

# Giant Multilocular Cystadenoma of the Prostate – A Potential Pitfall On Needle Core Biopsy

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## Abstract

Giant multilocular prostatic cystadenomas are rare tumors (only 20 cases reported so far) that may present as large abdominal masses causing symptoms of urinary frequency and urgency. Routine needle core biopsy can be misleading and reveals only benign prostatic tissue. We report the case of a 61-year-old male who presented with a large abdomino-pelvic mass and symptoms of an enlarged prostate. Following an inconclusive needle core biopsy a definitive diagnosis was only possible after examination of resected specimen. This article also reviews the current literature on this rare tumor and discusses the various investigations required to confirm the diagnosis.

## BACKGROUND

Giant multilocular prostatic cystadenoma is a rare, benign lesion and fewer than 20 cases have been described in English literature. The tumour is capable of growing to massive proportions and patients usually present with features of obstructive voiding dysfunction with or without an abdominal mass<sup>[1234567]</sup>. We report a case of giant multilocular prostatic cystadenoma which presented diagnostic difficulties on needle core biopsy.

## CASE REPORT

A 61-year-old man presented with complaints of urinary frequency and urgency. He was known to have an elevated prostate specific antigen (PSA) levels for the past ten years. However, a prostatic needle core biopsy performed at that time had been negative for malignancy. Clinical examination revealed a large pelvic mass extending up to the umbilicus and contiguous with the prostate. The patient had a raised serum PSA (696  $\mu\text{g/litre}$ ). Magnetic resonance imaging(MRI) revealed a non-invasive, partly cystic mass with a well defined capsule adherent to the posterior wall of the bladder (Figure -1).

## Figure 1

Figure 1: MRI scan showing sagittal section of the abdominal cavity and the extent of the tumor. Note the well-encapsulated appearance of the tumor.



Clinically, the possibilities of a low grade prostatic sarcoma or a massive benign prostatic hypertrophy (BPH) were considered. A repeat needle core biopsy of the mass was performed which showed benign prostatic tissue with no evidence of malignancy, and therefore the possibility that the biopsy was not representative was raised. The patient underwent laparotomy and the mass was excised in its entirety.

Grossly, the mass measured 29 x 22 x 15cm and weighed 3650gms, and was well encapsulated. The cut surface appeared pale with numerous small cysts (Figure-2).

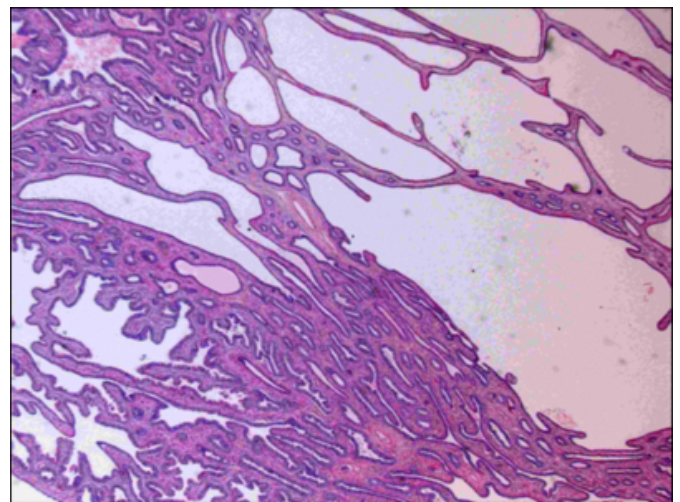
**Figure 2**

Figure 2: Gross appearance of the tumor – cut surface shows well encapsulated tumor with pale appearance and numerous small cysts containing pale fluid.



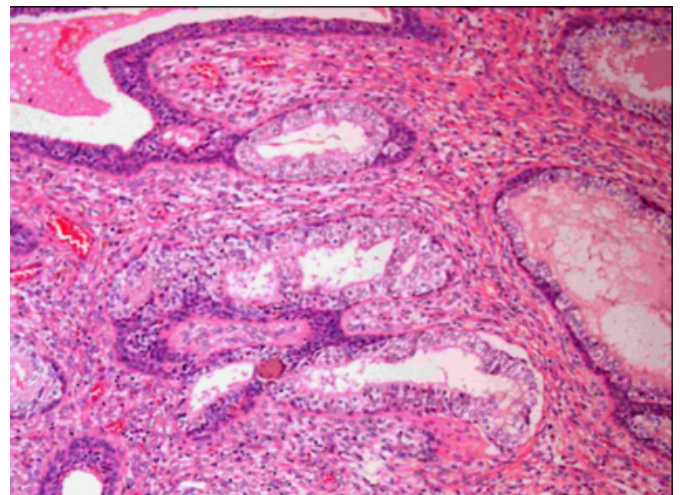
Histologically, the tumour exhibited a uniform morphology throughout and was composed of cysts of variable sizes and glands set in a hypocellular, fibrous stroma. Many of the glands exhibited convoluted outlines reminiscent of benign prostatic glands. The smaller cysts and glands exhibited a bilayered epithelium with basal, cuboidal cells and outer, columnar cells containing pale cytoplasm and bland basally located nuclei lacking nucleoli. Occasional papillary infoldings with a multilayered epithelium were noted. There was no prostatic intraepithelial neoplasia (PIN) or cytological atypia noted. The stroma was hypocellular with no atypia or mitoses. Occasional foci of foamy and hemosiderin laden macrophages were identified in the stroma.

**Figure 3**



**Figure 4**

Figure 3: Histopathological section showing variably sized cysts set in a hypocellular, fibrous stroma. Smaller cysts were lined by bilayered epithelium with a basal cuboidal cells and outer layer of columnar cells.



Immunohistochemical studies revealed positivity for PSA and prostatic acid phosphatase (PAP) within the epithelial lining confirming its prostatic origin. The epithelial cells also showed patchy moderate intensity granular circumferential cytoplasmic positivity for alpha-methylacyl coenzyme A racemase (AMACR). The basal cells were highlighted by extensive, intense CK5/6 staining. The pathological findings were consistent with those of a giant multilocular cystadenoma of the prostate.

The patient remains symptom free at six-month follow up with no evidence of tumour recurrence.

## DISCUSSION

The designation ‘giant multilocular prostatic cystadenoma’ was first proposed by Maluf et al [1]. These are rare benign tumours of the prostate which have been described in men of all ages [2]. Patients often present with features of urinary obstructive symptoms and an abdominal mass due to the large size of the tumor. Imaging modalities usually reveal a multicystic mass and may or may not confirm a prostatic origin [3] (because lesions may be attached only by a pedicle or be entirely separate from the prostate)[1]. Needle core biopsies of the prostate may be inconclusive, as in the current case, and usually reveal benign prostatic tissue. A definitive diagnosis can be made only on histopathological examination of the resected specimen.

Grossly the lesion is characterised by multilocular cysts of variable size. Masses up to 45cm and weighing up to 6500g have been reported [2]. Microscopically, the tumour is composed of prostate type glands and cysts lined by a double layer of cells with basal nuclei and pale cytoplasm and lacking cytological atypia. The epithelial lining usually exhibits positivity for prostate-specific antigen. The stroma is similar to the normal fibro muscular stroma of the prostate [1234567].

The tumour behaves in a benign fashion and complete surgical excision is the treatment of choice. Occasional recurrences have been described but these are related to incomplete excision [13]. Successful treatment of a recurrence with gonadotrophin-releasing hormone antagonists has been reported [4]. A recent case report describes high-grade prostatic intraepithelial neoplasia (PIN) within a multilocular prostatic cystadenoma[5].

The differential diagnosis includes Phyllodes tumour of prostate, seminal vesicle cystadenomas, pelvic mesotheliomas and multilocular peritoneal inclusion cysts.

Phyllodes tumour of the prostate belongs to a rare group of prostatic lesions currently designated ‘Prostatic Stromal Tumours of Uncertain Malignant Potential’ (PSTUMP) and has a growth pattern similar to phyllodes tumour of breast. It can be differentiated from prostatic multilocular cystadenoma by its hypercellular stroma which exhibits varying degrees of cytological atypia[8].

Seminal vesicle cystadenoma or cystic epithelial-stromal tumour is characterised by the presence of lipofuscin pigment and will be negative for PSA [9] as will be pelvic mesotheliomas and multilocular peritoneal inclusion cysts.

A recent report discusses ‘Aberrant’ AMACR positivity together with a heterogeneous presence of a basal layer in multilocular prostatic cystadenoma [6]. Our case showed a patchy positivity with AMACR immunostain and a prominent basal layer highlighted by CK5/6 positivity. This finding reiterates that AMACR is not specific for prostatic adenocarcinoma and can be positive in benign lesions including prostatic cystadenoma[6] and nephrogenic adenoma[1011].

## CONCLUSION

In conclusion, giant multilocular prostatic cystadenoma is an unusual prostatic lesion which can present a potential pitfall on needle core biopsy. An awareness of the lesion and its clinical presentation (a large abdomino-pelvic, multicystic and loculated mass arising from the prostate) is essential to provide an index of suspicion.

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