

A Rare Diagnosis in a Tertiary Institution: First Ever Case Report and Literature Review of Giant Cystadenoma of the Prostate at the University of Benin Teaching Hospital, Benin City, Edo state, Nigeria

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ABSTRACT

Background: Giant multilocular cystadenoma of the prostate (GMCP) is an extremely rare Benign tumour of the prostate gland. An extremely high index of suspicion is required to make a clinical diagnosis, however, in most cases, the diagnosis is made following radiologic investigations and histologic examination of resected specimen. We report the first case of GMCP seen at our institution and ever reported in Nigeria.

Case Description: Our patient was a 64-year-old man who presented to our facility, with painless lower abdominal swelling, storage and voiding lower urinary tract symptoms. He had several episodes of acute urinary retention relieved by a urethral catheterisation and had been catheter dependent. Abdominopelvic ultrasound scan (USS), abdominopelvic computed tomography (CT) scan and magnetic resonance imaging (MRI) done revealed a huge cystic pelvic mass of uncertain origin. Prostate specific antigen (PSA) done on several occasions ranged from 27ng/ml->100ng/ml. He had transrectal ultrasound (TRUS) guided prostate biopsy on 2 different occasions that revealed prostatic hyperplasia with no evidence of malignancy. He had an initial pelvic exploration which revealed a retroperitoneal mass with an abscess cavity. The mass was resected at that time, but reoccurred about 1 year after. He also had transrectal aspiration of the haemorrhagic cysts on 2 occasions due to pain, with cytology revealing haemorrhagic smear and no malignant cells. He subsequently had joint exploratory laparotomy with R2 resection of a huge cystic mass originating from the pelvis and infiltrating the bladder and surrounding abdominal viscera. Histology of the resected mass revealed the diagnosis of GMCP.

Conclusion: With no reported case of GMCP in Nigeria, we present our diagnostic dilemma of GMCP. A consideration should therefore be made for this entity in male patients presenting with huge cystic masses arising from the pelvis.

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INTRODUCTION

Giant multilocular cystadenoma of the prostate (GMCP) is an extremely rare, benign tumour that typically arises in the prostate gland but may also originate from the seminal vesicles (Chen et al., 2020; Wang et al., 2022). Despite its usual benign nature, GMCP can exhibit malignant behaviour, with adherence to contiguous structures such as the rectum and bladder. This presents significant diagnostic challenges, necessitating the use of multimodal imaging techniques. Definitive diagnosis is usually achieved through histologic examination of resected specimen, as biopsies frequently reveal benign prostatic tissue (Yuya et al., 2019; Wang et al., 2022; Mohammad et al., 2022). To date, fewer than 40 cases have been reported in the literature worldwide, with no documented cases in Nigeria (Maluf et al., 1991).

CASE DESCRIPTION

Our patient was a 64-year-old man, residing in Benin-City, Edo State, Nigeria. He presented with a one-year history of storage and voiding lower urinary tract symptoms, including frequency, urgency, nocturia, urge incontinence, hesitancy, intermittency, and a sensation of incomplete voiding, with no episodes of acute urinary retention. He reported no associated generalized abdominal pain, altered bowel habits, haematochezia, or vomiting. There were no symptoms of low back pain, weight loss, or anorexia, nor any chronic cough or drenching night sweats.

He had no known family history of prostate cancer or other malignancies. He was married with three children and had no desire for more. There was no history of erectile dysfunction. He had a smoking history of 10 packs a year for 15 years but quit 22 years ago and reported occasional alcohol use. The patient had a 15-year history of well-controlled hypertension.

On examination, the patient appeared healthy and was not in obvious distress. An abdominal examination revealed a non-tender, warm mass approximately 8 cm above the pubic symphysis. Genital examination was unremarkable, and rectal examination revealed a cystic mass in the anterior rectal wall. The prostate couldn't be felt separate from the mass. Initial PSA was 26.59 ng/ml. An abdominopelvic ultrasound revealed a hypoechoic mass anterior to the bladder, measuring 6.54 x 13.5 x 11.6 cm, with a volume of 429 mL and fluid levels. Due to the elevated PSA, a transrectal digital-guided prostate biopsy was performed, which revealed benign prostatic hyperplasia. The patient

subsequently developed post-biopsy sepsis, managed with antibiotics.

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He experienced an episode of acute urinary retention, which was relieved by urethral catheterization, and was started on tamsulosin. An initial assessment of a complex pelvic mass was made. He subsequently had urethrocytoscopy and open pelvic exploration, where a pyogenic cavity containing 1.2L of seropurulent effluent was identified, extending from the retroperitoneum. The cavity was distinct from the urinary bladder, which appeared normal, as did the rectum and both the small and large bowel. The abscess cavity was drained, and the wall was sent for histologic analysis.

Postoperatively, the patient developed a surgical site infection and was discharged on the 16th day post operation (post-op). Follow-up PSA levels increased to 61.2 ng/mL and exceeded 100 ng/mL three months post-exploration. He subsequently developed an incisional hernia at the site of the initial surgery, with the mass gradually increasing in size over the next year, accompanied by minimal lower abdominal pain.

After being lost to follow-up, the patient returned two years later with worsening lower urinary tract symptoms. Upon examination, a lower midline scar with multiple incisional hernias and an irregular mass approximately 20 x 24cm was noted. Rectal examination revealed a soft mass bulging into the rectal cavity. The prostate still wasn't felt distinct from the mass. A CT scan of the abdomen showed a complex pelvic mass indistinguishable from the bladder and prostate, with significant pelvic sidewall invasion and bladder calculi. The PSA level at this time was >100 ng/ml. A repeat transrectal prostate biopsy again reported benign nodular hyperplasia.

After another year of being lost to follow-up, the patient represented with worsening lower abdominal discomfort, and was referred to general surgery for consideration of a retroperitoneal tumour. A multidisciplinary team (MDT) meeting concluded that re-exploration was necessary due to the diagnostic dilemma presented by the case. The patient developed acute urinary retention and required urethral catheterization.

He ultimately sought care from another urologist (the author) after having the mass for seven years.

At this point, an abdominal examination revealed a full abdomen with a lower midline scar, multiple incisional hernias, and an irregular mass measuring approximately 28 x 20cm. Repeat abdominal ultrasound indicated a large cystic mass occupying the pelvis. A pelvic MRI revealed an extensive intra- and retroperitoneal multilocular mass with fluid levels, peritoneal lymphadenopathy, and soft tissue extension. He developed severe pain while sitting and rectal examination revealed tense cysts. Two sessions of transrectal aspiration yielded over 200mL of haemorrhagic fluid, which showed no malignant cells upon cytological analysis. He had a re-exploration, revealing a large retroperitoneal tumor with cystic and solid

components, adherent to the bladder and small bowel. A bladder calculus measuring 4cm x 4cm was also found. The mass was partially resected (R2), and cystolithotomy was performed, along with repair of a bladder laceration. Histological analysis of the mass confirmed features consistent with giant prostatic cystadenoma.

Postoperatively, the patient had an initially uneventful recovery and was discharged on the 11th day. Unfortunately, he returned two days later with signs of peritonitis and succumbed to overwhelming sepsis on the 15th postoperative day.

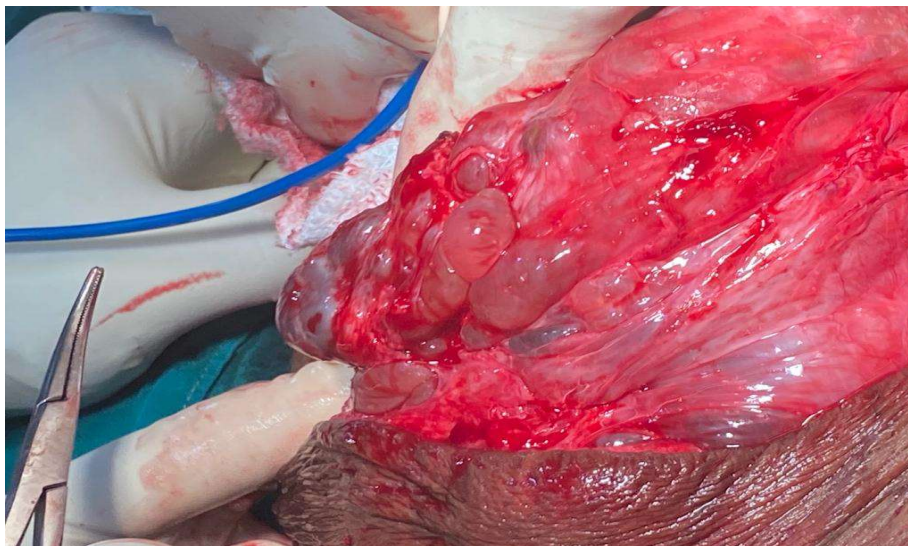


Figure 1: mass within the abdominal cavity showing multiple cysts, and adherence to the overlying skin

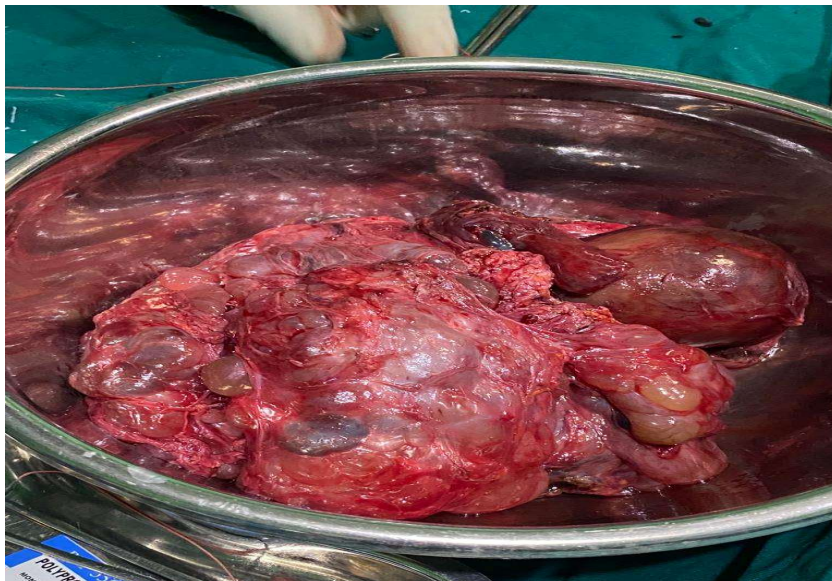


Figure 2: Resected specimen, with cystic and solid components

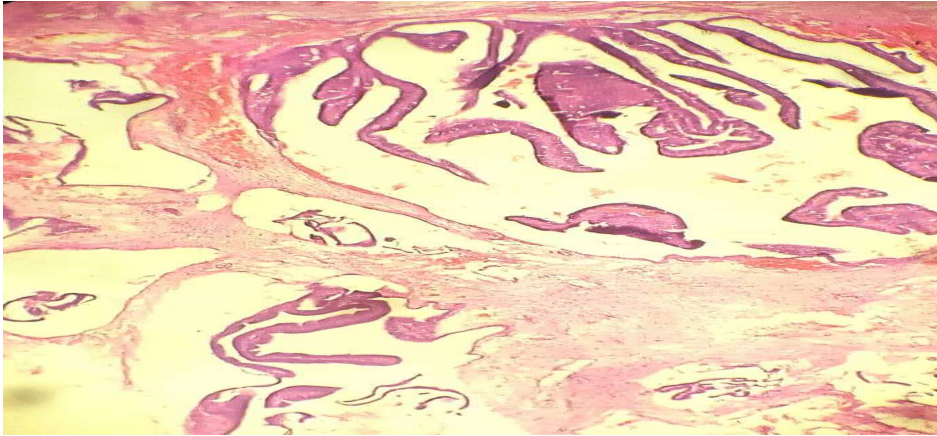


Figure 3: Photomicrograph of GMCP in our patient showing cystically dilated glands lined by cuboidal to low columnar epithelium, which forms well developed papillae, with fibromuscular cores. These papillary structures protrude into the cystic lumina. (H&E x 40)

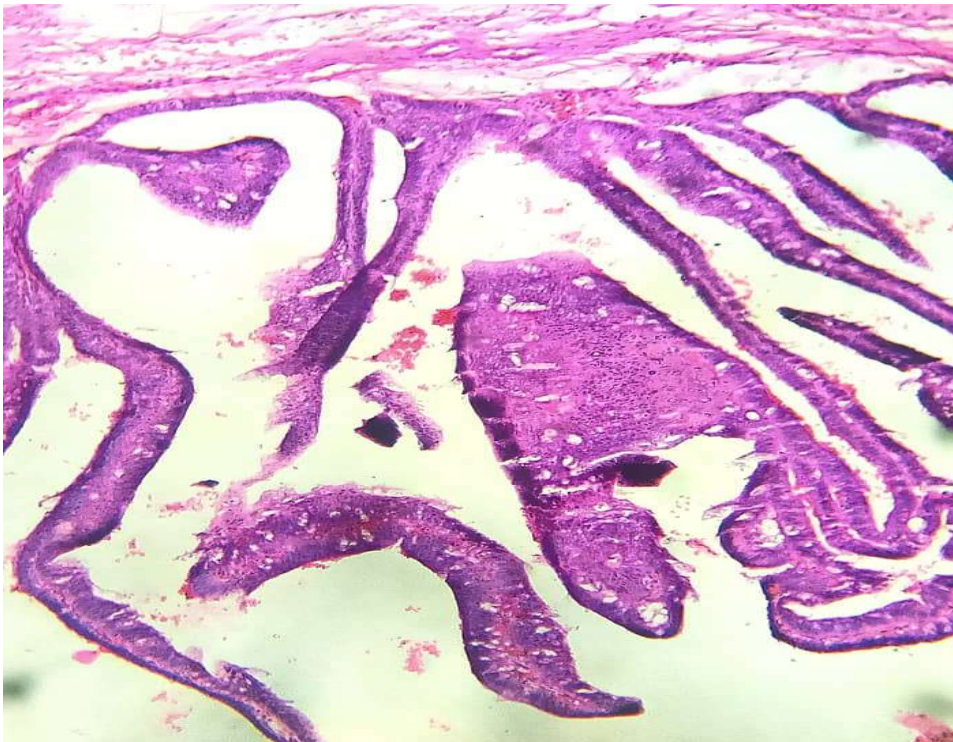


Figure 4: Higher magnification H&E x 100

DISCUSSION

The prostate gland is a pear-shaped accessory organ that has multiple common pathologies. However, it also houses some rarities. One of these, which most urologists will never encounter in their career, is Giant multilocular cystadenoma (GMCP) of the prostate.

Giant multilocular cystadenoma of the prostate (GMCP) is an exceptionally rare benign tumour, with fewer than 40 cases reported worldwide as of 2021 (Maluf et al., 1991). The first documented case

was by Watanabe et al. in 1990, and Maluf et al. further characterized the tumour in 1991. To the best of the authors' knowledge, no case has been reported in Nigeria prior to this report.

GMCP can occur at any age, with reported cases ranging from 16 to 80 years (Yuya et al., 2019). Our index case fell within this age group. Likely due to its rarity, the aetiology remains unknown, and no specific risk factors or racial predispositions have been identified thus far. GMCP typically presents as a multiloculated cystic-solid mass located in the pelvis, originating from the prostate and extending

into the space between the rectum and urinary bladder (Yuya et al., 2019; Chen et al. 2020; Wang et al., 2022). They are often identified and misdiagnosed as retroperitoneal tumours, especially when present with mixed densities (Yuya et al., 2019). Our index case was initially thought to be a retroperitoneal tumour, hence the referral to the general surgeons. Other differential diagnosis include: prostatic cysts, prostatic abscesses (especially when infected), prostate sarcoma, prostatic cystadenocarcinoma, mullerian duct cysts, cystic teratomas, phylloid variant of atypical prostatic hyperplasia, prostatic leiomyoma, pelvic mesothelioma, peritoneal inclusion cysts and lymphangiomas (Ines et al., 2019; Chen et al., 2020). At initial presentation to the urology unit, the index patient was thought to have a prostatic abscess, which was surgically drained, obscuring the true diagnosis. However, the abscess was likely as a result of infection of some of the cysts following prostate biopsy.

GMCPs are typically large, with the smallest reported case measuring less than 10cm and the largest reported by Yuya N. et al. measuring 33cm (Yuya et al., 2019; Wang et al., 2022). At the time of the second excision, our patient's mass measured 28cm, one of the largest documented cases. Due to its large size, patients frequently present with storage and voiding lower urinary tract symptoms due to pressure effects on the bladder, and they may also experience constipation (Chen et al., 2020). Our patient exhibited all the lower urinary tract symptoms, leading to an initial diagnosis of benign prostatic hyperplasia (BPH).

Although GMCP is benign, it can exhibit malignant characteristics, often adhering to adjacent structures, complicating complete surgical resection. Common structures involved include the rectum, urinary bladder, anterior rectal wall, and urethra. At the time of this patient's second exploration, his mass was attached to the bladder and the rectum. Hence, he had an R2 resection of the tumour, with an iatrogenic injury to the bladder that had to be repaired at the time of surgery.

Preoperative diagnosis of GMCP is challenging. Multimodal imaging techniques, including ultrasound (USS), computed tomography (CT), and magnetic resonance imaging (MRI), are often employed; however, radiologists must maintain a high index of suspicion, which can be difficult given the rarity of the condition. In our case, imaging studies were inconclusive, as the radiologists had not encountered this pathology before. The best assumption was a retroperitoneal tumour due to mixed density findings. The patient did not exhibit

features of malignancy and was otherwise healthy, aside from the lower urinary tract symptoms and the presence of an abdominal mass.

Prostate-specific antigen (PSA) levels may be elevated or within normal limits.¹⁻⁷ In our patient, the PSA level was persistently elevated, necessitating multiple transrectal prostate biopsies, which were either digital-guided or transrectal ultrasound (TRUS)-guided. Both biopsies revealed benign prostatic hyperplasia, which is commonly seen in GMCP cases (Yuya et al., 2019). Previous studies have not demonstrated a preoperative diagnosis made solely from biopsy (Chen et al., 2020). Cytologic examination of cyst aspirates is also typically unhelpful; in our patient, the aspirate revealed a haemorrhagic smear with no malignant cells.

Definitive diagnosis is achieved through histopathologic examination of resected specimens, which typically show multiple cysts within hypocellular fibrous stromal tissue, characterized by cuboidal cells lining the prostate glands and cysts, with positive immunohistochemical staining for PSA in the epithelial cells (Yuya et al., 2019; Chen et al., 2020; Wang et al., 2022).

Surgical resection is the primary treatment for GMCP, but complete resection is often difficult due to adherence to surrounding structures, leading to a high recurrence rate. Our patient initially underwent exploration and drainage of what was thought to be an abscess (Mohammad et al., 2022). Unfortunately, in 2015, diagnostic limitations at our facility, hindered accurate diagnosis. Following the second resection seven years later, a conclusive diagnosis of GMCP was finally made by the pathologist, resolving the diagnostic dilemma.

Some authors have reported the use of Gonadotropin releasing hormone analogs (GnRH) with varying success. GnRH was not utilized in this case due to the author's unfamiliarity with the pathology at the time of presentation (Yuya et al., 2019).

A multidisciplinary approach to patient care, involving the radiologists, pathologists, urologists and sometimes general surgeons is key to making an accurate diagnosis. The index case involved all of the above specialties, which eventually led to the above diagnosis.

CONCLUSION

GMCP is an extremely rare entity that poses significant diagnostic challenges. While clinical history and examination may suggest benign pathology, investigations can indicate possible malignancy. Histological examination of the entire

resected specimen is crucial for definitive diagnosis, as biopsies often yield nodular hyperplasia due to insufficient sampling of the solid areas. A multidisciplinary approach is essential for effective management of this complex condition.

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Conflicts of interest

The authors declare that they have no conflicts of interest.

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