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The anal canal is the fine line between "fecal incontinence and colitis" after a pull-through for Hirschsprung disease

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ABSTRACT

Background/purpose: Fecal incontinence after a pull-through is associated with different factors, although damage to the anal canal seems to be the most important. The objective of this article is to identify the variables related to the presence of fecal control and colitis in a homogeneous group of children after pull-through.
Methods: A retrospective cross-sectional study was performed in patients with HD for evaluation of post-operative problems from May 2014 to November 2016. The patients (39) had a transanal approach and were divided into two groups: Group 1 patients with fecal continence and Group 2 patients with fecal incontinence.
Results: Group 1 patients (13) had the anastomosis in the rectum, no damage to the anal canal, and a positive history of colitis. Group 2 (26) had the anastomosis at the skin, anoderm, pectinate line, or a combination of these and a negative history of colitis.
Conclusions: We demonstrated that patients with a technical error in the anastomosis have fecal incontinence, but not colitis. Preservation of the anal canal is associated with fecal control and colitis because it is a high-pressure zone. Education for proper identification of the anal canal during a pull-through is an absolute necessity.
Type of study: Retrospective Comparative Study.
Level of evidence: Level III.

The rectum is always at least partially aganglionic in Hirschsprung's Disease. Thus, the surgical treatment requires either a proctectomy [1], a rectal mucosectomy [2], or a bypass with preservation of the rectum [3]. When surgeons decide on a rectal mucosectomy or proctectomy, they can initiate these procedures through either an abdominal [1,2] or transanal approach [4,5]. A fully ganglionated bowel segment is then anastomosed to the rectum, 2 cm above the upper limit of the pectinate zone preserving the anal canal. [6]. The ideal outcome after a well performed pull-through for Hirschsprung's is regular bowel movements with fecal continence. However, some patients may have constipation or colitis.

Most patients with Hirschsprung disease possess all of the anatomical elements necessary to achieve fecal continence after pull-through. Thus, fecal incontinence should not occur after a pull-through. When it happens, it can be a devastating problem for the patient and the family [7]. In the literature, some degree of fecal incontinence can occur in anywhere from zero to 88% of cases [8]. Multiple factors have been involved in the etiology of the fecal incontinence, although damage to the anal canal seems to be the most important [9–12], making it a "preventable, irreparable, and irreversible complication".

Before the 1990's, Swenson and Soave operations started in the pelvis, using a laparotomy. Clear identification of the anal canal and the position of the anastomosis in the rectum 2 cm proximal to the end of the pectinate area was an essential step during these procedures. Clearly defining the anatomy was facilitated by prolapsing the rectum or the rectal mucosa through the anus [1,2] (Fig. 1). When surgeons subsequently moved to the transanal approach, they then needed to begin the dissection starting in the opposite direction, without prolapse of the rectum [4,5,13–15].

We thus hypothesize that during the transanal approach, an inaccurate identification of the level of the anastomosis and failure to protect the entire anal canal and 2 cm of the rectum can result in catastrophic consequences in postoperative fecal control.

This article discusses the variables that dictate the degree of fecal control in an otherwise homogeneous group of children undergoing an endorectal pull-through with transanal mucosectomy.

1. Methods

A cross-sectional retrospective study approved by our Institutional Review Board was performed (PRO 15030254). We included patients

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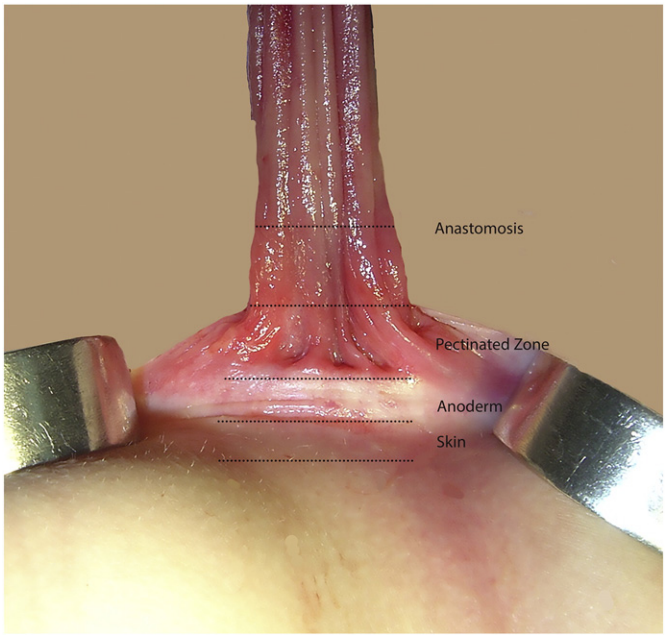

Fig. 1. Rectal prolapse during an open Soave procedure showing the limits of the anal canal consisting of the anoderm and the pectinate zone. Also, it shows the limit of the skin and the anoderm, and the proposed line of the anastomosis.

with Hirschsprung disease referred to the Colorectal Center for Children at Children's Hospital of Pittsburgh between May 2014 and November 2016. All patients were referred because of problems after the pull-through. We included only patients with aganglionosis of the rectum and rectosigmoid with a history of a Soave pull-through in whom the surgeons performed a transanal mucosectomy. We excluded patients with syndromic Hirschsprung or with long segment or total colonic aganglionosis. All patients underwent our "protocol for patients with Hirschsprung and postoperative functional problems" which includes the following: (1) History and physical. (2) Review of the medical record-when the pull-through was performed in another institution, we obtained consent from the parents for the release of health information. (3) Contrast enema performed without bowel preparation, discontinuing any medical treatment 3 days before the study and administering the contrast by hand injection with low pressure and fluoroscopic control. (4) Anorectal examination under anesthesia to map the line of the anastomosis. To facilitate the identification and record of the line of the anastomosis, we created a diagram (Fig. 2). (5) Biopsy of the bowel used for the pull-through. Once the protocol was completed, the patients began on our Bowel Management Program. After a minimum follow-up of 6 months, we could determine if the patients possessed fecal control and created two groups: fecal continent (Group 1) and fecal incontinent (Group 2).

Data obtained from medical records included gender, age at referral, diagnosis at referral, age at pull-through, type of pull-through (primary or staged), type of approach (open, laparoscopic or transanal), the length of the aganglionic segment obtained from the pathology report, and any history of colitis after the pull-through. The variables collected from our "protocol of study" included line of anastomosis, the presence of ganglion cells in the rectal biopsy, segment of bowel used for the pull through based on the residual control observed in the contrast enema, and the presence of dilation of the bowel in the pelvis.

1.1. Operational definitions

We defined "fecal continence" when the patient possesses the sensation of the urge of defecation, can differentiate at the anorectal area between gas, formed, and liquid fecal matter, and can hold the bowel movement for an appropriate amount of time to have a voluntary bowel movement when and where it is socially adequate. We defined "fecal incontinence" when the patient does not possess partial or total sensation of the urge of defecation, may or may not differentiate at the anorectal area between gas, formed, and liquid fecal matter, but is not able to hold the bowel movement for an appropriate amount of time to have a voluntary bowel movement when and where it is socially adequate.

We used the IBM SPSS 20.0 software for the statistical analysis. The statistical test included chi-square, Fisher's exact test, ANOVA, Mann–Whitney *U* test, and descriptive statistics where appropriate. A p-value <0.05 was considered significant.

2. Results

From May 2014 to November 2016, 53 patients were referred with problems after the pull-through. We excluded 14 patients. We included 39 patients with recto and rectosigmoid aganglionosis confirmed by contrast enema, operative, and pathology reports. All the patients had undergone a Soave pull-through with transanal mucosectomy. The referral diagnosis was encopresis ($n = 19$), constipation with bouts of colitis ($n = 13$), fecal incontinence ($n = 4$), and chronic diarrhea ($n = 3$). After the protocol of study and bowel management program, the functional diagnosis was fecal continence in 13 patients and fecal incontinence in 26.

2.1. Group 1: Fecal continent

The mean age at referral was 39 months [SD ± 13 months], and the median was 23 months [Range 10 months to 7 years and 6 months]. The referring diagnosis was obstructive symptoms or constipation with bouts of colitis in all patients. The mean age at the pull-through was 136 days [SD ± 151], and the median was 90 days. The mean length of aganglionosis was 11.7 cm [SD ± 4.6], and the median was 11 cm. The line of the anastomosis was in the rectum between 0.3 and 2.2 cm proximal to the pectinate area. The anal canal was preserved in all (Table 2).

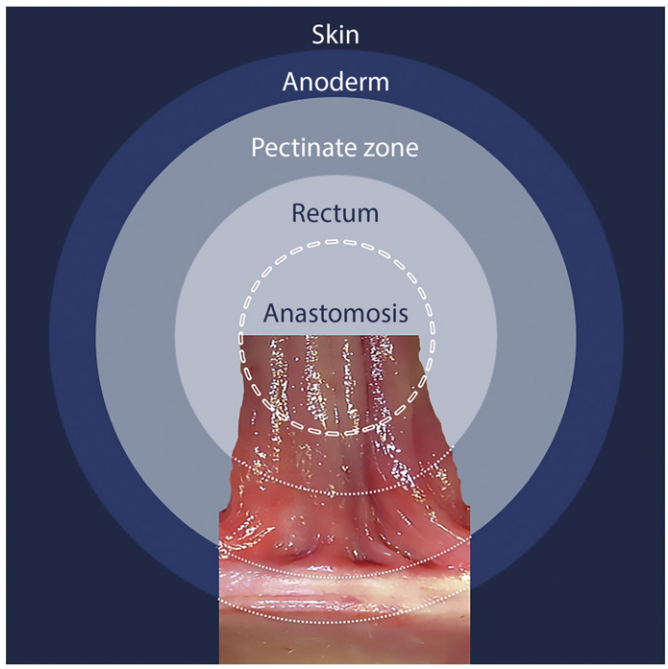

Fig. 2. Diagram used during the anorectal examination under anesthesia to determine the line of the anastomosis. The safe area for the anastomosis is represented with a white circular dotted line.

Table 1
Age at Soave pull-through, the length of the aganglionic segment in centimeters, number of operations, and type of approach of 26 patients with Hirschsprung disease and postoperative fecal incontinence.*

Number of patient	Age at pull-through	Aganglionic segment	Number of operations	Type of Approach
1	7 d	9	Primary	Transanal
2	2 d	Unknown	Primary	Transanal
3	12 d	5.5	Primary	Transanal
4	14 d	4.1	Primary	Transanal
5	7 d	12	Primary	Laparoscopic
6	3 d	22	Primary	Laparoscopic
7	6 d	11	Primary	Laparoscopic
8	4 d	14.5	Primary	Laparoscopic
9	8 m	9	Primary	Laparoscopic
10	24 d	8	Primary	Laparoscopic
11	4 m	9	Primary	Laparoscopic
12	1 m	7.8	Primary	Laparoscopic
13	20 d	Unknown	Primary	Laparoscopic
14	6 d	4	Primary	Laparoscopic
15	5 d	7	Primary	Open
16	7 w	Unknown	Primary	Open
17	3 m	10.5	Two stages	Laparoscopic
18	6 m	8.7	Two stages	Open
19	6 y	7	Two stages	Open
20	4 m	11	Two stages	Open
21	2 m	11	Two stages	Open
22	3 m	10	Two stages	Open
23	10 m	11	Two stages	Open
24	7 m	8.4	Two Stages	Open
25	13 m	6.5	Two Stages	Open
26	7 m	5	Two Stages	Open

* d = days, w = weeks, m = months, y = years.

2.2. Group 2: Fecal incontinent

The mean age at referral was 9 years [SD ± 44 months], and the median was 7 years and 10 months [range: 3 years 9 months to 17 years and 6 months]. The diagnosis at referral was encopresis in 19 patients, fecal incontinence in 4, and chronic diarrhea in 3 patients. Mean age at the pull-through was 219 days [SD ± 504 days], and the median was 39 days. The mean length of aganglionosis was 9.8 cm (SD ± 14.3 cm), and the median was 9 cm. We did not receive the pathology report of the pull-through of three patients (Table 1). The line of the anastomosis was at the same level circumferentially in 11 patients: two at the skin, seven at the anoderm, and two in the pectinate zone. Fifteen patients had an anastomosis that was not at the same level circumferentially: 1 on the skin, anoderm, and pectinate area, 11 on the anoderm and pectinate area, 2 on the anoderm, pectinate area, and rectum, and 1 on the pectinate area and rectum. All patients had an incorrect level of the anastomosis. None of the anastomoses were performed entirely in the rectum with an adequate margin from the pectinate area. The clinical pictures of some of these lines of anastomosis are shown in Fig. 3, and the diagrams recorded during the examination under anesthesia of five patients are shown in Fig. 4. Sixteen patients (61%) had a patulous anus. The bowel in the pelvis was non-dilated. It was narrow with no fecal reservoir in 23 patients, and the bowel in the other 3 patients was mildly dilated: one due to stenosis of the anastomosis and two with residual aganglionosis (Table 2).

2.2.1. Variables with no statistical significance

The gender did not show statistical difference that influenced the fecal control ($p = 0.456$). The surgical procedure with a primary or staged pull-through did not show differences ($p = 0.633$). There was no difference between the two groups when we analyzed the age at the pull-through ($p = 0.531$). There was no difference between the two groups when we analyzed the length of the aganglionosis ($p = 0.217$). The histopathology of the colon was not significant ($p = 0.414$).

2.2.2. Variables with statistical significance

The type of surgical approach showed more incontinence after a laparoscopic procedure ($p = 0.046$). The analysis of the residual colon showed a difference between the two groups ($p = 0.026$). The presence of a non-dilated bowel in the pelvis in patients with fecal incontinence compared with dilated bowel in patients with fecal continence was significant ($p < 0.0001$). The line of the anastomosis in all the patients of Group 1 was in the rectum. In Group 2 the line of the anastomosis was performed distal to the rectum ($p < 0.0001$) All patients with a history of colitis after the pull-through had fecal continence, and the patients with fecal incontinence did not have any history of colitis ($p < 0.0001$) (Table 2).

3. Discussion

Well-ganglionated colon, a well-performed pull-through, and an adequately located anastomosis are the main three elements required to achieve a technically correct operation for Hirschsprung disease. Even so, a group of patients did have postoperative obstructive colitis. When one or more of these elements are flawed, the patients are likely to present with postoperative complications. Fecal incontinence is one of these complications, and it is preventable, irreversible, and irreparable.

The line between colitis and fecal incontinence in the surgical treatment of Hirschsprung disease is very fine. We found in all the patients with fecal control the line of the anastomosis on the rectum, 0.3–2.2 cm from the end of the pectinate zone, which is a good technical operation. All had a history of episodes of postoperative colitis. In contrast, all the patients with fecal incontinence had damage of the anal canal because the line of the anastomosis was incorporated at the pectinate zone, the anoderm, the skin, or a combination of these structures. None of them had postoperative colitis (chi-square $p < 0.0001$), even when five patients had residual aganglionosis or transitional zone (this is discussed in detail below). This relationship between fecal control and postoperative colitis, and fecal incontinence and no postoperative colitis was demonstrated previously [10,11,16].

When we analyzed the surgical approach, we obtained a statistically significant difference between the group of patients with fecal control and fecal incontinence (46% vs. 13% transanal, 23% vs. 50% for laparoscopic and 30% vs. 38% open). Even though the laparoscopic approach

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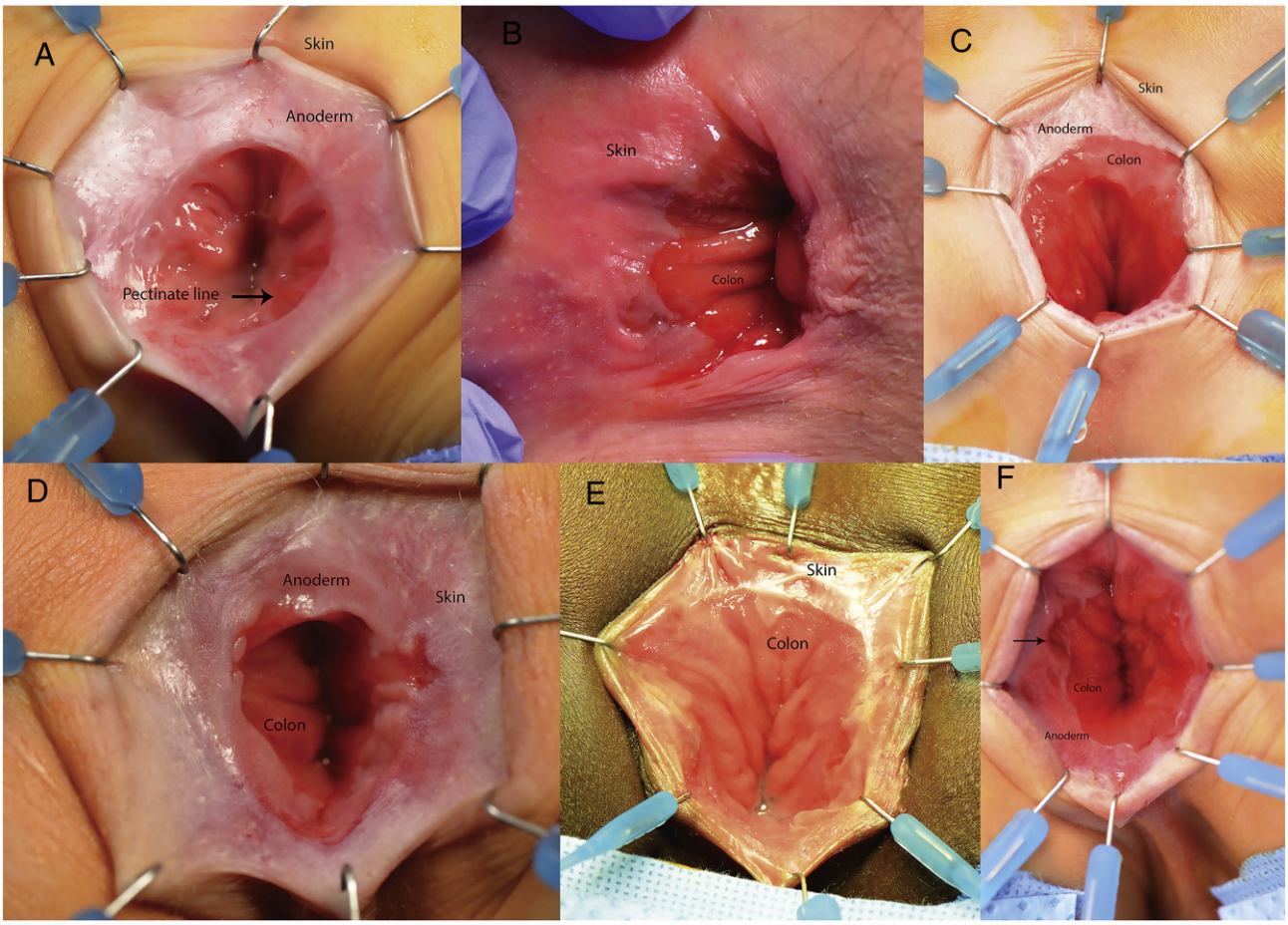

Fig. 3. (A) Normal anal canal obtained from a patient 6 months old after a transanal endorectal pull-through with history of colitis. (B) Anastomosis at the skin. (C) Anastomosis in the anoderm. (D) Asymmetric anastomosis in the anoderm and skin. (E) Anastomosis on the skin. (F) Asymmetric anastomosis on the anoderm. The arrow shows a small area of residual pectinate zone.

showed more patients with fecal incontinence (chi-square $p = 0.046$), we believe that it is not a contributing factor to fecal incontinence.

All patients in this series had aganglionosis of the rectum or rectosigmoid. In the group of fecal continence, the sigmoid and distal descending colon was utilized for the pull-through. However, in the group of patients with fecal incontinence, in three patients due to ischemia of the bowel and in two because of an intraoperative report of transitional zone, the surgeons performed a large resection, then utilizing the transverse colon in four patients and the ascending colon in the other for the pull-through. Again, even though there was a statistical difference (chi-square $p = 0.026$), we believe it is not a contributing factor to fecal incontinence.

A significant finding in the contrast enema between the two groups (chi-square $p < 0.0001$) was the presence of dilation of the colon in the pelvis in the fecal continent group and the absence of dilation in the group with fecal incontinence. These findings have been reported previously [9–11].

In the group with fecal continence all patients came because the history of colitis. We did not find any mechanical obstruction in these patients. Only three patients had residual aganglionosis. We re-operated on these patients performing a transanal Swenson. We could preserve the anal canal in two patients, and they have fecal control. However, in one patient the previous line of the anastomosis was in the rectum at 0.4 cm above the pectinate zone, and we used part of the anal canal for the anastomosis. This patient is fecal incontinent. When there are residual aganglionosis and the anal canal is preserved, but there is a very short rectum, the Duhamel technique is a suitable method for a reoperation to avoid a low anastomosis over the anal canal and produce fecal incontinence [17–20].

In the group of patients with fecal incontinence, the histopathology in the rectal biopsy of the colon anastomosed in the "anal canal" is aganglionic in 3 patients and transitional zone in 2. This residual abnormal histopathology causes persistent obstruction when the anal canal was preserved [21]. However, residual aganglionosis associated with damage of the anal canal does not seem to matter for potential obstructing anatomy. We do not indicate a reoperation in patients with residual aganglionosis and fecal incontinence due to damage of the anal canal, because we will not change the outcome. Other variables with no statistical significance between these two groups were the gender, age at pull-through, the number of operations, and length of aganglionosis (Table 2).

Where is the optimal site for the colorectal anastomosis during the pull-through for Hirschsprung? This important and frequent question has different anatomical references and lengths creating confusion (Table 3). Clearly, the goal is a colorectal anastomosis, so the target should be "the rectum." The importance in preserving the anal canal and 1.5 cm of rectal mucosa to assure fecal continence is that these segments possess great sensitivity, thus allowing the patient to differentiate between solid, liquid, gas, pain, pressure, heat, cold, longitudinal, or rotatory movement [1,22]. Our recommendation is to preserve 2 cm of the rectum. Another cause of fecal incontinence seems to be overstretching of the sphincters, leading to a patulous anus [12]. In our series, we had 16 patients with a patulous anus. It is important to minimize the overstretching of the sphincters during the transanal approach. Our recommendation is to begin the operation with progressive dilation of the anus before placement of the Lone Star Retractor® or other similar retractors. We stop dilations when we achieve two

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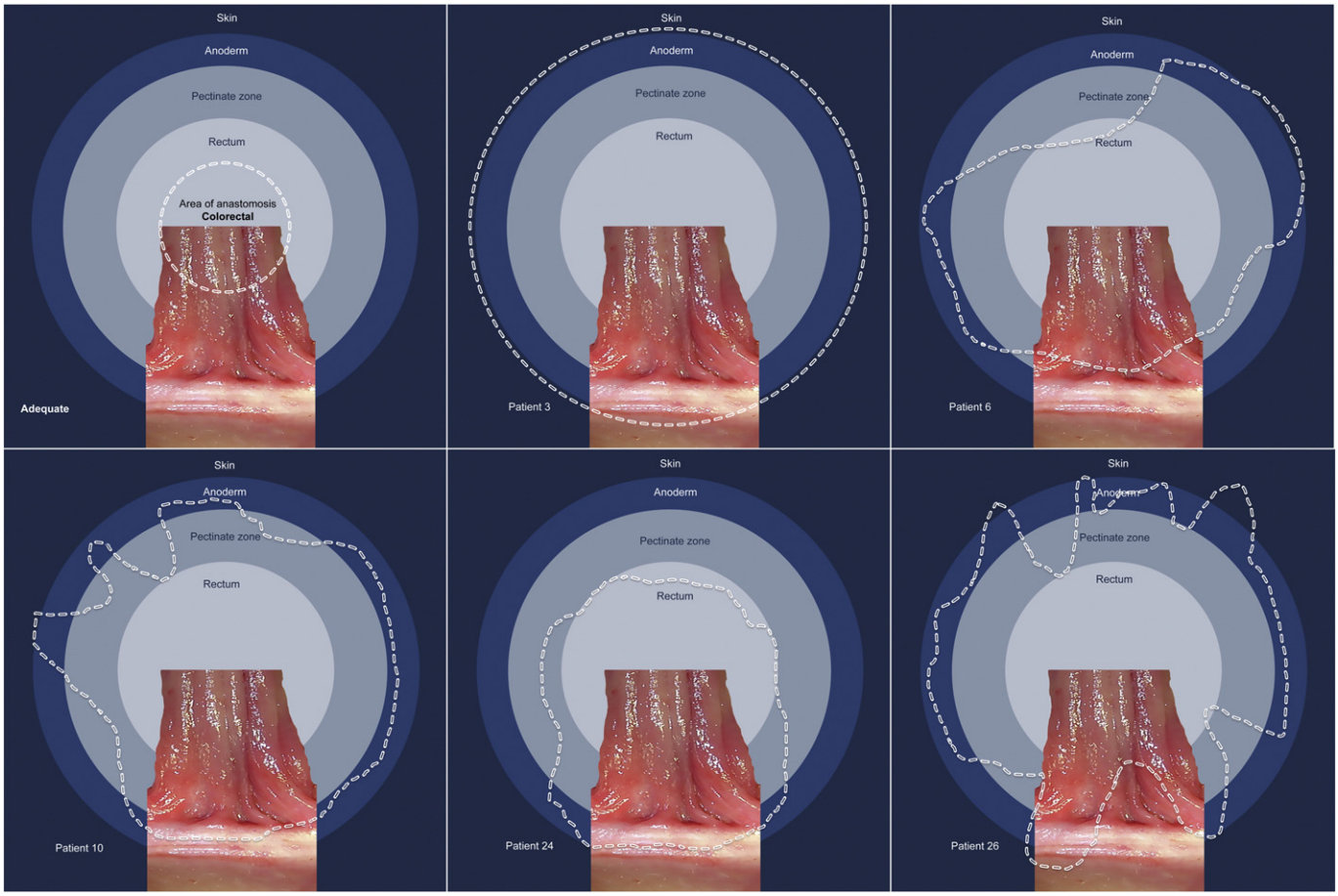

Fig. 4. Diagrams used to record the level of the anastomosis during the anorectal exam under anesthesia. First diagram shows with white dotted line an adequate anastomosis with the proper safe distance from the anastomosis to the pectinate zone. Patient 3 had a circumferential anastomosis on the skin. Patients 6, 10, 24 and 26 showed asymmetric anastomoses to different levels of the anal canal and skin.

Hegar dilators (2 mm) bigger than the number that corresponds to the age of the patient. For example, a newborn will need a Hegar 14. Then, we place four hooks at 3,6,9, and 12 o'clock in the line between the skin and the anoderm. This step permits precise identification of the complete pectinate zone and the rectum. Subsequently, between these first four hooks, equidistantly, we anchor another four hooks on the rectum at 2.2 cm from the end of the pectinate zone. These two extra millimeters permit the placement of the multiple traction sutures and the area for the circular incision in preparation for the anastomosis. Finally, we reposition the first four hooks on the rectum. In the newborn, we place the hooks at 1.2 cm, and it is a very delicate procedure.

We believe that the origin of technical errors during the treatment of Hirschsprung that causes fecal incontinence is the lack of identification of the anal canal and the line of the anastomosis. Identification of these areas was easier when we initiated the Swenson or Soave from the pelvis during the laparotomy, and we could prolapse the rectum. This last step permits the "external visualization" of this area (Fig. 1). In the 1990s the introduction of minimally invasive techniques to treat Hirschsprung

Table 2
Demographics and comparative statistic of the variables analyzed between Group 1 patients with fecal control and Group 2 patients with fecal incontinence.

Endpoint fecal control	Fecal continence (13 patients)	Fecal incontinence (26 patients)	*p* value
Gender (*male–female*)	11–2	20–6	0.456
Age at pull-through (*Range in days*)	4 to 420	2 to 2190	0.531
Primary/2-stages pull-through	8/5	16/10	0.633
Length of aganglionosis (*Range in cm*)	4.5 to 20.7	4 to 22	0.217
Histopathology of the colon (*ganglionated/aganglionosis/transitional zone*)	10/3/0	21/3/2	0.414
Open/Laparoscopic–assisted/transanal	4/3/6	10/13/3	0.046*
Colon used for the pull-through (*Sigmoid/Descending/Transverse/Ascending*)	6/7/0/0	18/3/4/1	0.026*
Radiologic colon in pelvis (Dilated/No dilated)	13/0	3/23	0.000*
Anastomosis on the rectum	13	0	0.000*
Anastomosis on the anal canal and/or skin	0	26	0.000*
History of colitis	13	0	0.000*

Table 3
Different proposed anatomic references and lengths to perform the anastomosis during the pull-through for Hirschsprung disease.

Reference	Year Author	Site of anastomosis
[1]	1948 Swenson	1.5 cm from anal skin
[23]	1949 Swenson	2.5 cm from anal skin margin
[24]	1956 Duhamel	Anocutaneous line
[2]	1964 Soave	1 cm above the mucocutaneous junction in the intranal mucosa
[25]	1977 Martin	1 cm above the mucocutaneous junction
[13]	1993 Rintala	3–5 mm above the dentate line
[15]	1995 Georgerson	5–10 mm above the dentate line
[4]	1998 De la Torre	1 cm above the dentate line on the rectal mucosa
[26]	1999 Langer	approximately 5 mm from the dentate line
[27]	2000 Shankar	3–5 mm above the dentate line
[6]	2002 Swenson	2 cm above the anal canal
[28]	2003 Rintala	3–5 mm above the dentate line
[29]	2011 Hebra	approximately 1.0 cm above the pectinate line
[5]	2013 Levitt	1.0 cm proximal to the dentate line
[30]	2014 Oancea	In neonates and infants at 0.5–1 cm above the dentate line and at 1–2 cm in older children

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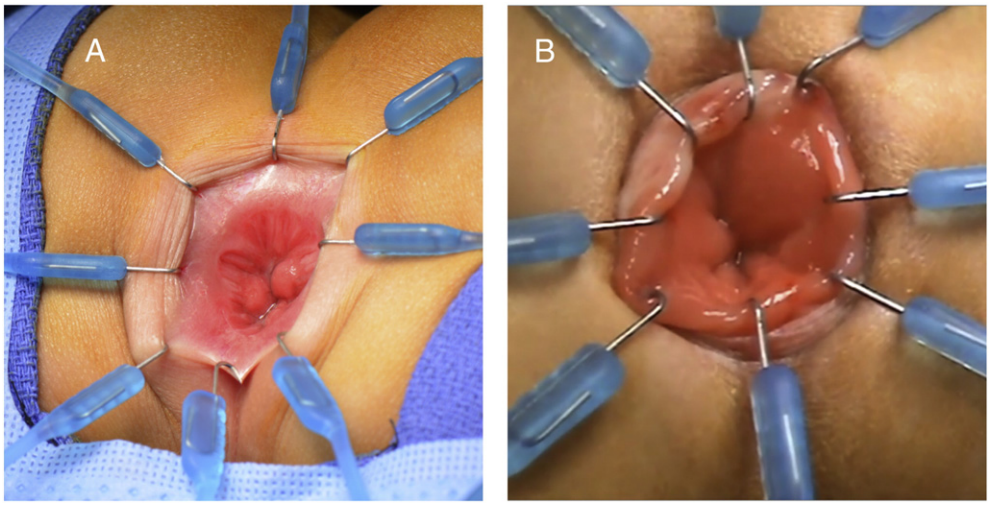

Fig. 5. (A) Internal visualization of the anal canal during a transanal approach, the hooks are anchored between skin and anoderm. (B) Internal visualization of the rectum, the hooks are anchored 2 cm from the end of the pectinate zone.

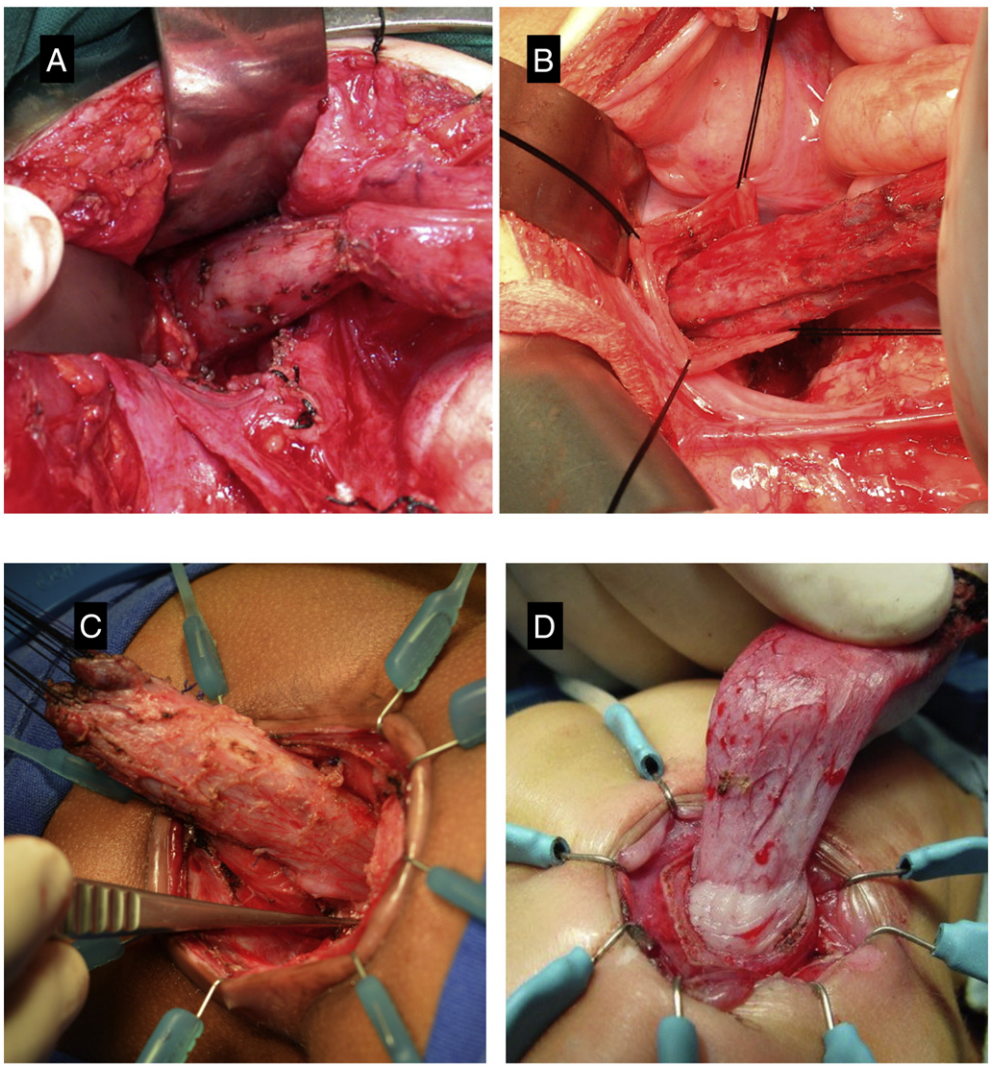

Fig. 6. Intraoperative images of (A) Open Swenson and (B) Open Soave, the dissection begins in the pelvis and ends with the prolapse of the rectum or rectal mucosa [see Fig. 1] (C) Transanal full-thickness (Swenson) and (D) Transanal endorectal (Soave), the dissection begins in the rectum and there is no prolapse of the rectum.

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disease changed the manner of these operations. The transanal endorectal pull-through is one of the more often utilized techniques. This change calls for an "internal visualization", making the identification of these critical structures more challenging (Figs. 5 and 6). Twenty years ago, when surgeons started to do this operation [4], they did not know the potential damage that this technique may produce when performed in a flawed manner. The transanal endorectal mucosectomy or proctectomy require proceeding with extreme care, following all of the recommendations reported in this document and others already published [12].

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