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International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report

Hemangioma of the prostate with retrograde ejaculation - a case report

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ARTICLE INFO

Keywords: Hemangioma Prostate Retrograde ejaculation Infertility

ABSTRACT

Introduction and importance: Hemangioma of the prostate is rarely reported. We here describe a hemangioma of the prostate in a 31-year-old man.

Case presentation: The history, imaging characteristics, treatment and one year follow-up results were well documented. The chief complaint was retrograde ejaculation. A $3.1~{\rm cm}\times2.9~{\rm cm}$ mass in the prostate was detected by ultrasound. Transurethral resection of the prostate (TURP) was performed.

Clinical discussion: Pathological examination revealed the mass was hemangioma. Immunohistochemical study found the tissue was SMA, CD34, CD31 positive, but D2–40 negative. Imaging feature combined with pathological result suggests the diagnosis of hemangioma of the prostate. One year follow-up revealed the patient was infertile

Conclusion: We suggest TURP should be performed to remove the hemangioma. Combined treatment is necessary to resolve the patient's infertility.

1. Introduction

Hemangioma is a common disorder pathologically characterized as capillary hemangioma, cavernous hemangioma or racemose hemangioma [1]. The expression of the disease varies substantially regarding affected organs. Hemangioma in the prostate is the most rarely reported hemangioma in urinary system [2]. We report a case of hemangioma of the prostate in a 31-year-old man found by ultrasound with a chief complaint of retrograde ejaculation. The treatment and short-term follow-up results were well documented. This work was reported in line with the SCARE 2020 criteria and the PROCESS criteria [3,4]. The patient gave informed consent for publication.

2. Case report

A 31-year-old male was admitted to hospital complaining of retrograde ejaculation. Sperm density of the patient was merely1.0 million/mL, which is extremely low referring to the normal value. Oligospermia is defined as a sperm density of less than 20 million/mL [5]. The total number of sperm was only 8.1 million (the normal value is 39 million or more). A large amount of sperm was found in the urine during the first

urination after ejaculation, which was in consistent with his chief complaint [6]. The urinary sperm count after ejaculation was 128,000/mL. He also suffered voiding symptoms as acraturesis and weak urine flow. Hematuria, urinary retention, frequency, urgency, or renal function insufficiency was not found in this case. The total prostate specific antigen (PSA) in venous blood was 1.89 mg/L, which was lower than the upper limit of 4 mg/L.

2.1. Imaging characteristics

Transrectal ultrasound: Transrectal ultrasound revealed a 3.1 cm \times 2.9 cm mass in prostate (Fig. 1a). The uneven isoechoic mass with abundant blood flow protruded into the bladder. The venous plexus around the prostate was dilated. The bilateral testicles were in smaller size, 35.6 mm \times 17.5 mm \times 24.0 mm on the left and 37.5 mm \times 16.4 mm \times 22.6 mm on the right, with normal position and blood flow. The size and shape of bilateral epididymis were normal. Bilateral spermatic vein were normal with the widest inner diameter of 2.2 mm on the left side and 2.1 mm on the right side. No reflow was found during supine position on both side. Reflow was detected during Valsalva maneuver on the right side. The reflow time was more than 6 s. Bilateral seminal

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https://doi.org/10.1016/j.ijscr.2022.107102

Received 25 February 2022; Received in revised form 15 April 2022; Accepted 15 April 2022 Available online 19 April 2022

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vesicles, vas deferens and ejaculatory ducts were normal.

Magnetic resonance imaging (MRI): MRI found an irregular nodule in the central lobe of the prostate protruded to the bladder. The nodule was about 21 mm \times 8 mm \times 21 mm in size, showing isointensity on T1-weighted images (Fig. 1b). The mass was hyperintense on T2-weighted images and fat suppression images. Thin layer of low signal capsule with slight enhancement was observed around the nodule. Multiple low signal spots, unclear boundary and enhancement were also observed in T2-weighted images. The prostate capsule was intact, and the surrounding fat tissue was clear. The size and shape of bilateral seminal vesicles were normal. No enlarged lymph node or effusion were found in pelvic cavity. The shape and signal of rectum were normal.

2.2. Management

After spinal anesthesia, the patient was placed in lithotomy position. Resectoscope was inserted transurethrally. Elevated bladder neck was observed (Fig. 1c). Hemangioma at the apex of the prostate was obviously protruding to the bladder cavity. The hemangioma was resected following routine transurethral resection of the prostate (TURP) procedure. No adjuvant management was given after TURP. Urinary catheter was removed 1 weak after surgery. The patient accepted the management strategy and shared no other perspective on the treatments.

2.3. Pathological findings

2.3.1. Pathological examination

A 1 cm \times 1 cm tissue taken from the prostate was monitored. Abundant smooth muscle and sinus like vessels were observed in the tissue. The tissue section was reviewed by two pathologists. The pathological diagnosis was hemangioma (Fig. 1d).

2.3.2. Immunohistochemical study

Immunohistochemical staining exhibited SMA, CD34 and CD31 were highly expressed in the tissue, but D2–40 was negative. The expression

pattern was consistent with the pathological diagnosis of hemangioma.

2.4. Follow up

The patient was discharged from the hospital with satisfactory urinary stream. At 1-year follow-up, the patient didn't complain of any voiding problem. Transrectal ultrasound showed no recurrence of the hemangioma in prostate. But he still suffered from low semen volume and oligospermia caused by retrograde ejaculation. The urinary sperm count after ejaculation was 94,000/mL. The patient's sperm density increased to 6.5 million/mL in the 1 year follow-up. Infertility was diagnosed after 1-year sexual activity after the surgery without any form of conception control. Thus, assisted reproductive technology was recommended.

3. Discussion

Due to the scarcity of hemangioma in the prostate, diagnosis and management remain difficult for practitioners. In all reported cases, diagnosis was elusive until made by pathologists [2,7–9]. Surgical complication even lead to one patient's death caused by uncontrollable bleeding [9]. Its nature history and management decision making merit further study.

The chief complaint of the present case was retrograde ejaculation. Various clinical manifestation has been reported, including urine retention [2], nocturia, hematuria [9], hematospermia or postejaculation urethral bleeding [8]. No specific manifestation distinguishes hemangioma of the prostate from benign prostatic hyperplasia (BPH). Hematospermia and urethral bleeding are commonly observed in infectious conditions or malignant disease. So the diagnosis had to be made according to pathological examination in previously reported cases. The young age (31 years) of our patient might distinct the mass from BPH. Normal PSA level was helpful to distinguish the mass from malignant lesion.

Due to the lack of featured complaint, cystoscopy and transrectal power Doppler ultrasonography were recommended to be employed for

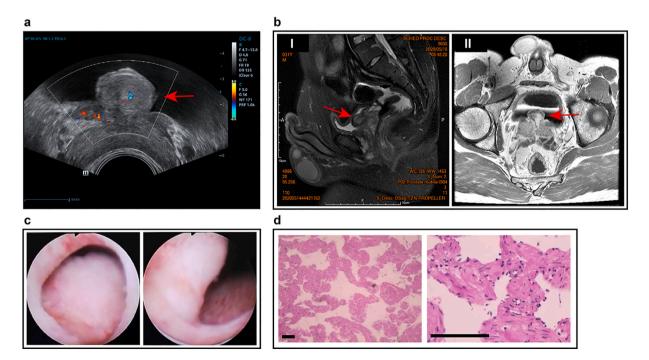


Fig. 1. MRI and transrectal ultrasound were applied to the patient. TURP was performed followed by histological examination. a. Transrectal ultrasound found the uneven isoechoic mass of prostate protruded to the bladder. b. MRI showed the mass in central lobe of the prostate. I. T2-weighted image. II. Enhanced T1-weighted image. c. Cystoscope showed elevated bladder neck. d. HE staining revealed the vessel structure of the dissected prostate tissue. Scale bar = $100 \, \mu m$. Red arrows indicate the hemangioma. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

diagnosis. Cystoscopy showed bladder trabeculation and injected bladder mucosa in one case [9]. The mass protruding into bladder cavity was observed during TURP performed in our case. Although infectious bleeding or bladder cancer can be discerned by cystoscopy, we found it difficult to distinguish hemangioma from BPH. Transrectal power Doppler ultrasonography and MRI are able to show the difference of benign prostatic hyperplasia and hemangioma. In addition, the resected tissue was evaluated by immunochemistry which demonstrated positive expression of smooth muscle marker (SMA), mesenchymal marker (CD34) and vascular endothelial marker (CD31), and the expression of mesothelial marker (D2–40) was negative. This particular expressing pattern and irregular vasculature containing structure along with prostate tissue in histological section gave rise to the diagnosis of hemangioma originated from prostate.

TURP was performed for the patient in our report. Neither intraoperative nor postoperative complication occurred. In previous reports, patients were treated as BPH by perineal intracapsular prostatectomy or transurethral resection [9]. Postoperative urine retention was found in one patient subjected to transurethral resection, followed by selfcatheterization [8]. Continuous bleeding of surgical wound on the prostate occurred in the patient treated by perineal intracapsular prostatectomy, which resulted in the patient's death [9].

In the present case, TURP relieved the voiding symptom. But the follow up results revealed removal of the hemangioma was not enough to improve the sperm density suggesting the anatomical cause of retrograde ejaculation had not been restored. The hemangioma may have affected the internal sphincter near the bladder neck, resulting in retrograde ejaculation. It should be noted that the thermal effect from the full resection of a hemangioma can also damage the internal sphincter, causing an ongoing instability of the internal sphincter after the surgery. Despite the fact that the patient still reported low semen volume after surgery, the sperm density was higher than it was before surgery. Retrograde ejaculation is the main cause of 1.2% cases of infertility [10]. The benefit from treatments that include electroejaculation [11,12], penile vibratory stimulation [13] and oral medication is controversial [6]. Therefore, assisted reproductive technology might be the recourse for the patient in our case.

4. Conclusion

We present a rare case of hemangioma affecting the prostate with chief complaint of infertility. We recommend TURP should be performed to remove the hemangioma completely. A complete resection prevents recurrence, and relieves the voiding symptoms. In addition, tissue for histological examination can be obtained, which is essential for a clear diagnosis. Because of the complexity of etiology, combined treatment is necessary to resolve the patient's infertility.

Funding

The authors did not receive support from any organization for the submitted work.

Ethics approval

This study was performed in line with the principles of the

Declaration of Helsinki. Approval was granted by the Ethics Committee of the First Affiliated Hospital, Sun Yat-Sen University.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Conceptualization: Zi Wan, Chun-hua Deng. Investigation: Zi Wan, Bi-cheng Yang. Methodology: Yan Guo, Chun-hua Deng. Project administration: Zi Wan, Hai-ming Cao. Supervision: Jin-huan Wei, Chun-hua Deng. Writing – original draft: Zi Wan, Bi-cheng Yang. Writing – review & editing: Zi Wan, Chun-hua Deng.

Declaration of competing interest

None.

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