Pregnancy and Delivery in Women with Lower Urinary Tract Reconstruction: A National Multicenter Retrospective Study from the French-Speaking Neuro-Urology Study Group (GENULF) and the Neuro-Urology Committee of the French Association of Urology

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Purpose: Management of pregnancy and delivery in women with lower urinary tract reconstruction is challenging and the currently available literature is insufficient to guide clinical practice. We report pregnancy and delivery outcomes in this specific population.

Materials and Methods: We conducted a national multicenter retrospective study (16 centers) including 68 women with 96 deliveries between 1998 and 2019. These women had at least 1 successful pregnancy and delivery after augmentation enterocystoplasty, catheterizable channel creation and/or artificial urinary sphincter implantation. Maternal and fetal complications during pregnancy and delivery were reported, as well as postpartum functional outcomes, according to the delivery mode. The chi-square test and Student's t-test were used to compare categorical and continuous variables, respectively.

Results: Overall 32% of reported pregnancies were complicated by febrile urinary tract infections, 13.5% by renal colic and 14.6% required upper urinary tract diversion. In addition, 10% of patients reported transient self-catheterization difficulties and 13.5% reported de novo or increased urinary incontinence. The preterm delivery rate was 35.3%. Elective C-section was performed in 61% of pregnancies. Twenty complications occurred during delivery (20%), including 19 during elective C-section. Urinary continence at 1 year was unchanged for 93.5%

Abbreviations and Acronyms

AE = augmentation enterocystoplasty
AUS = artificial urinary sphincter
CCC = catheterizable channel creation
LUTR = lower urinary tract reconstruction
SCI = spinal cord injury
UTI = urinary tract infection
WA = weeks of amenorrhea

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of deliveries. Delivery mode (p=0.293) and multiparity (p=0.572) had no impact on urinary continence.

Conclusions: In this population C-section appeared to be associated with a high risk of complications. In the absence of any obstetric or neurological contraindications, vaginal delivery should be proposed as the first line option to the majority of these women.

Key Words: urinary sphincter, artificial; urinary diversion; delivery, obstetric

THE management, treatment options and care given to women with neurological disease or born with a complex urological malformation have dramatically improved in recent decades. A growing number of these women now reach adulthood with good quality of life and a desire for childbearing, although this has often been eluded in the past and was often considered a complete fantasy in view of the woman's disability. Nevertheless, it is difficult to change misconceptions about the incidence of fetal malformations and parental abilities.[1,2]

Management of pregnancy and delivery in women with a history of lower urinary tract reconstruction is challenging. Recent studies have allowed a more reassuring position in relation to maternal and fetal issues of pregnancy in this situation while highlighting the frequent complications that may occur, including febrile urinary tract infections, urinary incontinence, self-catheterization and urinary diversion difficulties, therefore requiring special medical attention.[1,3–11] As no evidence-based recommendations can be drawn from the available literature, we conducted this study in order to assess morbidity and functional outcomes of pregnancy and delivery in a cohort of women with neurogenic bladder or bladder exstrophy, and a history of AE, CCC (Monti or Mitrofanoff principle) and/or artificial urinary sphincter.

MATERIALS AND METHODS

Study Design and Population

We conducted a national retrospective study in 16 tertiary referral centers after obtaining French data protection agency approval to collect the available historical data (CNIL No. 2207432). We included all women older than 18 years with a neurological disease (spinal dysraphism, SCI or multiple sclerosis) or bladder exstrophy, and a history of AE, CCC and/or AUS implantation, who reported at least 1 successful pregnancy. These techniques imposed themselves as the international standard of care in the management of many neuro-urological diseases for adults able to perform intermittent self-catheterization, justifying the special attention given to their consequences on pregnancy and delivery. Women who underwent concomitant bladder neck closure, bladder neck reconstruction and/or fascial sling were also included in the study.

All women who had undergone another type of urological reconstruction were excluded, ie other continent pouches, noncontinent ileal conduit or ureterosigmoidostomy. Data collected included underlying disease, previous surgery, voiding mode, continence (urinary and fecal), parity, pregnancy and delivery outcomes, and postpartum data (including continence).

Statistical Analysis

Data are expressed as mean and SD or frequency and percentage for continuous and categorical variables, respectively. Statistical significance was set at $p < 0.05$. The chi-square test and Student's t-test were used to compare categorical and continuous variables, respectively. When the application conditions were not met we used Fisher's exact test for categorical variables and Wilcoxon's test for continuous variables. The normality of the distribution was verified by the Shapiro-Wilk test. We used analysis of variance (ANOVA) to compare means among 3 groups. Statistical analysis was performed using R Studio software V.1.0.143.

RESULTS

Population

Overall 68 women with a total of 96 deliveries between 1998 and January 2019 were included. Median age at delivery was 29 (18-39). Patient characteristics are summarized in table 1. All women performed intermittent self-catheterization. Fifty percent of women had spinal dysraphism, 30% had SCI and 15% had bladder exstrophy. One woman with bladder exstrophy had a bicornuate uterus and another woman had undergone complex vaginoplasty.

Most women had undergone AE (92.6%), 27 (39.7%) of whom had undergone concomitant CCC. At the beginning of pregnancy 13 women (19%) were treated with anticholinergic drugs and 14 (20%) received botulinum toxin injections for overactive bladder.

The majority of these women were continent before pregnancy, in terms of urine (91%) and feces (84%). Urological followup was disrupted at adolescence for many of these women. Urodynamic data before pregnancy were often missing and/or too scarce to be reported, with mostly a gap of almost 10 years between pregnancy and the last complete neuro-urological assessment.

Pregnancy Outcomes

A total of 45 women (55.5%) received antibiotic prophylaxis for UTI and 27 women (32%) reported at least 1 febrile UTI during pregnancy. Details about prophylaxis and urinary tract infections are reported in table 2. No significant differences in terms of rate of pyelonephritis, premature birth or fetal weight at

Table 1. *Patient characteristics*

No. pts	68	
No. deliveries	96	
Mean age (range)	29 (18–39)	
No. underlying disease (%):		
Spinal dysraphism:	33	(48.5)
Spina bifida	20	(29.4)
Spina lipoma	6	(8.8)
Sacral agenesis	7	(10.3)
Spinal cord injury:	20	(29.4)
Paraplegic	9	(13.2)
Tetraplegic	11	(16.2)
Multiple sclerosis	2	(2.9)
Bladder exstrophy	10	(14.7)
Others (1 anorectal malformation, 1 complex ureterocele)	2	(2.9)
No. early miscarriage (%)	12	(17.6)
No. ambulatory status (%):		
Walking	43	(63.2)
Wheelchair	25	(36.8)
No. syringomyelia (spinal dysraphism only) (%)	4	(5.9)
No. ventriculoperitoneal shunt (spinal dysraphism) (%)	8	(11.8)
No. urinary tract reconstruction type (%):		
Augmentation enterocystoplasty	63	(92.6)
Catheterizable channel (Monti or Mitrofanoff):	31	(45.6)
Isolated	4	
Associated to AE	27	
Rt iliac fossa	13	
Umbilical	9	
Unknown position	9	
Artificial urinary sphincter	6	(8.8)
Bladder neck closure	4	(5.9)
Bladder neck reconstruction (Goebell Stoeckel, Young Dees, fascial sling)	18	(26.5)
No. urinary incontinence before pregnancy (%)	6	(8.8)
No. fecal incontinence before pregnancy (%)	11	(16.2)
No. parity (%):		
P1	47	(69.1)
P2	14	(20.6)
P3	7	(10.3)
Twins	1	(1.5)

birth were observed between the antibiotic prophylaxis and no antibiotic prophylaxis groups.

The main complications during pregnancy are presented in table 3. Thirteen women (13.5%) experienced renal colic and 14 women (14.6%) required ureteral stenting or nephrostomy tube during pregnancy. Three women (3%) reported worsening of fecal incontinence and 13.5% experienced de novo or more severe urinary incontinence. Ten women experienced difficulties in intermittent self-catheterization during late pregnancy and none experienced difficulties in the manipulation of their AUS.

Delivery

The main mode of delivery was C-section in 64% of cases (61), with 51% (49) of elective C-sections, 10% (9) of emergency C-sections for spontaneous labor before elective C-section and 3% (3) of emergency C-section after attempted vaginal delivery. The main reported reason for elective C-section was to avoid urological complications. Vaginal delivery occurred in 36% of cases (35). Of these 31% were eutocic (30) and 5% required instrumental help by forceps or suction cup (5). A urologist was present in 50% of cases of C-section.

Only 34 women (35%) received epidural or spinal anesthesia during labor, while the other women gave birth with classic analgesics alone (19, 20%) or under general anesthesia (30, 31%, missing data 13).

Twenty complications occurred during delivery and are described in the supplementary table (https://www.jurology.com). Of these, 19 occurred during C-sections (95%). Twelve complications were urological and at least 1 additional surgical procedure was required to repair the iatrogenic complication in 8 cases. Three surgical reconstructions had to be completely abandoned after C-section. A urologist was present during delivery in 6 of the 12 cases of urological complications (50%).

The 6 women with an AUS presented 11 deliveries, including 5 C-sections, 5 vaginal deliveries and 1 instrumental vaginal delivery. Two major complications were reported and are detailed in the supplementary table (https://www.jurology.com). The other 4 women were continent after delivery.

Table 3. *Pregnancy outcomes*

	No. (%)
Pyelonephritis*	27 (33.3)
Acute renal colic	13 (13.5)
Urinary diversion during pregnancy (ureteral stenting or nephrostomy)	14 (14.6)
Alteration of fecal continence during pregnancy	3 (3.1)
De novo urinary incontinence	13 (13.5)
Difficulties in intermittent self-catheterization:	10 (10.4)
Via native urethra	6
Via continent channel	4
Site of continent channel:	
Rt iliac fossa	2/13
Umbilical	1/9
Unknown	1
Indwelling bladder catheter during pregnancy	5 (5.2)
Total No.	96

* Missing data in 15.

Obstetric Outcomes

Maternal and fetal outcomes are presented in table 4. Elective C-section and C-section performed because of

Table 2. *Urinary tract infections during pregnancy*

	Antibiotic Prophylaxis	No Antibiotic Prophylaxis	p Value (Student's t-test)
No. pyelonephritis (%)	16 (34.8)	11 (31.4)	0.751
Mean term (wks of amenorrhea) (SD)	36.57 (3.55)	36.38 (2.57)	0.798
Mean gm birthweight (SD)	2,662 (690)	2,678 (676)	0.920
Total No. pregnancies (%)*	45 (55.5)	36 (44.5)	Not applicable

* Missing data in 15.

Table 4. *Maternal and fetal outcomes*

	Total	Elective C-Section	Vaginal Birth (eutocic or instrumental)	Emergency C-Section for Fetal Distress	p Value
No. deliveries (%)	96	58 (60.4)	35 (36.5)	3 (3.1)	Not applicable
			Women's underlying disease		
No. spinal dysraphism (%)	49 (51.0)	27	20	2	Not applicable
No. spinal cord injury (%)	24 (25.0)	15	8	1	Not applicable
No. bladder exstrophy (%)	15 (15.6)	13	2	0	Not applicable
No. others (%)	8 (8.3)	3	5	0	Not applicable
			Fetal outcomes		
Median term of delivery (WA) (min-max)	36 (21-41)	36.4	37	34	0.220
No. full term greater than 37 WA (%)	55 (64.7)	30	24	1	Not applicable
No. moderate to late preterm (32—37 WA) (%)	26 (30.6)	19	5	2	Not applicable
No. very preterm (28—32 WA) (%)	3 (3.5)	2	1	0	Not applicable
No. extremely preterm (less than 28 WA) (%)	1 (1.2)	0	1	0	Not applicable
No. stillborn	3	0	2	1	0.547
Median gm birthweight (min-max)	2,740 (429-4,090)	2,663	2,824	1,953	0.123
No. initial intensive care unit stay (%)	18 (18.8)	14 (24.1)	3 (8.5)	1 (33.3)	0.097
No. congenital anomaly	5	5	0	0	Not applicable
			Maternal outcomes		
No. intrapartum complications (%)	20 (20.8)	19 (32.8)	1 (2.8)	0	<0.05
No. urological complications (%)	12 (12.5)	11 (19.0)	1 (2.8)	0	<0.05
No. presence of urologist at birth (%)	29 (30.2)	29 (50.0)	0	0	Not applicable
No./total No. urinary continence (%):					
6 Mos after delivery	72/85 (84.7)	39/50 (78.0)	32/34 (94.1)	1	0.293
1 Yr after delivery	67/80 (83.8)	36/47 (76.6)	30/32 (93.8)	1	0.077
3 Yrs after delivery	47/59 (79.7)	25/34 (73.5)	21/24 (87.5)	1	0.275
No./total No. fecal incontinence (%)	14/68 (20.6)	6 (10.3)	8 (22.9)	0	0.482
No. surgical procedure for secondary urinary incontinence (%)	8 (10.4)	8 (13.8)	0	0	0.012

spontaneous labor before elective C-section are grouped in the "elective C-section" column in table 4, as we assume they did not represent an obstetric emergency due to fetal distress. In this setting the surgical approach is usually slow and controlled as in elective C-section.

The preterm birth rate was 35.3%, without significant difference in terms of delivery mode ($p=0.220$). Of the 96 newborns 3 died at birth in a context of maternal pyelonephritis and premature birth. Five babies presented a congenital anomaly (1 cardiac malformation, 1 neurofibromatosis, 3 spina bifida).

Functional Urological Outcomes

Only 6% of the included women reported de novo, persistent or more severe urinary incontinence 6 months after delivery, without significant difference according to delivery mode ($p=0.263$). The 4 women who gave birth by instrumental vaginal delivery were all continent after delivery. The continence rate (urinary and fecal) did not appear to be impacted by either delivery mode (table 4) or repeated pregnancies and deliveries (table 5). These results were maintained over time, as 47 of the 59 women with a minimum followup of 3 years after delivery were still continent at last followup (median followup 7 years). There was no significant difference in terms of 3-year continence rates between women who gave birth vaginally or by C-section ($p=0.275$).

Eight women required secondary urological surgery to treat urinary incontinence after delivery (mainly mid urethral slings) and 4 women required surgery to treat vaginal prolapse (1 bladder exstrophy and 3 spina bifida, after 3 elective C-sections and 1 vaginal delivery).

DISCUSSION

To the best of our knowledge, the present study represents the largest cohort reporting pregnancy

Table 5. *Impact of multiparity on urinary continence*

	No./Total No. (%)			
	First Delivery	Second Delivery	Third Delivery	p Value
No. urinary continence (%):				
6 Mos after delivery	46/54 (85.2)	19/23 (82.6)	7/8 (87.5)	0.572
1 Yr after delivery	42/49 (85.7)	18/23 (78.2)	7/8 (87.5)	0.472
3 Yrs after delivery	28/35 (80.0)	14/18 (77.8)	5/6 (83.3)	0.860
Surgical procedure for secondary urinary incontinence	4/49 (8.2)	4/22 (18.2)	0/6 (0)	0.247
Fecal incontinence	9/45 (20.0)	3/19 (15.8)	2/7 (28.6)	0.837

and delivery issues in women with LUTR. According to the literature, symptomatic lower UTIs are observed in 45% to 100% of pregnancies in neurourological patients and febrile UTIs are observed in about 30% of pregnancies.[1,2,4] Similar rates were observed in our study.

Half of the study population received antibiotic prophylaxis during their pregnancy, as recommended in this population since publications about weekly oral cyclic antibiotic (WOCA) prophylaxis, showing that prophylactic treatment of asymptomatic bacteriuria in women performing intermittent self-catheterization was effective to prevent symptomatic or febrile UTI and low birth weight.[12–14] The results of our study did not demonstrate any impact of antibiotic prophylaxis on the pyelonephritis rate during pregnancy, on fetal weight or term of delivery, probably because of the variable protocols used in this cohort, not all strictly following the WOCA protocol, as some women were included prior to protocol publication.

In our series 10% of women experienced self-catheterization difficulties during pregnancy, which is lower than the 30% usually reported.[6,14,15] In the literature difficult intermittent self-catheterization does not appear to be related to the umbilical position of the continent channel. On the contrary, this difficulty seems to be more common when the continent channel is located in the right iliac fossa.[6,15,16]

Overall 13% of the women included in this study reported de novo or more severe urinary incontinence during pregnancy, which can be explained by treatment modifications, especially as some anticholinergic drugs and botulinum toxin are contraindicated during pregnancy. This problem usually resolves spontaneously after delivery.[4,7,10]

Our results are consistent with the literature, reporting that emergency upper urinary tract diversion during pregnancy is required in 10% to 40% of these women.[4,6,16–18]

The main delivery mode was C-section, mostly elective. Due to the multicenter, retrospective design of this study, the decision to perform C-section vs vaginal delivery was not standardized. In some centers elective C-section was systematically performed in all of these women. In the available literature elective C-section still represents the predominant mode of delivery in this population.[4,5] The main argument is to avoid prolapse, deterioration of urinary and fecal continence, or to avoid injury of the precious urinary reconstruction.[7,10] From an obstetric point of view, emergency C-section could be made difficult by the presence of intraperitoneal scarring causing obstruction to prompt delivery of a baby in distress. From the urological point of view, injury to the reconstruction may be more frequent in the case of emergency C-section. In our study only 3 emergency C-sections were reported for fetal distress after attempted vaginal birth. No complication was reported but data are too limited to make any conclusion on the overall urological risk of emergency C-section. Therefore, C-section, even when it is elective and accompanied by a full surgical team, including a urologist, appears to be at higher risk than vaginal delivery. In our study a urologist was present in 50% of the C-sections, but probably not always the most experienced, explaining the high rate of complications. The presence of an experienced reconstructive urologist may decrease this rate.

Therefore, we believe that vaginal delivery should be proposed as a first line option in all women who have undergone LUTR,[10,17,19–23] unless there is any medical contraindication to vaginal delivery, including failure of epidural anesthesia (particularly in patients with SCI at risk of autonomic dysreflexia), neurological contraindication as syringomyelia, cephalo-pelvis disproportion or major orthopedic limitation of abduction. However, C-section may be preferable in women with bladder exstrophy as the genital and pelvic malformations observed in this population can result in more difficult vaginal delivery. Urologists may be warned in advance, even in cases of attempted vaginal delivery, in order to promptly react in case of emergency C-section.

In our cohort 64% of newborns were full term. Three died at birth, all in a context of threatened premature birth related to maternal pyelonephritis. Prematurity is reported in about a quarter of cases, with a significant difference between women with bladder exstrophy and women with neurogenic bladder.[2,11] The higher incidence of prematurity in the bladder exstrophy group can be explained by the high prevalence of associated uterine malformations, and early and severe genital prolapse in this population.[11] In the study population prematurity was usually secondary to threatened premature birth associated with febrile UTI.

Urinary continence did not appear to be markedly modified after delivery, as an average of 84.7% of women were considered continent 6 months after delivery vs 91% before pregnancy, similar to the rates observed in the general population.[10,11,15,17,24–26] These results did not appear to be impacted by the mode of delivery ($p=0.293$) or multiparity ($p=0.572$). It should be noted that the average birth weight in our cohort was 2,740 gm, which may be responsible for less perineal damage than a normal birth weight.

Eight women in the study population had a ventriculoperitoneal shunt with no shunt dysfunction reported during pregnancy. Vaginal delivery is recommended in this situation in order to avoid the infectious complications related to any form of intraperitoneal surgery.[5,27,28]

The C-section complication rate in women with an AUS was particularly high (1/3). Systematic elective C-section in the presence of bladder neck closure or AUS has been recommended for decades due to the fear of impairing the urinary continence of these women.[5,7,24] Several recent publications have been unanimously reassuring about the outcome of vaginal delivery in the presence of an AUS.[9,10,16,29] They recommended deactivation of the device at around 37 WA in the case of elective delivery in a nonexpert center or at the beginning of labor in the case of elective delivery in an expert center.[9,29]

More than a quarter of the study population had undergone surgical closure or reconstruction of the bladder neck, and 64% of these women gave birth by C-section and 36% gave birth by vaginal delivery. Only 4 of these women reported urinary incontinence after delivery. The literature reports a similar attitude toward bladder neck closure, with no justification for elective C-section in order to preserve continence.[6,7,22,25]

Four women required surgical management of genital prolapse. The incidence of genital prolapse during pregnancy is high in women with bladder exstrophy. Nevertheless, the incidence and severity of genital prolapse after birth have been reported to be similar after vaginal delivery and after C-section.[6,10,11,26,30]

This study presents a number of limitations, the first being its retrospective design, responsible for missing data. Pregnancy and delivery remain rare in this population, accounting for the small number of cases studied despite the long inclusion period and the multicenter design. The multicenter design is a strength and a weakness due to the marked differences in clinical practices and choices, particularly concerning the mode of delivery, as some medical teams strongly encourage vaginal delivery while others prefer systematic elective C-section for the same women. Therefore, the conclusions of this study must be interpreted cautiously. Larger prospective cohorts are necessary in order to establish strong evidence-based guidelines.

CONCLUSIONS

Pregnancy and delivery are challenging in women with LUTR, as they are associated with specific complications requiring multidisciplinary management in an expert center. Our study highlights the high surgical risk associated with C-section in this population. Delivery mode does not appear to have any impact on urinary continence or pelvic organ prolapse. In light of these findings, vaginal delivery can be proposed as first line option in women with LUTR in the absence of any major genital malformation, obstetric, anesthetic or neurological contraindication.

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