

Abdominal Endometriosis Arising in an Exstrophy Patient

Michael S Floyd (Jr),¹ Rahul Mistry,² Charles T Burrows,² Andrew David Baird¹

Departments of Urology ¹ and Pathology ², Aintree University Hospital, Lower Lane, Liverpool, L9 7AL, United Kingdom.

Corresponding Author:
Michael S Floyd (Jr), Specialist Registrar, Department of Urology, Aintree University Hospital, Lower Lane, Liverpool L9 7AL, United Kingdom.

Tel: +44 151 5293775
E-mail: nilbury@gmail.com

Received August 2013
Accepted February 2014

Keywords: abdomen; surgery; endometriosis; etiology; complications; bladder exstrophy.

INTRODUCTION

Bladder exstrophy is a rare congenital anomaly. Associations with complete duplication of the genitourinary and gynecological systems have been reported. We describe a case of an abdominal swelling in an exstrophy patient which revealed endometriosis.

CASE REPORT

A 29 years old nulliparous female presented to the urology outpatients with a four months history of an intermittently enlarging abdominal swelling located over a scar. Her past history was remarkable for bladder exstrophy for which she underwent primary bladder closure on the third day of life. This had been followed by a succession of urological procedures over seven years. She had initiated self-intermittent catheterization following this. Her menarche had been at the age of thirteen. On presentation she described episodic swelling over her inferior abdominal wall beneath her laparotomy scar. It was unrelated to her menses and no systemic upset occurred.

On examination a firm indurated area was noted beneath the skin within the rectus sheath. Subsequent magnetic resonance imaging (MRI) confirmed the presence of a 3.5 cm × 2.7 cm × 3.9 cm enhancing mass within her abdominal wall which was separate from the peritoneum (**Figure 1**).

Under general anesthesia an excisional biopsy was performed. Intraoperatively a mass was found within the reconstructed rectus sheath. The peritoneum was opened and a segment from the dome of the bladder was removed as the mass was attached to it inferiorly. The remainder of the bladder was unremarkable.

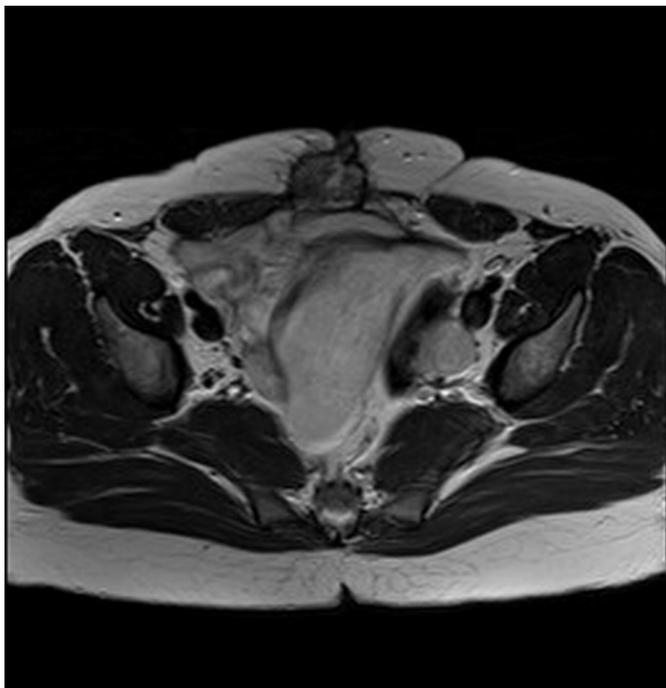


Figure 1. Abdominal magnetic resonance image showing a mass located on the inferior abdominal wall which enhanced following the administration of contrast.

She was discharged after 48 hours following removal of a suprapubic catheter. Outpatient review at 2 months revealed no recurrence and she remained asymptomatic.

Final histopathological examination confirmed a 40 g specimen consisting of fibrous tissue containing glands typical of endometriosis (**Figure 2**).

DISCUSSION

Variants of classical bladder exstrophy have been described by Marshall and colleagues⁽¹⁾ and include male and female epispadias with or without incontinence, superior vesical fissures, duplicated exstrophy and cloacal exstrophy. Duplicate bladder exstrophy consists of either an anterior-posterior or side by side alignment.⁽²⁾ Pseudoexstrophy patients have normal functioning bladders and external genitalia but have the skeletal and abdominal findings of exstrophy.⁽³⁾ In adults,

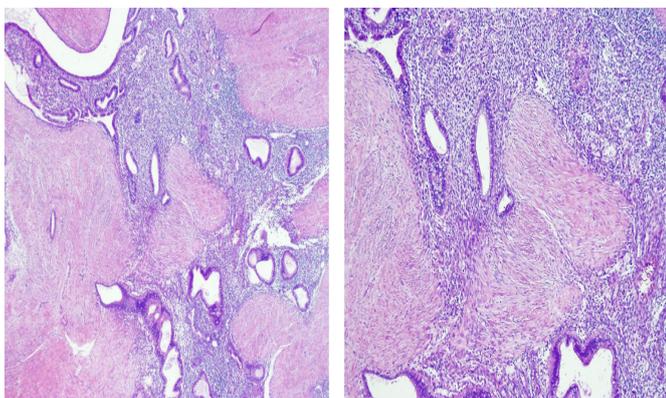


Figure 2. Photomicrographs of a section from the excised mass demonstrating endometrial glands and stroma in fibrous tissue (Hematoxylin and Eosin; left $\times 40$, right $\times 80$).

skin covered double bladder exstrophy has been reported along with complete duplication of the mullerian structures⁽⁴⁾ and also separately with colonic sequestration with a normal hindgut.⁽⁵⁾

Following progression into adulthood the reconstructed female exstrophy patient may face problems with parturition, sexual health and gynecological concerns. Krisloff and colleagues have found that multiple surgical procedures with scarring in the abdominopelvic area may have a detrimental effect on body image and sexual function.⁽⁶⁾ Successful pregnancies have been reported in patients who have undergone exstrophy repair⁽⁷⁾ but caesarean delivery is recommended⁽⁸⁾ Gynecological complications of exstrophy repair include mucocolpos and vaginal stone formation.^(9,10) Burbige and colleagues studied female exstrophy patients and found that none had endometriosis.⁽¹¹⁾ Our patient was nulliparous and denied dyspareunia or endometriosis.

We describe an abdominal mass in a corrected female exstrophy patient, which when excised, revealed endometriosis. Reports exist of dual pathologies in exstrophy patients who underwent reconstruction in adulthood.⁽¹²⁾ Kitajima and colleagues reported a case of scar endometriosis in a 26 years old exstrophy patient who underwent repair as an infant.⁽¹³⁾ Our case differs, however, as the ectopic deposit was painless and unrelated to her menstrual cycle.

CONCLUSION

We believe this to be the second reported case of scar related abdominal endometriosis in an exstrophy patient. As the long term follow up of this population is predominantly urological, female exstrophy patients with gynecological problems may present to the urologist. Therefore, the differential diagnosis of an abdominal mass in a female exstrophy patient should include gynecological pathologies such as ectopic endometrial tissue.

CONFLICT OF INTEREST

None declared.

REFERENCES

1. Marshall VF, Meucke EC. Variations in exstrophy of the bladder. *J Urol.* 1962;88:776-84.
2. Sheldon GA, McLorie GA, Khoury A, Churchill BM Duplicate bladder exstrophy: a new variant of clinical and embryological significance. *J Urol.* 1990;144:334-6.
3. Ahmed S, Abu Daia J. Exstrophic abdominal wall defect without bladder exstrophy (pseudoexstrophy). *Br J Urol.* 1998;81:762-3.
4. Berkowitz J, Warlick C, North A, Gearhart JP. Duplicate bladder exstrophy with complete duplication of Mullerian Structures. *Urology.* 2007;70:811.e15-7.
5. Kajbafzadeh AM, Aghdas FS, Tajik P. Complete covered duplication of the bladder, urethra, vagina, uterus and visceral sequestration. *Int J Urol.* 2006;13:1129-31.
6. Krisloff M, Puchner PJ, Tretter W, Macfarlane MT, Lattimer JK. Preg-

- nancy in women with bladder exstrophy. *J Urol*. 1978;119:478-9.
7. Sharma D, Singhal SR, Singhal SK. Successful pregnancy in a patient with previous bladder exstrophy. *Aust N Z J Obstet Gynaecol*. 1998;38:227-8.
 8. Mathews RI, Gan M, Gearhart JP. Urogynaecological and obstetric issues in women with the exstrophy-epispadias Complex. *BJU Int*. 2003;91:845-9.
 9. Zorn KC, Spiess PE, Salle JL, Jednak R. Mucocolpos associated with bladder exstrophy; a case report. *Can J Urol*. 2005;12:2614-5.
 10. Eyk NA, Grover S, Fink AM. Vaginal calculus as a late complication of bladder Exstrophy. *J Pediatr Adolesc Gynecol*. 2003;16:285-7.
 11. Burbige KA, Hensle TW, Chambers WJ, Leb R, Jeter KF. Pregnancy and sexual function in women with bladder exstrophy. *Urology*. 1986;28:12-4.
 12. Quiroz-Guerrero J, Badillo M, Munoz N, Anaya J, Rico G, Maldonado-Valadez R. Bladder augmentation in a young adult female exstrophy patient with associated omphalocele; An extremely unusual case. *J Pediatr Urol*. 2009;5:330-2.
 13. Kitajima T, Inoue M, Uchida K, Otake K, Kusunoki M. Scar endometriosis in a patient with bladder exstrophy. *Int Surg*. 2013;98:145-8.