

## Successful Pregnancy Outcome and Surgical Approach in Women with Repaired Bladder Exstrophy or Cloacal Exstrophy – Experience from a quaternary paediatric and adolescent gynaecology centre in Australia

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### Abstract

**Background:** Women with repaired bladder/cloacal exstrophy face unique challenges throughout their pregnancy. Limited obstetric care information available in the current literature.

**Aims:** To review perinatal issues, pregnancy outcomes and surgical techniques for five pregnancies in three women with repaired bladder exstrophy and cloacal exstrophy.

#### Materials and Method

**Design:** A case series and review of the literature.

**Setting:** A quaternary obstetric and paediatric and adolescent gynaecology service in Queensland, Australia.

**Participants:** Women with repaired bladder exstrophy and cloacal exstrophy, managed by the service over a ten-year period from January 2004 to January 2014.

**Main Outcome Measures:** Identification of perinatal and surgical complexities for this cohort of women.

#### Results

Antenatal issues in these women with repaired bladder exstrophy or cloacal exstrophy include miscarriage (3/3), malpresentation (3/3), urological complications such as recurrent urinary tract infection (UTI) (3/3), pyelonephritis (1/3), urosepsis (1/3), renal calculi and hydronephrosis (1/3) and stress incontinence (1/3). Other issues identified were hypertension, uterine prolapse, threatened preterm labour, cervical insufficiency, and low lying placenta with antepartum haemorrhage. Surgical planning and foresight of anticipated difficulties at delivery in women with repaired bladder exstrophy and cloacal exstrophy is critical.

#### Conclusions

A Women with repaired bladder exstrophy and cloacal exstrophy present additional challenges for those caring for them in the perinatal period. Surgical approach to delivery is best achieved through elective caesarean section with a high midline skin incision to avoid damage to the reconstructed bladder, which overlies the usual transverse suprapubic skin incision made for a lower segment caesarean section.

**Keywords:** Bladder exstrophy; Omphaloceleexstrophy; Imperforate anus; Urogenital abnormalities, Pregnancy complications; Caesarean section

## Introduction

Bladder exstrophy is a rare congenital anomaly, occurring in 2-3.3/100,000 births, with a male to female ratio of 3-4 to 1 [1]. Cloacal exstrophy is even rarer compared to bladder exstrophy, with a prevalence of 0.76/100,000 births, with male to female ratio of 1:1.14. [2]. Classical bladder exstrophy is characterised by a defect in the closure of the lower abdominal wall and bladder, and is always associated with pubic bone diastases [3,4]. Whereas classical cloacal exstrophy typically refers to the combination of omphalocele, bladder exstrophy and imperforate anus, cloacal exstrophy can also be associated with renal malformations and spine defects [2]. Multiple surgeries are usually required to correct the congenital defects in those born with bladder exstrophy or cloacal exstrophy. Common surgeries include initial bladder closure, bladder neck reconstruction, enterocystoplasty, and neobladder creation with an ileal conduit. The Mitrofanoff procedure is commonly utilised method in which the appendix or the bowel act as a conduit between the bladder and skin surface. Continence is usually maintained with self-catheterisation [1,5-15]. When women with repaired bladder exstrophy or cloacal exstrophy become pregnant, they face some unique challenges throughout the antenatal, peripartum and postnatal period. Antenatal issues include reduced fertility, higher rate of miscarriages compared to the general population, stillbirth, premature birth, pre-eclampsia and placental abruption, urinary tract infection and other urinary tract issues including urinary retention and urinary calculus. Postnatal complications include genital prolapse, temporary urinary incontinence and vesicocutaneous fistula [1,3,9-13,16-21]. Management of pregnant women with repaired bladder exstrophy and cloacal exstrophy must therefore include attention to their additional risks and complications. Elective caesarean section is recommended for delivery at or before 37 weeks with multidisciplinary teams including paediatric urologists experienced in this condition present during surgery [18]. Vaginal delivery is recommended by some authors for uncomplicated pregnancy and with an experienced obstetrician and urologist available [18,19]. There is limited information in the current literature on the topic of the best caesarean section technique for women with repaired bladder exstrophy or cloacal exstrophy [Table 1] [1,9-12,16-20,22-24]. In this study, we aim to explore the perinatal course and surgical techniques used for pregnant women with repaired bladder exstrophy and cloacal exstrophy.

## Materials and Methods

This is a retrospective case series. Cases were identified through the registry of the obstetric cases in a paediatric and adolescent gynaecology service in a quaternary Australian hospital. Review of the medical notes was carried out to collect detailed information on demographics, medical and surgical history, antenatal history, operation details, and pregnancy outcomes. Consent was obtained from the patients at the time of their treatment for use of their medical notes for medical research and education. Ethics approval was obtained from the local hospital ethics committee to carry out the study and publish the results.

**Table 1:** Literature summary of the pregnancy outcomes of the women with congenital bladder exstrophy or cloacal exstrophy

A total of 4 women were identified from the obstetric registry of the quaternary obstetric and paediatric and adolescent gynaecology centre. One woman with repaired bladder exstrophy had moved to another city after initial consultation in preparation for her pregnancy and is not included in the discussion. The remaining 3 women had their perinatal care carried out in the centre. There were 5 pregnancies among the three women. Due to the small numbers of cases identified, no formal statistical analysis was carried out. Results are summarised in table 2.

### Perinatal problems in women with bladder exstrophy or cloacal exstrophy:

Common antenatal issue faced by our patients include miscarriage in 3/3 women. They were first trimester miscarriages. No causes or histopathology for these miscarriages were documented in the notes. Urological antenatal complications were very common in our patient cohort; all 3 women had recurrent urinary tract infections (UTI) throughout their pregnancy. Antibiotic prophylaxis was used in all women after their first episode of UTI. Weekly monitoring of their urine samples were carried out and any signs of UTI were treated with extended courses of antibiotics as advised by urologists. The patient in case 1 who had had renal calculi throughout her life, had more urological complications during her pregnancy. She had two episodes of pyelonephritis leading to urosepsis, and requiring hospitalisation for her treatments. Malpresentation was another common issue faced by our patient cohort. Two women had bicornuate uterus, which was a likely contributor to the malpresentation. The other woman presented with threatened preterm labour at around 29 week of gestation, and a few days later, had emergency caesarean section due to cervical shortening as a consequence of uterine contractions. The baby was in breech presentation. Other issues were hypertension, uterine prolapse, stress incontinence, low lying placenta and antepartum haemorrhage.

### Surgical techniques:

All of our patients were recommended and planned to have elective caesarean section at around 35-37 weeks of gestation, however, 4 deliveries out of 5 were carried out in a semi-emergent to emergent manner due to development of additional antenatal complications. The reasons for emergency caesarean section include cervical shortening with minor uterine activity, antepartum haemorrhage and preterm labour. Operations were carried out by an experienced obstetrician, who is also the head of paediatric and adolescent gynaecology, with a paediatric urologist present. For the patient in case 2 the original paediatric urologist was in attendance at her two caesarean sections, whereas for the patient in case one the on call urologist was in attendance. The two patients who had had bladder exstrophy repairs both had abdominal incisions which were above the site of the previous abdominal wall repairs.

Study (Arthors)	Year	Number of women	Number of pregnancies	Outcomes	Gestation at delivery	Mode of delivery	Ref
Dy et al.	2015	12	22	Live birth 14 (64%), Terminations 4(18%), Spontaneous abortion <24 weeks 4(18%).	Mean gestational age at delivery: 36 weeks	Caesarean section in all 14 cases	22
Deans et al.	2012	52	57	Live birth 34 (56%), Miscarriage 21 (35%), Termination 1 (2%), Stillbirth 4 (7%)	Median gestational age of all live births was 37 weeks.	All patients delivered by cesarean section of which 3 were emergencies. 3 classical cesarean sections.	18
Ebert et al.	2011	2	3	Live birth 2 (67%); Miscarriage 1 (33%)	one at 39 weeks, one at 33 weeks	1 by elective LUSCS, 1 by emergency LUSCS at 33+4/40 due to preterm labour	19
Rubenwolf et al.	2016	12	17	Live birth 16 (94%); Miscarriage 1(6%)	Not mentioned	All delivered by elective caesarean section	24
Eswara et al.	2016				Mean gestational age at delivery: 36 weeks		23
Schumacher et al.	1997	6	7	Live birth 7(100%)	Not mentioned	All delivered by caesarean section	12
Mathews et al.	2003	6	11	Live birth 7 (63%), Miscarriage 2(18%), TOP 2 (18%)	Not mentioned	6 by caesarean section, 1 had uneventful vaginal birth	11
Gezer et al.	2011	1	1	Live birth 1 (100%)	36 weeks	Emergency Classical C/Section due to preterm labour	20
Giron et al.	2011	14	22	Healthy babies 17 (77.2%); Miscarriage 4 (18.1%); Neonatal death 1 (4.7%)	Not mentioned	All babies were delivered through caesarean section with the exception of one that had a premature normal birth	9
Bildirin et al.	2012	1	1	Live birth 1 (100%)	34	Vaginal birth	16
Greenwell et al.	2003	20	37	Live birth 29 (78%), Miscarriage 6 (16%), TOP 2 (5%)	Mean gestational age at delivery: 36.9 weeks (35-40)	Elective Caesarean section 13 (43%), Emergency Caesarean 7 (23%); vaginal birth 10 (34%)	10
Burbigi et al.	1986	5	7	Live birth 6 (86%), 1 TOP (14%)	Not mentioned	3 caesarean sections (50%); 3 vaginal birth (50%)	17
Mantel et al.	2000	3	6	Live birth 2 (33%), Miscarriage 4 (66%)	35 and 36 weeks	Emergency Caesarean sections for malpresentation in labour (100%)	1

	Congenital anomaly	Perinatal complications	Delivery method	Outcome
Case 1	Bladder exstrophy	Miscarriage X1 Renal calculus Hydronephrosis Recurrent UTIs Pyelonephritis Urosepsis Threatened preterm labour Cervical insufficiency	Emergency Classical caesarean section due to preterm labour	Nil significant maternal complications Fetal: Prematurity Respiratory distress
Case 2a	Bladder exstrophy	Miscarriage X1 (twins) Recurrent UTIs Hypertension Malpresentation Uterine prolapse Bicornuate uterus	Elective caesarean section	Maternal: small bowel injury Fetal: respiratory distress
Case 2b	2 <sup>nd</sup> pregnancy for case 2 Bladder exstrophy	Recurrent UTIs	Elective caesarean section	Maternal: minor bladder serosal tear Fetal: Minor respiratory distress
Case 3a	Cloacal exstrophy	Miscarriage X1 Malpresentation Bicornuate uterus Recurrent UTIs	Elective caesarean section	Nil significant complications
Case 3b	2 <sup>nd</sup> pregnancy for case 3 Cloacal exstrophy	Recurrent UTIs Low lying placenta Stress incontinence APH Malpresentation	Emergency caesarean section	Maternal: Indwelling catheter pushed through a fold into vagina Fetal Nil

The patient in case 1 had an upper left paramedian skin incision to avoid the right-sided Mitrofanoff stoma and neobladder, which was also adhered to suprapubic area of her anterior abdominal wall. This patient's caesarean section was done by an obstetrician other than the paediatric and adolescent gynaecologist. A high transverse upper segment caesarean section, was used preferentially over a routine lower segment caesarean section, under an emergent circumstance of preterm labour. The patient in case 2 had an upper midline skin incision to avoid the neobladder which was found to be adhered to the suprapubic area of her anterior abdominal wall (figure 1). The uterine incision for this patient was lower segment after dissection down of the bladder base from the lower uterine segment as is routine with lower segment caesarean sections. This patient also had a bicornuate uterus with the baby in a transverse position, back up, but was delivered without difficulty through the lower segment incision. Pfannenstiel incision was used in both of the deliveries of the same woman who had repaired cloacal exstrophy. Both of these operations were done by obstetricians on duty after hours and not by the experienced paediatric and adolescent gynaecologist. They were difficult operations due to adhesions and scars encountered in the lower abdomen.



**Figure 1:** Abdominal scars and mitrofanoff stoma prior to caesarean section.

Case2.

A: Mitrofanoff stoma, B: Midline laparotomy scar, C: Umbilicus, D: High incision for Caesarean section indicate by the red line.

Surgical complications include a minor small bowel laceration with one elective caesarean section, a bladder serosal tear of adherent neobladder with abdominal wall, in the patient in case 2 with bladder exstrophy repair. An indwelling catheter was inadvertently inserted through the urethral fold into the vagina by the after-hours obstetrician in the patient with cloacal repair with short term injury to the urethra. This was repaired by the duty urologist with full recovery of urethral function.

## Discussion

This study highlights the common antenatal issues in women with repaired bladder exstrophy or cloacal exstrophy, perinatal management and surgical techniques utilised in these cases. Patients with bladder exstrophy or cloacal exstrophy often have symphysis diastasis, anterior abdominal wall deficiency and pelvic floor deficiency as a result of their congenital condition [13,18,19]. Following their multiple operations to correct the congenital issues, they are often left with an augmented bladder, a conduit for catheterising and some expected adhesions [19]. The enlarging gravid uterus therefore can have some significant impact on the urinary system and surrounding structures in these women. A few issues were highlighted in the literature, in particular, urinary tract obstruction requiring stenting or nephrostomy, urinary tract infections, catheterisation difficulties particularly in the third trimester, urinary incontinence, and uterine or cervical prolapsed [18,19]. Our findings are similar to the literature. In our patients, all had pubic diastasis: the patient in case 1 had an 11cm gap in her anterior pelvis, and the patient in case 2 had a 9cm pubic diastasis. The patient in case 1 had shortened cervix with funnelling at 29 weeks of gestation, and subsequently had an emergency caesarean section due to preterm labour. The patient in case 2 had uterine prolapse, and also had preterm labour requiring an emergency caesarean section. None of the patients developed significant urinary obstruction requiring stenting or nephrostomy, however, hydronephrosis and urosepsis was a significant issue in one of our patients. Recurrent urinary tract infections were experienced by all our patients during their pregnancies. Urinary incontinence was a significant problem in one of our patients.

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She also needs to proactively completely empty her bladder at regular intervals to ensure minimal residual urine in the bladder. This reduces the chances of urinary stasis and infections in these women. The issue of prophylactic antibiotics should be discussed in light of its significant disadvantages, particularly its potential effect on the developing fetus.

Surgical planning and foresight of anticipated difficulties at delivery in women with repaired bladder exstrophy repair is critical [19]. Appreciating the abdominal wall adhesions from childhood surgery to create the neobladder, along with the repair of the anterior abdominal wall defect is important in avoiding damage to the neobladder. Access to the lower segment of the uterus is not difficult with a high midline abdominal skin and wall incision for the important reason of avoiding inadvertent surgical damage to the neobladder and its conduit often in the form of the Mitrofanoff abdominal ostomy. Having a planned delivery at late preterm gestations after steroid cover for the baby is a reasonable approach under the circumstance of multiple perinatal considerations. These include the increased risk of uterine prolapse, the risk of preterm labour, any associated uterine anomalies and considerations of surgical access. Ideally the management of these patients is by an obstetrician who is also experienced in paediatric and adolescent gynaecology; consequently these patients are best managed at a quaternary centre with availability of these specialist skills.

A collaborative approach between an experienced obstetrician with skills in paediatric and adolescent gynaecology and a urologist with experience of congenital anomaly reconstructive surgery is recommended for planned delivery in a centre with availability of these specialists, including a suitable neonatology service on site [Table 3] [12,13,18,23-25].

This small case series highlights some important surgical principles and considerations for this rare cohort of women during pregnancy and delivery of their babies.

**Table 3:** Summary of recommendations of pregnancy management and delivery

Perinatal issues	Perinatal management
High risk for multiple complications	Pregnancy to be considered high risk, collaborative approach between an experienced obstetrician with skills in paediatric and adolescent gynaecology and a urologist with experience in congenital anomaly reconstructive surgery for antenatal care and delivery planning
Fertility issues(1)	Aware and management as per conventional treatment for sub fertility
1:70 risk of congenital bladder exstrophy in the fetus which is a 500 fold increase over the general population(2)	Prenatal USS for detection
Urinary tract obstruction; Hydronephrosis	USS of the renal tract, first at 16 weeks, thereafter Q6-8 weeks, stenting or nephrostomy if obstructed;(3) recommend percutaneous drainage of the kidneys for the following indications: pain, progressive hydronephrosis, rising serum creatinine, febrile UTIs, recommend not removing the nephrostomy tubes until 1 week postpartum. Biggest risk period for obstruction appears to occur between 20 and 28 weeks of gestation, patient at risk may require USS every 2 weeks during that time(4)
Urinary tract infections	Baseline renal function and urine microscopy; screening for asymptomatic bacteriuria weekly; adequate hydration; proactively completely empty bladder at regular intervals advised; prophylactic antibiotics if recurrent UTIs, treat only febrile UTIs (3)
Stomal prolapse or parastomal hernia	Typically resolve after delivery
Worsening renal function, development of end stage renal failure (4)	Close monitoring, regular renal consultation.
Catherisation difficulties	Indwelling catheter or change the type and length of the catheter used (e.g. Coude tip catheter)(4)
Cervical incompetence	Monitor cervical length
Preterm labour	Having a planned delivery at late preterm gestations after steroid cover
Miscarriage	Early USS to confirm gestation, identify anomalies
Malpresentation	Planned delivery, aware of the presentation at the time of the delivery
Renal stone	Adequate hydration, proactively completely empty bladder at regular intervals, if suspicious of renal stone, USS and urology management
Urinary incontinence	Generally recover to pre-pregnancy status postpartum
Uterine prolapse including cervical prolapse	Cervical pessary, a diaphragm fitting ring, bed rest for late gestation(5)
Hypertensive disorders	Monitoring of the blood pressure and blood biochemistry and urine protein
Low lying placenta with antepartum haemorrhage	USS for detection, management according to standard low lying placenta management protocol
Delivery	Elective Caesarean section in the exstrophy population was the clear consensus (4). Planned caesarean section at late preterm gestations after steroid cover; Surgery to be done by experienced obstetrician with urologist in theatre, with suitable neonatology service on site; High midline abdominal skin and wall incision, lower segment uterine incision or classical uterine incision pending on status of adhesion. Vaginal birth only is considered in uncomplicated pregnancies (6).

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