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Resection of the extravertebral portion of the filum terminale with posterior internal sphincterotomy, a surgical technique for managing chronic constipation and encopresis in children

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Abstract

Objective: Constipation is a frequent digestive complaint in children and can be refractory to treatment in approximately 30% of patients. Etiologies that are linked to this pathology include rectal hyporeflexia, achalasia of the internal anal sphincter, and neurological disorders such as tight filum terminale syndrome. For these conditions, the resection of the extra vertebral portion of the filum terminale and posterior internal sphincterotomy has shown potential benefits for management.

Materials and methods: We conducted a cross-sectional descriptive study between 2020 and 2021. We collected data from 219 children's medical records at Saint Petersburg State Pediatric Hospital, Russia, that fulfilled the diagnostic criteria and were managed surgically. We analyze their demographic characteristics and clinical outcomes.

Results: We found that 83.6% (n=183) of participants showed improvement, while 16.4% (n=36) experienced no improvement or complications. Patients in the 16-17 age group had the highest proportion of lack of improvement or complications, with 12.8% and the 1-5 age group exhibited a 100% improvement rate. Male patients exhibited a decreased probability of experiencing complications in the postoperative period.

Conclusions: Resection of the extravertebral portion of the filum terminale with posterior internal sphincterotomy has shown potential benefits in managing chronic constipation and encopresis in pediatric patients. However, further research is warranted to investigate the benefits and risks of this surgical technique.

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Introduction

Constipation is a frequent digestive complaint in the pediatric population, with a wide prevalence ranging from 0.7% to 29.6% (1). Research findings have shown that over 40% of pediatricians' office visits are attributed to this complaint, making it challenging for physicians to face this condition in their daily practice (2). Effective management depends on many factors, including a correct initial assessment, particularly when determining the cause (3). Chronic refractory constipation poses a challenge for approximately 30% of patients experiencing constipation (4). It can be linked to various underlying or coexisting conditions, with poor treatment compliance being a common factor. Additionally, conditions such as Hirschsprung's disease, celiac disease, cow milk allergy, primary hyperparathyroidism, and connective tissue disorders may be associated (4). Rectal hyporeflexia, resulting from structural abnormalities affecting rectal innervation in the parasympathetic center of the spinal cord, can also contribute to these etiologies (5). Another contributing factor is achalasia of the internal anal sphincter, where the lack of parasympathetic innervation at the internal anal sphincter hinders proper emptying during defecation (6,7). Additionally, neurological disorders such as Tight filum terminale syndrome can result in spinal flexion with a rigid filum terminale, stretching the nerves that innervate the rectum and bladder. This can lead to ischemia, causing degeneration of nerve elements and reduced conduction of nerve impulses (8,9). In contrast to chronic constipation, encopresis is a disorder characterized by involuntary stool loss caused by a stricture at the internal anal sphincter. The prevalence of encopresis varies across different stages of childhood, with higher rates observed among preschoolers, estimated to be around 1-3% (10).

In the case of chronic constipation and encopresis, a surgical procedure that aims to resect the extravertebral portion of the filum terminale has shown potential for management (11–13). This procedure objective is to release the tension exerted on the sacral plexus in the parasympathetic pathway (S2-S4) (8,14). Additionally, a posterior internal sphincterotomy is performed to alleviate the narrowness of the anal canal while preserving structures important for anal continence.

A surgical technique based on the patent issued by the Russian Federation, known as "Method of surgical treatment of chronic constipation and encopresis in children" (2019), number 2717216, developed by Dr. Kolesnikova N. G., Dr. Komissarov I. A., and Dr. Kovalev F. S, has shown potential benefits for managing chronic constipation and encopresis, demonstrating promising outcomes in treatment (15).

The aim of this study is to evaluate the efficacy and safety of a novel surgical technique, the resection of the extravertebral portion of the filum terminale combined with posterior internal sphincterotomy,

Table 1: Chi-square tests for diagnostic categories by age and gender

		Diagnosis			
Characteristics	Total n (%)	Achalasia of the internal anal sphincter n (%)	Encopresis n (%)	Tight Filum Terminale Syndrome n (%)	p-value*
Age (year)					
1 - 5	86 (39.3)	21 (24.4)	23 (26.7)	42 (48.8)	0.105
6 - 10	75 (34.2)	32 (42.7)	11 (14.7)	32 (42.7)	
11 - 15	24 (11)	12 (50)	3 (12.5)	9 (37.5)	
16 - 17	34 (15.5)	13 (38.2)	8 (23.5)	13 (38.2)	
Gender					
Male	92 (42.0)	26 (28.3)	21 (22.8)	45 (48.9)	0.623
Female	127 (58.0)	52 (40.9)	24 (18.9)	51 (40.2)	
Total	219 (100)	78 (35.6)	45 (20.5)	96 (43.8)	

* Chi-square test with statistical significance at <0.05.

in the management of chronic constipation and encopresis in pediatric patients.

Materials and methods

We conducted a cross-sectional descriptive study between 2020 and 2021. Prior to obtaining authorization and informed consent from parents or legal representatives, we collected data from 219 children's medical records at Saint Petersburg State Pediatric Hospital, Russia to analyze their demographic characteristics and clinical outcomes. We selected patients who met the inclusion criteria and were diagnosed with either chronic constipation refractory to treatment or encopresis. The causes of their chronic constipation were later subdivided into rectal hyporeflexia, achalasia of the internal anal sphincter, and tight filum terminale syndrome. They also fulfilled the criteria for surgical management and underwent surgical treatment involving resection of the extravertebral portion of the filum terminale, posterior internal sphincterotomy, and a combination of both. Patients were followed during a 6-month window. We categorized them based on their clinical outcomes and gastrointestinal transit tests. We observed two groups: patients who showed improvement and those who did not. Additionally, any complications that arose during this period were monitored and recorded. We utilized Microsoft Office™ Excel to collect the data, and the statistical analysis was performed using STATA version 17. The data analysis involved chi-square tests and logistic regression. A $p<0.05$ was considered statistically significant.

Results

The data analysis involved 219 patients, with females accounting for 58% (n=127) and males representing 42% (n=92) of the sample. The age distribution revealed the highest prevalence in the 1-5 year age group (39.3%, n=86), followed by the 6-10 year group (34.2%, n=75), 11-15 year group (11.0%, n=24), and 16-17 year group (15.5%, n=34). The most prevalent condition observed was Tight filum terminale syndrome, and neurogenic hyporeflective dysfunction of the rectum collectively accounting for 43.8% (n=96) of all cases, followed by achalasia of the internal anal sphincter, and encopresis with 35.6% (n=78), and 20.5% (n=45) respectively. Statistical analysis indicated no significant associations between age groups and diagnoses (p=0.105). Likewise, no notable gender-based differences were found in the distribution of diagnoses (p=0.623) Table 1.

The most frequent surgical procedure performed was resection of the extravertebral portion of the filum terminale with posterior internal sphincterotomy accounting for 53.4% (n=117) of cases. Posterior internal sphincterotomy was performed in 21% (n=46) of cases, while resection of the extravertebral

Table 2: Chi-square tests for surgical procedures by age and gender

		Surgical Procedures			
	Total	1.Extravertebral Resection of the Terminal Filum	2.Posterior internal sphincterotomy	3.Extravertebral Resection of the Terminal Filum + Posterior internal sphincterotomy of rigid terminal filum	
Characteristics	n (%)	n (%)	n (%)	n(%)	p-value*
Age (year)					
1 - 5	111 (50.7)	29 (26.1)	11 (9.9)	71 (64)	**<0.01**
6 - 10	43 (19.6)	10 (23.3)	13 (30.2)	20 (46.5)	
11 - 15	30 (13.7)	9 (30)	10 (33.3)	11 (36.7)	
16 - 17	35 (16)	8 (22.9)	12 (34.3)	15 (42.9)	
Gender					
Male	117 (53.4)	30 (25.6)	30 (25.6)	57 (48.7)	0.164
Female	102 (46.6)	26 (25.5)	16 (15.7)	60 (58.8)	
Total	219 (100)	56 (25.6)	46 (21.0)	117 (53.4)	

* Chi-square test with statistical significance at <0.05.

Table 3: Chi-square tests and Odds Ratio for surgical outcomes by age and gender

Characteristics	Total n (%)	Results: No improvement and/or Complications n (%)	Results: With improvement n (%)	p-value*
Age (year)				
1 - 5	98 (44.7)	0 (0)	98 (100)	**<0.01**
6 - 10	73 (33.3)	2 (27)	71 (97.3)	
11 - 15	9 (4.1)	0 (0)	9 (100)	
16 - 17	39 (17.8)	34 (872)	5 (12.8)	
Gender				
Male	113 (51.6)	17 (15)	96 (85.0)	**0.565**
Female	106 (48.4)	19 (179)	87 (82.1)	
Total	219 (100)	36 (16,4)	183 (83.6)	

* Chi-square test with statistical significance at <0.05.

portion of the filum terminale, was observed in 25.6% (n=56) of cases. Statistical analysis using Chi-square tests revealed a significant association between age groups and surgical procedures (p<0.01). The highest prevalence of extravertebral resection of the filum terminale was found in the 1-5 age group (64%, n=71), while the combination procedure was more common in the 16-17 age group (42.9%, n=15).

No significant gender-based differences were found in the distribution of surgical procedures (p=0.164). Both males and females underwent procedures 1 and 2 to a similar extent Table 2.

```
ogistic regression                      Number of obs     =        112
                                        LR chi2(7)        =     115,79
                                        Prob > chi2       =     0,0000
og likelihood =  -12,43283              Pseudo R2         =     0,8232
```

complic	Coef.	Std. Err.	z	P>|z|	[95% Conf.	Interval]
sex						
Hombre	-14,67013	770,8035	-0,02	0,985	-1525,417	1496,077
interv						
1 - 5 años	0	(empty)				
6 - 10 años	-58,17891	1538,304	-0,04	0,970	-3073,2	2956,842
11 - 15 años	0	(empty)				
16 - 17 años	0	(omitted)				
diag1						
Acalasia del esfinter anal interno	38,61306	1,70e+07	0,00	1,000	-3,34e+07	3,34e+07
diag2						
Encopresis	67,57464	1,70e+07	0,00	1,000	-3,34e+07	3,34e+07
diag3						
indrome de filum terminal rígido y disfunsión nerogénica hiporref..	11,06038	1,70e+07	0,00	1,000	-3,34e+07	3,34e+07
cirug1						
Resección extravertebral del filum terminal	2,534067	1088,524	0,00	0,998	-2130,935	2136,003
cirug2						
Esfinterotomía interna posterior	15,68403	770,8036	0,02	0,984	-1495,063	1526,431
cirug3						
esección extravertebral del filum terminal + esfinterotomía inter..	15,76799	770,8047	0,02	0,984	-1494,981	1526,517

Figure 1: Estimated logit regression model

Logistic regression — Number of obs = 112; LR chi2(7) = 115,79; Prob > chi2 = 0,0000; Log likelihood = -12,43283; Pseudo R2 = 0,8232

complic	Odds Ratio	Std. Err.	z	P>|z|	[95% Conf.	Interval]
sex						
Hombre	4,25e-07	,0003279	-0,02	0,985	0	.
interv						
1 - 5 años	1	(empty)				
6 - 10 años	5,41e-26	8,32e-23	-0,04	0,970	0	.
11 - 15 años	1	(empty)				
16 - 17 años	1	(omitted)				
diag1						
Acalasia del esfinter anal interno	5,88e+16	1,00e+24	0,00	1,000	0	.
diag2						
Encopresis	2,22e+29	3,79e+36	0,00	1,000	0	.
diag3						
Sindrome de filum terminal rígido y..	63600,41	1,08e+12	0,00	1,000	0	.
cirug1						
Resección extravertebral del filum ..	12,60467	13720,49	0,00	0,998	0	.
cirug2						
Esfinterotomía interna posterior	6478698	4,99e+09	0,02	0,984	0	.

Figure 2: Matrix of coefficients of partial regressions (betas) ODDS

From our findings on clinical outcomes, 83.6% (n=183) of participants showed improvement, while 16.4% (n=36) experienced no improvement or complications. We established a significant link between age groups and clinical outcomes ($p<0.01$). Notably, the 16-17 age group had the highest proportion of patients with no improvement or complications at 12.8% (n=5). Conversely, the 1-5 age group exhibited a 100% improvement rate (n=98) (Figure 1).

The odds ratio analysis reveals that males have a protective factor compared to females, indicating a higher probability of not experiencing complications. Similarly, patients aged 6-10 years have a protective factor. Patients diagnosed with encopresis have a higher probability of postoperative complications compared to other diagnoses. Resection of the extravertebral portion of the filum terminale alone has the highest risk, with an odds ratio of 12.60, of postoperative complications (Figure 2).

The logistic regression model employed in this study demonstrated statistical significance, with the Maximum Likelihood statistic (LR Chi2=115.79) surpassing the threshold of 3 and a p-value of less than 0.05. In terms of the impact of variables, gender was a significant factor. Male patients exhibited a decreased probability of experiencing complications in the postoperative period (-14.67), suggesting a protective effect compared to females. Age also played a role, with patients aged 6 to 10 years showing reduced chances of complications (-58.18). Diagnoses of encopresis were associated with a higher probability (67.57) of lack of improvement or complications, emphasizing the challenges faced by patients with this diagnosis.

Regarding surgical procedures, Resection of the extravertebral portion of the filum terminale alone and posterior internal sphincterotomy was found to have a higher incidence of poor improvement or complications.

Discussion

Resection of the filum terminale has been used to treat various conditions, including occult tight filum terminale syndrome, part of the tethered cord syndrome (16). Patients with this syndrome can exhibit a range of clinical manifestations, such as sensory-

motor disturbances and parasympathetic instability leading to fecal incontinence, chronic constipation, and bladder instability (16). Sectioning the rigid filum terminale has shown benefits for patients experiencing fecal incontinence and chronic constipation (17). A study by Wehby et al. demonstrated that this procedure resolved fecal incontinence in 56% of patients and showed improvement in 41% of those with tight filum terminale syndrome and bowel disturbances (17). Similarly, our results indicate an overall improvement in most patients (83.6%) after a 6-month follow-up period. While these findings are promising, further research is necessary to determine the efficacy, risks, and long-term outcomes of this procedure. There is limited scientific information available regarding the use of this procedure alone for the treatment of chronic refractory constipation. Therefore, caution and a thorough evaluation of patients who may benefit from this procedure are recommended, especially in the pediatric population. Currently, there is no clear indication for surgical management in cases of chronic constipation, and careful consideration is needed before proceeding with this intervention.

Conclusions

We observed a potential benefit in patients who underwent the resection of the extravertebral portion of the filum terminale with posterior internal sphincterotomy for the management of chronic constipation and encopresis in the pediatric population. The majority of patients experienced improvement after the procedure, but we found a relationship between minimal improvement and specific diagnoses before the surgery, such as encopresis.

Age and gender play significant roles in the outcomes, with younger age groups, particularly the 1-5 year group, showing higher improvement rates than other age groups. Early detection can play a role in improving management outcomes. On the other hand, gender influenced the results, with males having a lower likelihood of postoperative complications than females.

Furthermore, the choice of intervention affected clinical results. Notably, extravertebral resection of the filum terminale with posterior internal sphincterotomy was the most frequently performed procedure with better outcomes. However, further research is necessary to explore this procedure's effectiveness and potential risks.

Given these findings, a multidisciplinary approach is needed to manage chronic constipation refractory to treatment and encopresis, considering individual patient characteristics, including age, gender, and specific diagnoses. Additionally, further research is warranted to investigate the benefits and risks of this surgical technique, which may contribute to beneficial outcomes in children.

Conflict of interest

The authors report no conflict of interest.

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Ethical approval

The study was conducted in accordance with the Declaration of Helsinki and approved by the Local Independent Ethics Committee of the National Research Center.

Informed consent

Written informed consent was obtained from all individual participants and/or their gaurdians.

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Peer-review

Externally. Evaluated by independent reviewers working in at least two different institutions appointed by the field editor.

Data availability

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

Contributions

Research concept and design: **FAAV, VSEV**

Data analysis and interpretation: **VSEV, FAAV, JMM, JCM, PJCH**

Collection and/or assembly of data: **FAAV, VSEV**

Writing the article: **FAAV, VSEV**

Critical revision of the article: **VSEV, FAAV, JMM, JCM, PJCH**

Final approval of the article: **VSEV, FAAV, JMM, JCM, PJCH**

All authors read and approved the final version of the manuscript.

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