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Comparison of musculoskeletal anatomic relationships, determined by magnetic resonance imaging, in postpubertal female patients with and without classic bladder exstrophy

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Abstract

- To characterize pelvic musculoskeletal anatomy in postpubertal females with classic bladder exstrophy, and to compare this with females without bladder exstrophy.
- The authors reviewed the medical records of all females in our institutional review board-approved bladder exstrophy database of 1078 patients and identified those with classic bladder exstrophy who underwent pelvic magnetic resonance imaging (MRI) after the age of 12 years.
- Indications for MRI included haematuria, adnexal lesion, perineal fistula, non-pelvic cancer staging, abdominal wall hernia and vaginal stenosis.
- Age- and race-matched female patients without exstrophy who underwent MRI evaluation for similar indications were included for comparison.
- The MRI protocol included axial, sagittal and coronal T1- and/or T2-weighted imaging.
- The study included 30 patients with a median (range) age of 22.5 (12--55) years at time of MRI. Ten patients had bladder exstrophy while 20 control patients did not.
- A smaller percentage of levator ani was located in the anterior compartment of the pelvis in patients with bladder exstrophy compared with controls.
- The iliac wing angle, puborectalis angle, ileococcygeous angle, levator ani width, symphyseal diastasis, erectile body diastasis, posterior bladder neck distance and posterior anal distance was greater in patients with bladder exstrophy than in those without.
- The ischial angle and obturator internus angle were narrower in patients with bladder exstrophy than in those without, and there was no significant difference between levator ani surface area, sacral anal angle, sacral bladder neck angle and bladder neck erectile body distance between the two patient groups.
- In postpubertal females with bladder exstrophy, significant deviations from normal pelvimetry exist, including posterior location of the majority of the levator ani muscle, a wider ileococcygeous angle and a wider symphyseal diastasis.

- These differences are similar to those described in previous comparisons of younger children with bladder exstrophy and control children.

Keywords

magnetic resonance imaging; bladder exstrophy; pelvic floor; anatomy; prolapse

INTRODUCTION

Classic bladder exstrophy is a rare congenital disorder involving gross anomalies of the genitourinary tract, abdominal wall and musculoskeletal structures of the pelvis and pelvic floor. The traditional aims of surgical repair during childhood are to close the exstrophied bladder, re-approximate the abdominal wall and pelvic ring, protect renal function, achieve urinary continence and reconstruct cosmetically and functionally acceptable external genitalia.[1] While there has not traditionally been a focus on approximating normal pelvic floor support, there is growing interest in understanding the differences in pelvic floor configuration between patients with bladder exstrophy and patients without. Several studies in the paediatric literature have characterized the pelvic musculoskeletal anatomy of infants and children with bladder exstrophy using MRI and three-dimensional CT.[2--5].

These studies have shed light on the marked differences in the configuration of the pelvis of children with bladder exstrophy and normal children. Some of the well described features of the exstrophy pelvis include a wider pubic symphyseal diastasis, obtuse iliac wing angle, wider puborectalis and ileococcygeus angle, and positioning of the majority of levator ani muscle in the posterior half of the pelvic floor. Some authors have investigated pelvic anatomic features in infants and toddlers with bladder exstrophy before and after closure and have still found significant differences between these patients and non-exstrophy controls. They noted persistently a wider symphyseal diastasis, obtuse iliac wing, puborectalis angle and ileococcygeus angle after closure.[2]

As females with bladder exstrophy age and go through puberty, they manifest disorders of pelvic floor dysfunction, particularly pelvic organ prolapse (POP), urinary incontinence, and faecal incontinence. Indeed, these patients have a higher prevalence of POP than the general population. In some series, the prevalence is reported to be 18% at a median age of only 16 years[6]. Others looking at older exstrophy patients with a mean age of 39 years have reported POP rates as high as 52% [7]. By contrast, a prevalence of 6% has been reported in the general population of women at a mean age of 56.5 years.[8] They may also develop prolapse while nulliparous. Thus, comprehensive characterization of the pelvic floor anatomy in the postpubertal exstrophy population is critical to understanding their predisposition to POP. Recently, some authors have used three-dimensional perineal ultrasonography to characterize some of the pelvic floor structures in postpubertal patients with bladder exstrophy.[9] Others have reported measurement of some pelvic floor variables using both three-dimensional perineal ultrasonography and MRI with comparison of the two imaging methods.[10]

The present study aims to characterize comprehensively the pelvic anatomy of postpubertal females with classic bladder exstrophy by measuring 15 pelvic floor variables previously described in a younger population with bladder exstrophy.

PATIENTS AND METHODS

Institutional review board approval was obtained for this study. The authors reviewed the medical records of all females in our bladder exstrophy database of 1078 patients, and identified those with classic bladder exstrophy who had undergone pelvic MRI after the age

of 12. Indications for MRI included haematuria, adnexal lesion, perineal fistula, non-pelvic cancer staging, abdominal wall hernia and vaginal stenosis. We obtained from the department of radiology, a list of age- and race-matched female patients (two controls for each patient with bladder exstrophy) who underwent MRI evaluation for similar indications; these patients were included for comparison. The MRI protocol included axial, sagittal and coronal T1- and/or T2-weighted imaging. A paired *t*-test analysis was used to compare variables between patients with exstrophy and age-matched controls, while a rank-sum test was used to compare variables among patients with exstrophy who had previously undergone pelvic osteotomy and those who had not. A *P* value < 0.05 was considered to indicate statistical significance. Statistical analysis was performed using STATA 11.0 software. The same investigator performed all MRI measurements according to a standardized protocol.[2, 3]

Fifteen features were measured. The surface area of the levator ani muscle, width of the levator ani muscle, and percentage of levator ani muscle located anterior to the rectum were measured as described by Stec et al. [3]. The iliac wing angle, ischial angle, obturator internus angle, puborectalis angle, symphyseal diastasis, ileococcygeous angle, sacral anal angle, posterior anal distance, posterior bladder neck distance, erectile body diastasis, bladder neck erectile body distance and sacral bladder neck angle were measured as previously described by Gargollo et al. [2]. Figure 1 shows representative MRI measurements of some of the variables evaluated.

RESULTS

The study included 30 patients with a median (range) age of 22.5 (12--55) years at time of MRI. Ten patients had bladder exstrophy while 20 patients did not and formed the control group. Table 1 shows the demographic and clinical variables of the patients with bladder exstrophy and those without. Of the 10 patients with bladder exstrophy, seven were closed in infancy, while three had early urinary diversion without bladder preservation (one ureterosigmoidostomy, one ileal conduit and one catheterizable pouch). Of the seven patients that were closed, four underwent pelvic osteotomy. Four patients were voiding from the urethra and were continent, while three had a catheterizable continent stoma. Six patients had undergone an exteriorizing perineal vaginoplasty for vaginal stenosis.

As shown in Table 2, there was a smaller percentage of levator ani located in the anterior compartment of the pelvis in patients with bladder exstrophy compared with those without. The iliac wing angle, puborectalis angle, ileococcygeous angle, levator ani width, symphyseal diastasis, and erectile body diastasis were greater in the patients with bladder exstrophy than in those without. Patients with bladder exstrophy also had a greater posterior bladder neck distance and posterior anal distance. The ischial angle and obturator internus angle were narrower than in patients without bladder exstrophy, but there was no significant difference between levator ani surface area, sacral anal angle, sacral bladder neck angle and bladder neck erectile body distance between the two groups.

In comparing patients with bladder exstrophy who had pelvic osteotomy with those who did not have osteotomy, we found that patients who underwent osteotomy had a significantly wider obturator internus angle and narrower levator ani width. There was a strong trend towards smaller symphyseal diastasis in patients who underwent osteotomy, but this did not reach significance (3.03 vs 7.83 cm, *P* = 0.088).

DISCUSSION

The last two decades have brought greater clarity to our understanding of the anatomy of the pelvis and pelvic floor of infants born with bladder exstrophy. They have also brought continued challenges in the management of POP in these patients, as the ageing female population with bladder exstrophy continues to develop POP at a younger age than those without bladder exstrophy, [6] and may be more likely to have recurrence after surgical repair of prolapse. Since it is not clear how the anatomical configuration of the pelvic floor changes in these women over the years leading into puberty and adulthood, it is necessary to characterize comprehensively the pelvic floor of postpubertal females with bladder exstrophy, and to compare this with the anatomy of postpubertal females without bladder exstrophy. In the present study, we found substantial differences in the pelvic floor of postpubertal females with bladder exstrophy compared with age- and race-matched control females, and some of these differences mirror those described in infants.[3]

Importantly, the majority of the levator ani muscle continues to be located posterior to the rectum in female patients with bladder exstrophy, leaving a disproportionately smaller muscle support for the anterior compartment which houses the uterus, vagina and bladder. The levator ani muscle was more divergent in the axial plane and flatter in the coronal plane in patients with bladder exstrophy compared with controls. As expected, we found a markedly wider symphyseal diastasis in bladder exstrophy patients compared with controls, and the degree of diastasis was larger than that reported in younger patients with bladder exstrophy.[2] This confirms findings from other studies where progressive widening of the pubic diastasis occurs as patients with bladder exstrophy age.[11] These significant aberrations in pelvic musculoskeletal anatomy demonstrate less anterior compartment support and probably contribute to the predisposition of bladder exstrophy patients to POP.

In the subset analysis of patients with bladder exstrophy closed with osteotomy vs those who did not undergo osteotomy, patients who underwent osteotomy had a significantly wider obturator internus angle, similar to those without exstrophy. The osteotomy group also had a narrower levator ani width, similar to female patients without exstrophy, and consistent with the findings of Stec et al. [3] who reported a narrower width of the levator ani muscle in control patients compared with that in patients with bladder exstrophy before closure. In our analysis of patients with bladder exstrophy closed with and without osteotomy, there was also a strong trend towards narrower symphyseal diastasis in patients who underwent osteotomy, but this did not reach significance. This result may be limited by the small sample size for this sub-analysis. The re-approximation of the pubic bones may add longevity to the pelvic ring closure, keeping the diastasis from recurring and keeping the muscles in a more anatomic relationship.

A limitation of the present study is the retrospective nature of its design, which could introduce a selection bias because bladder exstrophy patients without an indication for pelvic MRI were not evaluated. Finally, a larger sample size would allow more robust statistical analysis, especially for the subset analysis. Nevertheless, the present study represents an initial attempt to provide a comprehensive comparison of pelvic musculoskeletal variables in postpubertal patients with and without bladder exstrophy using pelvic MRI.

In conclusion, in postpubertal female patients with bladder exstrophy, significant deviations from normal pelvic anatomy exist. Some of these deviations include posterior location of the majority of the levator ani muscle, a wider ileococcygeous angle and wider symphyseal diastasis. The enhanced understanding of the anatomy of postpubertal exstrophy pelvis

provided by the present study could pave the way for improved surgical techniques focusing on the prevention and durable correction of POP in this challenging population.

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Abbreviation

POP pelvic organ prolapse

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What's known on the subject?

Several studies in the paediatric literature have characterized the pelvic musculoskeletal anatomy of infants and children with bladder exstrophy using MRI and three-dimensional CT. The pelvic floor anatomy of female patients with bladder exstrophy who have undergone somatic growth and puberty is less well described.

What does the study add?

This study uses MRI to characterize comprehensively the pelvic anatomy of postpubertal females with classic bladder exstrophy by measuring 15 pelvic floor variables previously described in younger children with bladder exstrophy.

Fig. 1.

Measurement of: 1.1 symphyseal diastasis; 1.2 puborectalis angle; 1.3 ischial angle; 1.4 obturator internus angle; 1.5 posterior bladder neck distance; 1.6 posterior anal distance; 1.7 iliac wing angle; 1.8 proportion of levator ani muscle in anterior vs posterior compartment; and 1.9 ileococcygeous angle in **A**, a patient with bladder exstrophy and **B**, a patient without bladder exstrophy.

Table 1

Demographic characteristics of patients with and without exstrophy

	Patients with bladder exstrophy, <i>n</i> =10	Patients without bladder exstrophy, <i>n</i> =20	<i>P</i> *
Mean (SD) age, years	28.4 (15.11)	28.4 (14.71)	0.72
Race			--
White	10	20	
Black	0	0	
Other	0	0	
No. of pregnancies			0.77
0	9	16	
1	0	1	
2	1	1	
3	0	1	
4	0	1	
No. of vaginal deliveries			0.59
0	10	18	
1	0	1	
2	0	1	

* *P* values represent rank-sum test for mean (SD) values and chi-squared test for proportions.

Table 2

Values of measured variables in patients with and without bladder exstrophy

	Patients with bladder exstrophy	Patients without bladder exstrophy	<i>P</i>
Levator ani located in anterior compartment, <i>n</i> (%)	45.20 (0.11)	56.00 (0.09)	0.027
Levator ani width *, cm	9.04 (2.04)	7.75 (1.79)	0.035
Levator ani surface area *, cm ²	21.02 (6.7)	20.37 (5.26)	0.66
Iliac wing angle *, degrees	112.17 (11.33)	99.91 (13.37)	0.023
Ischial angle *, degrees	42.79 (18.67)	75.21 (15.87)	0.0035
Obturator internus angle *, degrees	29.84 (22.94)	65.43 (8.99)	0.014
Puborectalis angle *, degrees	86.05 (14.45)	57.80 (15.86)	0.0020
Symphyseal diastasis *, cm	5.90 (4.39)	0.43 (0.15)	0.0036
Ileococcygeous angle *, degrees	120.32 (22.36)	88.52 (15.52)	0.0021
Sacral anal angle *, degrees	48.20 (16.07)	48.24 (15.11)	0.80
Posterior anal distance *, cm	4.15 (1.07)	2.37 (1.06)	0.0046
Posterior bladderneck distance *, cm	6.94 (1.17)	5.83 (0.77)	0.020
Erectile body diastasis *, cm	2.79 (2.61)	0.10 (0.18)	0.011
Bladder neck erectile body distance *, cm	1.33 (1.46)	1.73 (0.66)	0.46
Sacral bladder neck angle *, degrees	39.61 (6.80)	43.34 (8.55)	0.20

* Mean (SD) values.

Table 3

Values of measured variables in patients with bladder exstrophy who did or did not undergo osteotomy

	Osteotomy	No osteotomy	P
Levator ani located in anterior compartment, <i>n</i> (%)	41.0 (0.58)	48.6 (0.13)	0.46
Levator ani width *, cm	7.35 (1.05)	10.17 (1.73)	0.011
Levator ani surface area *, cm ²	18.63 (5.72)	22.94 (7.52)	0.54
Iliac wing angle *, degrees	108.88 (14.66)	114.36 (9.38)	0.52
Ischial angle *, degrees	53.14 (14.72)	35.89 (18.84)	0.20
Obturator internus angle *, degrees	51.86 (14.36)	15.16 (13.31)	0.011
Puborectalis angle *, degrees	80.88 (16.50)	89.50 (13.29)	0.39
Symphyseal diastasis *, cm	3.03 (1.12)	7.83 (4.79)	0.088
Ileococcygeous angle *, degrees	108.4 (6.10)	128.27 (26.23)	0.088
Sacral anal angle, degrees	51.36 (4.74)	46.10 (20.92)	0.67
Posterior anal distance *, cm	3.63 (0.81)	4.50 (1.13)	0.34
Posterior bladder neck distance *, cm	6.39 (0.74)	7.30 (1.32)	0.20
Erectile body diastasis *, cm	1.40 (1.04)	3.71 (3.01)	0.39
Bladder neck erectile body distance *, cm	1.89 (2.08)	0.87 (0.67)	0.62
Sacral bladder neck angle *, degrees	39.65 (8.81)	39.58 (6.04)	0.83

* Mean (SD) values.