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# Case Report

# Giant multilocular prostatic cystadenoma, a diagnosis to consider in large pelvic male masses

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#### ABSTRACT

Giant multilocular prostatic cystadenomas are rare benign prostatic tumor, usually presenting as a large pelvic mass, compressing the adjacent organs but with no clear aggressive features, frequently causing obstructive voiding symptoms. Nowadays, imaging plays an important role on the adequate characterization of these lesions, not only on depicting their different internal components, but also the relationship with the adjacent structures, therefore providing the best preoperative surgical planning. Here we present a case of a 62year-old patient with recurrent obstructive voiding symptoms due to a giant multilocular prostatic cystadenomas, with histologically correlation, posteriorly excised.

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## Introduction

Giant multilocular prostatic cystadenomas (GMPC) was first described in 1991 and is a relatively rare benign clinicpathologically neoplasm [1–6] and accounts for one of the unusual prostatic tumors diferential [2]. These lesions are commonly unnoticed until the patient develops obstructive voiding or retention symptoms. GMPC's can achieve large dimensions and cause significant mass effect on the adjacent structures, but despite that they do not demonstrate an invasive behavior or aggressive features [1–6].

Imaging modalities allow not only the establishment of the prostatic origin of these lesions, provide the accurate measurements, but also demonstrate their well-defined, multicystic, and multiseptated nature [3–5]. We present a case of a 62-year-old patient with recurrent obstructive voiding symptoms due to a GMPC, and its histological correlation.

#### **Case presentation**

A 62-year-old male presented at the emergency department with intermittent urinary obstruction symptoms over a 3-month period, with 2 previous episodes of acute urinary obstruction, with the need of catheterization. Laboratory analysis did not demonstrate any significant abnormalities, with prostate-specific antigen (PSA) within normal range. At ultrasound a complex pelvic mass was depicted. To better and further characterize this lesion, an abdominal and pelvic computed tomography (CT) scan before and after I.V. contrast

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Fig. 1 – Axial CT scan before contrast administration demonstrating an enlarged and heterogeneous pelvic mass, with its most posterior and peripheral aspect revealing a spontaneous hyperdense area, in keeping with hemorrhagic/hyperdense protein contents (blue dotted line).



Fig. 2 – Sagittal reconstruction CT scan after contrast administration. The bulky mass appeared to be in close relation with the prostatic region, particularly considering the catheter positioning. It pushed the bladder superiorly and anteriorly and seemed to push the rectum posteriorly, though its definite relationship with the rectal wall was somehow difficult to appreciate.



Figs. 3 and 4 – Axial CT scans before and after I.V. contrast administration. Before contrast administration (figure above), some variable sized septa were depicted (yellow arrow), as well as some low attenuation areas, representing the cystic component (asterisks). After contrast administration (figure below), no abnormally enhancing nodular areas nor abnormally nodular enhancing septa were seen. The previously known hyperdense area (orange dotted line) and the central cystic area (asterisk) were still visible. (Color version of figure is available online.)

was performed. CT revealed a bulky and heterogeneous mass in the rectovesical space, measuring  $15\times10\times8$  cm, compressing the bladder anteriorly and the rectum posteriorly, with the Foley catheter going through the lesion. The prostate and seminal glands were indistinguishable from the mass (Figs. 1 and 2). The lesion was well-defined with a considerably large peripheral spontaneously hyperdense area, probably related to hemorrhagic/hyperdense proteinaceous (Fig. 1). It also demonstrated a complex and multilocular structure, with hypodense/cystic areas and numerous septa of different thickness, only mildly enhancing after contrast administration (Figs. 3 and 4). Despite the septations, no nodular solid enhancing components were evident nor signs of lymphadenopathies or metastatic disease. Delayed phases were attempted, however due to the compression effect of the lesion, the adequate opacification of the urinary tract and bladder was not successful (Figs. 5 and 6). CT-guided biopsy targeting septations was performed (Fig. 7) with the histolog-



Figs. 5 and 6 – Axial CT scan on delayed phase and coronal MPR demonstrating opacification of the caliceal urinary tract, but with failure in the opacification of the bladder. Despite this fact, the lesion kept showing absence of abnormal areas of enhancement, even within the septa, favoring a benign etiology.



Fig. 7 – Axial CT scan without contrast during biopsy, prior to surgical excision.



Fig. 8 – Photomicrograph with low power magnification of the biopsy specimen after H&E staining. Revealing some cystic variable-sized dilated glandular spaces, surrounded by dense fibromuscular stroma. Some areas of hemorrhagic components are also visible, correlating to the spontaneously hyperdense areas depicted on the CT (black arrow).



Fig. 9 – Magnification view of the dilated cystic glandular space, which demonstrates a lining of low cuboidal and columnar epithelial cells with basal nuclei.

ical analysis depicting cystically dilated glandular prostatic epithelium, with fibrotic stromal tissue and a considerable amount of hemorrhage (Figs. 8 and 9). No signs of malignancy were noted. The mass was surgically excised with postoperative histological findings in keeping with GMPC. During surgical procedure, there was an accidental small laceration of the anterior rectal wall, due to the close proximity and mass effect that the lesion was causing against this structure. Through the course admission after surgery, the patient developed fever with elevated inflammatory markers. A CT scan was performed documenting a fluid collection with





Figs. 10 and 11 – Axial and sagittal CT scan after contrast administration contrast administration revealing a thick and enhancing wall collection in the surgical bed around the catheter (arrows), in keeping with a postsurgical infectious complication – abscess.



Fig. 12 – Axial CT scan after retrograde contrast opacification of the bladder via Foley catheter, documenting a fistulous tract adjacent to the posterior left side of the bladder (arrow), leading to passive rectal opacification.



Fig. 13 – Axial CT scan with rectal contrast and intravenous contrast administration denoting complete resolution of the previous pelvic collection and absence of extravasation of the rectal administrated contrast, indicating closure of the previous leak.

enhancing walls in the surgical bed around the Foley catheter, in keeping with abscess (Figs. 10 and 11). Considering the suspicion of a rectal leak another follow-up scan with retrograde bladder contrast administration was requested, confirming a small fistulous tract, that was surgically corrected (Fig. 12). The patient kept improving after the second surgery with the predischarge CT scan demonstrating complete resolution of surgical bed collection and fistula (Fig. 13)

## **Discussion and conclusions**

Despite well-known, GMPC's are rare benign prostatic tumors. They have the potential to attain large dimensions and subsequently most patients present with obstructive urinary symptoms similar to those of benign prostatic hypertrophy, such as obstructive voiding symptoms, incomplete voiding, nocturia, increased frequency. Other symptoms related to the mass effect will include acute urinary obstruction or defection problems. The age of presentation is wide, varying between 20 and 80 year old [1,3,4,6,7]. Physical examination, including digital rectal evaluation, denotes an enlarged prostatic gland [1,3–7], often with no suspicious nodules. A palpable abdominal mass has also been reported as a clinical finding [3,4]. PSA value may or may not be elevated [3].

In general, cystic prostatic lesions can be divided according to their location: median, paramedian, or lateral. Besides the location assessment, further classification can be done according to the relationship with the prostatic urethra, as either prostatic or periprostatic. The majority of prostatic cystic lesions will include utricle cysts, Müllerian cysts, ejaculatory cysts, and seminal gland cysts, with the 2 formers being commonly median and the 2 others lateral. Compared to GMPC, these usually present as smaller and simpler cysts, lacking its common multilocular appearance [6]. There are some reports of a GMPC occurring completely separate from the prostatic gland, as a result the differential diagnosis of this lesion will also include rectovesical and retroperitoneal cystic lesions [5]. Among the differential one should include the phylloid variant of atypical prostatic hyperplasia, prostatic leiomyoma, prostatic sarcoma, cystic prostatic carcinoma, infection (hydatic cysts, echinococcosis, or cavitary prostatitis with or without abscesses), pelvic mesothelioma, lymphangioma, peritoneal inclusion cyst, and teratomas [1–6].

At imaging they frequently present as large multiloculated masses, composed of variable-sized cysts, usually with some thin enhancing septations and an outer thin enhancing wall [3-6]. In fact, these findings were demonstrated in the presented case (Figs. 3 and 4). Few solid enhancing areas can be found within the lesion, representing stromal components [3], posing some diagnostic difficulties. However, one distinguishing feature is that GMPC's are in fact well-defined noninfiltrative masses of indolent growth [8], therefore, if a complex multicystic pelvic mass is depicted on imaging demonstrating aggressive and structural invasive behavior, another diagnosis should be considered. Yet again, in the presented case, the lesion had sharp marginated borders, appearing to push the adjacent structures away and not invading into them, favoring this diagnosis (Figs. 1 and 2). Imaging studies are also helpful in establishing the origin of these large lesions and GMPC's should be considered when a retroperitoneal mass is located between the bladder and rectum [6]. Seldom CT or magnetic resonance imaging can demonstrate a prostatic pedicle [3–5]

Histological analysis provides the definite diagnosis, demonstrating dilated/cystic prostatic gland, but with the typical lining of columnar and cuboidal cells, pale cytoplasm, and basally located nuclei, in a background of hypocellular fibrous stroma, positive for PSA staining [1,6,7]. High-grade prostatic intraepithelial neoplasia within the GMPC's though not common has already been described.

Complete surgical excision is the optimal treatment option. Incomplete excision can lead to tumor regrowth and recurrence [3–6], which can have a surgical approach (mass excision or pelvic exenteration) or with gonadotropinreleasing hormone agonist [3,4].

In conclusion, radiologists should be aware of this condition when confronted with large pelvic masses of unknown history or that possibly maybe related to the prostate, with the multiseptations and the lack of enhancing nodules hinting the diagnosis. Despite biopsy being and surgery are often necessary in most of the cases, raising the possibility of a benign lesion might help avoiding unnecessary patient's distress.

### **Declaration of Competing Interest**

No conflicts of interest.

#### REFERENCES

- Kirsch A, Newhouse J, Hibshoosh H, O'Toole K, Ritter J, Benson M. Giant multilocular cystadenoma of the prostate. Urology 1996;48(2):303–5. doi:10.1016/s0090-4295(96)00178-1.
- [2] Chang J, Lee H, Lee S, Byun S, Choe G, Kim S, et al. Unusual tumours involving the prostate: radiological-pathological findings. Br J Radiol 2008;81(971):907–15. doi:10.1259/bjr/68294775.
- [3] Baad M, Ericson K, Yassan L, Oto A, Eggener S, Nottingham C, et al. Giant multilocular cystadenoma of the prostate: AIRP best cases in radiologic-pathologic correlation. Radiographics 2015;35(4):1051–5. doi:10.1148/rg.2015140316.
- [4] Olgun D, Onal B, Mihmanli I, Kantarci F, Durak H, Demir H, et al. Giant multilocular cystadenoma of the prostate: a rare cause of huge cystic pelvic mass. Korean J Urol 2012;53(3):209. doi:10.4111/kju.2012.53.3.209.
- [5] Rusch D, Moinzadeh A, Hamawy K, Larsen C. Giant multilocular cystadenoma of the prostate. Am J Roentgenol 2002;179(6):1477–9. doi:10.2214/ajr.179.6.1791477.
- [6] Nakamura Y, Shida D, Shibayama T, Yoshida A, Matsui Y, Shinoda Y, et al. Giant multilocular prostatic cystadenoma. World J Surg Oncol 2019;17(1). doi:10.1186/s12957-019-1579-7.
- [7] Hauck EW, Battmann A, Schmelz HU, Diemer T, Miller J, Weidner W, et al. Giant multilocular cystadenoma of the prostate: a rare differential diagnosis of benign prostatic hyperplasia. Urol Int 2004;73(4):365–9. doi:10.1159/000081602.
- [8] Patriarca C, Zucchini N, Corrada P. Giant multilocular prostate cystoadenoma: an entirely benign prostate neoplasm with some phenotypic features of malignancy. Am J Surg Pathol 2005;29(9):1252–4. doi:10.1097/01.pas.0000170347.62927.d9.