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Toilet training achievements in children born with posterior urethral valves

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ABSTRACT

Objective Boys born with posterior urethral valves (PUV) often experience bladder development issues due to urethral obstruction during gestation. Despite early surgical intervention, bladder dysfunction can persist, leading to detrusor overactivity and vesical overdistension. Successful toilet training is key in managing bladder function and controlling intravesical and upper urinary tract pressures. This study aims to evaluate toilet training outcomes in children with PUV.

Study design This single-centre ambispective cohort study included 142 children aged 4 years and older who underwent PUV ablation between January 2003 and December 2021. The patients were categorised into two groups: those with cognitive comorbidities (group A), including autism spectrum disorder, attention deficit hyperactivity disorder and trisomy 21, and those without (group B).

Results Among the 142 children, 128 (90.1%) achieved daytime continence at a median age of 37 months (range 33–49 months) and 115 (81.0%) attained night-time continence at a median age of 42 months (range 34–60 months). Children in group A showed significantly lower rates of continence compared with group B (daytime: 74.2% vs 94.6%, p=0.0026; night-time: 67.7% vs 84.7%, p=0.0412). Additionally, children in group A completed toilet training significantly later than those in group B (daytime: 64 vs 36 months, p=0.0002; night-time: 60 vs 42 months, p=0.0139).

Conclusion Boys with PUV can achieve daytime and night-time continence at ages comparable to those reported by their peers; the presence of cognitive comorbidities is associated with delays in achieving these milestones.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Posterior urethral valves (PUVs) are a common cause of bladder dysfunction in children.
⇒ Toilet training in children with PUV can be delayed due to bladder dysfunction and poor sensation.
⇒ Cognitive comorbidities can further delay toilet training and continence achievement.

WHAT THIS STUDY ADDS

⇒ Boys with PUV can achieve daytime and night-time continence at similar ages to healthy peers when encouraged with proper bladder management.
⇒ Cognitive comorbidities (eg, autism spectrum disorder, attention deficit hyperactivity disorder, trisomy) are linked to delayed toilet training milestones in patients with PUV as much as in children without PUV.
⇒ Bladder and bowel medications are useful for continence achievement in selected boys with PUV.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ Early toilet training can be achieved in children with PUV (including those with cognitive comorbidities) in order to enhance long-term bladder and kidney function.

INTRODUCTION

With a reported incidence of 1 in 7000–8000 live births, posterior urethral valves (PUV) represent one of the most common causes of lower urinary tract obstruction in children.[1 2] Being characterised by flaps arising from the verumontanum to the posterior urethra, PUV create a valve-type obstruction that can impact urine elimination and cause harmful effects on the urinary bladder and the upper urinary tract development.[3 4] Histological modifications and increased bladder wall thickness accompany the functional changes induced by mechanical stress. Bladder structure alterations reflect directly in bladder dysfunction, a long-lasting pathological process that, with age, can evolve in functional changes ranging from detrusor overactivity to vesical overdistension and myogenic failure. Moreover, the intense inflammatory pathway that characterises chronic obstructive uropathy functionally manifests as haemodynamic and structural alterations in the developing kidney.[5–7] These mechanisms highlight the complex interaction between bladder and kidney and can lead to chronic kidney disease in about 20–65% of patients.[1 8]

Normal urinary continence in boys is reported as the ability to control bladder emptying and stay dry during the day and night. However, the age at which boys achieve urinary continence can vary widely, depending on cultural aspects. While in some Asian and African cultures, children are trained as infants, training at 2–3 years is more typical in Western countries.[9–12] The pathway to urinary continence involves several physical and psychological development stages.

Toilet training is a fundamental developmental step for children and is even more critical in boys born with PUV, as it allows easier control of bladder function. Only a few studies have been published

analysing toilet training achievements in children born with PUV.[11]

We aimed to evaluate the natural history of continence achievement in a cohort of boys born with PUV and followed up in our institution with particular attention to the presence of associated cognitive comorbidities and the requirement for bladder/bowel medications.

PATIENTS AND METHODS

A single-centre ambispective (ie, retrospective and prospective data collection) cohort study was performed, including all children who underwent PUV ablation between January 2003 and December 2021 (January 2003 to February 2019 for retrospectively collected data and March 2019 to December 2021 for prospectively collected data) and were followed up at our institution (audit number 17042). Inclusion criteria required patients with PUV to be at least 4 years old at the last follow-up and to have documented information regarding their toilet training age.

Patients were excluded from the study if they were younger than 4 years at the last follow-up, if there were missing data on the child's age of toilet training, if the child's age at PUV ablation was not recorded or if PUV ablation was performed after completing the toilet training.

These children's main steps and achievements in toilet training were recorded and analysed.

Urinary continence was considered to have been obtained if children became aware of the need to eliminate urine and the ability to use the toilet/potty. Age at toilet training achievement and presence and frequency of urinary leaks (small or large) during daytime or at night at the last follow-up were registered together with other patients' characteristics that could interfere with the acquisition of bladder control and the completion of the toilet training process, such as age at surgery, presence of constipation, the requirement of bladder and bowel medications and type of medication used.

Data were analysed and compared between children with (group A) and without (group B) associated cognitive comorbidities, including autism spectrum disorder (ASD), attention deficit hyperactivity disorder (ADHD), syndrome-associated developmental delay and chromosomal defect (eg, trisomy 21). Identifying patients' cognitive comorbidities was based on documented medical diagnoses in the patient's clinical records. Qualified healthcare professionals, including paediatric neurologists, developmental specialists or psychologists, made these diagnoses. The documentation included information from multidisciplinary evaluations and was supplemented by parental reports where applicable.

Statistical analysis

Statistics were performed using Stata V.16.1 (StataCorp LLC) and SPSS V.29.

Descriptive statistics were performed for all the variables. For quantitative variables, summary statistics were reported as median and first–third interquartile (q_1–q_3) whereas were based on absolute and relative frequencies for qualitative variables. Comparisons between the two groups of patients were performed using the non-parametric Mann-Whitney U-test or the Wilcoxon signed ranks test on a set of matched samples. Frequency data were analysed using the Pearson χ^2 or Fisher exact test in cases of expected frequencies lower than 5.

Cross-tab with χ^2 analysis was used to assess the relative proportions of categorical parameters in the potty-trained and not-trained groups. Binary logistic analysis was used to test the collective predictive power of all relevant parameters.

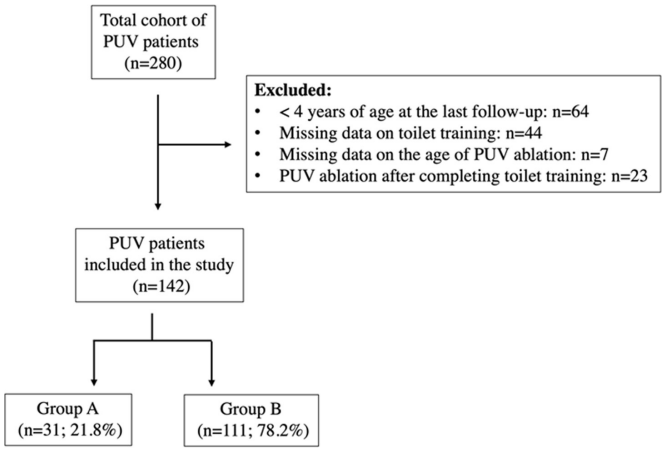

Figure 1 Study design. PUV, posterior urethral valve.

RESULTS

During the analysed period, 280 patients underwent PUV resection at our institution and 142 met the inclusion criteria and were included in the study (figure 1).

The median age at valve ablation was 49 (17–307) days, and the median follow-up period was 9 (6–12) years (table 1). At the last follow-up, 128 (90.1%) were toilet trained during the day and 115 (81.0%) during the day and the night. Daytime and night-time continence were achieved at a median age of 3.1 (2.8–4.1) years and 3.5 (2.8–5.0) years, respectively.

In total, 31/142 (21.8%) children had associated comorbidities and were included in group A: 9 (29.0%) had ASD, 5 (16.1%) had ADHD and 11 (35.5%) had cognitive delay associated with trisomy 21. The remaining 6 (19.4%) had other specified cognitive difficulties.

In group A, 23 (74.2%) out of 31 patients completed toilet training during the day and 21 (67.7%) completed toilet training during the day and the night. In group B, 105 (94.6%) out of 111 patients completed toilet training during the day and 94 (84.7%) completed toilet training during the day and the night. There was a statistically significant difference in the proportion of children who reached daytime (p value: 0.0026) and night-time (p value: 0.0412) continence between the two groups of patients (figure 2). Children in group A completed their daytime toilet training at a median age of 64 (38–103) months compared with 36 (32–46) months in group B (p value: 0.0002). With regard

Table 1 Patients' demographics and outcomes

Patients' demographics	
Age valve ablation, days (median; IQR)	49 (17–307)
Follow-up period, years (median; IQR)	9 (6–12)
Patients' outcomes	
Age at completion of toilet training daytime, median (months; IQR)	37 (33–49)
Age at completion of toilet training night-time, median (months; IQR)	42 (34–60)
Daytime toilet-trained patients with daytime urine leaks (n; %)	25 (19.5)
Daytime and night-time toilet-trained patients with night-time urine leaks (n; %)	18 (15.7)
Patients on bladder medications (n; %)	31 (21.4)
Patients on bowel medications (n; %)	41 (28.9)

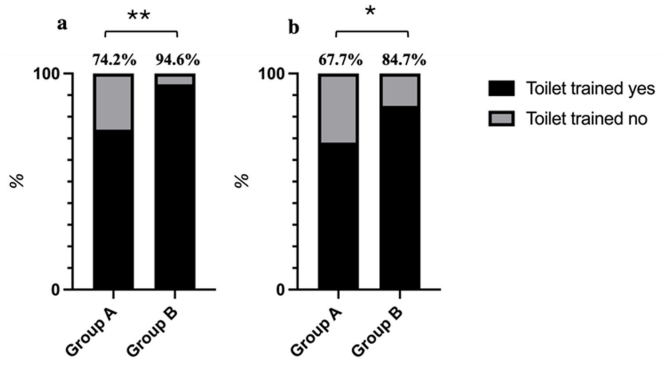

Figure 2 Posterior urethral valve children aged ≥4 years with (group A) or without (group B) associated cognitive impairment potty trained during the day (a) and during the night. * p-value < 0.05; **p-value: < 0.01.

to night-time continence, children in group A achieved it at a median age of 60 (36–96) months compared with 42 (33–52) months in group B (p value: 0.0139) (figure 3).

The use of bladder medications was not found to be significantly associated with the achievement of continence both during the day and at night (21.1% vs 28.6%; p value: 0.5060 and 20.0% vs 29.6%; p value: 0.3039, respectively) (figure 4a,b). No significant association was also found for the need for bowel medications (30.4% vs 42.9%; p value: 0.2289 and 27.0% vs 37.0%; p value: 0.3469) (figure 4c,d) or anti-hypertensive drugs (6.3% vs 7.1%; p value: 1.000 and 3.5% vs 3.7%; p value: 1.000) (figure 4e,f).

Table 1 shows the relative proportions of toilet-trained and non-trained patients and the associated proportion of patients on bladder and bowel medications, as well as clean intermittent catheterisation (CIC). These parameters were not statistically significantly different between the groups (p>0.05). Binary logistic regression analysis did not identify any significant predictors.

Looking at all cohorts of patients, out of the 128 boys with PUV who achieved toilet training during the day, daytime accidents were reported in 25 (19.5%) patients (small leaks: n=23; large leaks: n=2) while out of the 115 boys toilet trained during

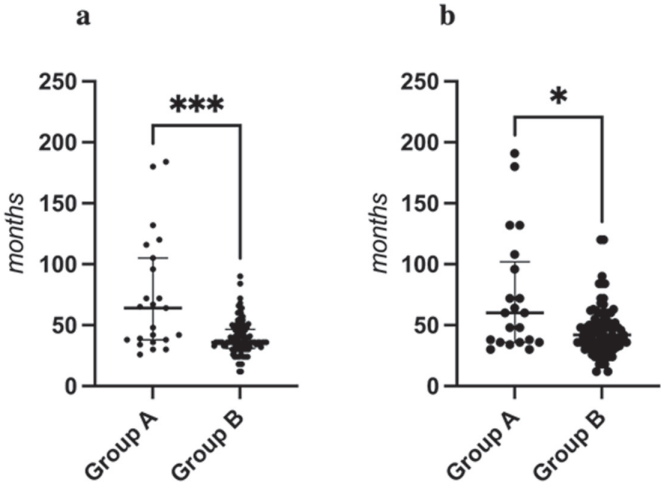

Figure 3 Age at daytime (a) and night-time (b) urinary continence in our cohort of boys with posterior urethral valve, with (group A) or without (group B) associated cognitive impairment. * p-value < 0.05; ***p-value: < 0.001.

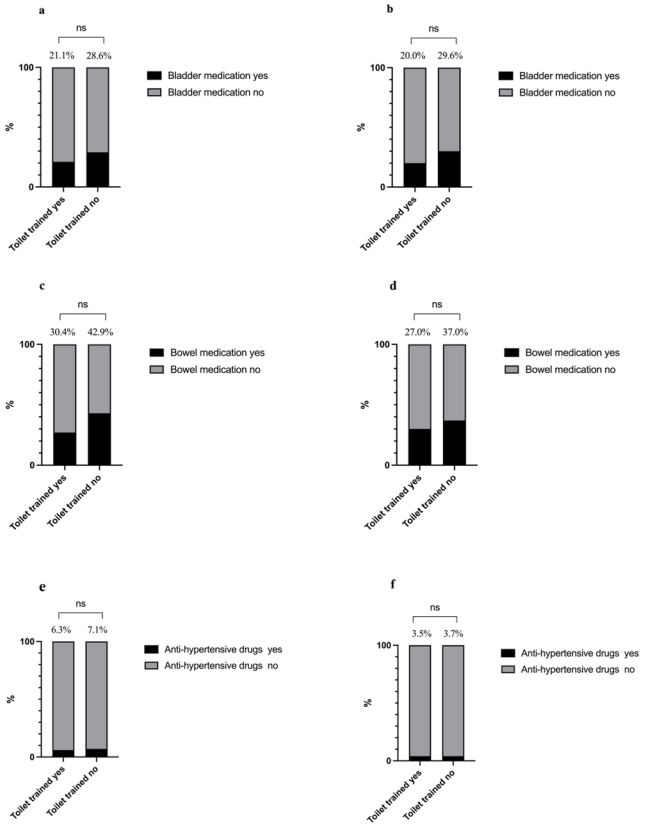

Figure 4 Relationship between toilet training achievements reached during the day and the night and the need for bladder (a, b), bowel medications (c, d) and hypertensive drugs (e, f). ns: not statistically significant.

both day and night, night-time accidents were reported in 18 (15.7%) patients (small leaks: n=17; large leaks: n=1) (table 1 and online supplemental figure 1). No association between the use of bladder or bowel medication or antihypertensive drugs and urinary accidents was identified (online supplemental figure 2).

DISCUSSION

Toilet training is a significant milestone in early childhood development. It marks the transition from dependency on diapers to mastering control over bodily functions. Toilet training is a complex process that occurs with normal physical and psychosocial growth and is influenced by various factors, including physiological maturation, cognitive readiness, social norms and parental strategies. While it is celebrated as a milestone, it can also be a source of stress and frustration for children and parents alike. It can be affected by anatomical, physiological and behavioural conditions.

Children are usually considered ready to start toilet training when they are emotionally mature and interested in the process. They have the neurologic capability to cooperate and show some degree of bowel and bladder control, identified by the ability to suppress bladder and bowel reflexes.[13] A child is considered fully toilet trained when he/she is aware of his/her own need to eliminate urine and stool and initiates the act without being reminded or prepared by parents or caregivers.[13]

Multiple factors can influence toilet training, such as society and customs, parental education level, mother's working status, single parenthood,[14] family's socioeconomic

status, race, gender and prematurity.[13 15] Cultural expectations and practices play a significant role in these milestones. For instance, in Western cultures, toilet training is generally delayed until the child exhibits readiness cues, such as staying dry for extended periods, showing awareness of bladder and bowel urges, expressing interest in the toilet/potty or desire to stop wearing nappies. Conversely, in many Asian and African societies, early toilet training—sometimes initiated even within the first year of life—is customary, reflecting a broader focus on caregiver-led training strategies.[10 16] Studies also indicate that boys typically achieve continence milestones later than girls; this may reflect differences in physiological development and set parental expectations/actions.[17] The American Academy of Pediatrics and the Canadian Paediatric Society recommend starting toilet training in healthy children when they are 18 months of age.[1] However, in the past 50 years, the average age at which children with normal neuropsychomotor development start and complete toilet training has been postponed from 18 to 24–36 months and from 24 to 36–39 months, respectively.[13]

Several studies have shown that once bladder control is achieved, the physiological conditions of the bladder change. Bladder capacity increases while the coordination between the bladder and sphincters matures, improving bladder-emptying ability.[18 19] Early bladder control has been associated with reduced risks of dysfunctional voiding and urinary tract infections, likely due to shorter periods of bladder-sphincter dyscoordination.[20]

Therefore, despite being a great challenge for children, parents and physicians, ensuring adequate and successful toilet training is an important stepping stone in a child's development.

A recent survey reported that children born prematurely have a 2.7 times greater chance of achieving potty training at a later age (> 3 years).[14]

Late achievement of continence has also been associated with a higher degree of dysfunctional voiding with unclear cause–effect relationships.[20]

In boys born with PUV, toilet training represents an even more critical step for the impact that the development of bladder dysfunction can have on renal function. Interference with normal bladder contraction starts during gestation as a response to the urethral obstruction, and even in cases in which early valve ablation is performed, the voiding pattern is characterised by extreme variability.[10] Historically, it was believed that it is more likely to find bladder hypercontractility at the early stages of bladder development, which subsequently resolves. In some cases, the pattern changes toward overdistension.[5–7]

Even though there has been much discussion on bladder dysfunction over time, little research has been done to define toilet training performance in children born with PUV.

We investigated toilet training achievements in our cohort and explored the effectiveness of the process in boys with associated neurodevelopmental comorbidities. An interesting finding in the present study is that boys born with PUV completed their toilet training at similar ages compared with what has been reported for the general population of otherwise healthy children.[1 9 13 21]

This data contrasts with what was reported in 2019 by Jalkanen *et al*.[22] In their study, boys with a diagnosis of PUV achieved urinary continence significantly later than their peers from the general population (daytime and night-time urinary continence: 5.5±3.3 and 5.4±3.0 years vs 2.3±0.5 and 2.9±1.2 years, respectively; p<0.001).

Their data, however, included patients with a wide age range at resection (from 0 to 11 years) and were collected through a retrospective hospital database review of adults with a median age of 35 years at the time of the study. This could generate obvious bias and possible inaccuracies.

On the contrary, our cohort is much larger, more selected and with less possible bias.

Since bladder dysfunction in boys with PUV is often noted at an early age[10] and treatment may reduce the risk of deterioration of kidney function, we believe that it is crucial to institute bladder treatment as early as possible and, therefore, we encourage parents to start toilet training at an early age (after the second birthday).

Whether this approach has contributed to the difference in the results achieved is unclear.

Another benefit of achieving continence early on is the possibility of investigating bladder emptying and intervening with behavioural strategies (eg, time voiding and double voiding) to ensure correct bladder emptying.

Large post-void urine residuals expose children to an increased risk of urinary tract infections but can also aggravate the dysfunction of the detrusor-sphincter complex. The possibility of teaching a boy to routinely perform a second void within seconds/minutes (double voiding) would minimise the residuals and reduce such risk.

In our cohort of patients, associated cognitive comorbidities were associated with difficulties in reaching these goals, which were achieved but at older ages.

This is in line with previous literature that suggests a late achievement of continence for cognitively impaired children, regardless of the presence of PUV.[17–20]

Children with cognitive comorbidities represent a not negligible group of our cohort, and the number of children receiving such diagnoses has increased in recent years. The presented data highlights the important message that children with such comorbidities can still achieve continence, even at a later age. Children with trisomy 21, for example, represent a subgroup, and we feel it is important to highlight that parents should still aim to achieve continence and that, in case of failure, bladder-specific investigation is recommended to avoid dangerous outcomes, as in the case report of a bladder rupture that occurred in a boy with trisomy 21 and undiagnosed PUV.[23]

In our institution, we investigate bladder emptying during infancy.[24] CIC is considered when emptying difficulties are documented. The need for CIC is then reevaluated throughout the child's growth. In some boys, CIC is discontinued when they complete potty training, as it can be substituted by voluntary double voiding.

Boys with bladder/detrusor overactivity may require detrusor-relaxing drugs. In our cohort, 31/142 (21.4%) boys were on one or two bladder medications (n=30 antimuscarinics, n=8 alpha-blockers) during potty training. However, the need for bladder medication does not negatively influence the ability to achieve continence; this data emphasises the importance of establishing correct bladder management as early as possible.

The role of constipation in achieving continence is controversial. Some authors report a significant impact,[20] while others suggest the opposite.[14]

We are inclined to agree with the former and systematically inquire about bowel function during outpatient consultations and advocate medical support for boys with signs of constipation. In our cohort, we have found no correlation between potty training achievement and the use of bowel medications. This suggests that adequate bowel control would allow the boys to achieve their continence normally.[25]

Several studies have demonstrated that the increasing age of bladder control means that the period of dyscoordination between the bladder and sphincter has been prolonged.[18 19]

Therefore, showing that boys born with PUV can reach bladder control at similar ages to their peers might benefit the overall preservation of their bladder and kidney function.

The limitations of this study are mainly associated with its design. Some of the data were collected through a review of charts initially not designed to collect data for research. Therefore, some information was bound to be missing. Selection and recall biases also affect the results, and differences between patients and lost follow-ups can often not be ascertained and may lead to bias; we feel, however, that the large cohort attenuates the influence of those potential biases analysed. While a post hoc power calculation was not performed, the study's findings are based on a substantial sample size and robust statistical analyses. However, future studies incorporating predefined power analyses and larger cohorts would further strengthen and validate these results.

Furthermore, while we acknowledge that the absence of a control group for toilet training achievements and medication comparison is a limitation, this study provides meaningful data within the constraints of its design, and we believe the findings contribute valuable insights to the field. We also recognise that the ethnic makeup of the population could influence the interpretation of our findings, as cultural and societal factors significantly impact toilet training milestones. Finally, we acknowledge the possibility that some patients in group B may have unidentified or emerging cognitive comorbidities that were not apparent during the study period. This is an inherent limitation in studies where neurodevelopmental conditions may manifest later in life. However, the large cohort analysed likely attenuates the influence of these biases.

Given these limits, we suggest that boys born with PUV can achieve daytime and night-time continence with appropriate bowel and bladder management, including support for toilet training. While the study did not include data on the timing of toilet training initiation or direct assessments of bladder-emptying efficiency, these outcomes remain important areas for future research to determine the potential impact of early interventions on bladder and kidney health.

CONCLUSION

Our findings suggest that children born with PUV who receive proactive bowel and bladder management can achieve daytime and night-time continence at similar ages to what has been reported for their peers. However, associated cognitive comorbidities, like in the general population, lead to late continence achievement. The effect of early potty training in children with PUV deserves better attention. Further studies are needed to explore its impact on preserving long-term bladder and kidney function.

Correction notice Since this article first published, an open access licence has been added.

Contributors MG is responsible for the overall content as the guarantor. IP and MG conceived the presented idea. IP and MG developed the theory and performed the computations. MG and ES verified the analytical methods. JCC encouraged MG to investigate the nephrological aspect and supervised the clinical findings of this work. IP and MM contributed to the data collection.

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