

Quality of Life in Children with Spina Bifida: A Cross-Sectional Evaluation of 102 Patients and their Parents

Zegers SHJ^{1*}, Houterman S², Uiterwaal CSPM³, Winkler-Seinstra PLH⁴, Kimpen JLL⁵ and de Jong-de Vos van Steenwijk CCE⁶

¹Pediatrician, Department of Pediatrics, Máxima Medical Center, Veldhoven, Netherlands

²Epidemiologist, Catharina Hospital, Eindhoven, Netherlands

³Clinical Epidemiologist, Julius Center for Health Sciences and Primary Care, University Medical Center Utrecht, Netherlands

⁴Pediatric Researcher, Wilhelmina Children's Hospital, University Medical Center Utrecht, Netherlands

⁵Professor in Pediatrics, Wilhelmina Children's Hospital, University Medical Center Utrecht, Netherlands

⁶Pediatrician, Wilhelmina Children's Hospital, University Medical Center Utrecht, Netherlands

Abstract

Background: In the so-called SPIN UTI study on 176 patients with spina bifida and clean intermittent catheterization (CIC) we have studied the influence of antibiotic prophylaxis (AP) on the number of yearly urinary tract infections (UTIs) and bacterial resistance to commonly used antibiotics. We have proven that stopping AP increased the number of yearly UTIs only slightly and clinically irrelevant patients would have to take AP for two years to prevent one extra non-febrile, non-scarring UTI. We have also shown that bacterial resistance improved over time when AP was stopped.

In this article we study the influence of the recommended discontinuation of AP on Health Related Quality of Life (HRQL) in spina bifida.

Methods: Participating children aged 4-16 years and their parents were asked to fill out the validated KINDL-R questionnaire, combined with specific spina bifida questions. Scores in study participants were compared with healthy controls. Also, the impact of AP, gender, level of spinal lesion, hydrocephalus, methods of catheterization and defecation, mobility and level of schooling on HRQL was evaluated.

Results: One hundred and two of the 133 eligible children (77%) filled out the KINDL-R. Patients and their parents had significantly lower HRQL scores than healthy controls ($p < 0.001$). Parents had significantly lower scores than their children, especially in the psychological domains. Severity of co-morbidity was not associated with differences in HRQL. There were no differences between children who continued and stopped AP.

Conclusions: HRQL scores are lower in spina bifida patients compared to healthy children. Parents have even lower scores, especially in the psychological domains. Stopping antibiotic prophylaxis does not adversely affect HRQL, and should therefore be pursued to improve bacterial resistance patterns in spina bifida patients.

Keywords: Health related quality of life; Spina bifida; Antibiotic prophylaxis

Introduction

Despite periconceptional administration of folic acid and prenatal ultrasound screening, spina bifida is the second most common congenital birth defect [1]. Due to genetic and environmental factors, prevalence varies throughout the world, with approximately one in every two thousand live births in the Western World [2]. After the innate postnatal surgical morbidity, there is a lifelong burden of neurological, urological and orthopedic consequences and severely disabling emotional and behavioral problems [3-6].

In many studies the Health Related Quality of Life (HRQL) was assessed in children with spina bifida [3,6-12]. A wide variety of HRQL questionnaires has been developed, addressing the psychological and physiological aspects of various domains of everyday life activities, either with or without specific spina bifida related questions. Most studies show that parental expectancy, patient mobility and urinary continence are the most important factors to predict HRQL in patients with spina bifida [6,9,11].

We recently conducted the so-called SPIN UTI randomized controlled trial to assess whether stopping antibiotic prophylaxis (AP) in 176 patients with spina bifida on clean intermittent catheterization (CIC) influenced the number of urinary tract infections (UTIs) [13]. Stopping prophylaxis increased the number of yearly UTIs only slightly.

Patients would have to take prophylaxis for two years to prevent one extra non-febrile, non-scarring UTI. Therefore it was concluded that most spina bifida patients on CIC can safely discontinue AP. Bacterial resistance is common in every day AP use (REF). In our study on bacterial resistance patterns in continuing and stopping AP, bacterial susceptibility for commonly used antibiotics improved when AP was stopped. Discontinuation of AP should therefore be pursued to improve bacterial susceptibility for antibiotics as long as this has no negative effect on the number of UTIs and perceived HRQL in spina bifida patients on CIC. Our aim in this study is to investigate the HRQL in our cohort of 133 eligible children, and whether stopping AP either increased HRQL due to less daily medication or decreased HRQL due to loss of perceived antibiotic protection.

***Corresponding author:** Zegers SHJ, Máxima Medical Center, Department of Pediatrics, P.O. Box 7777, 5500 MB, Veldhoven, Netherlands, Tel: 0031 40 8888270; Fax: 0031 40 8889609; E-mail: b.zegers@mmc.nl

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Method

Study population

From 2005 through 2008, 176 patients with spina bifida on CIC were included in the SPIN UTI study and randomized to either continue or stop daily AP; primary outcome was the number of urinary tract infections over an 18 month study period. Details and results of the study are described elsewhere [13]. Out of the 176 participants, 133 were between age 4 and 16 years old and therefore eligible for the HRQL sub-study.

Procedure

At the end of the study period, the 133 eligible patients and their parents were asked to fill out the validated KINDL-R questionnaire [12]. KINDL-R consists of two separate lists: one list with general questions on six domains (physical well-being, emotional well-being, self-esteem, family, friends and school), each domain containing four specific questions, and one chronic disease module with extra questions for the parents. We also added the KINDL-R list of 37 specific spina bifida related questions for parents [12]. The first two lists were validated for both chronic disease and children with spina bifida, with 1501 healthy German primary school children as historical controls. The added specific spina bifida questionnaire has not been validated yet. The results were compared with the results of previous studies of HRQL observations in both healthy controls and spina bifida children and their parents [5,11,14,15]. The answers to the questions were calculated into a combined total HRQL score and scores per domain.

Demographic data and current functional status

To determine the influence of gender and severity of disability in the participating patients, gender, level of spina bifida lesion, manner

of micturation and defecation, mobility and level of schooling were also evaluated, using regression analysis.

Statistical analysis

First, general characteristics of the patients were tabulated by intervention (stop or continue antibiotic prophylaxis). Only a subgroup of children in the trial was age-eligible for KINDL-R (133 of 176) of whom 77% did participate in this quality of life study. Therefore, the randomization principle was violated and we tested for group differences in general characteristics using Chi-square tests or Fisher's exact tests where appropriate. Subsequently, differences in HRQL scores between the intervention groups were tested for both patients and parents with independent samples T-tests. Univariable linear regression analyses were used to correct for the effect of differences in general characteristics that might confound the association between intervention and HRQL. Additionally, we intended to test for differences between patients' and parents' perceived quality of life. This was done in the total patient group, and we intended to use paired t-tests in case of no treatment effect or general linear models in case of a treatment effect on HRQL. As all analyses of subdomains of HRQL are part of one overriding hypothesis, we did not account for multiple testing. All statistical analyses were performed using SPSS, version 19.0 (IBM Company).

Results

Subject responses and baseline characteristics

One hundred and two (77%) of the 133 eligible children and their parents filled out the KINDL-R questionnaire at the end of the trial period between 2006 and 2010. Patient characteristics are described in Table 1. There were no significant differences in the relevant variables

		Total group		Continue prophylaxis		Stop prophylaxis		p-value
		N=102		N=54		N=48		
		N	%	N	%	N	%	
Gender	male	45	44	25	46	20	42	0.64
	female	57	56	29	54	28	58	
Age	4-7 years	49	48	26	48	23	48	0.03
	8-12 years	23	23	17	32	6	13	
	12-16 years	30	29	11	20	19	40	
School	regular	65	64	37	69	28	58	0.98
	special	28	27	16	30	12	25	
	missing	9	9	1	2	8	17	
Level of lesion	thoracal	11	11	7	13	4	8	0.16
	lumbal	60	59	27	50	33	69	
	sacral	31	30	20	57	11	23	
Hydrocephalus	no	32	31	21	39	11	23	0.08
	yes	70	69	33	61	37	77	
Mobility	ambulatory	65	64	35	65	30	63	0.81
	wheelchair	37	36	19	35	18	38	
CIC	vesicostoma	13	13	7	13	6	13	0.94
	transurethral	89	87	47	87	42	88	
Defecation	spontaneous	23	22	14	26	9	19	0.41
	colon cleansing	69	68	36	67	33	69	
	manual evacuation	1	1	1	2	0	0	
	stoma	9	9	3	6	6	13	

CIC=Clean Intermittent Catheterization

Table 1: Patient characteristics of evaluated children with spina bifida on clean intermittent catheterization.

between the stopping and continuing group, with the exception of the division of children over 8 years of age in the stop and continuing group ($p = 0.03$).

Quality of life scores

Compared to healthy controls, both spina bifida children and their parents have significantly lower HRQL scores on every domain except disease, as well as in the total score (Table 2). Patients generally had higher HRQL scores than their parents (62.0 vs. 56.8, $p < 0.001$), specifically in the domains of disease ($p < 0.001$), emotional well-being ($p = 0.005$), self-esteem ($p < 0.001$), friends ($p = 0.002$) and school ($p = 0.02$). Regression analyses revealed no significant influence of gender, level of spinal lesion, presence of hydrocephalus, route of catheterization, manner of defecation or level of mobility and schooling on HRQL.

There was also no difference between the sub-domain HRQL scores of the group stopping daily AP and the group that continued AP in any specific domain (Table 3). Also, the total HRQL score, both in patients

(62.0 in stop group vs. 62.3 in continuing group, $p = 0.92$) and parents (57.2 vs 54.8, $p = 0.16$) did not differ significantly between stopping or continuing prophylaxis (Table 3).

Discussion

Spina bifida patients and their parents have lower HRQL scores than healthy controls, and parents have lower HRQL scores than their children. Severity of the spinal lesion and stopping daily AP for urinary tract infections in children with spina bifida on CIC has no significant effect on HRQL.

This is in accordance with prior studies [11,14], and may be explained by the fact that children appreciate life as they know it: their answers reflect current and short term social functioning, while they disregard potential future threats. Parents however tend to incorporate future outcome in their responses, and therefore have a less optimistic view of social status, relationships and employment possibilities of their offspring. Another explanation could be that children want to protect their parents by concealing their true feelings. Janse et al. showed that

	Parents		Children		Reference		p-value ¹
	mean	SD	mean	SD	mean	SD	
physical well-being	57.86	14.6	60.61	14.2	73.78	3.53	0.34
emotional well-being	60.38	12.3	67.88	11.5	81.64	1.76	0.005
self-esteem	45.85	8.65	59.15	13.5	63.91	3.75	< 0.001
family	60.00	11.6	63.21	13.8	81.18	3.81	0.22
friends	48.90	16.3	58.70	14.5	78.16	0.11	0.002
school	56.46	12.5	62.44	8.52	70.28	4.17	0.021
disease	41.88	15.9	63.04	9.15	62.54	1.94	<0.001
total score	56.76	6.13	61.97	8.52	75.94	1.98	<0.001
¹ estimated with paired t-test							

Table 2: HRQL scores in spina bifida children, their parents and healthy controls.

Parents	Continue		Stop		p-value ¹
	mean	SD	mean	SD	
physical well-being	56.94	15.00	59.24	12.15	0.41
emotional well-being	59.26	13.58	63.37	10.33	0.10
self-esteem	46.04	8.79	47.34	8.07	0.44
family	63.11	11.82	60.94	11.47	0.35
friends	49.53	14.94	52.20	14.36	0.39
school	55.33	12.49	58.60	9.72	0.17
kiddy parents	51.04	9.28	50.07	6.57	0.75
disease	41.15	15.75	37.54	13.42	0.39
spina	62.98	9.05	62.51	7.57	0.84
total score	54.83	7.88	57.24	7.16	0.16
Children	Continue		Stop		p-value ¹
	mean	SD	mean	SD	
physical well-being	60.20	14.89	61.80	14.06	0.70
emotional well-being	66.07	13.43	69.80	8.35	0.24
self-esteem	57.68	14.17	60.80	12.88	0.41
family	60.71	16.26	66.00	9.90	0.16
friends	56.61	16.67	61.74	10.72	0.21
school	63.26	9.72	63.10	8.44	0.95
disease	61.81	10.90	65.19	7.52	0.27
total score	62.25	8.89	62.03	7.53	0.92

¹ estimated with paired t-test

Table 3: Differences in HRQL scores in continuing and stopping antibiotic prophylaxis in spina bifida patients and their parents.

pediatricians tend to misunderstand perception of HRQL in patients' parents [15]. Therefore, systematic assessment of HRQL is needed in patients as well as their parents, as this may differ from the assumed HRQL by the health care professional. Proper assessment of HRQL thus contributes to a tailored psychosocial and somatic counseling by health care professionals treating spina bifida patients.

The level of the spinal lesion, presence of hydrocephalus, manner of both micturation and defecation, gender and level of mobility and schooling does not influence the HRQL scores in either patients or their parents. This is in accordance with previous studies [5]. Padua et al. found that patients with high disability have lower scores in physical domains, whilst patients with milder lesions have more emotional problems, due to their incontinence problems. Spina bifida itself is therefore the main factor contributing to perception of HRQL, and not so much the severity of the morbidity.

In our study, stopping AP neither improved nor worsened HRQL scores at the end of the study period. Although there is a slight increase in the incidence of UTIs in the stopping group, this increase was clinically irrelevant because every patient would have to use daily antibiotics for more than two years to prevent one extra non-febrile UTI [13]. Also, bacterial resistance for commonly used antibiotics improved in the months after stopping AP (REF). Thus, stopping daily AP does not only result in a decrease in medication consumption and an improvement of bacterial susceptibility to antibiotics: it can be done without decreasing the experienced HRQL in children with spina bifida.

Conclusion

Spina bifida patients have lower HRQL scores than their healthy peers. Parents have even lower scores, especially in the psychological domains. Stopping daily antibiotic prophylaxis in children with spina bifida on clean intermittent catheterization can be done safely to improve bacterial resistance for antibiotics without influencing their quality of life.

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