Abstract

Lymphangioma of the urinary bladder is a very rare tumour in adulthood. Robotic partial cystectomy is evolving for treatment of a limited number of bladder tumours. We describe a case of an adult woman with a bladder dome lymphangioma for which robotic partial cystectomy was carried out.

Introduction

The presence of intra-abdominal lymphangioma is rare in adults. Very few cases of lymphangioma of the urinary bladder have been reported. These lesions are usually quiescent. The importance of these bladder masses is that they mimic bladder carcinoma. Robotic partial cystectomy is evolving for the management of bladder tumours.

Case report

A 27-year-old woman presented with painless gross hematuria for 6 months. Occasionally she had suprapubic pain. She had history of abdominoplasty, mammoplasty, gastric banding and bronchial asthma. There was no history of recurrent urinary tract infection, fever, diabetes, hypertension or allergy.

Physical examination showed a midline scar of previous abdominoplasty. Complete blood count and renal profile were normal. Urine analysis showed red blood cells notable for >50 per high power field and a negative urine culture. Computerized tomography (CT) scan showed a heterogeneously enhancing mass arising from or involving the anterior urinary bladder wall measuring 3.6 × 3.8 cm, causing some stranding of the adjacent fat (Fig. 1, panel A). Cystoscopy under general anesthesia showed a solid mass at the dome of the bladder covered by a normal mucosa; the mass was mobile under bimanual examination. Transurethral resection biopsy of the mass was carried out and histopathology revealed only fibrosis with chronic inflammation.

Using the da Vinci Surgical Robotic System (Intuitive Surgical, Inc., Sunnyvale, CA), an elective partial cystectomy was carried out for complete excision of the mass via an intraperitoneal approach (Video 1). In the modified low leg lithotomy position, 4 trocars were inserted: a 12-mm trocar for the camera in the supraumbilical area using an open technique; 2 trocars for the right and left robotic arms brought 8 cm lateral and inferior to the camera trocar; and a 10-mm assistant trocar inserted parallel to the camera trocar. The procedure started by identifying an evident mass, located superior to the urinary bladder. Using blunt and sharp dissection and electrocautery, the peritoneal reflection was incised over the mass anteriorly, laterally and posteriorly. The mass was completely mobilized except where attached to the superior vesical wall. The bladder was mobilized from the side wall of the pelvis. No cystoscopic guidance was used for the cystostomy. The wall of the bladder was incised with a 1-cm safety margin and the mass removed with the covering peritoneum and underlying bladder wall. The bladder was closed in 2 layers with running absorbable sutures in a water tight fashion. The midline incision was extended to remove the mass and the abdominal wall was closed.

Pathological examination showed that the removed specimen was 7 × 4 × 4 cm in size. The excised bladder wall area measured 2.5 cm in maximum diameter. The cut section showed a solid tumour 4 cm in diameter with dilated vessels in some areas. Histopathology of the mass showed a benign lymphangioma on hematoxyline and eosine staining arising within the wall of the urinary bladder with a negative surgical margin (Fig. 2, panel A). Immunohistochemistry with CD31+ and D2-40+ antibodies confirmed the abundance of lymphatic endothelium lining of the vascular spaces of the mass (Fig. 2, panels B and C). At the site of transurethral biopsy, reactive urothelium with subepithelial chronic inflammation, foreign body giant cell reaction and calcifica-
tion were seen. No malignancy was found and the remaining bladder mucosa was normal. At follow-up at 6 months, the patient was asymptomatic, urine analysis was normal and CT showed normal urinary bladder (Fig. 1, panel B).

**Discussion**

Lymphangioma is a benign lesion resulting from abnormal development of the lymphatics. Lymphangiomas are rare and affect mainly the head and neck; most are diagnosed before 5 years of age. More recently, the use of colonoscopy has identified a significant number of large intestinal lymphangiomas in adults in Japan. However, bladder lymphangioma are extremely rare worldwide. Lymphangiomas of the neck exhibit tumour-like pathogenesis owing to the high expression of angiogenic inducers and low expression of inhibitors. Areas of inflammation and fibroplasia are sometimes associated with lymphangioma. The initial transurethral biopsy showed chronic inflammation and fibrosis. In the final pathology of the excised mass, the presence of inflammatory reaction in the bladder mucosa was associated with the transurethral biopsy site. Immunohistochemistry using D2-40 and CD31 antibodies confirmed that the excised lesion was lymphangioma. D2-40 is a novel monoclonal antibody which is highly sensitive and a specific marker for lymphatic endothelium in normal tissue, Kaposi sarcoma, Dabska tumour and lymphangioma. CD31 antibody, on the other hand, is less specific for lymphangiomas and cross reacts with hemangiomas.

The bladder mass was domal and covered with intact mucosa raising the suspicion of an adenocarcinoma probably of urachal origin. Transurethral biopsy was not conclusive and an elective surgical excision was planned. Partial cystectomy has been used safely to treat selected cases of malignant bladder tumour. Laparoscopic partial cystectomy is evolving, with better control of suture closure of the bladder incision.

A simultaneous cystoscopy is sometimes used with laparoscopic or robotic partial cystectomy to define the boundaries of the bladder lesion and guide the cystotomy. Transillumination, transurethral cold knife incision of the bladder mucosa around the tumour or combined transurethral resection of bladder lesion have been described. We did not use the cystoscopy-assisted cystotomy because the bulk of the tumour was outside the bladder. By freeing the dome of the bladder and tumour from the pelvic wall side, we were able to apply traction on the mass cephalad facilitating placement of the bladder wall incision with a 1-cm safety margin away from the tumour.

**Conclusion**

We report a rare case of bladder lymphangioma in an adult. We successfully managed to completely excise the tumour.
with robotic partial cystectomy unassisted with transurethral endoscopy.

Competing interests: None declared.

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References


Correspondence: Dr. Mohammed F. Al-Otaibi, King Faisal Specialist Hospital & Research Center, P.O. 3354 (MBG33), Riyadh, Saudi Arabia 11211; fax: +966-01-442-4301; otaibim@kfshrc.edu.sa

Fig. 2. Histopathology of the bladder tumour (10×). A: H&E stain showing dilated proliferating lymphatic vessels within the muscularis propria B: D2-40 marker is positive for lymphatic endothelium. C. CD31 endothelial marker is positive in both blood and lymphatic vessels.