

Giant dumbbell-shaped prostatic cystadenoma presenting as pelvic and scrotal mass

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Abstract

This study reports the case of an elderly man with a large tumour of the pelvic cavity and scrotum which was once diagnosed as a prostate cyst. Imaging studies considered the source of the tumour to be prostate, and the tumour was ultimately diagnosed by confirmed tissue expression of prostate specific antigen (PSA) and prostate acid phosphatase (PSAP) after surgery. This is the first report about dumbbell-shaped prostatic cystadenoma with invasive growth and even urethral damage, but there was no evidence of clear malignancy. Early diagnosis and treatment are crucial in such kinds of diseases.

Keywords: Prostatic cystadenoma, Prostate specific antigen, Pelvic mass, Scrotal mass, Benign prostatic hyperplasia cyst.

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Introduction

Prostatic cystadenoma is a roundish polycystic rare benign tumour with slow growth, distinctly demarcated from the surrounding tissue.^{1,2} It can be either attached to the prostate or entirely separate from it. Histologically, it is composed of glands and cysts without any atypical cells even though several cases have reported malignant prostatic cystadenoma arising in the prostatic cystadenoma. The definitive diagnosis is reliant on postoperative pathology with positive expression of prostate-specific antigen (PSA).

The patient usually feels comfortable, though sometimes presents with obstructive voiding which requires histopathological examination and barely any malignant tissue components are discovered.

We submit the report of a huge pelvic cystic or solid mass that entered the scrotum through the genitourinary diaphragm, reaching the hip adipose tissue at the

deepest point. To the best of our knowledge, this is the first report about dumbbell-shaped prostatic cystadenoma with invasive growth and even urethral damage but no evidence of clear malignancy. Early diagnosis and treatment are crucial in such cases

The study received informed consent of the patient and was approved by the IRB committee.

Case Report

A 66-year-old male patient, without any discomfort, was admitted to the Department of Colorectal Surgery of the Cancer Hospital affiliated to the University of Chinese Academy of Sciences (Zhejiang Cancer Hospital) in November 2017 because of a huge pelvic cystic mass. He was diagnosed with a prostatic hyperplasia cyst 18 years ago and relapsed two months after transurethral resection of the prostate (TURP) surgery. The therapy including ultrasound-guided prostate cyst puncture, drainage, while ethanol sclerotherapy was accepted later, which was effective in the treatment of dysuria. A huge mass in the left scrotum was revealed on physical examination, while rectal examination result was negative. His PSA level was 0.67 ng/ml. The enhanced computer tomography (CT) result revealed a large pelvic and scrotum mass and showed oppressive signs (Figure-1a, d). There were no indecisive boundaries between the lesion and the prostate (Figure-1b). The pelvic mass protruded downward into the perineum, with the deepest reaching the hip adipose tissue (Figure-1c). There was no evidence of involvement of any lymph node or any free fluid existing in the pelvic or peritoneal cavity. Considering that the pelvic and left scrotum lesion were derived from the mesenchymal tissue, which most likely arose from the prostate, it was speculated that it may be an aggressive angiomyxoma (AA). The patient was transferred to our hospital and received intraoperative exploration on December 4, 2017.

Consistent with the imaging results, a pelvic giant tumour (15 × 10.5 × 6 cm in size, Figure-2b) was found, which was closely related to the prostate, connected to the scrotal lesion (13 × 10 × 4.5 cm in size, Figure-2c) through the pelvic urethral genital tract. The lesion area implicated the urethra, bladder neck and the pelvic floor muscle, the

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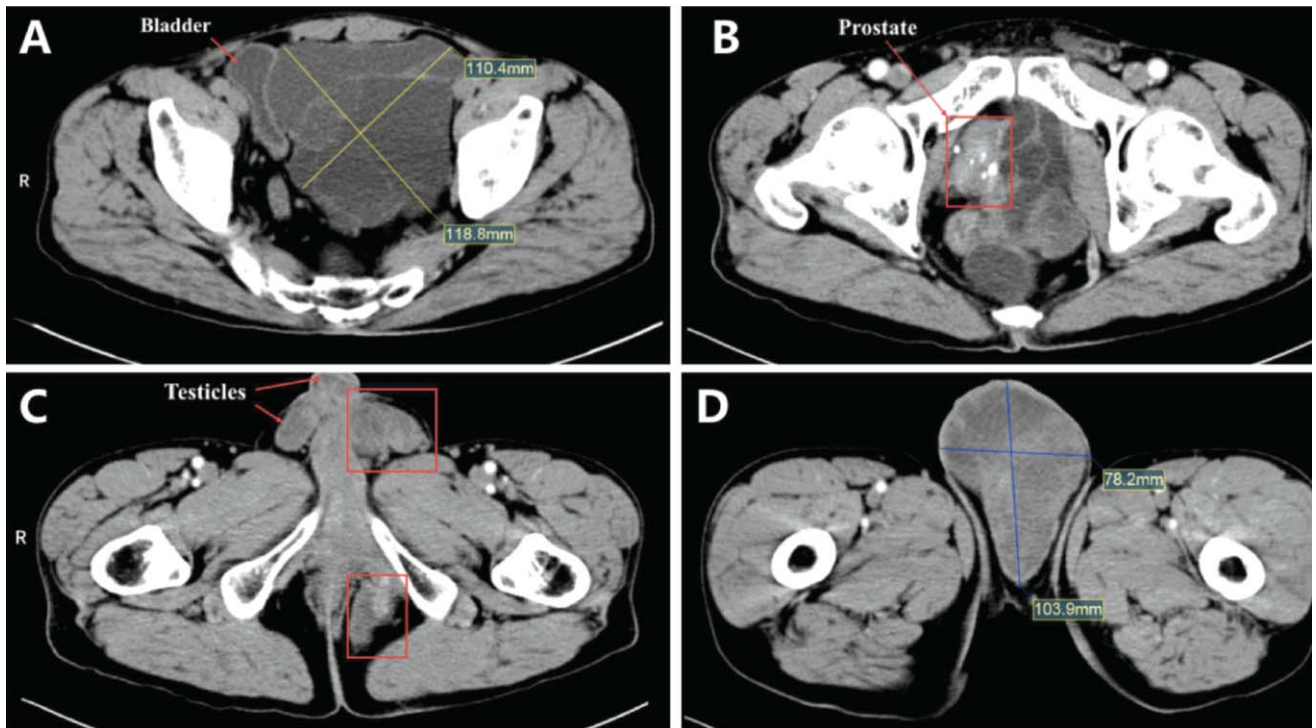


Figure-1: The results of contrast-enhanced computed tomography (CECT) for the pelvic and perineal floor. (a) Abdominal/Pelvic CT revealed a multi-cystic prostatic mass (dimension, $11.88 \times 11.04 \text{ cm}^2$) with a low-density shadow, which pushed the bladder to the right. (b) Prostatic cystadenoma contained multiple compartments and had unclear boundaries with prostate. (c) Prostatic cystadenoma passed through the urogenital diaphragm and invaded the fat of the buttocks and the urethra, in which the scrotum mass pushed the testicles. (d) CT showed a mixed-density shadow in the scrotum with a dimension of $10.39 \times 7.82 \text{ cm}^2$.

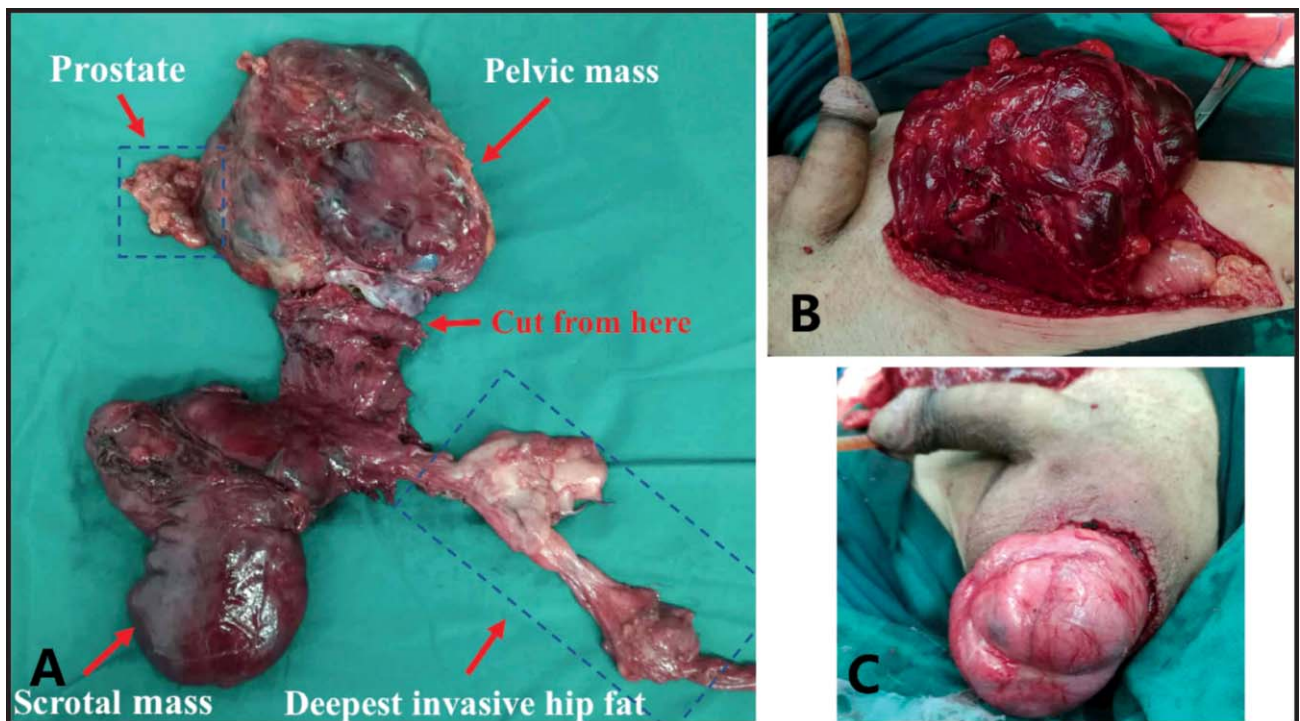


Figure-2: Macroscopic view of a large prostate tumour. (a) It was cut into two parts from the urogenital diaphragm. (b) The cystic pelvic mass was $15 \times 10.5 \times 6 \text{ cm}^3$ in size and adhered to part of the prostate tissue without clear boundary. (c) The scrotal mass was mainly composed of solid tissues with a size of about $13 \times 10 \times 4.5 \text{ cm}^3$, and another part was extended to the buttock tissue.

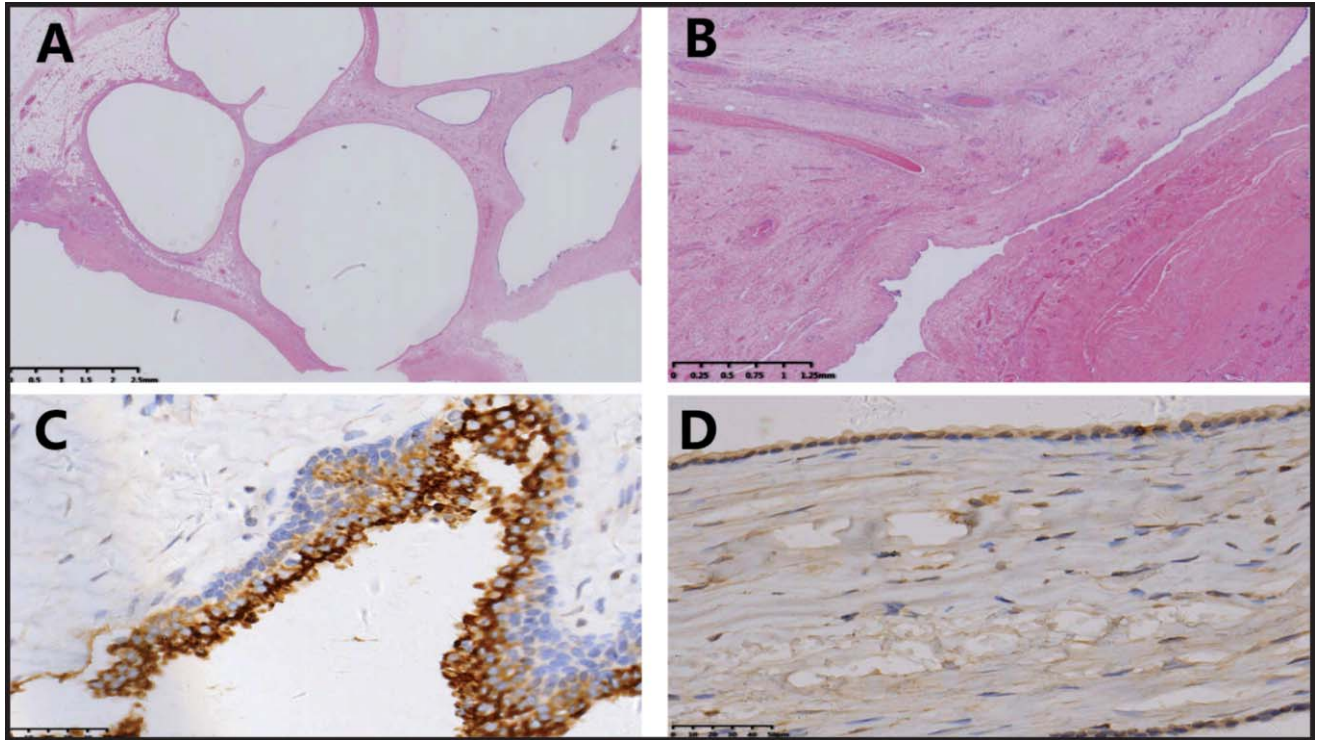


Figure-3: Histological observation of a large prostate tumour. (a) Photomicrograph showed the cystic dilated glandular structures in a pelvic mass (x20) lined by cuboidal and columnar epithelial cells, and the nuclei located in the base without any cellular atypia or mitosis. (b) The scrotal mass (x20) was mainly composed of solid structure, and interstitial cells were diffusely filled with smooth muscle tissues and new vessels. (c) Strong cytoplasmic staining of the epithelial cells for PSA antibody (x400). (d) Strong cytoplasmic staining of the epithelial cells for PSA antibody (x400).

deepest reached hip adipose tissue (Figure-2a). The tumour was separated, resected from the pelvic floor, and finally the broken urethra was repaired. The patient recovered well and the urethral catheter was removed five weeks later. There was no sign of recurrence on one-year follow-up. However, urinary incontinence persisted for a long time, and two to three diapers per day were needed.

The final pathology results confirmed the diagnosis of prostatic cystadenoma (Figure-3a) and the solid scrotal mass was filled with fibroplasia and neovascularisation (Figure-3b). Immuno-phenotypically outcome, the PSA (Figure-3c), prostate acid phosphatase (PSAP) (Figure-3d) and Prostein\PS01S were positive, expressed in epithelial cells, proving that the tumour was arising from the prostate gland. Besides, there were different degrees of expressions of CD34 in vascular endothelium and Ki67, and was 20% positive expressed on Ki67 which may be associated with cellular proliferation.

Discussion

Giant multilocular cystadenoma of the prostate is a rarely encountered tumour, with less than 30 cases having been

reported. The problem may cause urinary tract obstruction, even azoospermia due to mass effect.^{2,3} In some cases, it presents as a palpable abdominal mass, while the patient carries the growing tumour for over a decade without any discomfort. Considering both the previous medical history and the slow-growing characteristic of a benign tumour, it might be misdiagnosed or combined with a simple cyst. Unfortunately, the previous post-operative pathological results were not available and thus it was hard to figure out. Furthermore, the scrotal lesion's invasive growth pattern and pathology of fibrous tissue tumour-like hyperplasia might be the result of gene mutation, relating to vascular myofibroblastoma and ethanol sclerotherapy. It has been reported that ethanol could activate endothelial cells and promote the formation of fibrous tissues and blood vessels, increasing the risk of metastasis.⁴ But the potential relationship between ethanol and tumour progression was complicated and remained to be further investigated.

The serum PSA released from the columnar secretory tumour cells and expression level could be normal or increased. There is no evidence to prove that the PSA level

was associated with tumour size or related to the degree of malignancy. In our case, even though a huge benign tumour was discovered, the PSA level remained low. Up to now, it remained less than 1ng/ml.

Prostatic cystadenomas provided lots of diagnostic challenges, which should be differentiated from retroperitoneal malignant mass, prostatic hyperplasia cyst, prostatic abscess, teratoma, Müllerian cyst, seminal vesicle cyst and so on.^{2,5,6} Imaging examinations such as ultrasound, CT and magnetic resonance imaging (MRI) could determine the shape, size, location and the relationship between mass and surrounding tissues, and be helpful with differential diagnosis and therapeutic plan. In our case, it was conjectured as a prostate-derived malignant soft tissue tumour considering the different morphologic and clinic-pathological features of a scrotal lesion comparing the pelvic mass. The pathology report finally determined the diagnosis and showed no evidence of existence of malignant cells, even though some experts have reported cases of prostatic cystadenoma with high-grade prostatic intraepithelial neoplasia (PIN)6 and cystadenocarcinoma.⁷⁻⁹

Complete surgical excision by an open or laparoscopic approach was considered curative, but some others thought it was unnecessary. Dr Datta¹⁰ first reported medical treatment, gonadotropin-releasing hormone antagonist (GnRH antagonist), showed an excellent result against biochemical failure, pathologic recurrence of giant prostatic cystadenomas. However, it may not be useful for patients with low PSA levels. Incomplete TURP surgery could not have treated the prostatic cyst or cystadenoma effectively but could lead to a rapid recurrence to worsen urinary tract symptoms. Sclerotherapy can be effective for simple cysts and abscesses, but cannot cure the cystadenoma, in fact can make it "go into hiding" and delay the treatment. The patient finally accepted open surgery for the removal of the mass. The main post-operative complication was urinary incontinence, which reduced the quality of life to some extent. In general, considering the tumour size, growth pattern and pathology, even the possibility of transition to malignancy henceforth, radical excision was of benefit and it was believed that continued post-operative function

rehabilitation training could help the patient deal with the lengthy convalescence and final outcome.

Conclusion

Prostatic cystadenoma is a complex benign tumour that is easily misdiagnosed. The possibility of malignant transformation and combination of adenocarcinoma requires more attention in clinical practice. Surgery is still the main means of treatment, and drugs or interventional treatments need to be further explored.

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