



Standard Urotherapy for Children with Neurodevelopmental Disabilities: A Systematic Review

Maayke van Galen^{1,2} · Bibi Huskens² · Alexander von Gontard^{3,4} · Robert Didden^{1,5}

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Abstract

Objectives Standard urotherapy (SU) is regularly used as a first-line intervention for children with incontinence. It is suggested that SU is also effective for children with neurodevelopment disabilities (NDs) when altered to the specific needs of these children. The aim of the review was to answer the following research questions: (a) what was the effectiveness of SU interventions in children with ND?, (b) which procedures and protocols were utilized during SU?, (c) what was the role of parents during SU?, and (d) what was the current evidence base regarding SU and children with NDs?

Methods Four electronic databases (i.e., Embase, PsychInfo, PubMed, Web of Science) were searched in accordance with the JBI methodology for mixed methods systematic reviews (MMRS) in the Joanna Briggs Institute (JBI) manual for Evidence Synthesis. The Mixed Method Appraisal Tool (MMAT) from JBI was utilized to determine the methodological quality of the studies.

Results The search resulted in the inclusion of five articles related to SU in children with ND, indicating that research relating to SU and NDs is scarce. Results of the review indicate that SU resulted in a complete or partial positive response in 30% of the children with NDs ($N = 114$) based on the criteria listed by the International Children's Continence Society (ICCS). When using criteria by the authors of the included articles, the results indicate that 59% of the children achieved full continence. Most importantly, the evidence suggests that most children showed improved symptoms (e.g., reduction in urinary/fecal accidents, schedule-dependent continence) even if full continence was not obtained.

Conclusions The review indicated that SU could be beneficial as a first-line treatment for children with NDs. Individualizing treatment to the needs of the child, parental involvement, follow-up appointments, and adding adjuncts when SU alone is not effective seem to be beneficial for this specific population. However, research relating to this topic is scarce and therefore more research needs to be completed regarding the role of parents and effective guidelines specific to different NDs. Implications for future research are discussed.

Keywords Standard urotherapy · Neurodevelopmental disorders · Incontinence · Children · Systematic review

According to the DSM-5, neurodevelopmental disorders (NDs) typically have an early onset in the developmental period of a child's life (American Psychiatric Association, 2013). The developmental deficits are characterized by a wide variety of impairments pertaining to social, personal, academic, or occupational functioning. The DSM-5 categorizes the following NDs: intellectual disability (ID), (social) communication disorders, autism spectrum disorder (ASD),

attention-deficit/hyperactivity disorder (ADHD), specific learning disorders, motor disorders, stereotypic movement disorder, and tic disorders. NDs often co-occur, but they also have a high comorbidity with other conditions such as elimination disorders (EDs). In fact, individuals with NDs have a higher risk of all types of ED (van Laecke, 2008; von Gontard et al., 2016).

The DSM-5 describes specific criteria for EDs. However, the International Children's Continence Society (ICCS) classification system and Rome-IV state that the DSM-5 criteria do not fully reflect ED and should be revised (Schaefer and Diamond, 2014; von Gontard, 2011; von Gontard & Kuwertz-Bröking, 2019). There is international and interdisciplinary consensus upon using terminology of ICCS and

✉ Maayke van Galen
mgalen02@seyscentra.nl

Extended author information available on the last page of the article

Rome-IV (von Gontard & Kuwertz-Bröking, 2019; von Gontard, 2013a, 2013b). According to the ICCS criteria, ED can be categorized in continuous urinary incontinence (UI) which entails constant urine leakage during day- and night-time and intermittent UI. Intermittent UI is divided in daytime UI (DUI) and night-time UI, i.e., nocturnal enuresis (NE) (Austin et al., 2016). Rome-IV criteria are now the most widely accepted guidelines for fecal incontinence (FI). There are two types of FI: functional constipation (FC) and non-retentive fecal incontinence (NRFI) (Bongers et al., 2007; Hyams et al., 2016). The minimum chronological age requirement for UI and FI is 5 and 4 years, respectively, or an equivalent neurodevelopmental level (American Psychiatric Association (APA), 2013; Austin et al., 2016).

A study by Francis et al. (2017) indicated that individuals with ASD are more likely to experience a wide variety of EDs such as NE and FI compared to the general population. In addition, the prevalence of DUI is higher in ASD compared to typically developing children with rates ranging between 13 and 55%, while the range of FI varies from 2 to 71% (median of 12%) (Niemczyk et al., 2018). When looking at the prevalence of ED in children with ADHD, research is limited. However, the current evidence base suggests that 25% of children are affected with DUI and 13% with FI (von Gontard & Equit, 2015; von Gontard et al., 2021). Furthermore, prevalence research pertaining to ID and ED indicates that the lower the IQ, the higher the incontinence rates are, with prevalence rates varying between 23 and 86% (van Laecke, 2008; von Wendt et al., 1990). These prevalence rates support the claim that there is high comorbidity between ED and NDs in children.

Achieving continence is an important milestone during the child's development. However, achieving continence can be difficult for some children, and these struggles can have a damaging impact on the child's overall health (Collins et al., 2019). Effective treatment for ED is socially significant as incontinence is often linked to behavioral and emotional problems, such as low self-esteem, low quality of life, anxiety, and depression (Gontard, 2011). However, ED treatment can sometimes be difficult to implement as NDs can present additional challenges that might arise during treatment (Austin et al., 2016; Gontard, 2013b; von Gontard et al., 2021). For example, lower compliance with the procedures and treatment outcomes are worse for children with ADHD, while children with ASD often require more treatment time with more structure during treatment (Crimmins et al., 2003; von Gontard & Equit, 2015). Furthermore, an intensive behavioral approach in children with ASD seems to be desired in order to increase treatment outcomes (Francis et al., 2017; Hanney et al., 2013; van Oorsouw et al., 2009). In addition, research indicated that individuals who are not successful in attaining toileting skills as a child have an increasingly difficult ability to obtain successful toilet skills

as an adult (Lohmann et al., 1967; Smith & Smith, 1977; van Laecke, 2008). Since the prevalence rates of DUI and FI for children with NDs are higher compared to the general population, and considering the high impact of incontinence on the child's overall health, it is especially important for this population to get access to effective treatment.

The general consensus within the field indicates that the assessments pertaining to incontinence should be straightforward, clinically based, and least intrusive (von Gontard & Kuwertz-Bröking, 2019; von Gontard, 2013a). In addition, simple interventions should be introduced first within the treatment of incontinence. If there is a combined disorder (e.g., DUI and FI), FI should be treated first (von Gontard, 2013a). It is advised that children with NDs should receive the same access to the diagnosis and treatment of incontinence compared to typically developing children (van Laecke, 2008; von Gontard et al., 2016). Urotherapy and medication are basic treatments for ED (von Gontard, 2013a). Urotherapy implies counseling, provision of information, charts, change of toileting and drinking habits, cognitive-behavioral interventions, alarm treatment, relaxation methods, and biofeedback. A meta-analysis revealed standard urotherapy (SU) as a first-line intervention for daytime incontinence (Schäfer et al., 2018). According to the ICCS, SU is a first-line treatment effective for most functional disorders, while specific urotherapy requires an indication and is specifically effective for lower urinary tract disorders (von Gontard, 2013a). SU includes disorder specification, instructions to establish better micturition, guidelines for fiber and fluid intake, documentation of all symptoms, and continuous monitoring by the treatment team (Schäfer et al., 2018). Current research indicates that SU could and should be implemented as a first-line treatment for incontinence in all children, including children with NDs. Several practical guidelines are stated within the literature, such as an interdisciplinary approach for children who show treatment resistance (Caldwell et al., 2018). This is in consensus with a document issued by the ICCS, indicating that treatment should be altered according to the needs of children with NDs. A multidisciplinary approach is therefore crucial in order to provide effective treatment (Gontard et al., 2021). In addition, several other strategies related to the three most common NDs are also listed in this overview (von Gontard et al., 2021).

Results of several studies support the use of SU as an effective treatment for DUI, regardless of the subtype, while the effectiveness for children with FI is lower (Schäfer et al., 2018). However, it is important to determine current evidence base regarding the effectiveness of SU in children with NDs. Even though current literature suggests some practical guidelines in order to alter incontinence treatment to the needs of children with NDs, it is necessary to investigate the evidence related to these alternations in SU for

children with NDs. Especially the following data related to the effectiveness of treatment will be useful for professionals: the duration of treatment, frequency of contact with the treatment, protocols/procedures utilized during treatment related to specific NDs, and parental involvement (Assis et al., 2019). This is especially relevant considering each ND is different and therefore might require different alternations to SU in order to be effective.

A systematic literature review was conducted with the aim of answering the following research questions: (a) what is the effectiveness of SU interventions in children and adolescents with ND?, (b) which procedures and protocols are being utilized during the application of SU for children and adolescents with ND and are specific childhood disorders distinguished?, (c) what is the role of parents during SU?, and (d) what is the current evidence base regarding (including the methodological quality of published articles) SU and children with NDs?

Methods

The systematic review was conducted in accordance with the JBI methodology for mixed methods systematic reviews (MMRS) in the Joanna Briggs Institute (JBI) manual for Evidence Synthesis (Lizarondo et al., 2020). A mixed methods systematic review was selected since this review will consider qualitative, quantitative, and mixed-method studies.

Inclusion and Exclusion Criteria

All articles included in this review met the following inclusion criteria: (a) data were collected within (non) randomized controlled group designs, quasi-experimental designs, qualitative studies, or observational studies; (b) the study population consisted of children and/or adolescents (age 5–18 years); (c) at least one of the participants was diagnosed with DUI or FI; (d) the participants included in the original study had at least one neurodevelopmental disorder; (e) at least one component of standard urotherapy was utilized during treatment; (f) comparative studies consisted of two or more experimental groups which included at least an SU (alone) condition and control condition; (g) selected articles are published in English and/or Dutch; (h) articles were available in full text or obtained from the authors upon request.

Furthermore, the following exclusion criteria were utilized during the selection process: (a) articles related to the adult population; (b) one or more of the included participants in the original study did not have a listed ND; (c) solely poster abstracts were available; (d) comparative studies without an SU-alone condition; (e) studies without an

intervention component; (f) articles in languages other than English and Dutch.

Search Procedure

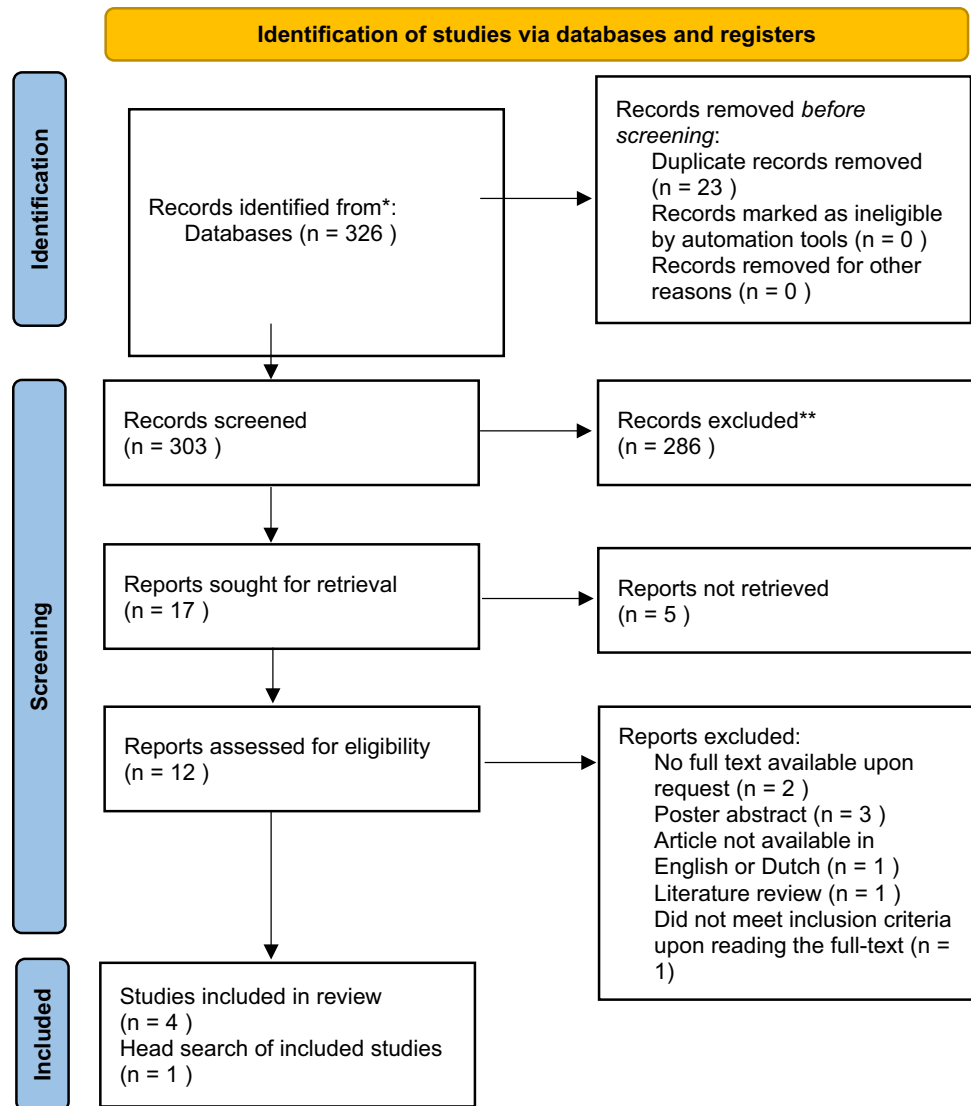
Four electronic databases (i.e., Embase, PsychInfo, PubMed, Web of Science) were utilized for the selection of articles. The selection was not restricted by the year of publication. Four key concepts were used during the search process which included incontinence, standard urotherapy, neurodevelopmental disorder, and child and/or adolescent. Within each database, the search was completed by entering the following algorithm with parenthetical terms into the keyword field: (incontinen* or “bladder bowel dysfunction” or “urination dis*” or enuresis or encopresis or “dysfunctional voiding” or constipation) AND (child* or adolescen*) AND (urotherapy or “standard urotherapy” or “behavioral interventions” or “incontinence treatment” or “bladder training”) AND (“neurodevelopmental dis*” OR “developmental dis*” OR ADHD OR ASD OR autism* OR “learning dis*” OR “intellectual dis*” OR “mental retardation” OR “IQ < 70” OR “Attention Deficit Hyperactivity Disorder” OR “mental dis*”). The search in the relevant databases took place in September 2021.

Study Selection

All identified abstracts were collected in a Microsoft Excel document, stating the article’s author, publication date, title, and abstract. This information was then screened by two independent reviewers (i.e., the first author of this study and a university master student of educational science) for assessment against the inclusion and exclusion criteria. Next, the full text of selected citations were obtained and assessed in detail against the inclusion and exclusion criteria by the two independent reviewers (Lizarondo et al., 2020). Finally, hand searches were completed regarding the references of all included articles. The results of the search are reported in the PRISMA flow diagram (McKenzie et al., 2020) (see Fig. 1).

During the first database search, 326 articles were found. After removal of all duplicates, 303 remaining articles were assessed by the two independent reviewers based on the inclusion criteria. The resulting list of abstracts was compared between the two reviewers. There was 96% agreement (i.e., agreement was obtained on 291 of the 303 studies) on the abstracts eligible for inclusion, resulting in a total of 17 studies. These studies were further screened by obtaining and reading the full article of the articles. An additional 13 articles were excluded based on listed inclusion and exclusion criteria (see Fig. 1). This resulted in inclusion of four studies with 100% agreement between the reviewers. Subsequently, a hand search was completed regarding the reference list of the selected articles. This search identified two

Fig. 1 PRISMA flow chart showing the study selection (McKenzie et al., 2020)



additional articles, of which only one article was included since the full text of one of the articles was not available in English or Dutch. Ultimately, five studies were included in this systematic review.

Data Extraction

The included articles utilized quantitative and qualitative data to report the results. Therefore, both quantitative and qualitative data was extracted from the included studies by the first author using Microsoft Excel. The data extracted from the quantitative studies typically included descriptive statistical data that provided information regarding the sample size and statistical significance indicating the effectiveness of the procedures. In addition, the qualitative studies entailed descriptive data. All data relevant to the research questions was extracted from the articles and summarized based on the context of the current

review (Lizarondo et al., 2020). The information from the included articles was therefore summarized using the following categories: (a) participant characteristics, (b) dependent variable(s) of the study, (c) research question, (d) intervention procedures, (e) SU component(s), (d) intervention outcomes, and (g) methodological quality. In order to facilitate interpretation and comparison between the quantitative studies and qualitative study, a data transformation took place in which the quantitative data was converted into qualitized data by providing textual descriptions or narrative interpretation of the results as to respond directly to the research questions.

Regarding the characteristics of participants, the following information was extracted from each article: (a) number of participants, (b) type of incontinence problems, (c) neurodevelopment disorder(s) listed, (d) parental involvement during SU, and (e) SU component(s) utilized during treatment as described by Schäfer et al. (2018).

Finally, the methodological quality of each study was assessed using the JBI critical appraisal instrument from JBI SUMARI (Hong et al., 2018). The Mixed Method Appraisal Tool (MMAT) utilized for this systematic literature review is a critical assessment tool for the appraisal of systematic mixed studies reviews. This tool was chosen based on the possibility to appraise all types of studies presented in this review (i.e., quantitative non-randomized, quantitative descriptive, and qualitative case study). The MMAT distinguishes between five categories that could be assessed by utilizing the tool: qualitative research, randomized controlled trials, non-randomized studies, quantitative descriptive studies, and mixed-method studies. For each of these categories, the MMAT provided a checklist with a description of the criteria, which allowed a sensitive analysis of methodological quality of each study. Depending on the format of the study, the corresponding MMAT checklist was

used for this review. The results are presented in percentages based on the percentage of methodological quality criteria met (see Table 1).

In order to assess the methodological quality of each study (after data transformation), the following data were extracted from the article: research question, dependent variable, study design (i.e., qualitative research, quantitative non-randomized studies, quantitative descriptive studies), and methodological quality score in percentages. All studies were assessed by the previously mentioned reviewers, using the critical appraisal instrument from JBI. There was 93% agreement between the reviewers on the quality criteria for the included articles. The methodological quality of four out of five studies has been rated sufficient by both reviewers as the methodological quality scores ranged between 71 and 100%. The scores were based on the number of positive responses related to seven methodological quality criteria

Table 1 Critical appraisal studies based on the MMAT (Hong et al., 2018)

Citation	Dependent variable(s)	Research Question	Category of study design	Quality of the study MMAT (0–100)%
Eliezer et al. (2021)	Percentage of symptom reduction (based on the ICCS research criteria) measured by obtaining the post-void residual (PVR), the Akbal score and Pediatric Incontinence Quality of Life (PinQ)-score	Evaluate the efficacy of providing SU to children with treated behavioral disorders, describe indications for combination therapy, report on both objective and subject bladder outcomes and to identify barriers for treatment of bladder dysfunction	Prospective cohort study; quantitative non-randomized	100%
Hanney et al. (2013)	Frequency and latency data regarding the following target behaviors: successes, accidents, accident/success conversions, and initiations	The purpose is to summarize the processes and outcomes during implementation of the LeBlanc et al. (2005) protocol at a university based outpatient clinic for children with ASD	Quantitative descriptive (archival) study	71%
Kaye and Palmer (2010)	Percentage of symptom reduction (based on the ICCS research criteria) (PinQ)-score	Could voiding dysfunction in children with ADHD be successfully treated with if the therapy was tailored to their needs?	Quantitative descriptive study	71%
Smith et al. (2000)	The dependent variables are not clearly stated in this study	The research questions is not explicitly stated, which makes it difficult to state how the collected data relates to the research question	Case study; qualitative	0%
Van Laecke et al. (2010)	Maximum voided volume, fluid intake quantity and urinary incontinence Questionnaire scores	What is the prevalence of urinary incontinence in a group of intellectually and physically disabled children, while looking at causing factors and evaluating therapeutic options What is the efficacy of adequate fluid intake as part of the a treatment strategy	Prospective study; quantitative descriptive study	86%

(see Table 1) (Eliezer et al., 2021; Hanney et al., 2013; Kaye & Palmer, 2010; van Laecke et al., 2010). In addition, both independent reviewers rated the methodical quality of one article as insufficient since both reviewers scored 0% on the methodological quality of that article (Smith et al., 2000). The main methodological concern was the lack of a clear research question. Furthermore, the dependent variable of the present study was not clearly stated which made it difficult to determine how the collected data addressed the research question. Due to the limited amount of research related to this topic, this article does contribute to the current base of evidence related to the literature on this topic. Therefore, the article was included in this review. However, the results of this study should be interpreted with caution based on the low methodological quality of this study.

Results

Table 2 summarizes all the included articles based on the following components: (a) participants characteristics, including number of participants, type of incontinence problems and relevant NDs, (b) SU component(s) utilized as described by Schäfer et al. (2018), (c) parental involvement, (d) other intervention procedures utilized, and (e) intervention outcomes.

Participant Characteristics

Specific child characteristics including relevant NDs were reported in all five articles. In total, 258 children participated in all five studies combined. The sample size ranged from 3 to 111 participants, which included one case study ($n=3$) (Smith et al., 2000), while the remaining four studies had a sample size of 30 or higher (Eliezer et al., 2021; Hanney et al., 2013; Kaye & Palmer, 2010; van Laecke et al., 2010).

Of the 258 participants, 158 (61%) participants were boys and 100 (39%) participants were girls. The ages of the participants ranged from 3.6 to 16.0 years ($M=8.5$ years). Of the 258 children, 94 (36%) had ADHD, 73 (28%) had a combined ID and physical disability, 42 (16%) had an ASD diagnosis, 22 (9%) participants were motor disabled, and 16 (6%) participants had ID (i.e., 51% had a mild intellectual impairment, 36% were moderately intellectually impaired, 10% had a severe intellectual impairment, and 3% were profoundly intellectually impaired). The remaining disorders mentioned included ODD (6%), anxiety (5%), non-PDD developmental disability (2%), learning disability (1%), and fragile X syndrome (<1%).

Three studies reported the child's disorder specification related to ED (Eliezer et al., 2021; Kaye & Palmer, 2010; van Laecke et al., 2010). Of the 225 participants related to the three studies, 91 (40%) participants had DUI, 41 (18%)

children had DUI and nocturnal enuresis (NE), 104 (46%) participants had NE, 75 (33%) children had urgency, 66 (29%) reported frequency, and 3 (1%) children had bladder and bowel dysfunction. Smith et al. (2000) reported that one of the three cases had nocturnal enuresis and no specifics were reported regarding the other two children. Hanney et al. (2013) did not report specific incontinence disorders.

Dependent Variable

As indicated previously, Smith et al. (2000) did not report about the dependent variable measured during the study. Two studies reported their findings using the ICCS guidelines regarding percentage of symptom reduction (Eliezer et al., 2021; Kaye & Palmer, 2010). Symptom reduction was measured by obtaining the post-void residual (PVR), the Akbal score (Eliezer et al., 2021), and Pediatric Incontinence Quality of Life (PinQ) score (Eliezer et al., 2021; Kaye & Palmer, 2010). Van Laecke et al. (2010) measured the maximum voided volume, fluid intake quantity, questionnaire scores, and urinary incontinence. Solely one study measured frequency and latency data regarding the following target behaviors: successes, accidents, accident/success conversions, and initiations (Hanney et al., 2013).

SU Components

According to previous studies, SU interventions should include the following components (Schäfer et al., 2018): (1) disorder specification, (2) instructions to establish better micturition, (3) guidelines for fiber and fluid intake, (4) documentation of all symptoms, and (5) a continuous monitoring by the treatment team.

The range of SU components utilized during the included studies varied from one component (i.e., instructions to establish better micturition) by Smith et al. (2000) to all five components by Eliezer et al. (2021) and van Laecke et al. (2010). Kaye and Palmer (2010) utilized two of the five components (i.e., disorder specification and instructions to establish better micturition), while the remaining study (Hanney et al., 2013) applied four out of the five components, as no information was provided regarding the disorder specification.

Another important finding is that the studies had a different execution regarding some of the components. Eliezer et al. (2021) gave a disorder specification by providing a detailed history regarding the child's incontinence, completed an examination and uroflow, and obtain PVR, Akbal, and PinQ scores. Kaye and Palmer (2010) executed the disorder specification by completing a physician examination, renal and bladder ultrasonography with PVR, urine volume measurement, urinalysis, uroflowmetry with EMG, and PVR. In addition, the treatment of VD was explained. Van

Table 2 Summary of included articles

Citation	Participant characteristics	SU component(s)	Parental involvement	Intervention procedures	Intervention effectiveness
Eliezer et al. (2021)	Children: <i>N</i> = 39 (27 boys and 12 girls); <i>M</i> age = 10.3 years; ADHD (<i>n</i> = 34), ASD (<i>n</i> = 15), ODD (<i>n</i> = 15), anxiety (<i>n</i> = 14); DUI alone (<i>n</i> = 5), monosymptomatic nocturnal enuresis (<i>n</i> = 10), non-monosymptomatic nocturnal enuresis (<i>n</i> = 14), bladder and bowel dysfunction (<i>n</i> = 3)	Disorder specification Instructions micturition Documentation symptoms Monitoring team Guidelines for fluids	Not mentioned	Response assessed at 3 month visit: children with no response classification received combination therapy – adjunct specific urotherapy or pharmacotherapy	Complete response (urotherapy = 9 and adjuncts = 4), partial response (urotherapy = 3 and adjuncts = 7), no response (urotherapy = 5 and adjuncts = 4)
Hanney et al. (2013)	Children: <i>N</i> = 30 (23 boys and 7 girls); <i>M</i> age = 5.5 years; ASD (<i>n</i> = 25), PDDNOS (<i>n</i> = 1), non-PDD developmental disability (<i>n</i> = 4); incontinence disorder not specified	Instructions micturition Guidelines for fluids Documentation symptoms Monitoring team	Treatment protocol gradually transferred to parents Instructions, modeling and performance feedback to implement protocol by parents	Treatment protocol gradually transferred to parents Instructions, modeling and performance feedback to implement protocol by parents	Full continence (<i>n</i> = 14) <i>M</i> days of treatment 14.4 days, schedule-dependent continence (<i>n</i> = 3), partial continence (<i>n</i> = 3)
Kaye and Palmer (2010)	Children: <i>N</i> = 75 (39 boys and 36 girls); <i>M</i> age = 7.8 years; ADHD (<i>n</i> = 60); DUI (<i>n</i> = 75), urgency (<i>n</i> = 75), frequency (<i>n</i> = 66) and sleep enuresis (<i>n</i> = 65)	Disorder specification Instructions micturition	Stating parental expectations Ask if parents were mentally or logistically able to commit Individualized treatment based on parental input	- Anticholinergic pharmacotherapy contingent upon successful behavior modification + continence not yet achieved + normal uroflow findings - Weekly or bi weekly animated biofeedback by DV	Complete resolution of symptoms (<i>n</i> = 17) with behavior therapy (<i>n</i> = 7) with anticholinergics (<i>n</i> = 2) and biofeedback (<i>n</i> = 8) Partial symptoms (<i>n</i> = 30) with behavior therapy (<i>n</i> = 9) with anticholinergics (<i>n</i> = 15) and biofeedback (<i>n</i> = 6) No improvement (<i>n</i> = 9)
Smith et al. (2000)	Children: <i>N</i> = 3 (3 boys), range 8 to 12 years; ASD (<i>n</i> = 1), learning disability (<i>n</i> = 3), fragile X syndrome (<i>n</i> = 1); nocturnal enuresis (<i>n</i> = 1)	Instructions micturition	Not mentioned	- Positive reinforcement - shaping - fading - stimulus control transfer	Successful stimulus control transfer (<i>n</i> = 2), treatment time 54 weeks and 51 days. Defecation in the toilet without nappy (<i>n</i> = 1 with ASD), treatment time 130 weeks

Table 2 (continued)

Citation	Participant characteristics	SU component(s)	Parental involvement	Intervention procedures	Intervention effectiveness
Van Laecke et al. (2010)	Children: N = 111 (66 boys and 45 girls), M age = 9.1 years; motor disabled (n = 22), mentally disabled (n = 16), intellectually and physically disabled (n = 73); DUI and nocturnal enuresis (36.9%), DUI (9.6%), nocturnal enuresis (13.5%)	Disorder specification Instructions micturition Guidelines for fluids Documentation symptoms Monitoring team	Parental advice given on fluid quality and quantity	- After 6 weeks: medical treatment, anticholinergics and a wetting alarm - After 3 months with no change: videourydynamics	Complete continence (67%), independence of urinary incontinence decreased from 46.9% to 18.4% Continence (n = 11) with combined anticholinergics and fluid intake, not effective (n = 2) with daytime alarm, successful (n = 2) with nocturnal enuresis and bedwetting alarm

Laecke et al. (2010) provided the maximum voided volume, IQ, mental age, verbal capacity, mobility and postural stability, functional autonomy, clinical examination, uroflowmetry, and PVR.

Regarding the instructions to establish better micturition, Eliezer et al. (2021) gave a review of symptoms and utilized a bladder diary for the children. Furthermore, they gave instructions which included an explanation of how bladder filling and emptying occurs and factors that make it more difficult. They advised children to aim for bladder emptying every 3 h during the day and gave specifics regarding the voiding position for boys (i.e., standing with pants down and making a hissing sound) and girls (i.e., seated with feet flat or elevated and leaning forward). Hanney et al. (2013) used an intensive and progressive sitting schedule, differential reinforcement for urination, urine sensor and alarm, positive practice for accidents, and communication training. Kaye and Palmer (2010) started with behavior modifications including timed voiding, bowel management, and positive reinforcement charts. Children started with small compliance goals and progressed to more difficult goals. Smith et al. (2000) used positive reinforcement, shaping (gradually increasing the proximity to the toilet), fading (reducing the presence of the nappy), and a stimulus control transfer. Van Laecke et al. (2010) described using a daytime alarm, wetting alarm, and the toilet that was adapted to the individual needs of the children to optimize a stable toilet position and maximum pelvic floor relaxation.

The execution of guidelines for fiber and fluid intake entailed increased fluids (Hanney et al., 2013) and a fluid schedule, which was individualized based on giving children a target of drinking 30–50 ml/kg per day, and children were advised to avoid drinking within 2 h of bedtime. To avoid irritants, they advised drinking solely water and milk

(Eliezer et al., 2021). Eliezer et al. (2021) also provided recommendations to increasing fiber in the child's diet with grains, cereal, fruit, and vegetables. They advised adding Metamucil and laxatives if there was no improvement.

The documentation of symptoms was completed by providing a bladder diary (Eliezer et al., 2021), or a description of the maximum voided volume, fluid intake quantity, and degree of incontinence (Van Laecke et al., 2010). Furthermore, Hanney et al. (2013) utilized frequency and latency data on successes, accidents, accident/success conversions, and initiations. Kaye and Palmer (2010) solely indicated that they registered during the first visit, but no further details were provided.

Finally, the monitoring by the treatment team was described by Eliezer et al. (2021) as an initial visit, a 3-month visit, and a 6-month visit. The child's responses were assessed at the 3-month visit. Children with a "no response" classification received combination therapy, adjunct specific urotherapy, or pharmacotherapy. Hanney et al. (2013) described that they transferred the procedures to parents on the first treatment day and provided phone consultation during the following weekend. The implementation at school happened the following Monday, while all therapeutic support was eventually faded as the clients progressed. Services were provided by 20 graduate students. Van Laecke et al. (2010) described that initially the study consisted of 6 weeks with regularly repeated uroflows and bladder scans. Furthermore, additional treatment procedures were considered after a minimum of 6 weeks of treatment. After 3 months with no change, videourodynamics were performed. Kaye and Palmer (2010) did not specify if and how the treatment team monitored the child, besides mentioning the initial visit. The treatment team consisted of a pediatrician, psychiatrist, and/or

psychologist. In addition, Smith et al. (2000) also provided no details regarding continuous monitoring of the treatment team, except stating the involvement of an assistant psychologist.

Additional Intervention Procedures

Three out of the five articles started with SU interventions, and additional interventions were implemented after a specific timeframe if SU alone was not sufficient in obtaining the desired result regarding the child's incontinence (Eliezer et al., 2021; Kaye & Palmer, 2010; Van Laecke et al., 2010).

First of all, Eliezer et al. (2021) indicated that they completed a review of symptoms at the 3-month visit. Based on the child's response at that time, the child's progress was maintained on urotherapy alone or adjuncts were considered. Combination therapy was based on the underlying bladder disorder, availability of treatment and patient, and the preferences of the caretaker(s). The adjunctive therapies included desmopressin, alarm training, physiotherapy, anticholinergics, and alpha blockers. Another evaluation took place at the 6-month visit.

Next, Kaye and Palmer (2010) indicated that they did not use a standard treatment approach. Instead they started with applying behavior modification (i.e., timed voiding, bowel management, and positive reinforcement charts) and moved to more advanced goals as the child progressed. If behavior modifications were successful in the sense of performance, but continence was not yet achieved (with normal EMG-PVR findings), anticholinergic pharmacotherapy (i.e., oxybutynin 0.1 mg/kg 3 times daily) was given. Animated biofeedback was given to children demonstrating DV on the EMG-PVR.

Finally, Van Laecke et al. (2010) indicated that after a minimum of 6 weeks of treatment, medical treatment, anticholinergics, and a wetting alarm were considered as additional therapies. After 3 months with no change, vid-eourodynamics were considered.

The remaining two articles described procedures related to specific behavioral modifications. Smith et al. (2000) used positive reinforcement, shaping, fading, and stimulus control transfers, while Hanney et al. (2013) utilized intensive and progressive sitting schedules, increased fluids, differential reinforcement of urination, urine sensor or alarm, positive practice for accidents, and communication training.

Parent Involvement During SU

Hanney et al. (2013) stated that the implementation of the treatment protocol was gradually transferred from the graduate students to the parents and school staff during the first

day of treatment at the clinic. In addition, parents and school staff implemented the treatment protocol during all subsequent days in the home or at school. In order to ensure that parents and school staff were able to implement the procedure, parents and school staff were trained on implementing the protocol by the treatment team using instructions, and modeling and performance feedback was given.

Kaye and Palmer (2010) involved parents during the first treatment sessions by stating clear expectations for treatment by explaining that the treatment of incontinence is a process (especially in children with ADHD), and therefore no immediate results should be expected after one visit or with one sole intervention. They clearly asked parents if they were willing to continue treatment and indicated that if parents were mentally or logistically unable to commit at that time, they could opt to return at a later time. Furthermore, treatment was individualized based on the input of the parents, and they concluded that parent involvement (as well as the involvement of a pediatrician and psychologist) was essential for successful treatment.

Van Laecke et al. (2010) indicated that parents were involved during treatment by giving parents and caretakers guidelines on fluid quality and quantity. The remaining articles (Eliezer et al., 2021; Smith et al., 2000) did not mention if and how they involved parents during treatment.

Intervention Effectiveness

An updated report was released by the ICCS regarding the use of certain terminology and definitions of treatment outcomes (Austin et al., 2016). According to these guidelines, researches should report initial study findings as follows: (a) no response, < 50% symptom reduction; (b) partial response, 50–99% symptom reduction; (c) complete response, 100% reduction. It is important to state that one article reported the findings in accordance with these guidelines which made it possible to compare this study based on a uniform standard (Eliezer et al., 2021). Eliezer et al. (2021) stated the following study outcomes after the 6-month visit: (a) nine children (24%) had a complete response to SU alone and four (11%) with adjuncts, (b) three children (8%) had a partial response with SU alone and seven (19%) with adjuncts, and (c) five children (14%) with SU alone had no response, while four children (11%) with adjuncts had no results. This indicated that at the 6-month follow-up, 32% of the children had a complete/partial response to SU alone. Type of underlying behavioral disorder did not influence the outcome.

Kaye and Palmer (2010) also reported their findings according to the ICCS criteria at the time; however, these requirements have been updated since the publication of that specific study. The results of that study according to the guidelines at the time of publication were as follows:

(a) of the 56 patients with ADHD, 17 (30%) achieved a complete response—with behavior therapy alone ($n=7$), with anticholinergics ($n=2$), and biofeedback ($n=8$); (b) 30 patients with ADHD (54%) achieved partial improvements—with behavior therapy alone ($n=9$), with anticholinergics ($n=15$), and biofeedback ($n=6$); finally, (c) 9 of 56 patients with ADHD (16%) had no results even with all three interventions (behavior therapy, anticholinergics, and biofeedback). This entails that at the end of the study, 29% of the children had a complete/partial response to SU alone. The authors suggests that noncompliance by the parents may have been reduced by asking parents if they were mentally and logistically able to commit to treatment and therefore the success of treatment might have been affected by the patient selection bias.

Van Laecke et al. (2010) reported the outcome findings based on the ICCS definitions regarding continence and determined the statistical significance in continence patterns at the start and end of the study. At the end of the study, 69 children (67%) were completely continent (day and night). Fourteen children (14%) remained urinary incontinent (day and night), while five children (5%) had DUI and fifteen children (15%) had NE. No significant differences in continence results were found among the study subgroups (i.e., mentally disabled, motor disabled, and mentally and motor disabled). As this study additionally looked at adequate fluid intake, it was found that 66% of the children drank at least 25% less than the physiologically necessary quantity.

Hanney et al. (2013) described the findings of the study in terms of the level of continence: (a) full continence— independent initiation of successful voids with no accidents, (b) partial continence—inconsistent successful voids, (c) schedule-dependent continence—successful voids based on prompts (every 3–4 h), and (d) unknown—insufficient data. Of the initial 30 participants, 14 children with ASS (47%) obtained full continence with an average of 14.4 days of treatment, 3 children with ASS (10%) obtained partial based continence, and another 3 participants with ASS (10%) were schedule-dependent continent at the end of the study. Overall, for all participants, the mean time off the toilet steadily increased during treatment.

Finally, the results from Smith et al. (2000) should be interpreted with caution as the critical appraisal of this article was insufficient. Individual findings of the three cases were reported in the study. Child 1 completed the program in 54 weeks and at the end of treatment no soiling occurred at home or at school. Child 2 was in treatment for 130 weeks at which point the child was able to defecate on the toilet without a nappy. Finally, child 3 completed the program in 51 days when the child was able to urinate in the toilet without a nappy. Important to note is that this study suggested that children with learning disabilities might be more prone to treatment resistance which may explain the need for a

longer treatment time for these children. The hypothesis is that this resistance to change is not solely due to simply failure to learn or a skill deficit; therefore, other factors should be considered during treatment (e.g., anxiety).

Discussion

This systematic literature review was conducted in order to obtain more information relating to the effectiveness of SU interventions for children with NDs, to identify which protocols and procedures are being utilized, to attain information regarding the role of parents during treatment, to determine the current base of evidence regarding SU for children with NDs, and to propose suggestions for future research. The systematic review identified five studies, indicating that research related to SU for children with NDs has been very limited. Despite a growing body of literature related to the effectiveness of standard urotherapy, articles specifically related to the SU and children with NDs are sparse.

A mixed-method literature was required in order to include both quantitative and qualitative studies. In addition, the inclusions criteria utilized for this review were broad (e.g., at least one component of standard urotherapy was utilized during treatment instead of all five components (Schäfer et al., 2018)) with the aim of including as many relevant articles as possible. Therefore, some of the included articles arguably used treatment procedures that do not fully reflect SU. For example, Smith et al. (2000) utilized solely one SU component, without clear baseline data, which is potentially insufficient to be classified as standard urotherapy as described in the standardization of terminology in the ICCS report (Austin et al., 2016). However, the low methodological quality of one of the included articles and perhaps limited utilization of all the SU components of some of the other articles do state the necessity of further research regarding the effectiveness of SU for children with NDs, especially considering the high prevalence rates of incontinence related to this target population and significant impact of incontinence on the child's overall health (von Gontard et al., 2021; von Wendt et al., 1990). Nevertheless, these articles were included as part of the limited body of research articles pertaining to NDs and incontinence (treatment) as they do provide essential information regarding some of the components utilized during SU treatment for children with NDs. However, this literature review does amplify the need for more research related to this topic.

SU for Children with NDs

The results of this literature indicate that SU could successfully be implemented for children with a wide variety of

NDs (e.g., ADHD, ASD, physical disability, and ID). Of the NDs represented in the studies, children with ADHD were studied more often compared to children with ASD and ID. However, ADHD is a more common ND and therefore more likely to be represented higher within the research population. It is suggested in previous literature that urotherapy should be adjusted to the needs of children with NDs. Specific helpful strategies are given for the three most common NDs during incontinence treatment (von Gontard et al., 2021). The results of this literature review indicate that in the clinical practice, SU is implemented as a first-line treatment, including one or more follow-up appointments. During these appointments, a review of symptoms is often times conducted in order to determine if combination therapies are required for the individual. The results suggest that these combination therapies could increase the success rate of therapy if SU alone is not effective at these follow-up appointments. However, there are many differences stated in the identified articles regarding the SU components utilized and the way these procedures were executed. The ICCS provides specific guidelines regarding SU and therefore it is recommended that these guidelines should be followed more closely in the clinical practice (Schäfer et al., 2018; Gontard et al., 2021).

In addition, very limited information has been found regarding the specific strategies used for children with NDs during SU within the clinical practice. The strategy most often listed in the included articles states the importance of increased fluid intake during treatment. Not only does this increase the amount of learning opportunities for children, it may also benefit bladder sensation. Furthermore, the use of (differential) reinforcement is listed in some of the articles as a helpful strategy during treatment. This entails access to preferred items, such as TV, social praise, and toys/edibles, or the avoidance of an undesired stimulus such as escape from the toilet upon successful elimination. Other strategies listed included communication training, stimulus control transfers, shaping, fading, positive practice, intensive and progressive sitting schedules, parental involvement, and modifications of training intensity/duration (varying from 14.4 days to one report of treatment lasting 130 weeks). Compared to the all the helpful strategies listed in the ICCS overview (von Gontard et al., 2021), the strategies utilized in the clinical practice as reviewed by this literature review are limited. Therefore, it is beneficial for practitioners to utilize the tools available as listed in the ICCS overview (von Gontard et al., 2021).

Furthermore, literature related to SU in children with NDs suggested that it would be beneficial to utilize an interdisciplinary approach (Caldwell et al., 2018; von Gontard, 2021). The included articles provided very limited information regarding the use of an interdisciplinary approach. Solely one article mentioned that the treatment team consisted of a

pediatrician, psychiatrist, and/or psychologist. Therefore, it would be beneficial for the clinical practice to look into possibilities of providing a more interdisciplinary approach of treatment for this population. Future research should determine the effectiveness of this interdisciplinary approach of treatment in children with NDs.

Limitations and Future Research

This systematic review found evidence supporting positive outcomes for SU treatment for children with NDs. The two articles reported in accordance with the ICCS treatment outcome suggestions and indicated that SU alone provided a complete/partial response in 30% of the children with NDs. The remaining three articles also reported positive outcomes based on other criteria (e.g., toileting success, initiations, transfer of stimulus control, schedule-dependent continence), indicating that 59% of the children achieved full continence. Furthermore, the evidence suggests that most children will show improved symptoms even if full continence was not obtained, which further outlines the need for SU treatment within this population. However, these results are limited due to the small amount of included articles and lack of randomized controlled trials. Therefore, more research is needed to draw more firm conclusions about the outcome of SU for children with NDs.

As not all children seem to benefit from SU alone, more research is needed to determine why and which children with NDs do not benefit from SU. It might be helpful to differentiate between different NDs in order to obtain a better understanding relating to the specific needs of these children during SU treatment. Also, considering the fact that most articles report on ADHD and ASD, limited information is available regarding SU and the other NDs.

The evidence found suggests that it is beneficial for this population to individualize SU, such as increasing compliance for children with ADHD by clearly stating treatment expectations and asking parents (when applicable) if they are mentally and logistically able to commit to treatment at that time. Furthermore, future research and clinicians within the clinical practice could also individualize SU by focusing on the specific needs of children with different NDs in order to tailor SU treatment specific to the needs of each child. For example, the included articles state very different treatment times, indicating that some children might show resistance to change and therefore might require more therapy time compared to typically developing children. Research needs to determine which duration, frequency, and intensity of treatment is most suitable for this specific population as the review indicated that treatment times vary greatly (from 14 days to 130 weeks of treatment). In addition, specific behavioral

strategies could contribute to the success of treatment and should be implemented in the clinical practice in order to tailor therapy to the needs of the individual child.

Another way to individualize therapy might be to utilize combination therapies or adjuncts (e.g., SU combined with biofeedback, alarm therapy, anticholinergic therapy). This literature review found evidence for the use of adjunct therapies when SU alone is not sufficient in providing the desired results at a specific evaluation point. This again suggests the need of more individualized SU therapy for children with NDs adapted to the specific needs of each child. However, more research needs to be completed in order to determine at which point in time these adjunct therapies should be added to SU alone. In addition, future research should determine which adjunct therapies are effective for the individual child (e.g., based on specific NDs or IQ) as the included articles varied greatly regarding the timeframe of adding adjunct therapies, varying from implementing them at the start of treatment to adding adjuncts at the 3-month visit.

Furthermore, the role of the child and parent(s) during SU has been described in the literature for typically developing children as SU focuses highly on the responsibilities of the individual child and their contribution to treatment. However, these roles might be different for children with NDs and their parents based on the specific skills repertoire of the child. For example, parents might have different or additional obligations during treatment as children with lower cognitive abilities require more parental support (e.g., during data collection, completing diaries/charts, or when shaping desired behaviors). Some articles mentioned the involvement of parents during treatment; however, limited information was given. Some of the limited suggestions given in the literature involved the gradual transition of parents' involvement by providing instructions, modeling, and feedback. One of the articles even stated that the role of parents was crucial for treatment success. Furthermore, it was suggested that compliance could be increased by clearly stating expectations of treatment to parents. Future research should focus on the specific role of parents of children with NDs and how the treatment team could best assist parents during this process in order to increase treatment outcomes.

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Declarations

Conflict of Interest The authors declare no competing interests.

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Authors and Affiliations

Maayke van Galen^{1,2}  · Bibi Huskens² · Alexander von Gontard^{3,4} · Robert Didden^{1,5}

¹ Behavioural Science Institute, Radboud University, Nijmegen, The Netherlands

² SeysCentra, De Horst 12, 6581 Malden, The Netherlands

³ Governor Kremers Center, Department of Urology, Maastricht University Medical Centre, Maastricht, The Netherlands

⁴ Psychiatric Services Graubünden (PDGR), Outpatient Services for Child and Adolescent Psychiatry, Chur, Switzerland

⁵ Trajectum, Zwolle, The Netherlands