

# STROBE-anastomotic leakage after pull-through procedure for Hirschsprung disease

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## Abstract

This study was undertaken to explore the causes, diagnosis, and treatment of anastomotic leakage after pull-through (PT) procedure for Hirschsprung disease (HD).

A retrospective analysis of patients with anastomotic leakage after a PT procedure in the General Surgery Department of Beijing Children's Hospital from July 2013 to June 2016 was undertaken. The surgical characteristics, diagnosis, and treatment were retrospectively analyzed.

Among the 213 patients who underwent PT procedures, 5 patients had a documented anastomotic leakage. The median age of these 5 patients at the time of the PT procedure was 6.8 years old, and this was higher than those without anastomotic leakage (1.7 years old). In all patients, rectal examination in the lithotomy position revealed an anastomotic dehiscence at the 6 o'clock position. The abdominal ultrasonography demonstrated retrorectal pneumatosis with or without an abscess in 4 patients. All patients were treated with ileostomies and anastomotic resuturing. The median delay to management was 4 days (range: 1–29 days). Four patients (4/5, 80%) were cured, and 1 (delay, 29 days) of these 4 patients developed postoperative ileus. The remaining patient (delay, 9 days) was required to undergo a repeat PT procedure. For the 4 cured patients, the median follow-up time was 20 months (range: 15–37 months), and these patients defecated 3 times daily at most without soiling.

Older children with HD might be prone to anastomotic leakage. The findings of the rectal examination and ultrasonography were distinctive and useful for the diagnosis. Early ileostomy and resuturing of the anastomosis could be used to treat anastomotic leakage.

**Abbreviations:** CT = computed tomography, HD = Hirschsprung disease, PT = pull-through.

**Keywords:** anastomosis resuturing, anastomotic leakage, Hirschsprung disease, ileostomy, rectal examination

## 1. Introduction

Anastomotic leakage is one of the most serious complications after a pull-through (PT) procedure in patients with Hirschsprung disease (HD). The reported incidence of anastomotic leakage varies from 1.3% to 8.0%.<sup>[1–3]</sup> Some studies have reported that anastomotic leakage occurs frequently in children with late-diagnosed HD.<sup>[4,5]</sup> However, few studies have introduced early diagnostic methods. If anastomotic leakage is treated incorrectly, it would seriously affect the quality of life of patients, or even become life threatening.<sup>[6]</sup> In most studies, when patients developed early postoperative anastomotic leakage, enterostomy was considered and performed.<sup>[4,5,7–9]</sup>

Merely 1 study reported that the resuturing of the anastomosis was performed for treating anastomotic leakage.<sup>[10]</sup> After enterostomy, some patients would require another treatment, or even need to undergo a PT surgery. The present study aims to determine the causes and diagnosis of anastomotic leakage, and discuss the combined use of ileostomy and anastomosis resuturing for treating anastomotic leakage.

## 2. Materials and methods

A retrospective analysis of patients with anastomotic leakage after a PT procedure in the General Surgery Department of Beijing Children's Hospital from July 2013 to June 2016 was undertaken. The medical records of these patients were retrospectively analyzed with respect to the PT approach, diagnosis, treatment, and surgical outcomes of the anastomotic leakage. The patients and their guardians were contacted by phone or interviewed in the Outpatient Department to obtain information regarding the postoperative bowel movements of these patients. This study was conducted in accordance with the Declaration of Helsinki, and was approved by the Ethics Committee of our hospital. A written informed consent was obtained from each participant.

All patients with anastomotic leakage underwent ileostomies and resuturing of the anastomoses after confirmation of the anastomotic leakage. The resuturing was performed as follows. The patient was placed in the lithotomy position with the buttocks elevated. A thyroid retractor was used to open the anus. A standard catheter was inserted behind the retracted PT bowel. Diluted iodophor was injected by the catheter to irrigate the

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**Table 1****Details of pull-through procedures.**

Case no.	Numbers of PT	PT approach	Age at PT, y	Aganglionic segment
1	1	Transanal Soave with assistance of laparotomy	5.2	Rectum
2	1	Transanal Soave	6.8	Rectum
3	2	Transanal Soave with assistance of laparotomy	9.6	Rectosigmoid
4	1	Transanal Soave with assistance of laparotomy	7.7	Rectum to transverse colon
5	1	Transanal Soave	6.4	Rectosigmoid

PT = pull-through.

**Table 2****Presenting symptoms of anastomotic leakage.**

Case no.	Presenting symptoms
1	Fever, abdominal pain, bloody stools, sacrococcygeal pain
2	Fever, vomit, abdominal distention
3	Fever, abdominal pain, bloody stools, sacrococcygeal pain
4	Fever, abdominal pain, bloody stools, sacrococcygeal pain
5	Fever, abdominal pain, sacrococcygeal pain

abscess behind the PT bowel. Then, the anastomotic dehiscence was resutured using 3-0 or 4-0 Vicryl.

The treatment was confirmed to be satisfactory, and the stoma was closed when the patient met these 2 criteria: rectal examination: the examiner's finger could be smoothly inserted into the rectum, the anastomosis was smooth without stricture or dehiscence, and on removal of the examiner's finger, no blood was present on the fingertip and barium enema: the contrast agent did not spill from the bowel.

### 3. Results

#### 1.1. Surgical characteristics of patients

Among the 213 patients who underwent PT procedures, 5 patients had a documented anastomotic leakage. The 5 patients who underwent the Soave PT procedure were all male. Four of these 5 patients underwent surgery in our hospital, and the remaining patient was transferred to our hospital 1 month after surgery in another hospital. The incidence of anastomotic leakage in our hospital was 1.9% (4/212). Patient 3 underwent 2 PT procedures. The first procedure was a laparoscopic-assisted Duhamel PT procedure, which was performed in another hospital. A redo of the PT procedure was performed in our hospital due to postoperative constipation. Patient 4 had a long aganglionic segment. Because of misdiagnosis, the dilated segment, including the ileocecum and right hemicolon, was resected in other hospital. The age of these 5 patients at the time the PT procedure was performed ranged within 5.2 to 9.6 years old, with a median age of 6.8 years old. The age of these patients

was higher than the 208 patients without anastomotic leakage (1.7 years old, 1 month to 17.3 years old). Information on the PT procedures is summarized in Table 1.

#### 1.2. Clinical manifestations

The clinical presentations of patients included fever, abdominal pain, abdominal distention, vomiting, bloody stools, and sacrococcygeal pain (Table 2). The median interval between the PT procedure and onset of symptoms was 7 days (range: 1–12 days). Rectal examination in the lithotomy position revealed anastomotic dehiscence at the 6 o'clock position in all patients, and blood was present on the fingertip. Abdominal palpation revealed signs of peritonitis in all patients. At 5 days after the PT procedure, patient 1 had a retrorectal abscess of approximately 5.5 × 5.0 × 3.9 cm, as revealed by ultrasonography, and the hemoglobin concentration of this patient dropped from 110 g/L preoperatively to 74 g/L. However, after 7 days, repeated abdominal ultrasonography revealed pneumatosis with an abscess. Details regarding the anastomotic leakage in all patients are presented in Table 3.

#### 1.3. Treatment

All patients were treated with ileostomies and resuturing of the anastomoses. The median delay to management was 4 days (range: 1–29 days).

#### 1.4. Outcomes of surgery

The anastomoses were examined by rectal examination at 2 or 3 weeks after the ileostomies. Three patients had anastomotic dehiscences, among which, 2 had minor anastomotic dehiscences (<1 cm) in the back wall of the anastomoses. However, 3 months later, these minor dehiscences healed without stricture or further therapy. A barium enema was performed for all patients at 3 months, postoperatively. The contrast agent did not spill in 4 patients, but spilled in 1 patient (Fig. 1), and this patient continued to have anastomotic dehiscences, as determined by rectal examination. Therefore, the stoma was closed in the 4 patients who were cured, whereas the other patient (delay, 9

**Table 3****Details of anastomotic leakage.**

Case no.	Symptoms (post-PT/ds)	Ultrasonography	Stoma (post-PT/ds)	Interval between stoma and closure, mo
1	12	Pneumatosis with abscess	13	4
2	1	Not done	30	28
3	8	Pneumatosis	17	No closure
4	7	Pneumatosis with abscess	11	13
5	7	Pneumatosis with abscess	8	4

PT = pull-through.



**Figure 1.** Barium enema. A lateral radiograph shows that the contrast agent spilled into the presacral space. The arrow indicates the overflow contrast agent.

days) who was not cured required another PT procedure. The success rate of the ileostomies and resuturing of the anastomoses was 80%.

The median interval between the stoma formation and closure in 4 patients was 8.5 months (range: 4–28 months). The postoperative course was uneventful in 3 of these patients. The remaining patient (delay, 29 days) who developed adhesive ileus after closure of the stoma was cured after gastrointestinal decompression and the administration of parenteral nutrition for approximately 20 days.

The 4 patients with stoma reversal were followed up for a median of 20 months (range: 15–37 months). These 4 patients had no more than 3 bowel movements per day, with good fecal continence and no soiling.

#### 4. Discussion

HD is a common abnormality in children. Its incidence is significantly higher in men than that in women. Furthermore, the Soave PT procedure has been widely used in China in recent years. Therefore, the fact that all 5 patients with anastomotic leakage in the present study were men and underwent the Soave PT procedure was not unexpected.

##### 1.5. Reasons for anastomotic leakage

The incidence of anastomotic leakage in our hospital was 1.9%, and this was comparable to other studies.<sup>[1–3]</sup> In the present

study, 5 patients had anastomotic leakage, and the median age of these patients at the time of the PT procedure was 6.8 years old. The age of the patients was higher than patients without anastomotic leakage (1.7 years old) in our hospital. Hence, the investigators considered that older children with HD might be prone to anastomotic leakage, which was also supported by other studies. Stensrud et al<sup>[4]</sup> considered that early postoperative complications, especially anastomotic leakage, occur frequently in children with late-diagnosed HD. Levitt et al<sup>[5]</sup> reported that 2 patients had postoperative anastomotic leakage: 1 was diagnosed at 5 years old, whereas the other was diagnosed at 7 years old. Both patients had a severely dilated proximal bowel. Meanwhile, Stensrud et al<sup>[4]</sup> considered that complications, especially complications correlated to anastomotic dehiscence, appear to occur more frequently in patients with HD, who underwent operations after 3 years old, when compared with neonates and toddlers. In the present study, all 5 patients were older than 3 years at the time of the PT procedure. A plausible explanation for the leakage in patients with late-diagnosed HD may be problems associated with the severity of the dilated colon.<sup>[11]</sup> The bowel proximal to the aganglionic bowel, which tends to be severely dilated, increased the difficulties encountered during dissection, especially in patients who have a short aganglionic segment and underwent procedures solely via the transanal approach. Furthermore, older children have a deeper pelvis and larger mesenteric vessels, when compared to younger children, making the dissection more difficult. All of these factors may lead to inadequate hemostasis, which in turn may result in the

development of postoperative hematoma and secondary infection. In the present study, this process was verified in patient 1, who developed postoperative bleeding, retrorectal abscess, and subsequent anastomotic leakage. Furthermore, a severely dilated colon contributes to difficult dissection, and anastomosis in congruence, when not resected.<sup>[11]</sup>

The incidence of anastomotic leakage after the redo PT procedure is higher. Dingemans et al<sup>[12]</sup> reported that 3 of 16 patients (19%) who underwent a redo PT procedure developed postoperative anastomotic dehiscence. In the present study, patient 3 underwent 2 PT procedures, whereas patient 2 developed anastomotic leakage at the first day after the PT procedure. It was considered that this might have been correlated to the inaccurate placement of anastomotic sutures by the surgeon.

It was found that all 5 patients had anastomotic dehiscence at the 6 o'clock position. Furthermore, some adult patients developed presacral sinus secondary to anastomotic leakage after the colorectal surgery.<sup>[13,14]</sup> Their positions were similar to ours. In the present study, this may be correlated to the following factors. First, the reason for the anastomotic leakage at the 6-o'clock position may have been associated with the weakness of the tissue, which may have been correlated to the excessive splitting of the muscular cuff. Second, patients were maintained in the supine position after the PT procedure, allowing the 6-o'clock position to be the lowest point. This would allow liquid or blood to easily gather in this point, making the anastomotic leakage more likely to occur at this point.

### 1.6. Diagnosis

The accurate and timely diagnosis of anastomotic leakage after a PT procedure in patients with HD is very important. Because of the anastomotic leakage, intestinal contents enter the presacral space through the dehiscence and peritoneal cavity by peritoneal reflection, leading to fever, abdominal pain, abdominal distention, bloody stools, vomiting, and sacrococcygeal pain. The median time of symptom onset was 7 days after the PT procedure in the present study. Lu et al<sup>[2]</sup> reported that the anastomotic leakage occurred at <1 week, postoperatively, and its mean time was 5.7 days. Therefore, when patients develop these referring symptoms during the first week after the PT procedure, anastomotic leakage should be suspected. Hence, rectal examination should be immediately performed, because it can gain information relating to the routine healing of these anastomoses.<sup>[10]</sup> In addition, the diagnosis of leakage can be verified through radiological study, such as contrast enema and computed tomography (CT).<sup>[15]</sup> CT with contrast enema is accurate for diagnosing postoperative colorectal anastomotic leakage. Contrast extravasation is the most reliable sign.<sup>[16]</sup> In the present study, ultrasonography examination was used, and the specific manifestation was retrorectal pneumatosis with or without an abscess. A perianastomotic collection containing liquid and air was the only feature significantly more common in patients with a clinically important leak.<sup>[17]</sup>

### 1.7. Treatment

The early treatment of anastomotic leakage after a PT procedure in patients with HD is very important, to avoid more serious sequelae. If patients develop early postoperative anastomotic leakage, enterostomy should be considered.<sup>[4]</sup> It has been reported that colostomy was used to treat the anastomotic

leakage after a PT procedure.<sup>[7-9]</sup> Furthermore, enterostomy is essential for patients with postoperative anastomotic leakage for 2 main reasons. First, if patients who develop anastomotic leakage and have signs of peritonitis are not treated with enterostomy, they would develop life-threatening infectious toxic shock and other fistulas, or the retraction of the PT bowel. Quan et al<sup>[18]</sup> reported that 1 patient died of severe abdominal infection due to anastomotic leakage. Second, pelvic dense fibrosis can result from a previous PT procedure, which is more commonly caused by its complications (anastomotic leakage or dehiscence).<sup>[19]</sup> These pelvic dense fibrosis increases the difficulty of redo PT procedures. All 5 patients in the present study were treated with enterostomies at the early stage. The time when the enterostomy is performed is very important. The median delay to management in the present study was 4 days (range: 1–29 days). If this delay is long, it would affect the prognosis. First, a long delay could result in failure of anastomotic resuturing. Patient 3 had a delay of 9 days, and the anastomotic leakage failed to heal after ileostomy and resuturing of the anastomosis. Second, a long delay could lead to severe intra-abdominal inflammation and adhesion. Patient 2 had a delay of 29 days, and developed adhesive ileus after stoma closure.

Surgeons choose either colostomy or ileostomy as the enterostomy technique. The investigators have previously proposed that an ileostomy may be a good choice to retain a longer colon and avoid increasing the difficulty of the second PT procedure.<sup>[20]</sup> A loop ileostomy has a number of advantages over colostomy.<sup>[21]</sup> Ileostomies were chosen for all 5 patients in the present study.

The resuturing of the anastomosis was performed at the time of the enterostomy for all 5 patients, and 4 of these patients (80%) were cured. Jester et al<sup>[10]</sup> considered that rectal examination under general anesthesia enabled the surgeon to perform a thorough examination of the anastomosis, offering the possibility of reconstructing the anastomosis within the same operation. The 2 key points of resuturing the anastomosis are as follows: the retrorectal abscesses should be thoroughly eliminated. If a retrorectal abscess exists, it would affect the healing of the anastomosis and the recovery of systemic symptoms. The anastomotic dehiscence should be resutured. The PT bowel is fragile and could be easily torn due to inflammation. Thus, the anastomosis should be resutured using 3-0 or 4-0 Vicryl, instead of a too-thin absorbable suture. The early rectal examination after resuturing revealed that 2 patients had minor dehiscence (<1 cm) in the posterior wall. However, 3 months later, it is discovered that the dehiscence had healed. Although the resuturing of the anastomosis cannot ensure primary healing, it can prevent the further retraction of the PT bowel. As long as the retraction of the PT bowel is not severe, the anastomosis can heal after resuturing. Jester et al<sup>[10]</sup> reported 2 patients with symptomatic anastomotic leakage, in which one patient was cured by ileostomy without further problems, whereas the other patient was cured by resuturing of the anastomoses without any complications. In the present study, 2 methods were chosen at the same time. The success rate of the ileostomies and resuturing of the anastomoses was 80%. However, it was reported that most patients with anastomotic leakage were treated with enterostomy, and the reported median success rate was 50% (25%–100%)<sup>[4,5,7,8,22,23]</sup> (Table 4). When compared to studies on enterostomy, the combined ileostomy and resuturing of the anastomosis was a better and effective method.

The early resuturing of dehiscence might be helpful to prevent hazardous sequelae.<sup>[10]</sup> In the present study, 3 patients were resutured within 2 weeks, postoperatively, whereas the other 2 patients were resutured at >2 weeks (17 and 30 days,



**Table 4**

**The reported number of patients and success rate for prognosis of anastomotic leakage.**

	Patients	Success rate, %
Fortuna et al <sup>[23]</sup> (1996)	4	25
Wilcox et al <sup>[8]</sup> (1997)	2	50
Liem et al <sup>[22]</sup> (2008)	4	75
Stensrud et al <sup>[4]</sup> (2012)	2	50
Levitt et al <sup>[5]</sup> (2013)	2	50
Shahjahan et al <sup>[7]</sup> (2014)	4	100

respectively). Merely 1 patient was not cured. This patient underwent 2 PT procedures, whereas the other 4 patients only underwent 1 procedure. Although the time of resuturing can influence the healing of the anastomosis, this factor is not absolute. The number of PT procedures is also an important factor that influences the healing of the anastomosis. More PT procedures and severe scarring can affect the healing of the anastomosis.

Once the acute medical conditions improves and after the anastomosis has been proven to be healed, the stoma can be reversed.<sup>[24]</sup> Ileostomy closure (<3 months) is practical and safe.<sup>[25]</sup> However, in the present study, the median interval between stoma formation and closure was 8.5 months (range: 4–28 months). Furthermore, there are few studies on stoma closure in HD patients with anastomotic leakage. Early closure before 10 weeks of formation for necrotizing enterocolitis patients is associated with significant morbidity.<sup>[26]</sup> According to the experience of the investigators, when the healing of an anastomotic leakage was confirmed without severe abdominal adhesion, the most appropriate interval was 3 to 4 months.

In summary, the present study found that older children with HD might be prone to anastomotic leakage. Patients who underwent the combined ileostomy and resuturing of the anastomosis presented a better curative effect for the treatment of anastomotic leakage after a PT procedure for HD, when compared to patients who received enterostomy in other studies. The limitation of the present study was that there were only 5 cases due to the low incidence of anastomotic leakage, and no statistical analysis was conducted. Further prospective and contrastive studies may be needed to explore other approaches for treating anastomotic leakage.

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