Case Report

Giant multilocular prostatic cystadenoma in a 16-year-old male with difficulty in defecation: case report and literature review

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Abstract: Giant multilocular prostatic cystadenoma is a rare benign tumor that originates from the prostate gland. It usually is between the rectum and the bladder, and is composed of predominantly cystic enlarged prostatic glands in a fibrous stroma and spreads extensively in the pelvis. The mass usually causes a series of obstructive symptoms by compressing adjacent organs. Here we report a case of a giant multilocular prostatic cystadenoma in a 16-year-old patient, who is the youngest case reported up to now, complaining of difficulty in defecation.

Keywords: Giant multilocular prostatic cystadenoma, pelvic mass, prostatic neoplasm, difficulty in defecation, benign lesion

Introduction

Multilocular prostatic cystadenoma (MPC) is a rare benign tumor originating from the prostate gland [1]. It usually is between the rectum and the bladder, and is composed of cystic enlarged prostatic glands in a fibrous stroma. Typically, MPC does not invade adjacent organs due to its benign nature. However, in some rare cases, local aggressiveness and adherence to surrounding viscera has also been reported [2]. The age of patients with MPC ranges from 23 to 80 years according to literature reviewed [3]. Most cases reported symptoms of difficulty in urinary voiding, such as poor stream, intermittency, straining, and sensation of incomplete emptying. The less common symptoms include hematuria, a palpable abdominal mass, azoospermia, and difficulty with defecation [1]. Imaging examinations provide valuable information including tumor characteristics and adjacent structures. Definite diagnosis depends on histopathology. Here, we present a case of a 16-year-old patient, the youngest case published, complaining of difficulty in defecation which led to a diagnosis of a giant multilocular prostatic cystadenoma.

Case report

A 16-year-old boy presented with a 1-month history of difficulty in defecation leading to the discovery of a pelvic mass. He complained no other symptoms and was well in medical history before. Digital rectal examination revealed a cystic-solid mass. The results of laboratory tests were normal except for an elevated serum prostate specific antigen (PSA) level of 17 ng/ml.

CT scan was performed, which revealed a cystic-solid mass seemed arising from the left seminal vesicle gland or prostate in the pelvis, with a maximum cross section of about 8.0 cm × 8.8 cm (Figure 1). MRI confirmed a pelvic cystic-solid mass, located between the rectum and the bladder (Figure 2). A pelvic cystic-solid mass, measuring 8.0 cm × 7.8 cm × 6.3 cm, with an unclear delimitation from the prostate and the seminal vesicle, was demonstrated by
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PET-CT, with the standard uptake value (SUV) of 1.0 (maximum 2.8). B-ultrasound depicted an 8.6 cm × 6.7 cm × 6.0 cm cystic-solid mass on the left posterior side of the bladder, with a regular shape and less peripheral blood flow signal (Figure 3). Subsequent biopsy of the mass was performed with B-ultrasound guidance for a second time after a negative finding. Both of the biopsy findings showed normal prostatic tissue without cancer.

The patient underwent a laparoscopic resection of pelvic mass (Figure 4). In surgical exploration, a giant mass with a complete capsule was discovered in the pelvis. It was located between the rectum and the bladder, while the base of the tumor was attached to the prostate. The tumor did not invade the surrounding intestine, seminal vesicle, pelvic wall, ureter, bladder, or other organs. Intraoperatively, special attention was paid to protect the organs.

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**Figure 1.** CT scan. A. Sagittal CT image showed a multilocular cystic mass in the pelvis, compressing the rectum and bladder. B. The same section in an axial CT image showed a well-circumscribed, multilocular mass with multiple septations.

**Figure 2.** MRI. A. Sagittal MR imaging showed a large, multilocular, encapsulated mass with numerous thin septations and variable-sized cysts, located between the rectum and the bladder. B. The same section in an axial MR image showed a pelvic cystic-solid mass, displacing the bladder and compressing the rectum.
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Figure 3. B-ultrasound. A transabdominal ultrasound examination of the pelvis showed a cystic-solid mass on the left posterior side of the bladder, with a regular shape and less peripheral blood flow signal.

Figure 4. Intraoperative view. Intraoperative view showed a giant mass with a smooth surface in the pelvis, attached to the prostate.

and tissues related to reproductive and sexual function such as vas deferens, seminal vesicles, and nerves. The tumor was resected completely and successfully and was 6.0 cm × 7.5 cm × 4.4 cm in size and grayish pink. Multicystic spaces were revealed after sectioning. The cysts were filled with dark brown viscous liquid (Figure 5). Final histology indicated multilocular prostatic cystadenoma. Immunohistochemical staining was positive for PSA. The patient was followed up for two years with no complaint of voiding or defecation symptoms and there was no evidence of recurrence. Erection and ejaculation were as normal as before the operation.

Discussion

Giant multilocular prostatic cystadenoma (MPC) is such an extremely rare benign neoplasm originating from the prostate gland that only a few cases have been reported. In the previously reported cases [3], the age of patients ranged from 23 to 80, while our case was only 16 years old, the youngest case of MPC to date. This disease is prone to be misdiagnosed by clinicians not only for its rarity, but also by its insidious onset. These patients presented with no corresponding symptoms until the masses grew large enough to compress the adjacent organs [3]. Obstructive voiding symptoms and urinary retention, similar to the symptoms of benign prostatic hypertrophy (BPH), were mentioned almost in all the cases of prostatic cystadenoma. So, it is easily misdiagnosed as BPH or malignant neoplasm of prostate [4]. Other less common symptoms include hematuria, a palpable abdominal mass, azoospermia, and difficulty with defecation. However, dysdefecation as the exclusive symptom of MPC was only reported in our case. It suggests that we should pay attention to the diagnosis of MPC as well as digestive tract tumors in patients with a pelvic mass and rectal obstruction.

MPC can be located in the prostate or connected with prostate, or completely separated from
the prostate [5, 6]. Thus, the differential diagnoses include other prostatic cystic lesions such as cystic change of benign prostatic hyperplasia, prostatic retention cysts, müllerian cysts, and prostatic abscess [7]. Besides, some retroperitoneal cystic lesions such as lymphangioma and prostatic sarcoma should also be considered [8]. In addition, seminal vesicle cystadenoma also needs to be taken into consideration before the source of the mass is determined. Moreover, prostatic cystadenoma can also occur in heterotopic prostate. The prostatic tissue that occurs outside the normal part of the prostate is called heterotopic prostate which can occur in the bladder, vesicorectal space, rectal wall, and other organs.

PSA elevation has been reported in several cases [3-5, 8-11], the highest of which was 68.2 ng/mL [9]. In our case, the PSA level was 17 ng/ml. However, patients with MPC with normal PSA were also reported [1, 7, 12, 13]. Consequently, PSA level is not specific for diagnosis of MPC. At imaging, MPC presents as large multiloculated masses, composed of variable-sized cysts, generally with some thin enhanced septations [7]. The mass is usually located between the bladder and rectum as in our case, but Fariña-Pérez [13] reported a special case in which ultrasound examination revealed a cystic lesion in the midline of the prostate growing inside the bladder and causing high post-void residual. Because of the benign nature of this disease, the masses always oppress the adjacent organs, such as bladder and rectum, but generally MPCs do not invade surrounding tissues and organs. According to these imaging features, we can make a preliminary diagnosis of MPC before operation, although it has not been confirmed by pathology. At the same time, imaging findings can also provide useful information about the characteristics of the tumor and its relationship to adjacent organs, through which we can determine the appropriate surgical approach [3].

Although imaging studies may provide useful information and suggest the possibility of MPC, a clear and definitive diagnosis is possible only through histological means. Almost all the previous reports have described the pathologic features of MPC [1, 7, 12]. The
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tumor is composed of glands and cysts. The inner wall of the cysts is lined with cuboidal epithelial cells to columnar epithelial cells without obvious heteromorphism or mitosis, and the nucleus is located in the base of the cells. The stroma is composed of smooth muscle and fibrous matrix. Besides, metaplasia of squamous epithelium and urothelium can also be found. Our case confirmed these features (Figure 6). With regard to the results of immunohistochemical staining, as in our case, reports indicated that MPC was positive for PSA, which confirmed the mass originating from the prostate.

Complete surgical resection is the main treatment of MPC [3]. In our case, a successful laparoscopic resection of tumor was performed due to absence of invasion of adjacent organs. However, most of the reported resections received laparotomy. Adjacent organs which were difficult to separate or suspicious for malignancy invasion were excised [1]. In addition, some surgery-related complications have been reported. Nakamura [1] described a patient who developed a pelvic abscess due to urine leakage from the prostatic urethra, and the patient recovered conservatively. Pandi [12] described an accidental injury of the anal canal which required rectoraphy with protective colostomy. The patient underwent surgery for restoration of large bowel continuity two months later. Teixeira [7] described a rectal leak that was surgically corrected subsequently.

Some cases were partially excised due to benign nature; however, recurrences should not be ignored. Recurrences of tumor were reported to be treated by surgery generally, such as mass excision and pelvic exenteration. El Rahman [5] reported a case of a 74-year-old male who presented with a retrovesical recurrence of prostatic cystadenoma after 16 years, treated with a laparoscopic approach. It is also reported that gonadotropin-releasing hormone antagonist is effective for recurrence of giant MPC [1]. In some infrequent cases, MPC may coexist with a malignant lesion [6]. Lee [6] reported a case of prostatic cystadenoma harboring conventional prostatic adenocarcinoma. Uguen [10] diagnosed a multilocular prostatic cystadenocarcinoma associated with a cystadenoma treated with a total pelvic exenteration. After 2 years of follow-up, the patient did not present any tumor recurrence or PSA level elevation under hormonal therapy.

Most of the patients were completely asymptomatic and recovered well, with no complaints of any urinary disorders or sexual dysfunction at their follow-ups. El Rahman [5] reported a case with the longest follow-up. After 16 years of follow-up, the patient had a retrovesical recurrence of prostatic cystadenoma, and a laparoscopic approach was performed as salvage treatment. The patient was continuously followed up for another 48 months, during which he remained free of lower urinary tract symptoms without signs of recurrence.

We present our experience with the youngest person with giant multilocular prostatic cystadenoma. This disease is still difficult to diagnose for its relatively rare condition. When a retrovesical cystic lesion fills the pelvis in a male patient with voiding or defecation symptoms, the possibility of multilocular prostatic cystadenoma should be considered. Complete excision may preempt recurrence. Hormonal therapy is sometimes used, but its role needs more exploration in clinical practice.

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Disclosure of conflict of interest

None.
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