CASE REPORT

Laparoscopic excision of an inflammatory myofibroblastic tumour of the bladder disguised as deep infiltrating endometriosis

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SUMMARY

Inflammatory myofibroblastic tumour (IMT) of the bladder is a rare tumour of indeterminate malignant potential with myofibroblastic differentiation, with a generally benign but rarely aggressive behaviour. Vesical IMT is usually treated by transurethral resection or partial cystectomy. Herein we describe a case of a woman who underwent laparoscopic excision of an IMT of the bladder, initially diagnosed as deep infiltrating endometriosis.

BACKGROUND

Our main motivation for presenting this case is that inflammatory myofibroblastic tumour (IMT) of the bladder is a rare mesenchymal tumour by an uncertain malignant potential which was first described by Roth in 1980.¹ The most common site for IMT is the lung, however it may arise in almost every location of the body.² Though bladder location is rather uncommon, it arises from the bladder submucosal stroma as a polypoidal growth and can be easily mistaken for a malignant neoplasm or even a benign disease, such as in our case.^{3,4} Herein we describe a case of a woman who underwent laparoscopic excision of an IMT of the bladder, initially presumed as deep infiltrating endometriosis.

CASE PRESENTATION

A 46-year-old G2P2 woman presented with increased dysuria, suprapubic pain and dyspareunia for the past few months. She had no history of urinary tract infection, abdominal trauma or sexually transmitted disease. She had regular periods and a recent normal Papanicolaou smear. Bimanual uterine palpation evoked tenderness at the anterior vaginal fornix, in close proximity to the uterine corpus.

INVESTIGATIONS

Laboratory studies were normal, except for microscopic haematuria. Abdominal ultrasound raised the suspicion of an infiltrating mass in the bladder wall. MRI showed a 3×4 cm mass arising from the posterior bladder wall, with deep muscle invasion, in close proximity to the anterior uterine wall (figure 1). Cystoscopy revealed a broad-based, lobulated tumour which gave the macroscopic appearance of an endometriotic nodule. Cytological analysis of urine did not detect any malignant cells. A biopsy was not taken at the time of cystoscopy.

The patient was keen on having a minimal invasive operation and laparoscopic excision of the lesion was decided. An informed written consent was obtained from the patient prior to the operation. This case had an ethics approval for publication, obtained by our local ethics committee.

TREATMENT

A Foley catheter and ureteric stents were placed preoperatively and a urologist was present during the entire operation. The laparoscope was inserted into the abdominal cavity from an 11 mm supra-umbilical incision using the open access technique. Three additional trocars were introduced under direct vision. The uterus and both adnexa were normal with no visible endometriotic lesions. The bladder tumour was protruding just underneath the vesicouterine peritoneal fold (figure 2A). Bipolar scissors were used to dissect the peritoneum just around the lesion with the intention of achieving mobilisation of the bladder (figure 2B). Cystotomy was done with the monopolar hook. The multilocular mass was identified and the incision was made circumferentially around the tumour (figure 2C). Care was taken to excise the entire tumour (figure 2D,E). The specimen was removed from the abdominal cavity with a bag. The bladder wall was then closed in one layer with interrupted 2–0 monocryl sutures (figure 2F). A single layer is considered adequate to provide safe approximation of the bladder wall without unnecessary tension to the tissue. Water tightness of the bladder was

Figure 1 Red arrows indicate the infiltrating lesion. The mass was in close proximity to the bladder.

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Figure 2 (A) Arrows define the bladder lesion. (B) Dissection of the Retzius' space with the bipolar scissors and mobilisation of the mass. (C) Entering the bladder with the monopolar hook so the multilobular tumour is exposed. (D, E) The specimen is excised and the defect of the bladder is closed with one layer of interrupted 2–0 monocryl suture. Foley catheter and ureteric stents were away from the surgical plane. The defect was estimated to be <30% of the bladder wall. (F) Final laparoscopic view of the excised area.

confirmed by instillation of 250 mL of water into the bladder. The ureteric stents were removed at the end of the procedure and the catheter was removed on the 10th postoperative day. Histopathology revealed the fusiform or rounded cells arranged in a storiform pattern and the inflammatory infiltrate composed of scattered mononuclear cells and polymorphonuclear leucocytes. Histopathological features and also positive immunohistochemical reaction for ALK were indicative of a myofibroblastic tumour of the bladder (figure 3). The lesion was confined within the specimen margins which were free of disease.

OUTCOME AND FOLLOW-UP

Due to the indeterminate malignant potential of the lesion, the patient underwent a repeat cystoscopy, 4 months after the operation with no sign of recurrence.

DISCUSSION

IMT of the bladder is a rare tumour of indeterminate malignant potential, with a generally benign, nevertheless rarely aggressive behaviour.⁵ Roth has first described this tumour as 'an unusual pseudosarcomatous entity'. Although some cases of IMT are considered to represent an inflammatory response to infection, trauma or surgery, the aetiological factors are not clear and there are no known predisposing conditions. Differential pathological diagnosis from malignant spindle cell tumours is considered difficult as both tumours resemble morphologically and immunologically.⁶ Positive immunohistochemical reaction for ALK in up to 87.5% of the IMTs can be useful for the differentiation of IMTs from other malignant lesions.⁷

Vesical IMT most commonly presents with haematuria, dysuria, frequency and various degree of suprapubic pain.^{8,9} Our

Figure 3 (A) IMT of the bladder in relation to the urothelium (H&E $\times 40$). (B) Higher magnification of (A), revealing the fusiform or rounded cells arranged in a storiform pattern and the inflammatory infiltrate composed of scattered mononuclear cells and polymorphonuclear leucocytes (H&E $\times 100$). (C) Positive immunohistochemical reaction for ALK ($\times 100$). ALK, anaplastic lymphoma kinase; IMT, inflammatory myofibroblastic tumour.

patient presented with increased dysuria, suprapubic pain and dyspareunia. Although the lower urinary tract symptoms were suggestive of a bladder involvement, dyspareunia and the detection of a tender mass in close proximity to the uterine wall rose the suspicion of vesical endometriosis. MRI findings were indicative of an infiltrating mass which involved the muscular layer of the bladder and the anterior uterine wall. Cystoscopy is essential in cases where bladder involvement is suspected, in order to obtain a biopsy prior to surgery.⁸ Cystoscopy in our case showed a broad-based, papillary growth arising from the bladder dome. The macroscopic appearance of the tumour resembled that of an endometriotic nodule, hence no biopsy was taken at that point. In retrospect however, we consider biopsy as an essential component of the diagnostic approach of any infiltrating mass of the bladder as, in an earlier report by Nezhat and colleagues, one case of presumed infiltrating endometriosis of the bladder was proven to be a primary intravesical endometrioid adenocarcinoma.⁹

Based on our findings and with no preoperative histological confirmation of the lesion, we proceeded to laparoscopic excision of the mass. Transurethral resection or partial cystectomy is the usual mode of treatment in most cases due to the predominantly benign course of the tumour.^{10–12} Evidence derives mainly from case reports, suggesting that open cystectomy, or radical cystectomy is the preferred mode of treatment.^{13–14} To the best of our knowledge, there are only two recent reports that describe laparoscopic excision of the lesion.^{15–16} In any case, care should be taken in order for the entire mass to be excised, while preserving only healthy margins for repair. Removal of endometriotic nodules follows the same surgical principles. Although morcellation of such lesions is unnecessary and as in our case the lesion was removed with a bag, it is mandatory to avoid excessive manipulation of such tumours in view of their potential for unforeseen malignancy. Fortunately, in our case, the mass was away from both ureteral orifices, and the final histopathological report showed that the excisional margins were free of disease. Final diagnosis necessitated the need for further follow-up, as a significant recurrence rate of 10%–25% and rare malignant transformation have been reported.^{8–17} Therefore, routine cystoscopic surveillance and close clinical follow-up are mandatory.

Laparoscopic treatment of an IMT vesical tumour is uncommon, as most of these cases are treated by urologists abdominally or transurethrally. Although the outcome of the patient was uneventful, we ought to recognise the main limitation of our study which is the inaccurate preoperative diagnosis. The initial speculation that we were dealing with another case of deep infiltrating endometriosis was biased by the fact that we frequently treat cases of severe intestinal and vesical endometriosis. The patient's symptomatology and the preoperative imaging findings corresponded the disease most familiar to us. Even though a urologist was present in the operating room during the operation, a multidisciplinary preoperative assessment of the patient would probably alert the surgical team on the likelihood of a more sinister diagnosis. We feel however that the mode of treatment was in accordance to what has been described in the literature for similar cases, and overall, the laparoscopic route represents a safe surgical alternative.

Contributors All authors contributed equally to the manuscript. OK: writing and editing. ZA: collection of data. KP: histopathology, images and editing. GP: surgeon and supervision.

Competing interests None declared.

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Patient's perspective

I am glad that this misfortune of mine ended well, although I am aware that a long term follow up is necessary. My doctors presumed that I had endometriosis and that is why I had pain during intercourse. I know that they took all the necessary precautions to ensure my safety during surgery and that the urologist who performed the cystoscopy was also present during the operation. I feel lucky that my tumour was not malignant and that I am free of symptoms now. I hope that I will not need further treatment in the future, although my doctors explained to the rarity of my case.

Learning points

- ▶ Inflammatory myofibroblastic tumour (IMT) of the bladder is a rare tumour of indeterminate malignant potential with myofibroblastic differentiation, with a generally benign but rarely aggressive behaviour which is usually treated by transurethral resection or partial cystectomy.
- ▶ Due to the close proximity of the bladder to the uterus, symptoms may resemble those of a gynaecological disease and therefore a multidisciplinary team of both gynaecologists and urologist should be involved in the management.
- ▶ Cystoscopy and biopsy should be an essential component of the diagnostic process as vesical lesions with corresponding gynaecological symptoms may also represent an aggressive neoplasm.
- ▶ Laparoscopic excision of an IMT with clear margins is a safe alternative to transurethral or abdominal approach, although in all cases long-term follow-up is mandatory.

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