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Is daily dilatation by parents necessary after surgery for Hirschsprung disease and anorectal malformations?

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Abstract

Background/Purpose: Most surgeons recommend daily dilatation after surgery for Hirschsprung disease and anorectal malformations. Our goal was to critically evaluate the potential risks and benefits of this practice.

Methods: A retrospective chart review was carried out of all children undergoing repair of Hirschsprung disease or anorectal malformation over 5 years. Patients with long segment Hirschsprung disease or anal stenosis were excluded.

Results: There were 95 patients, of which 34 had Hirschsprung disease and 61 had an anorectal malformation. Postoperatively, 65 underwent routine dilatation by parents; and 30 underwent weekly calibration by the surgeon, with daily dilatation by the parents only if the anastomosis was felt to be narrow. Of the 30 children undergoing weekly calibration, only 5 (17%) developed late narrowing that required conversion to the daily parental dilatation approach. There were no significant differences between the 2 approaches with respect to stricture development, anastomotic disruption, perineal excoriation, or enterocolitis.

Conclusion: Weekly calibration by the surgeon is associated with similar outcomes to daily dilatation by the parents. Because this approach is kinder to the parents and the child, it should be seriously considered for the postoperative management of children with Hirschsprung disease or anorectal malformations.

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Surgical management of both anorectal malformations (ARMs) and Hirschsprung disease (HD) involve some kind of "pull-through" procedure in which the normal colon is brought down to the anus and an anastomosis is performed. In some cases, a preliminary colostomy is done, although

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there has been a move toward primary pull-through in recent decades, particularly for the treatment of HD [1-4].

Anastomotic complications, particularly stricture or leak, are uncommon, occurring with an incidence of 4% to 22% in reported series [4-10]. Because these complications can have devastating consequences and can impair the long-term functional quality of life for these children, most surgeons traditionally required diligent daily anal dilatation programs to avoid anastomotic stricture. The rationale for this practice has been to "stimulate the normal growth of the rectum,

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which, in turn, will gently distend the muscle structures, avoiding their rupture" [11]. Dilatations are usually done with metal or plastic dilators or a parent's finger, typically once or twice daily. Many surgeons feel that the dilatations will help to prevent strictures and thus prevent constipation and postoperative enterocolitis. However, there is also a theoretical risk to daily dilatations by parents, including inadvertent disruption or perforation of the anastomosis. In addition, both children and parents usually dislike the procedure, which can be very stressful for them. We therefore sought to critically analyze the need for daily dilatation on the outcomes after pull-through surgery.

1. Methods

A retrospective review was carried out of inpatient surgical records to identify children undergoing pull-through procedures for HD or ARMs between January 2005 and January 2010. Patients who were diagnosed with *long segment* HD (defined as a transition point proximal to the descending colon), those with anal stenosis who did not require surgical correction, and those with inadequate follow-up were excluded. Collected data included patient sex, gestational age, presence of other anomalies, age at treatment, presence of colostomy, postoperative dilatation regimen, and complications. Children were assigned to daily dilatation or weekly calibration based on the individual practice of the operating surgeon.

The groups were compared using either t test or χ^2 analysis, with a P value < .05 being considered significant. Research Ethics Board approval was obtained from the Hospital for Sick Children, Toronto, before beginning the study.

2. Results

One hundred twenty-four charts were identified through the initial search criteria. Of these, there were 54 with a diagnosis of HD (11 female and 43 male) and 70 with ARM (35 female and 35 male). All children with HD had biopsyproven aganglionosis, either on full-thickness or suction rectal biopsy.

Of the 54 with HD, 10 were found to have either long segment disease or hypoganglionosis; and a further 10 with short segment disease had inadequate follow-up records. Thus, a total of 34 Hirschsprung patients were included in the final analysis (5 female and 29 male). Included in the 70 ARM patients were 1 patient with anal stenosis and 8 who did not have adequate follow-up. These patients were all excluded, leaving a total of 61 patients (29 female and 32 male). The demographics of the study population can be seen in Table 1. Of the total of 95 patients, 65 underwent daily dilatation by the parents; and 30 underwent weekly calibration by the surgeon.

Table 1 Demographic data			
	Daily dilatation	Weekly calibration	P value
HD	n = 17	n = 17	
Male	15 (88%)	14 (82%)	.63
Mean gestational age (wk)	38.7	39.3	.27
Mean birth weight (kg)	3.14	3.28	.54
Mean age at pull-through (mo)	3.4	4.2	.7
Mean weight at pull-through (kg)	4.97	5.9	.34
Preliminary colostomy	2 (12%)	1 (6%)	.34
ARM	n = 48	n = 13	
Male	26 (54%)	6 (46%)	.76
Mean gestational age (wk)	37.3	36.9	.73
Mean age at pull-through (mo)	2.7	3.1	.03
Mean weight at pull-through (kg)	5.79	7.45	.008
Preliminary colostomy	24 (50%)	7 (54%)	.8
High malformations	36 (74%)	11 (84%)	.7
Laparoscopic approach	2 (4%)	2 (15%)	.2

Postoperatively, all patients were seen by the surgeon in the clinic for initial examination and calibration of the anastomosis. The average number of days postsurgery for this visit was 18 (± 5) in the daily group and 17 (± 8) in the weekly group. In the daily dilatation group, the parents were taught how to use Hegar dilators or a finger to perform dilatation daily or twice daily, depending on surgeon preference. Those who were treated by weekly calibrations were brought back to see the surgeon once per week for 6 weeks, then with decreasing frequency. Patients in this group were converted to a daily dilatation approach if they were deemed by the surgeon to be forming an anastomotic stricture or cuff narrowing. This occurred in only 5 cases (17%, 2 ARM and 3 HD). These patients remained in the weekly calibration group for the purpose of analysis. None of these patients required dilatation beyond 3 months, and none had any postoperative complications.

The outcomes that were of primary interest in this study were the development of late stricture requiring daily dilatations or surgical correction; anastomotic disruption or perforation; and, in the case of patients with HD, the occurrence of severe perineal excoriation and enterocolitis. These results are shown in Table 2. Three patients in the weekly dilatation group (1 HD and 2 ARM) developed late strictures (>3 months after cessation of dilatation). This 10% stricture rate is comparable to previously reported data. Interestingly, none of the patients who required conversion to daily dilatations developed "late strictures." One of the ARM patients who developed a late stricture required operative repair. Within the daily dilatation group, 2 patients (both ARM) developed late strictures; but neither developed further complications. Within the ARM group, there was no difference between the 2 samples with regard to the use of

	HD daily $n = 17$	HD weekly $n = 17$	P	ARM daily $n = 48$	ARM weekly $n = 13$	P
Late narrowing requiring dilatation	0%	6%	.31	4%	15%	.18
Late narrowing requiring surgery	6%	6%	1.00	2%	8%	.38
Anastomotic disruption or perforation	0%	6%	.31	2%	0%	.60
Severe perineal excoriation	12%	0%	.15			
>2 episodes of enterocolitis	18%	12%	.63			

colostomy. This is significant because proximal diversion may have allowed improved healing of the stricture and thus better outcomes. There was also no difference between the groups with respect to percentage with high malformations or use of laparoscopic approach.

There were 2 patients with anastomotic disruptions: 1 HD patient in the weekly group and 1 ARM in the daily group. Both were early disruptions likely related to use of the dilator, without any preceding evidence of stricture, and were treated operatively with repair and the creation of a loop colostomy. Neither had any further strictures or leaks following colostomy reversal. Mean length of follow-up was $16~(\pm 15)$ months in the daily dilatation group and $13~(\pm 9)$ months in the weekly calibration group. Three patients (4.6%) in the former group had long-term difficulties. One developed a recurrent rectourethral fistula, one had ongoing feeding and toileting issues, and the last required revision of the anastomosis owing to mechanical obstruction. One patient (3.3%) in the weekly calibration group required an emergency laparotomy for peritonitis following colostomy closure.

3. Discussion

Although ARMs and HD are distinct congenital abnormalities, both require surgical correction using one of a variety of procedures. Among the postoperative complications of these operations, anastomotic stricture or leak is one of the most difficult to manage. In some cases, an initial leak will lead to a subsequent stricture. There are a number of potential causes for anastomotic complications, including inadequate blood supply, excessive tension, technical error in suture placement, or physical injury to the anastomosis in the postoperative period. In addition, children with HD undergoing endorectal pull-through may develop narrowing because of the persistent rectal cuff. Attempts to minimize this complication have included splitting of the cuff and use of a very short cuff [12]. To prevent these anastomotic complications, most pediatric surgeons have recommended daily dilatation of the anastomosis by the parents. Our data, however, failed to document any benefit to this approach over an alternative protocol in which the surgeon calibrates the anastomosis on a weekly basis, with daily dilatation reserved for children in which the anastomosis is felt to be narrowing.

What are the reasons not to have parents perform daily dilatations? Firstly, there is concern that the parent, not being experienced with this technique, might accidentally perforate or disrupt the anastomosis. This is a rare event; and in our series, there was only one perforation in each of the groups. However, the second problem with the use of daily dilatations by the parents is the psychological stress for both parent and child. There is evidence that invasive anal procedures in children may affect them and their caregivers in a negative way [13,14]. Several studies have shown that patients who underwent prolonged daily dilatation over 2 to 4 years had more mental health and psychosocial issues than the general population [13]. In a study of ARM patients, anal dilatation became a chronic stressor for children and parents alike [14], with parents feeling guilty having to inflict pain on the child by anal dilatations and the dilatation creating a power struggle [13,14]. Both mental health and psychosocial functioning correlated significantly with duration of anal dilatation [13-15], and mental and psychosocial problems were high in comparison to those for the general child and youth population [13,14]. Of note, when patients had invasive procedures performed in the hospital setting by the medical professionals, parents were in a supportive role with respect to their children and thus were still cast in the role of protectors [14]. The aforementioned studies were conducted in older children who had been dilated at varying ages. The effect of dilatations during infancy remains to be proven. There is, however, no question that parents experience stress from doing daily dilatations in their children. The only way to justify this practice would be if there was clear evidence that it was beneficial in preventing anastomotic or cuff narrowing. The only potential advantage to daily dilatation over weekly is the extra cost incurred by the family for weekly trips to the surgeon's office.

This study is limited by its retrospective nature and small sample size but raises interesting questions regarding the ideal postoperative management of this patient population. Our results failed to document any difference in postoperative complications between the patients who underwent weekly calibration by the surgeon compared with those who received daily dilatation by the parents. Specifically, there was no significant difference in the development of late strictures or anastomotic leaks; and there was no increase in the incidence of severe perineal excoriation or enterocolitis in the children with HD. The latter is especially important

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because enterocolitis is one of the most serious complications of HD and it has been suggested that more regular dilatation may be important in prevention [16,17].

In summary, our data suggest that there is no difference in outcomes between weekly calibration by the staff surgeon and daily dilatation by the parents. Given the psychological stress placed on the family and the child by the daily dilatation regime, it is our belief that a weekly calibration approach should be seriously considered for routine postoperative management in these children.

References

- Levitt MA, Peña A. Anorectal malformations. Orphanet J Rare Dis 2007;2:33.
- [2] Langer JC. Repeat pull-through surgery for complicated Hirschsprung's disease: indications, techniques and results. J Pediatr Surg 1999;34:1136-41.
- [3] Langer JC, Minkes RK, Maxxiotti MV, et al. Transanal one-stage soave procedure for infants with Hirschsprung's disease. J Pediatr Surg 1999;34:148-52.
- [4] Langer JC, Durrant AC, de la Torre L, et al. One-stage transanal soave pull-through for Hirschsprung disease: a multicenter experience with 141 children. Ann Surg 2003;238:569-76.
- [5] Ruttenstock E, Puri P. Systematic review and meta-analysis of enterocolitis after one-stage transanal pull-through procedure for Hirschsprung's disease. Pediatr Surg Int 2010;26:1101-5.
- [6] Rouzrokh M, Khaleghnejad AT, Mohejerzadeh L, et al. What is the most common complication after one-stage transanal pull-through in infants with Hirschsprung's disease? Pediatr Surg Int 2010;26:967-70.

- [7] Sookpotarom P, Vejchapipat P. Primary transanal Swenson pullthrough operation for Hirschsprung's disease. Pediatr Surg Int 2009;25:767-73.
- [8] Teitelbaum DH, Ciley RE, Sherman NJ, et al. A decade of experience with the primary pull-through for Hirschsprung disease in the newborn period: a multicenter analysis of outcomes. Ann of Surg 2000;232: 372-80
- [9] Kim AC, Langer JC, Pastor AC, et al. Endorectal pull-through for Hirschsprung's disease—a multicenter, long-term comparison of results: transanal vs. transabdominal approach. J Pediatr Surg 2010;45:1213-20.
- [10] Dutta HK. Clinical experience with a new modified transanal endorectal pull-through for Hirschsprung's disease. Pediatr Surg Int 2010;26:747-51.
- [11] Peña A, Devries PA. Posterior sagittal anorectoplasty: important technical considerations and new applications. J Pediatr Surg 1982;17: 796-811.
- [12] Nasr A, Langer JC. Evolution of the technique in the transanal pullthrough for Hirschsprung disease: effect on outcome. J Pediatr Surg 2007;42:36-9.
- [13] Diseth TH, Emblem R, Solbraa IB, et al. A psychosocial follow-up of ten adolescents with low anorectal malformations. Acta Paediatr 1994;83:216-21.
- [14] Diseth TH. Dissociation following traumatic medical treatment procedures in childhood: a longitudinal follow-up. Dev Psychopathol 2006;18:233-51.
- [15] Diseth TH, Egeland T, Emblem R. Effects of anal invasive treatment and incontinence on mental health and psychosocial functioning of adolescents with Hirschsprung's disease and low anorectal anomalies. J Pediatr Surg 1998;33:468-75.
- [16] Obermayr F, Szavay P, Beschorner R, et al. Outcome of transanal endorectal pull-through in patients with Hirschsprung's disease. Eur J Pediatr Surg 2009;19:220-3.
- [17] Teitelbaum DH, Qualman SJ, Caniano DA. Hirschsprung's disease. Identification of risk factors for enterocolitis. Ann Surg 1988;207: 240-4.