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Anorectal physiology and colonic motility in children with a history of tethered cord syndrome

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Abstract

Objectives: The understanding of the impact of tethered cord syndrome (TCS) on the physiology of the colorectal area is limited. Our aim was to describe anorectal and colonic motility in children with TCS and compare the findings to those of children with functional constipation (FC).

Methods: We conducted a retrospective review of children with TCS who had an anorectal manometry (ARM) performed at our institution from January 2011 to September 2023. We recorded demographics, medical and surgical history, clinical symptoms, and treatment at time of ARM, ARM findings (resting pressure, push maneuver, rectal sensation, rectoanal inhibitory reflex [RAIR], and RAIR duration), and the final interpretation of colonic manometry (CM) if performed. We identified age and sex-matched control groups of children with FC.

Results: We included 24 children with TCS (50% female) who had ARM testing (median age at ARM 6.0 years, interquartile range 4.0–11.8 years). All children had constipation at time of ARM. Nineteen children had detethering surgery before ARM was performed. No significant differences in ARM parameters were found between children who had detethering surgery before ARM and children with FC. Among the 24 children, 14 also had a CM performed (13/14 after detethering surgery). No significant differences in colonic motility were found between children with a history of TCS and children with FC.

Conclusions: Anorectal physiology and colonic motility are similar between children with a history of TCS and children with FC, suggesting that the underlying pathophysiology of defecatory disorders in children with and without history of TCS is similar.

KEYWORDS

anorectal manometry, colonic manometry, detethering surgery, functional constipation

1 | INTRODUCTION

More than 95% of children with constipation have functional constipation (FC), which has a worldwide prevalence of 9.5% in children.[1,2] However, in some children an underlying organic cause responsible for constipation can be found. Spinal cord abnormalities, including tethered cord syndrome (TCS), have been described as a potential cause of constipation.[3] While rare, with an estimated incidence of 0.25 per 1000

births, TCS is one of the most common pediatric spinal disorders and is characterized by an abnormal attachment of the spinal cord to surrounding tissues, typically the lower end of the spinal column.[4,5] The postulated pathological mechanism in patients with TCS is that spinal cord stretching due to restriction of spinal cord movement leads to reduced blood flow and subsequent prolonged ischemia.[6] The presentation of TCS in children encompasses a variety of conditions, including back pain, scoliosis, lower extremity sensorimotor changes, neurogenic bladder dysfunction, and bowel dysfunction.[7] Fecal incontinence and constipation can be daily problems for children with TCS even after detethering surgery. Our understanding of the impact of TCS on the physiology of the colorectal area is limited. Studies evaluating anorectal function in children with TCS are sparse, while data on colonic motility are, to our best knowledge, nonexistent.[8,9] Therefore the aim of this study was to describe anorectal and colonic motility in children with TCS and compare the findings to those of children with FC.

What is Known

- The understanding of the impact of tethered cord syndrome (TCS) on the physiology of the colorectal area is limited.
- Even after detethering surgery, fecal incontinence and constipation are commonly seen in children with TCS.

What is New

- No significant differences in anorectal manometry findings and colonic motility were found between children with a history of TCS and an age and sex-matched cohort of children with functional constipation.
- The underlying pathophysiology of defecatory disorders in children with and without a history of TCS might be similar and not related to differences in colonic motor function.

2 | METHODS

We conducted a retrospective chart review of children ≤18 years of age with TCS who had undergone testing at Nationwide Children's Hospital between January 2011 and September 2023. The Nationwide Children's Hospital Institutional Review Board approved the study protocol (STUDY00000932). Children with additional congenital anomalies or spinal cord abnormalities other than TCS were excluded. Data on demographic information, medical and surgical history, clinical symptoms and treatment at time of ARM, and the results of ARM and CM testing (if CM was performed as well) were collected. Recorded measures of ARM included study condition (awake or under general anesthesia), resting pressure (reported as a numerical value and categorized as low [<39 mmHg], normal [55 ± 16 mmHg], or high [>71 mmHg] based on the normative values from Benninga et al.[10]), presence or absence of the rectoanal inhibitory reflex (RAIR), and, if present, RAIR duration defined as the time between a drop >15% in the anal canal resting pressure and the pressure returning to the basal pressure value. For children who completed ARM testing while awake, additional data on maximum squeeze pressure, push (or bear down) maneuver results, and rectal sensation (balloon volume in milliliters at first sensation, urge to defecate, and discomfort) were collected. ARM studies were considered as normal if in all recorded manometric data no abnormalities were found. Recorded measures of CM included the final interpretation of the study, as explained below. After identifying children with TCS, we identified a matched control group of children with FC and no other significant medical history. We matched children based on age at time of ARM testing and sex. For the children with TCS who also had a CM performed, we identified a separate matched control group of children with FC and no other significant medical history and matched them based on age at time of CM testing and sex.

2.1 | Manometry testing

ARM studies were performed using a high-resolution solid-state catheter. When deemed possible, the procedure was performed with the patient awake, to assess rectal sensory thresholds during balloon inflations, as well as to evaluate squeeze and push maneuvers. CM studies were performed with either a high-resolution solid-state catheter (Unisensor AG) or a water-perfused catheter (Medical Measurement Systems). On the day before the study, children completed a bowel clean out with a combination of polyethylene glycol 3350 and stimulant laxatives. The following day, the manometry catheter was placed either using colonoscopy or fluoroscopic guidance. After a recovery period, manometric data were recorded during the fasting phase, postprandial phase, and after administration of stimulant laxatives, typically one or two doses of bisacodyl 0.2 mg/kg (up to 10 mg) and an additional dose of glycerin (10–20 mL based on the child's age and size) if high-amplitude propagating contractions (HAPCs) were not identified after bisacodyl. To classify a CM study as normal, we required an appropriate gastrocolic response as well as HAPCs that propagate the entire length of the studied colon sparing only the rectum. Additionally, these HAPCs should be

accompanied by an urge to defecate and stool output. All ARM and CM studies were executed in accordance with current guidelines and following the protocol of our institution.[11] For the purpose of this study, all manometries were reanalyzed by one pediatric gastroenterologist specialized in gastrointestinal motility disorders using a commercially available manometric system (Solar GI HRM v9.1; Medical Measurement Systems).

2.2 | Statistical analyses

Data are presented using frequencies and percentages or medians and interquartile ranges. Differences between groups were analyzed using Mann–Whitney *U* test or Fisher's exact test as appropriate. *P*-values less than 0.05 were considered to be statistically significant. Statistical analyses were conducted using SPSS for Windows, version 28.0.0.0 (SPSS Inc.).

3 | RESULTS

We identified 52 children with TCS and constipation symptoms who underwent ARM testing. Of these, 28 children were excluded due to the presence of additional congenital anomalies or spinal cord abnormalities other than TCS, leaving a total of 24 children (50% female, median age at ARM 6.0 years, interquartile range [IQR] 4.0–11.8 years, 96% white) in this study. Patient characteristics at time of ARM are shown in Table 1. Children in the TCS group were more likely to have a developmental delay when compared to children with FC (38% vs. 13%, $p = 0.046$). In addition, children in the TCS group had more episodes of fecal incontinence per week (4 vs. 0.5, $p = 0.045$) at time of ARM testing. Other constipation symptoms as well as surgical history and treatment strategies were similar between the two groups at time of ARM testing.

Six of the 24 children had ARM testing performed before detethering surgery (Table 2). High resting pressure and an abnormal push maneuver was found in one, only an abnormal push in one, and no RAIR (normal rectal biopsy, subsequently diagnosed with internal anal sphincter achalasia) in one. Three (50%) studies were found to be normal. Of these six children, only five underwent detethering surgery with one of these five having repeat testing after detethering that did show a change. In this patient, before detethering no RAIR (normal rectal biopsy) was found, while after detethering the ARM was normal. Both ARMs were performed under general anesthesia and during ARM testing normal resting pressures were found.

In total, for 19/24 children, ARM was performed a median of 2.5 years (IQR 1.0–4.3 years) after

TABLE 1 Patient characteristics at time of anorectal manometry.

	TCS ($n = 24$)	FC ($n = 24$)	*P*-value
Female, *n* (%)	12 (50)	12 (50)	1.000
Age in years, median (IQR)	6.0 (4.0–11.8)	6.0 (4.0–11.8)	1.000
Race, *n* (%)			
White	23 (96)	19 (79)	0.188
Black/African American	1 (4)	3 (13)	0.609
Multiracial	0 (0)	2 (8)	0.489
Medical history, *n* (%)			
Autism	3 (13)	3 (13)	1.000
ADHD/ADD	5 (21)	5 (21)	1.000
Developmental delay	9 (38)	3 (13)	0.046*
Anxiety/depression	6 (25)	4 (17)	0.477
Surgical history, *n* (%)			
Malone appendicostomy	1 (4)	1 (4)	1.000
Cecostomy	1 (4)	0 (0)	1.000
Colonic resection	0 (0)	2 (8)	0.489
Colostomy	0 (0)	1 (4)	1.000
Sacral nerve stimulation	2 (8)	0 (0)	0.489
Constipation symptoms			
Bowel movements pw, *n/N*, median (IQR)	18/24, 7.0 (2.0–11.0)	16/24, 2.5 (2.0–6.0)	0.141
Fecal incontinence pw, *n/N*, median (IQR)	15/21, 4.0 (0–5.0)[a]	18/21, 0.5 (0–2.3)[a]	0.045*
Abdominal pain, *n/N* (%)	11/19 (58)	12/15 (80)	0.271
Distension, *n/N* (%)	8/17 (47)	8/12 (67)	0.296
Pain defecation, *n/N* (%)	5/14 (36)	8/13 (62)	0.180
Straining, *n/N* (%)	4/12 (33)	7/13 (54)	0.302
Hard stools, *n/N* (%)	5/20 (25)	7/19 (37)	0.423
Urinary symptoms, *n/N* (%)	13/19 (68)[a]	5/12 (42)[a]	0.141
Treatment, *n* (%)[b]			
Oral laxatives	16 (67)	19 (83)	0.179
Rectal laxatives	0 (0)	2 (8)	0.489
Oral and rectal laxatives	4 (17)	0 (0)	0.109

(Continues)

TABLE 1 (Continued)

	TCS ($n = 24$)	FC ($n = 24$)	*P*-value
Antegrade enemas	2 (9)	1 (4)	1.000
No treatment	1 (4)	1 (4)	1.000

Abbreviations: ADHD/ADD, attention deficit hyperactivity disorder/attention deficit disorder; FC, functional constipation; IQR, interquartile range; *n/N*, number/total number of data available; Pw, per week; TCS, tethered cord syndrome.

[a]$n = 3$ not yet toilet trained.

[b]$n = 23$; in both groups $n = 1$ not reported.

*$P < 0.05$.

TABLE 2 Anorectal manometry findings in children with anorectal manometry testing before detethering surgery.

	TCS ($n = 6$)
Performed under anesthesia, *n* (%)	3 (50)
Resting pressure, median (IQR)	53 (24–75)
Low resting pressure (<39 mmHg), *n* (%)	0 (0)
High resting pressure (>71 mmHg), *n* (%)	1 (17)
Low squeeze pressure, *n* (%)	0 (0)[a]
Abnormal push maneuver, *n* (%)	2 (66)[a]
First sensation (balloon volume in mL), median (IQR)	10 (10–10)[b]
Urge sensation (balloon volume in mL), median (IQR)	30 (25–60)[a]
Discomfort sensation (balloon volume in mL), median (IQR)	90 (65–90)[a]
No RAIR, *n* (%)	1 (17)[c]
RAIR > 15% duration in seconds, median (IQR)	13.0 (5.6–15.3)[d]
RAIR > 25% duration in seconds, median (IQR)	9.5 (2.0–14.2)[d]
Normal, *n* (%)	3 (50)

Abbreviations: IQR, interquartile range; RAIR, rectoanal inhibitory reflex; TCS; tethered cord syndrome.

[a]$n = 3$; $n = 3$ under anesthesia.

[b]$n = 2$; $n = 3$ under anesthesia, $n = 1$ not reported.

[c]Had normal rectal biopsy.

[d]$n = 5$; $n = 1$ had no RAIR.

TABLE 3 Anorectal manometry findings in children with anorectal manometry testing after detethering surgery.

	TCS ($n = 19$)	FC ($n = 19$)	*P*-value
Performed under anesthesia, *n* (%)	10 (53)	3 (16)	0.038*
Resting pressure, median (IQR)	54 (37–64)	68 (53–77)	0.031*
Low resting pressure (<39 mmHg), *n* (%)	0 (0)	0 (0)	-
High resting pressure (>71 mmHg), *n* (%)	4 (21)	7 (37)	0.476
Low squeeze pressure, *n* (%)	0 (0)[a]	0 (0)[d]	-
Abnormal push maneuver, *n* (%)	5 (56)[a]	8 (53)[d]	1.000
First sensation (balloon volume in mL), median (IQR)	40 (20–50)[a]	25 (10–40)[e]	0.462
Urge sensation (balloon volume in mL), median (IQR)	60 (35–90)[a]	35 (20–65)[e]	0.134
Discomfort sensation (balloon volume in mL), median (IQR)	120 (60–135)[a]	90 (80–120)[d]	0.809
No RAIR, *n* (%)	2 (11)[b]	0 (0)	0.486
RAIR > 15% duration in seconds, median (IQR)	12.8 (10.5–18.1)[c]	13.1 (9.6–17.8)	0.899
RAIR > 25% duration in seconds, median (IQR)	10.3 (9.1–14.0)[c]	11.5 (8.2–15.0)	0.662
Normal, *n* (%)	10 (53)	10 (53)	1.000

Abbreviations: FC, functional constipation; IQR, interquartile range; RAIR; rectoanal inhibitory reflex; TCS, tethered cord syndrome.

[a]$n = 9$; $n = 10$ were under anesthesia.

[b]$n = 2$ no rectal biopsy performed; $n = 1$ barium enema was normal, $n = 1$ loss to follow=up.

[c]$n = 17$; $n = 2$ had no RAIR.

[d]$n = 15$; $n = 3$ under anesthesia, $n = 1$ too young to perform.

[e]$n = 14$; $n = 3$ under anesthesia, $n = 1$ too young to perform, $n = 1$ not reported.

*$P < 0.05$.

detethering surgery (Table 3). Children with a history of TCS were more likely to have an ARM under general anesthesia when compared to children with FC (53% vs. 16%, $p = 0.038$). The median resting pressure was significantly lower in children with TCS compared to children with FC (52 mmHg vs. 67 mmHg, $p = 0.031$). However, no significant differences were found in the proportion of both low and high resting pressure between children with a history of TCS and children with FC. In children who had ARM performed awake, no significant differences in number of children with a low squeeze pressure or abnormal push maneuver were found between the two groups. In addition, no statistically significant differences were found in rectal sensation (first sensation, urge, and discomfort). Since balloon expulsion test was only reported in a limited number of children, we did not compare results between the two groups. RAIR durations were similar between the two groups (13.1s vs. 12.8 s, $p = 0.899$). Even with a more stringent cutoff of >25% relaxation, we did not find a significant difference in RAIR duration between children with a history of TCS and children

TABLE 4 Colonic manometry findings in children with colonic manometry testing after detethering surgery, *n* (%).

	TCS (*n* = 13)	FC (*n* = 13)	*P*-value
Total colonic dysmotility	2 (15)	0 (0)	0.480
Distal colonic dysmotility	0 (0)	4 (31)	0.096
Proximal colonic dysmotility	0 (0)	0 (0)	-
Normal colonic motility	10 (77)	8 (62)	0.673
Normal colonic motility with high dose stimulants[a]	1 (7)	1 (7)	1.000

Abbreviations: FC, functional constipation; TCS, tethered cord syndrome.
[a]After bisacodyl and an additional dose of glycerin.

with FC (10.3 s vs. 11.5 s, $p = 0.662$). In both groups, 10 studies (53%) were found to be normal.

3.1 | Colonic manometry

Among the 24 children with TCS, 14 children (50% female, median age at CM 7.5 years, IQR 5.0–10.3, 93% white) also had a CM performed. Two children had CM performed before detethering surgery; one had normal motility and one had distal colonic dysmotility. The child with distal colonic dysmotility had a repeat CM after detethering surgery and creation of a Malone appendicostomy (interval 11 months) and displayed normal colonic motility on the repeat study. In total, 13 children had a CM performed a median of 2.5 years (IQR 1.3–4.0 years) after detethering surgery. No significant differences in colonic motility between children with a history of TCS and children with FC were found (Table 4).

4 | DISCUSSION

In our cohort of children with TCS, most of whom had prior detethering surgery, we found that anorectal physiology and colonic motility were similar when compared to a matched cohort of children with FC.

In clinical practice, we often encounter children with TCS with continued defecatory symptoms after they have had detethering surgery. Persistent symptoms may vary depending on severity and duration at presentation. Pain and motor weakness seem to improve more after detethering surgery compared to improvement in sensorimotor and sphincter function.[12–14] One retrospective cohort study with 31 children showed that preoperative bowel and bladder dysfunction was associated with a significantly poor long-term prognosis after detethering surgery.[15] The clinical challenge that arises in patients with persisting symptoms after detethering surgery, is determining the extent to which the history of TCS plays a role in their current symptoms. Only one small prospective study from 1993 including nine children with a first manifestation of tethered cord, reported on ARM findings before and after detethering surgery, showing that ARM abnormalities, such as hyperactive rectum sensitivity and hyperactive internal and external sphincter pressure and activity, before detethering surgery persisted 6 months after surgery without clear clinical improvement.[9] However, this single study was performed over 30 years ago and used a Gaeltec micro-tip transducer (a 16–20 F one lumen catheter with transducers placed at a distance of 3–5 cm from one another; one in the rectum, one in the anorectum, and the third at the level of the internal anal sphincter), a separate balloon catheter for distension of the rectum, and perianal surface electrodes to assess the function of the external anal sphincter. Hence, new data are needed for this population of children.

In our cohort, we found that children with a history of TCS were more likely to receive an ARM under general anesthesia when compared to children with FC (53% vs. 16%, respectively). This disparity was primarily due to the higher prevalence of concurrent procedures being performed at the time of ARM in the children with a history of TCS. In four children, ARM was performed at the same time as CM catheter placement, in one child ARM was combined with anal botulinum toxin injection, and one had an MRI under general anesthesia due to age. Additionally, we found that the median resting pressure in children with TCS was significantly lower compared to children with FC (52 mmHg vs. 67 mmHg, respectively). This difference is likely attributable to the higher frequency of ARMs performed under general anesthesia in children with TCS, as general anesthesia is known to cause a decrease in resting pressure.[11,16,17] In addition, children who are awake during the procedure may experience increased anxiety, which can result in higher resting pressures.[18] Nevertheless, both resting pressures are still within the normal range. Furthermore, episodes of fecal incontinence per week were more prevalent in children of the TCS group at time of ARM testing compared to children of the FC group (4 vs. 0.5, respectively). Since low resting pressure was not more common in children with TCS and, despite small numbers, there was no significant difference in rectal sensation between children with detethering surgery before ARM and those with FC, this difference in fecal incontinence might be attributable to constipation.

Previous studies have described unique manometric characteristics in patients with spinal cord abnormalities. A shift in the RAIR dose–response curve to the left has been described in children with a variety of spinal cord abnormalities uncovered by MRI as well as in children with sacral agenesis.[8,19] A study in adults with complete traumatic supraconal spinal cord lesions described this same phenomenon of greater internal

anal sphincter relaxation in response to small balloon volumes.[20] In addition, they found that the duration of anal relaxation was greater at low balloon distension when compared to healthy controls.[20] A recent observational cross-sectional study comparing 54 children with myelomeningocele with 81 children with refractory FC, also found that the duration of the RAIR is significantly longer in children with myelomeningocele.[21] Although it is known that the presence of the RAIR is independent of the spinal cord, the exact mechanisms responsible for the more pronounced internal anal sphincter relaxation are not understood.[19] It is thought that the spinal cord might modulate the characteristics of the RAIR.[8] However, in our cohort, the duration of the RAIR was not found to be significantly different when children with a history of TCS were compared with children with FC. Furthermore, the duration of the RAIR of the six children who had ARM testing before detethering surgery did not appear to be longer to those who had ARM after detethering surgery. That being said, we did have one patient who did not have a RAIR with normal ganglion cells on biopsy before detethering surgery and was subsequently diagnosed with internal anal sphincter achalasia who following surgery did have a RAIR on repeat ARM. The abnormalities found on ARM in our patients with history of TCS status postdetethering surgery are findings also routinely seen in children with FC. Based on our data, these abnormal ARM findings are not more common in children with a history of TCS than in children with FC. One small case-control study, including 10 constipated children with spinal cord abnormalities detected by MRI, found that 60% exhibited anal spasm upon balloon distension.[8] In our cohort, we did not observe this finding, which may be because the majority of our children had already undergone detethering surgery.

To our knowledge, this is the first study evaluating colonic motility in children with TCS. We found that CM findings in children with a history of TCS were similar to those of a matched cohort of children with FC, suggesting that the underlying pathophysiology of defecatory disorders in children with and without history of TCS is not related to differences in colonic motor function. Colonic motility is mediated by the colonic musculature, a network of excitable cells, which consists of foremost smooth muscle cells providing the contractile force, and accessory cells that orchestrate contractions into functional patterns, with the Interstitial Cells of Cajal (ICC) being a very important one.[22,23] ICC act as pacemaker cells and determine the frequency, velocity, and direction of propagation for many colonic motor patterns, including HAPCs, the most recognizable motor pattern of colonic motility.[22] The enteric nervous system, an intrinsic nervous system that is entirely located within the walls of the gastrointestinal tract, coordinates colonic motility and can operate independently of the central nervous system, including the spinal cord.[23] Colonic motility will therefore most likely not be affected by TCS or a history of TCS, a theory which is generally supported by our findings. We did have one child who had colonic manometry testing before and after detethering surgery, with improvement from distal colonic dysmotility to normal colonic motility. However, this child also underwent creation of a Malone appendicostomy before repeat testing and we suspect that antegrade continence enemas led to improvement in colonic motility.[24]

The first limitation of our study is its single-center design. Other limitations are inherent to its retrospective design. We were unable to control ARM and CM test conditions to ensure they were consistent across all children in this study. Our data were based on what was documented in the medical charts. For our TCS group, most of whom became our patients after detethering surgery, we had no clear data if they had suffered from any other comorbidities often seen in TCS, such as muscle weakness or urinary problems. In addition, unfortunately, only two children with TCS had manometry testing performed before and after detethering surgery. Because the number of children who had ARM performed before detethering surgery was limited, we did not compare outcomes with those who already had detethering surgery at time of ARM. A prospective study with ARM and CM testing before and after detethering surgery would be of great interest to further evaluate the impact of TCS on the gastrointestinal motility. This might help to understand what it means when a child is diagnosed with TCS, especially as the existing literature regarding indications and outcomes of pediatric detethering surgery is highly variable.[25] An additional consideration is to include additional diagnostic tests that could evaluate for differences in pathophysiology not studied by ARM and CM. Both ARM and CM have inherent limitations, both related to their performance in children and also in the variables they measure. Lastly, we were not able to compare our age and sex-matched control groups of children with FC with our overall FC population who underwent ARM and CM to verify their representativeness.

In conclusion, we found no significant differences in anorectal physiology and colonic motility between children with a history of TCS and children with FC. Our findings suggest that the underlying pathophysiology of defecatory disorders in children with and without history of TCS are similar and not related to differences in colonic motor function. This supports that the management of children with a history of TCS should be similar to that of children with FC.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

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