis and microcystic changes.

Fig. 2. The tumor shows a typical cribriform arrangement and most of the cystic spaces are filled with mucoid material (H & E, ×100).
stony hard in consistency. The cut surface revealed several lobules separated by fibrous septa with discrete areas of hemorrhage, necrosis and cystic change (Fig. 1).

**Microscopic findings**

The tumor was not encapsulated with malignant cells invading the normal prostate tissue forming nodular growth. Variable patterns, that is, glandular, trabecular, cribriform and solid areas, were seen (Fig. 2) and within the lumen of the cribriform gland, eosinophilic amorphous material was found. The malignant cells had hyperchromatic nuclei with a few vacuoles within the cytoplasm that resembled myoepithelial cells, and frequent perineural invasion was observed (Fig. 3).

**DISCUSSION**

Adenoid cystic carcinoma was first described by Billroth in 1859 when he characterized a tumor with cribriform, glandular and basaloïd patterns which contain a mucoid material and called it cylindromas (Billroth 1859). Later, Spies named it adenoid cystic carcinomas (Spies 1930) and it was generalized by Foote and Frazell (1954), but it also called basiloma, adenocystic basiloïd carcinoma or adenoepithelioma. Adenoid cystic carcinoma is a slow growing, indolent tumor with frequent recurrence and invasion into the surrounding organs but with slow systemic dissemination (Spies 1930; Thackray 1974). It usually occurs in the salivary glands and comprised 15% of the tumors arising in the minor salivary glands, 5% in the major salivary glands and 1.2% in the parotid glands (Thackray 1974). It has been reported to arise in the breast, uterine cervix, Cowper's gland, lacrimal gland, ear canal, upper airway, esophagus and skin. However, it is extremely rare in the prostate gland and less than 0.01% of the malignant tumors arising in the prostate are adenoid cystic carcinoma (Tannenbaum 1975). Frankel and Craig first reported it in 1974 in the English literature and only 8 well-documented examples have been described, even though there are several other reports described in less detail.

The origin of this tumor is unknown; electron microscopy of adenoid cystic carcinoma from the human salivary gland revealed two kinds of cells, myoepithelial and secretory. The cytoplasm of the.
secretory cells contained basement membrane material and tonofilaments; with these substances it has been postulated that adenoid cystic carcinoma of the human salivary gland arises from the myoepithelial cells of the intercalated ducts of the salivary glands (Cavanza et al. 1969; Thackray 1974). However, as yet, there is no evidence of myoepithelial cells or myoepithelial differentiation in the human prostate gland. Dickman et al. (1973) reported that ectopia of the seromucinous glands within the prostatic stroma was similar to that found in the salivary glands (Ash et al. 1974), and thus it can be suggested that some tumors of the prostatic glands may have the same origin and histologic pattern as those of the salivary gland. Some authors insist that adenoid cystic carcinoma arises from the seromucinous glands of the external auditory canal, that is, the modified apocrine glands. Carpenter et al. (1971) reported a case of adenoid cystic carcinoma of the prostate gland and stated its origin as the Cowper’s gland near the urethra. However, the latter can be characterized by clinical symptoms of rectal pain, pain during defecation, and constipation which made it possible to differentiate it from carcinoma arising from the prostate gland.

There have been very few reported cases of adenoid cystic carcinoma originating from the prostate gland, so no information regarding proper treatment method or prognosis is available. It is thought that the adenoid cystic carcinoma from the prostate gland is similar to that arising from the other parts of the body (Frankel et al. 1974), and in our case, the lengthy duration of symptoms suggested a slow rate of growth and that distant metastasis would not appear until a rather late stage. Eby et al. (1972) stated that the tumor could be differentiated histologically into 2 groups by the predominant patterns of cribriform or solid arrangement and reported that the former had a better prognosis than the latter, Batsakis et al. (1979), on the other hand, reported that only the origin of the tumor and the spread of the tumor clinically could determine its prognosis. No histological parameters which can forecast recurrence or metastasis have been described as yet. Surgical excision and radiotherapy have been suggested as the treatment of choice despite the high recurrence rate and death. Estrogen treatment is known to have no effect. This case is thought to be one of the rare types of tumors arising from the prostate gland and it is hoped that this report will give insight to future cases for treatment and prognosis.

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