

The Tethered Spinal Cord in Patients With Anorectal Malformations

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● The aims of this study were to find the prevalence of tethered cord in patients with anorectal malformations; to determine if the presence of tethered cord relates to the severity of the anorectal defect, and to certain symptoms, signs, radiologic findings, and associated anomalies; and finally to determine whether tethered cord impacted on a patient's functional prognosis and whether surgical untethering improved the patient. The authors studied 934 patients with anorectal malformations, 111 of whom had magnetic resonance imaging (MRI) of the spine. We compared patients with and without tethered cord by using parametric and nonparametric statistical tests. Tethered cord occurred in 24% of the patients. The prevalence varied according to the type of anorectal defect from 43% in the complex group to 11% in patients with rectovestibular fistula. Patients with tethered cord had a lateral sacral ratio lower than that of patients without tethered cord (0.410 versus 0.702). Tethered cord was present in 90% of patients with myelodysplasia, 60% of patients with a presacral mass, 57% of patients with sacral hemivertebrae, and 56% of patients with a single kidney. The greater number of associated anomalies a patient had, the greater the risk of having tethered cord ($P < .05$ for all differences). The authors noted differences between patients with and without tethered cord in the presence of voluntary bowel movements (46% versus 70%), fecal soiling (91% versus 63%), constipation (21% versus 43%), and urinary incontinence (86% versus 42%). The data indicate that patients with tethered cord have a worse functional prognosis than patients without tethered cord. However, the incontinence in our patients was also predictable based on the type of anorectal defect and the character of the sacrum irrespective of the presence of tethered cord. Eighteen patients underwent surgical untethering of the cord, and none had any significant change in bowel or urinary function postoperatively. No patient with tethered cord experienced incontinence that could be attributed to the cord defect alone. This study suggests that tethered cord occurs more frequently in patients with severe anorectal defects, sacral hypodevelopment, myelodysplasia, presacral mass, sacral hemivertebrae, or a single kidney, or in those with an anorectal defect with poor functional prognosis. At present no solid evidence supports the concept that tethered

cord by itself affects the functional prognosis of patients with anorectal malformations. Also, there is no good evidence demonstrating that surgical untethering improves the prognosis.

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INDEX WORDS: Tethered cord, anorectal malformation, spinal cord, bowel control.

INTRAVERTEBRAL fixation of the filum terminale results in tethering of the spinal cord and is caused by various conditions that restrict the normal cephalad migration of the conus medullaris. According to the literature, patients with tethered cord may remain asymptomatic or can experience motor and sensory disturbances.¹⁻¹⁶ Some investigators note that the condition may provoke urinary^{2,6,7,10,13,14,17,18} and bowel^{1,2,6,10,13,17,19} dysfunction. Once tethering is identified, the decision to surgically untether the cord is controversial. Most investigators recommend surgery in a patient with motor and sensory symptoms,^{1,2,4-6,20} or urologic disturbances.^{1,4,7} Some authors advocate prophylactic surgery, particularly in the younger patient,^{8,11,19,21,22} and others advocate a conservative approach.⁵

The association of anorectal malformations and tethered spinal cord is well documented.^{1,8,19,23-25} Several investigators^{1,9} have suggested that the presence of tethered cord may have an effect on fecal and urinary continence. Therefore, it is becoming standard practice at many centers to screen patients with anorectal malformations for the presence of tethered cord^{1,8,19,21,26} and to surgically untether them.^{8,19,21} From our literature review we were unable to find good evidence that the presence of tethered cord worsens the functional prognosis of patients with anorectal malformations. We were also unable to find convincing evidence to support the practice of prophylactic surgical untethering of the cord.

We sought answers to the following questions. (1) What is the prevalence of tethered cord in patients with anorectal malformations? (2) Is there a direct relationship between the type of anorectal malformation and the prevalence of tethered cord? (3) Are there symptoms, signs, radiological findings, and associated anomalies that should alert the clinician to the presence of tethered cord and that should prompt its investigation? (4) Is there a direct relationship between the presence of tethered cord and a patient's functional prognosis, and does surgical tethering improve the patient?

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MATERIALS AND METHODS

Study Sample

In the senior author's personal series, 934 cases of anorectal malformations who have undergone surgical correction were reviewed. Of these, 111 patients underwent magnetic resonance imaging (MRI) of the spine. The diagnosis of tethered cord was established when the conus medullaris was located below the level of the L2 vertebra associated with anomalies of spinal dysraphism, such as a lipomenigocele, spinal lipoma, meningomyelocele, meningocele, or a thickened filum (Fig 1). The presence of tethered cord was analyzed in relationship to type of anorectal defect, quality of sacrum, the presence of associated defects, the degree of bowel and urinary control, and the effect of surgical untethering.

Type of Anorectal Defect

The frequency of tethered cord was determined for each type of anorectal malformation. The previously published²⁷ classification of anorectal malformations was used. Patients with cloacal exstrophy and those with complex defects were also included.

Quality of Sacrum

To evaluate the sacrum, the anterior-posterior and lateral sacral ratios were used, which were derived by measuring the radiologic image of the sacrum and comparing its size with fixed bony structures of the same patient's pelvis. The sacral ratio of a normal pelvis in an anterior-posterior (AP) film is 0.74.²⁷ A line is traced from the most upper portion of one iliac crest to the other. A second line is traced from the lowest point of one of the sacroiliac joints (posterior and inferior iliac spine) to the other. A third line runs parallel to the other two and touches the lowest radiologically visible point of the sacrum. A ratio is obtained by dividing the distance between the two lower lines by the distance between the two upper lines. Children with anorectal malformations show a spectrum of values; lower ratios represent different degrees of sacral hypodevelopment and are associated with defects that have poor functional prognosis. The sacral ratio in the lateral position in normal children is 0.77.²⁷

Associated Defects

The anomalies most frequently associated with anorectal malformations were tested for their association with tethered cord and therefore

for their value in predicting the defect. These included spinal, sacral, and urologic abnormalities. Myelodysplasia included conditions such as lipomenigocele, syringomyelia, spina bifida, spinal lipoma, or meningocele.

Bowel and Urinary Function

Functional status was assessed in four areas: the presence of voluntary bowel movements, fecal soiling, constipation, and urinary continence. Voluntary bowel movements were defined as the act of feeling the urge to have a bowel movement, the capacity to verbalize that, and the ability to hold the bowel movement until reaching the bathroom. Soiling was defined as the involuntary leaking of small amounts of stool that smeared the underwear. Constipation was considered the incapacity to empty the rectum spontaneously (without help) every day. Patients with urinary incontinence were those patients who were unable to hold the urine. Patients with mild dribbling and wetness at night were considered in the continent group.²⁷ Only patients older than 3 years of age were investigated for functional status, because there are normal children less than 3 years of age who are still not toilet trained.

Statistical Analysis

Univariate statistical techniques were used to improve our clinical understanding of risk factors associated with tethered cord. Both parametric and nonparametric tests were used to evaluate differences among groups. Differences in the prevalence of tethered cord between patients with and without associated anomalies and functional status limitations were evaluated by using 2×2 cross tabs and the Fisher's Exact test. Small cell sizes for a substantial number of comparisons indicated that a nonparametric approach would be more appropriate in these cases. Lateral and anterior-posterior sacral ratio differences by tethered cord status were evaluated by using standard unequal variance two-sample Student's *t* tests (sacral ratios were determined to be normally distributed with a Kurtosis and Skewness normality test, and unequal variances were established with an F test for variance).^{28,29} Differences in the probability of tethered cord by increasing severity of diagnosis (an ordinal scaled ranking) were assessed by using a Wilcoxon signed rank test.²⁸ Several maximum likelihood estimates of the association between the presence of multiple associated anomalies and tethered cord status were produced by using multivariate logistic regression models. For these models, statistical significance for individual effects was evaluated with a Wald test.³⁰

Given the hypothesis that prevalence of tethered cord is positively associated with presence of associated anomalies and functional status deficiencies, all statistical tests were one tailed. All tests were evaluated at the $\alpha = 0.05$ level of significance. Tabulations and statistical analyses were performed with version 6.10 of the Statistical Analysis Software (SAS 1995).

Literature Review

We found 12 references in the literature^{1,8,18,19,21,24-26,31-34} that specifically dealt with the issue of tethered cord in patients with anorectal malformations. Of these, there were 4 studies^{8,19,21,25} that mentioned specific patients with anorectal malformations who suffered from bowel and urinary functional problems and who underwent surgical untethering.

RESULTS

Of 934 patients with anorectal malformation in the current series, 111 patients underwent MRI of the spine; 27 of these patients had MRI evidence of tethered spinal

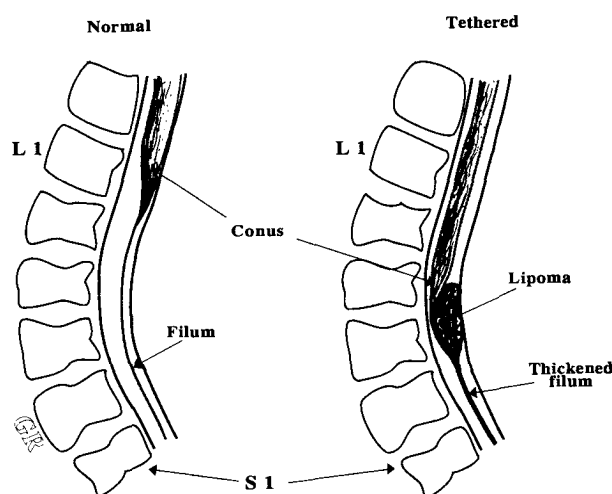


Fig 1. Schematic tethered cord.

cord. The overall prevalence of tethered cord among the 111 patients was 24%. There were 43 boys, and 68 girls in total. Ten boys (23%) and 17 girls (25%) had tethered cord.

Type of Anorectal Defect

The frequency of tethered cord varied from 43% in the complex group to 11% in patients with retrovestibular fistulas. (Table 1). The prevalence of tethered cord increased with increasing severity of the malformation, however this trend did not attain statistical significance. The overall prevalence in patients with a known anorectal diagnosis was 24%. There were seven patients in the tethered group and five patients in the nontethered group for whom the specific anorectal defect could not be determined because they underwent surgery previously elsewhere, and the original anatomic diagnosis was not known when the patients presented for reoperation.

Quality of Sacrum

Fifty-seven patients had lateral radiographs of the sacrum, and 67 patients had anterior-posterior (AP) sacral films. The average lateral sacral ratio in patients with tethered cord was 0.410, which was statistically significantly lower than for patients without tethered cord (lateral ratio 0.702, $P < .01$). The AP ratio in the tethered cord group was 0.344, which was statistically significantly lower than 0.597 ($P < .01$) in the nontethered group. Of 13 patients with tethered cord, 11 (85%) had a lateral ratio that was lower than 0.6, and the majority of the nontethered cord group had lateral ratios greater than 0.6. All patients with a lateral ratio greater than 0.8 were untethered. The two groups of patients, those with and those without tethered cord, seem to distribute into two distinct populations (Fig 2).

Tethered Cord and Associated Anomalies

Table 2 shows the associated anomalies and the prevalence of tethered cord in patients with each anomaly. The prevalence varied from 90% for patients with myelodysplasia to 10% in patients with vesicoureteral

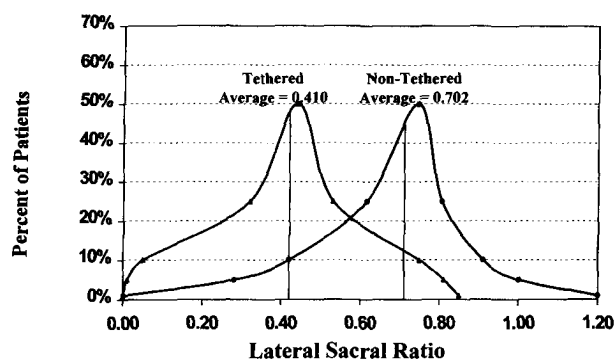


Fig 2. Lateral sacral ratio: Distribution by tethered cord status.

reflux. Of 17 patients with none of the associated defects, only 1 (6%) had a tethered cord. That patient had the diagnosis of persistent cloaca, with a common channel of less than 3 cm. A multivariate analysis of these associated anomalies showed that with increasing number of anomalies, the risk of tethered cord increases ($P < .01$).

Bowel and Urinary Function

Important differences were found in bowel and urinary function between patients with and without tethered cord. Fifty-four percent of patients with tethered cord did not have voluntary bowel movements compared with 30% in the nontethered group; 91% of patients with tethered cord suffered from fecal soiling as compared with 63% in the nontethered group. In contrast, constipation occurred more frequently in nontethered patients, 43% compared with 21%, and 86% of patients with tethered cord suffered from urinary incontinence compared with 42% in nontethered patients. Overall, these data suggest that functional limitations are greater among patients with tethered cord. The differences that attained statistical significance were for patients with fecal soiling and urinary incontinence.

In an effort to clarify the impact of tethered cord on the functional prognosis of our patients, we found three patients with tethered cord and anorectal defects with

Table 1. Tethered Cord in Anorectal Malformations

Diagnosis	No. of Patients	Tethered	%
Complex	7	3	43
Cloacal exstrophy	5	2	40
Bladder neck	6	2	33
Cloaca C Ch >3 cm	21	6	29
Cloaca C Ch <3 cm	9	2	22
Prostatic fistula	11	2	18
Bulbar fistula	11	2	18
Vestibular fistula	9	1	11
Total	79	20	25

Abbreviation: C Ch, common channel.

Table 2. Associated Defects and Tethered Cord

Defect	No. of Patients	Tethered	%
Myelodysplasia	10	9	90
Presacral mass	10	6	60
Sacral hemivertebrae	14	8	57
Single kidney	16	9	56
Ectopic kidney	4	2	50
Hypospadias	4	2	50
Spinal hemivertebrae	14	6	43
Hydroureter	5	2	40
Hydronephrosis	23	7	30
Dysplastic kidney	4	1	25
Reflux	21	2	10
None	17	1	6

good functional prognosis²⁷ (2 recto-urethral bulbar fistula, 1 vestibular fistula). All three of them were continent of stool and urine despite having tethered cord. In seven patients without tethered cord with a defect with poor prognosis (persistent cloaca with a long common channel), four (57%) had voluntary bowel movements (a similar result to the one expected based on type of defect)²⁷ and two (28%) had urinary incontinence (versus 69% expected for that type of defect).²⁷

Tethered Cord Release

Of the 27 tethered patients, 18 underwent surgical untethering. Of those who underwent untethering, five underwent that procedure as part of another operation (3 patients had myelomeningocele, one patient had a presacral mass, and one patient had a sacrococcygeal teratoma.) Three other patients had sensory or motor symptoms of the lower extremities; one improved and two were unchanged but did not worsen after surgery. Before surgery, nine patients had urinary incontinence and four had fecal incontinence. Six were asymptomatic and underwent prophylactic untethering. Of these six, four are under 3 years of age (therefore we do not have information concerning continence), and two are older. No significant changes were found in urinary or fecal continence postoperatively. No patient in our series who was doing well from the urinary and bowel continence point of view and then became incontinent was found to have tethered cord. Likewise, no patient with good functional prognosis based on the type of defect and with normal sacrum who had unexplained fecal or urinary incontinence was found to have a tethered cord. Of the nine patients who have not undergone untethering, six were older than 5 years of age; one had urinary incontinence and one had fecal incontinence, and four remain asymptomatic. Three were under 3 years of age; one had urinary incontinence, two are asymptomatic and are being considered for untethering.

Literature Review

Of the articles reviewed on tethered cord, three related to myelodysplasia,³⁵⁻³⁷ eight related to urologic prob-

lems,^{3,7,10,16,18,34,38,39} and 12 dealt specifically with anorectal malformations.^{1,8,18,19,21,24-26,31,34} Of these, there were four studies^{8,19,21,25} that mentioned specific patients with anorectal malformations who suffered from bowel and urinary functional problems and who underwent surgical untethering. We were impressed by the fact that from all of the reports, only four discussed patients meeting these criteria (Table 3). Of the reports cited, there were 47 patients with anorectal malformations who underwent surgical untethering. Eight patients had preoperative urinary difficulties, and there was mention of only one patient with fecal incontinence. Only two patients had some degree of improvement in urinary function postoperatively, and there was no mention of the outcome in the patient with fecal incontinence.

DISCUSSION

The association of tethered cord and anorectal malformations is well described.^{1,8,19,23-25} The prevalence stated in the literature was found to vary from approximately 10%^{3,19,26} to 52%.²¹ Our 24% prevalence is higher than most studies, perhaps because ours is a major referral center for anorectal malformations and tends to receive more complex defects. Warf's²¹ series included a large percentage of patients with cloacal exstrophy, and we as well as others^{8,40-42} have shown that such patients have a considerably higher risk of tethered cord. Other series specifically dealing with tethered cord and anorectal malformations are not subdivided by specific anorectal defect but rather use terms such as "high" or "low" imperforate anus.

Our data show an increasing prevalence of tethered cord as the severity of the anorectal defect increases. Although unable to demonstrate statistical significance between the prevalence of tethered cord in each individual defect, it is likely that with a larger sample size, the result would attain statistical significance. In the current series, complex defects were those that defied classification because of their severity and were usually associated with poor sacrum and muscles.⁴³ That group had the highest prevalence of tethered cord, followed by those with cloacal exstrophy. The 40% prevalence of tethered

Table 3. Anorectal Malformations and Tethered Cord Literature Review

Study	No. of Patients Untethered	No. of Patients With Preoperative Problems		Postoperative Changes	
		Urinary	Bowel	Urinary	Bowel
Warf et al ²¹	26	1	0	Mild improvement	—
Karrer et al ⁸	13	2	0	1 No change 1 Incontinence changed to stress incontinence	—
Davidoff et al ⁹	4	1	1	No change	No mention of result
Sato et al ²⁵	4	4	0	No change	—
Total	47	8	1	2 Improved	—
Levitt et al	18	9	5	No significant changes	No significant changes

cord in patients with cloacal exstrophy is consistent with several reports^{8,40,41} but less than in one series, which found tethering in all patients with cloacal exstrophy.⁴²

Three studies tried to establish a relationship between the type of anorectal defect and the presence of tethered cord.^{8,19,21} In the largest series²¹ the investigators were unable to establish a relationship and included only three patients with imperforate anus; the other patients had cloacal exstrophy, "VATER association," or "complex" anomalies including penoscrotal transposition and persistent cloaca. Karrer⁸ found that patients with a high imperforate anus had more significant spinal lesions than those with low lesions, but this series had only four patients with high imperforate anus, four with a low defect, and six with cloacal exstrophy.

The sacral ratio was found to be a strong predictor of tethered cord. Other investigators have noted a high prevalence of spinal cord anomalies when there is radiographic evidence of a sacral anomaly,^{12,24} but they do not use an objective method in their assessment of the sacrum. We feel the sacral ratio²⁷ is an accurate and objective value that, in our series, was helpful in predicting the presence of tethered cord. We have noted previously²⁷ that high sacral ratios correspond with good functional prognosis, and low sacral ratios predict poor prognosis. In our series there has never been a fecally continent patient with a lateral sacral ratio less than 0.3. The lateral sacral ratio is more reliable because the AP ratio will vary depending on the tilt on the AP axis of the pelvis when the radiograph is taken.

In our literature review, we did not find any mention of the specific prevalence of tethered cord and associated anomalies in patients with anorectal malformations. Tethered cord has been noted to be associated with myelodysplasia⁷ and with sacral anomalies,¹ but no description of its relationship with urologic problems was found. We determined that the presence of myelodysplasia, a single kidney, sacral and spinal hemivertebrae, and a presacral mass were important risk factors for tethered cord. Other urologic abnormalities such as hydronephrosis, dysplastic kidney, and vesicoureteral reflux were not as important.

Patients with tethered cord were more likely to have fecal soiling and urinary incontinence. An opposite trend for constipation was observed, which is consistent with a prior finding of more frequent constipation in patients with less severe anorectal defects.²⁷ We agree with the general statement in the literature that fecal^{1,2,6,7,10,13,17-19} and urinary^{2,6,7,10,13,14,17,18} abnormalities appear to be more prevalent in patients with tethered cord. However, in our literature review, we found very few cases in which patients with tethered cord were described with fecal or urinary difficulties. In fact, although several au-

thors^{1,2,6,10,13,17-19} mention that fecal incontinence may result from tethered cord, only one patient who experienced it is mentioned,¹⁹ but the author does not report the impact of untethering on fecal continence.

The obvious question is whether it is the tethered cord that is impacting on the patient's continence or is it only an incidental finding? Unfortunately, the number of patients in our series does not allow us to definitively answer this question. As shown in our results, when dealing with a benign defect, the presence of tethered cord does not seem to change the prognosis. On the other hand, in patients with a poor prognosis type of defect, when tethered cord was absent, urinary function was slightly better than expected, but the presence of voluntary bowel movements was not different. It is vital therefore to take into consideration the severity of anorectal defect and the quality of the sacrum when one is analyzing the impact of tethered cord on functional prognosis. Larger prospective series should seek to elucidate this question.

Once tethered cord is diagnosed, the decision to proceed with surgical untethering is controversial. In our series, 18 of 27 patients underwent untethering of the cord. One patient showed improvement in sensory and motor symptoms postoperatively and in two the symptoms did not worsen. No patient demonstrated a significant change, either positively or negatively, in bowel or urinary function. Of our nine patients who remain tethered, two have urinary and one has fecal incontinence, the rest are asymptomatic. Many of them are from other countries in which the option for a neurosurgical procedure is not readily available. Our series does not help to clarify the question concerning surgical untethering. We saw isolated cases of sensory and motor improvement after surgery from which we could not draw any definitive conclusions. We know from the literature that it is not clear that sensory and motor symptoms will worsen if the cord is left tethered^{26,36,44,45} and that surgical untethering has a potential risk for causing neurological damage.^{39,46} Preoperative and postoperative MR scans have shown that the position of the conus is unchanged by surgery,³⁷ and retethering can occur.⁵

In the presence of an anorectal defect and sacrum that imparts a poor prognosis on a patient, tethered cord may play an insignificant role. In patients with anorectal malformations, we could find only 47 patients in four studies^{8,19,21,5} who underwent surgical untethering (Table 3). Of these, 10 patients were considered to have improvement in sensory and motor symptoms of the lower extremities after surgery; two patients had improved urinary function but postoperatively still had some degree of urinary incontinence. There was mention of only one patient who had any alteration in fecal

continence, but the author did not report any postoperative clinical information.¹⁹ In addition, these studies do not mention what the specific anorectal diagnosis was in the patients who improved or the quality of their sacrum, factors we have shown impact on functional prognosis.²⁷ Clearly, a prospective study is needed to answer this question.

It is desirable to screen every patient with an anorectal malformation for the presence of tethered cord. However, based on our study, mandatory and recommended indications were determined for screening those patients we found to have a considerably higher risk of having tethered cord (Table 4). On the basis of our results, we recommend the above screening criteria. Also, we agree with the recommendation for screening noted by others^{19,26} of patients with new onset of sensory, motor, or urologic symptoms. Even though we did not study

Table 4. Indications for Screening

Mandatory
Myelodysplasia
Lateral sacral ratio <0.6
AP sacral ratio <0.4
Complex defect
Cloacal exstrophy
Recommended
Recto-bladder neck fistula
Cloaca C Ch >3 cm
Presacral mass
Sacral hemivertebra
Single kidney
Ectopic kidney
Hypospadias
Spinal hemivertebra
Good prognosis, poor result

Abbreviation: C Ch, common channel.

orthopedic and cutaneous associations with tethered cord, others have shown that findings such as limb shortening, gait disturbances, foot defects, scoliosis,⁸ cutaneous malformations like a blemish on the midline of the back (lump, nevus, sinus, or angioma), hypertrichosis, skin dimples, or sacral lipomas,^{8,13} should alert the clinician to the possibility of an associated tethered cord.

We were impressed by the scarcity of clinical data in the literature concerning patients with anorectal malformations who have undergone surgical untethering and for whom clinical data concerning bowel and urinary function are available. Many authors make definitive statements such as "intervention in asymptomatic patients is indicated because the natural history of tethered cord is one of deceptively gradual worsening to a fixed neurologic deficit"⁸ or "patients often have difficulty with micturition [and] sphincter disturbances"¹ and "early detection and repair may improve their ability to achieve continence."¹ "We consider the finding of a tethered cord, even in an asymptomatic patient, to be an indication for surgery."¹⁹ Many investigators quote each other and visa versa. When one searches for hard evidence documenting fecal or urinary difficulties in patients with anorectal malformations resulting from tethered cord or improvements noted after surgical untethering we were able to find only the data summarized in Table 3. Untethering may play a role in arresting the progression of sensory and motor symptoms^{1,2,4-6,20} and in urologic symptoms in patients with myelodysplasia^{7,38} and spina bifida.³ The relationship between tethered cord and continence in patients with anorectal malformation so far is not firmly established. Prospective investigation is necessary to answer the question.

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