



Error traps and culture of safety in Hirschsprung disease

L. De La Torre^{a,*}, L.A. Wehrli^b

^a Colorectal and Hirschsprung Center, Pediatric Colorectal Surgery, Children's Hospital of Pittsburgh, University of Pittsburgh Medical Center, 4401 Penn Avenue, Pittsburgh, PA 15224, USA

^b Pediatric Colorectal Surgery, Children's Hospital Lucerne, Switzerland

ARTICLE INFO

Keywords:
Hirschsprung
Megacolon
Safety culture
Fecal incontinence
Colitis

ABSTRACT

Hirschsprung disease affects many children every year around the world. Currently, there is an extensive menu of diagnostic methods, and surgical treatments. This situation compels the physicians to follow the rationale of these interventions. The comprehensive diagnosis and treatment of Hirschsprung disease need singular procedures. The clear understanding of how to perform each of these techniques, as well as to read the results is mandatory. Otherwise, the medical team may perform unconscious errors and fall into traps. Many errors still happen in patients with Hirschsprung, resulting in a spectrum of problems; from delayed diagnosis to unnecessary colectomies. In other patients, the damage to the anal canal results in fecal incontinence. When this is established, it is an unreversed and devastating social problem. This article describes why these errors occur and how to prevent them.

© 2019 The Author(s). Published by Elsevier Inc.
This is an open access article under the CC BY-NC-ND license.
(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Error trap #1. False aganglionosis due to inadequate biopsies

The gold standard for the diagnosis of Hirschsprung disease is the proper study of an adequate rectal biopsy. Presumption of a diagnosis of Hirschsprung disease in patients with idiopathic constipation because of a false rectal aganglionosis is not uncommon, because of which these patients may have to undergo unnecessary pull-through or colostomy. The two most common situations causing this error are a small biopsy and an improper histopathological study.

A rectal biopsy sample is obtained using a suction tool or surgically through a transanal approach. The suction rectal biopsy possesses two possible error traps that can end in a diagnosis of false rectal aganglionosis. The first error is to take biopsy sample from the skin, anoderm, or pectinate area, which are regular aganglionic segments. Performing a suction rectal biopsy is a blind procedure. The surgeon introduces the instrument into the rectum to a depth of 1, 2, or 3 cm. However, the specimen could be obtained from a more distal zone (anal region) and not from the rectum. The cause of this proximal specimens is because the surgeon or gastroenterologist does not consider the length of the anal canal (1–2 cm from the skin) or stretches the rectal mucosa while introducing the instrument. The second possible error trap is obtaining

a superficial biopsy sample without enough submucosa. Reports from such biopsies could be incorrectly read as “aganglionic,” “Hirschsprung disease,” “biopsy without ganglion cells,” or “compatible with Hirschsprung disease,” whereas, in reality, the report should read “the sample is inadequate for diagnosis.” Same situations may arise with excisional biopsy samples obtained from the rectum through the transanal approach or from the colon during laparotomy or laparoscopy (Figs. 1–5).

A safety plan for obtaining an adequate biopsy sample

Suction rectal biopsy

This procedure should not be underestimated. Prepare the patient for the procedure with rectal irrigations using warm saline solution. This clean-out assures an empty rectum. The presence of feces makes it difficult to obtain a proper biopsy. Then, spreading the buttocks, insert the instrument (gun) to a depth of at least 3 cm from the anus with the hole of the capsule facing the posterior rectal wall, bypassing the anoderm and the pectinate area to assure a rectal specimen. Then, apply gentle pressure on the rectum with the gun and create adequate negative pressure, maintaining it for 10 s. This continuous negative pressure allows the suction of a sufficient amount of mucosa and submucosa into the instrument. Then, activate the blade by pulling the trigger of the device and do not release it. Remove the tool from the rectum and take out the specimen from the capsule. It is essential to confirm the presence of an adequate amount of submucosa (whitish tissue),

* Corresponding author.

E-mail address: cirugia.pediatrica@mac.com (L. De La Torre).

<https://doi.org/10.1053/j.sempedsurg.2019.04.013>

1055-8586/© 2019 The Author(s). Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license.
(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

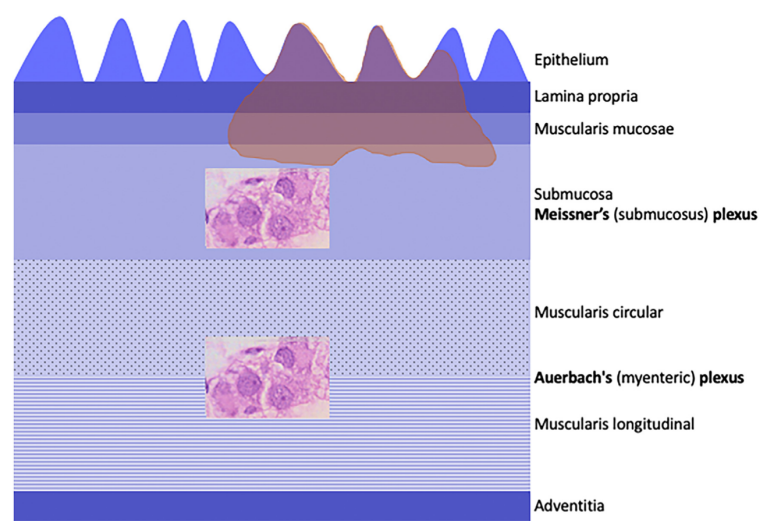


Fig. 1. Inadequate transanal rectal biopsy (obtained through suction or open procedure). Mucosa and a tiny fragment of submucosa can be seen. This type of biopsy is inadequate for the study and is a risk because it can lead to a result reported as aganglionic.

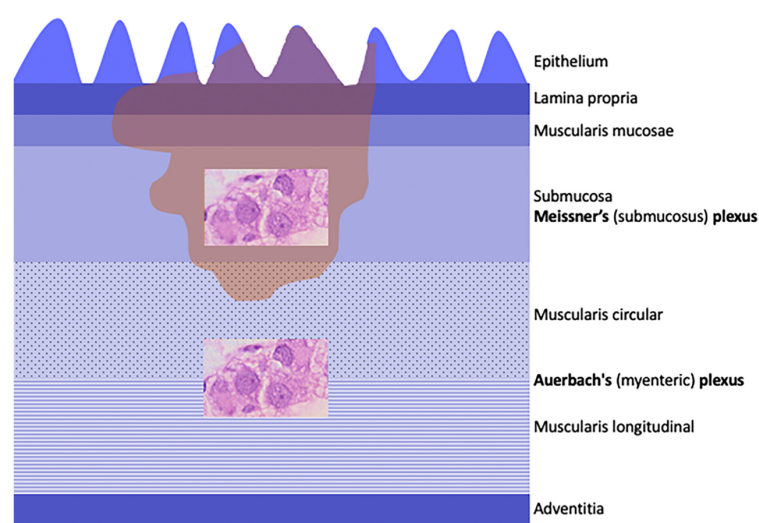


Fig. 2. Adequate transanal rectal biopsy sample. The specimen possesses a sufficient amount of submucosa. The adequate size of these biopsy samples is 2–3 mm.

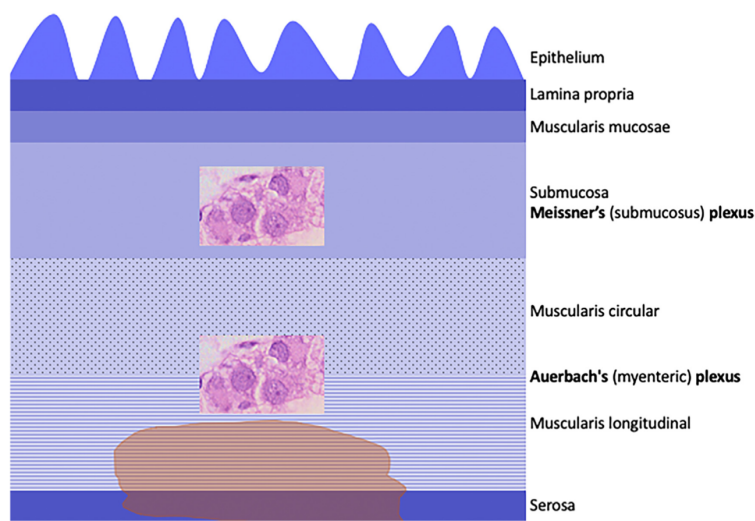


Fig. 3. Inadequate biopsy sample of the colon (obtained by laparoscopic or open procedure). The sample contains only serosa and the longitudinal layer of the muscularis propria. The myenteric plexus is deeper. This type of biopsy is a risk because the result can be reported as aganglionic.

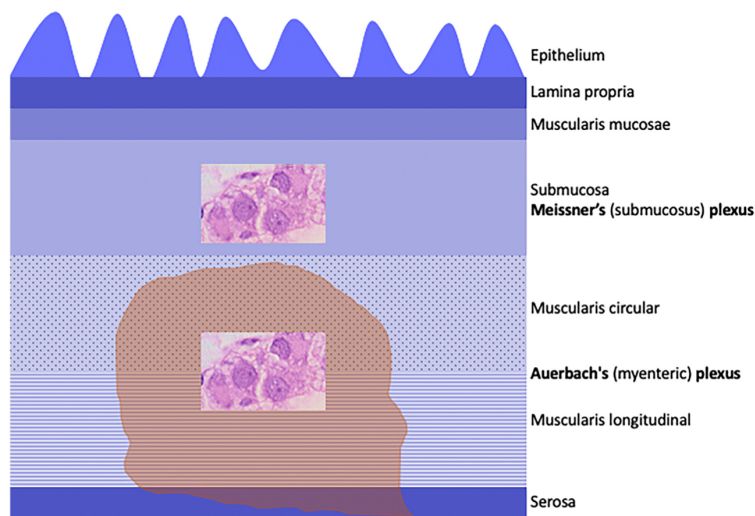


Fig. 4. Adequate biopsy sample of the colon. The sample contains all of the external muscular layer containing the myenteric plexus between the outer longitudinal and the internal circular layer.

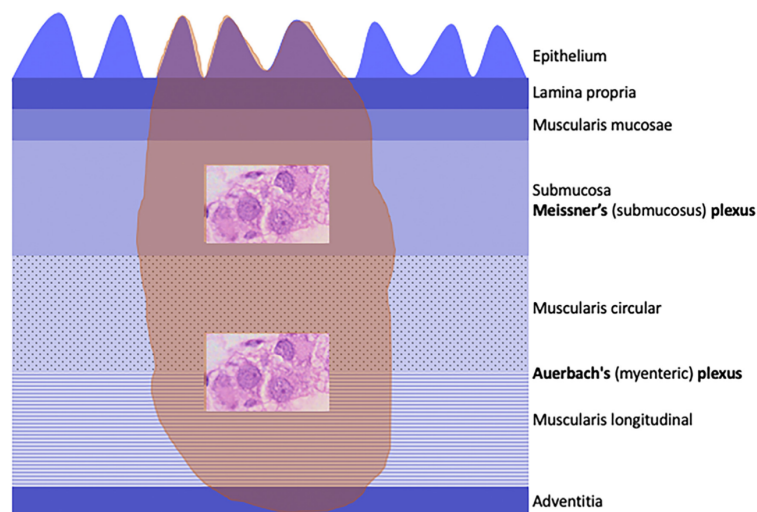


Fig. 5. A full-thickness biopsy sample. This sample includes both plexuses. This type of biopsy is recommended for intraoperative study. The ganglion cells in the myenteric plexus are more identifiable compared with those in the submucosal plexus.

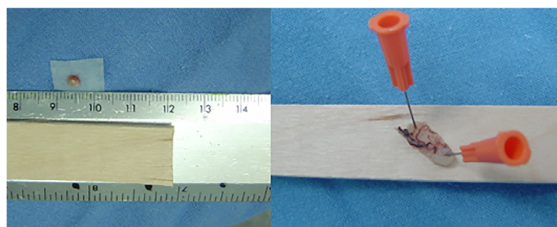


Fig. 6. An adequate suction rectal biopsy sample possessing a sufficient amount of submucosa (white tissue) and measuring 2–3 mm. A full-thickness biopsy sample of the colon is recommended for intraoperative studies. It is advisable to keep the specimen flat on a piece of wood or plastic.

which is different from the mucosa (red tissue) (Fig. 6). Place the sample on a piece of filter paper with the mucosal surface facing the paper. Introduce the paper with the specimen into the container with formalin. If the pathologist is planning ancillary histological stains, such as acetylcholinesterase or other histochemical stains, send the biopsy sample fresh or with Roswell Park Memorial Institute medium. Do not use saline as a medium. This type of rectal biopsy is ideal in newborns, but it is suitable for patients less than three months.

Transanal approach for rectal biopsy of full-thickness biopsy

This procedure should not be blind. Adequate exposure of the anal canal and rectum is required. We highly recommend the use of the Lone-Star® colorectal retractor (Cooper Surgical, Inc.) or similar. If it is not available, a rhinoscope or nasoscope is a suitable alternative. The “3-suture technique” is an easy and reproducible technique for obtaining a good sample from the rectum. It allows controlling the thickness of the sample and distance

from the pectinate area. After a thorough examination of the anal canal, place a 5/0 silk traction suture on the rectum 1 cm from the pectinate area and snap it (suture #1). Next, applying traction with suture #1, place another 5/0 silk suture on the rectum 2 cm from the pectinate area (suture #2) and snap it; this suture marks the location of the biopsy. Then, applying traction with suture #2, place a 5/0 Vicryl suture on the rectum 3 cm from the pectinate area (suture #3) and tie it. To obtain the biopsy sample, apply traction with sutures #1 and #3 in opposite directions and lift suture #2. Then, create a vertical incision on the rectum between sutures #1 and #2 and, then, another vertical incision between sutures #2 and #3. Continue cutting until a good sample with enough mucosa and-submucosa or a full-thickness sample is obtained. Send the specimen to the laboratory for pathological analysis as indicated above. Finally, close the wound with suture #3 using a locked running suture. (video at <https://youtu.be/ZEmZ5m3fu1U>). This “3-suture technique” is also used to obtain an adequate biopsy sample of the colon during laparotomy or laparoscopy.

A safety plan for performing a reliable biopsy sample study

The biopsy is studied initially using hematoxylin and eosin (HE) staining. If definitive ganglion cells are observed, a diagnosis of Hirschsprung disease is ruled out. However, if ganglion cells are not observed, the biopsy requires 50–75 sections for examination. Some protocols require 60 sections from the beginning.^{1–3} The histopathological diagnosis of Hirschsprung disease relies on the demonstration of the absence of ganglion cells within the nerve plexus of the intestinal wall using HE staining. Other helpful histological findings may include hypertrophic nerve trunks (twigs) replacing the nerve plexus and an abnormal pattern on acetylcholinesterase histochemical staining. Currently, the use of immunohistochemistry is an ancillary method used by pathologists in the diagnosis of Hirschsprung disease. The lack of calretinin in the lamina propria of the mucosa and positive choline transporter staining result support the diagnosis.^{4,5} It is essential to work with an experienced pathologist in the diagnosis of Hirschsprung disease.⁶ Unfortunately, it is not uncommon for many pediatric surgeons to receive an unreliable pathological report. In these situations, false-positive and -negative reports may lead to incorrect decisions. A second or third opinion with another pathologist is mandatory.

Error trap #2. – Deficient decompression of the colon due to inadequate colorectal irrigations. Failure to verify the success of colorectal irrigations

Colorectal irrigations, as non-surgical management for Hirschsprung disease, were first described by William Osler in 1892.⁷ Since then, several operative techniques have been described. The goals of the rectal irrigations are to relieve the obstruction, keep the colon decompressed, and treat the colitis until corrective surgery is performed, particularly when a one-stage pull-through is planned. If the colorectal irrigations are insufficient and the patient presents intermittent colonic distension, subclinical colitis may develop and may progress to live threatening colitis. Lack of verification of the success of daily colorectal irrigations may cause medical treatment failure.

A safety plan for achieving sufficient decompression of the colon

Perform proper irrigations and detect signs of colon distention in a timely manner. In case of insufficient colorectal irrigations, the clinical signs might be subtle and can be missed if not looked for actively. The evaluation of effective decompression of the colon is clinical. If there is a doubt that the irrigations have not decompressed the colon, an abdominal radiograph is necessary to confirm that the entire colon is not distended. Frequency and volume of saline used for colorectal irrigations are patient dependent (e.g., some patients need irrigations every 24 h, while others need them every 4 h). Appetite, weight gain, and abdominal softness are clinical signs of successful irrigations. Conversely, nausea, vomiting, abdominal distension, and irritability are signs of unsuccessful irrigations. The most common cause of unsuccessful irrigations is infrequent irrigations.

Volume: The volume of warm saline needed for colorectal irrigations is patient dependent. The irrigations are performed until the return of the irrigation fluid is clear.

Frequency: The irrigations are performed as frequently as needed. Communication with parents about the clinical condition of the patient is essential to regulate the schedule of irrigations.

Technique: To perform irrigations, the parents of the patient need an 18-French Foley catheter, water-based lubricant, warm saline, and a 20-cc syringe. The parents insert the catheter 5–10 cm into the rectum and allow the release of gas and stool. Then, they

inject 20 mL of warm saline gently and disconnect the syringe to allow spontaneous run out of the fluid. This process is repeated several times until the irrigation fluid that runs back is clear. The parents can insert the catheter further, but care is needed to ensure that the colonic wall is not damaged. The total amount of saline for each irrigation is patient dependent but can vary between 60 mL and 500 mL (no upper limit exists) each time.

Error trap #3. Failure to demonstrate the transition zone because of an inadequate contrast enema

The goal of the contrast enema in Hirschsprung disease is to demonstrate the transition zone. The technique to perform this study is different from that for other contrast enemas (e.g., for the reduction of an intestinal intussusception). The following three factors increase the chance of identifying the transition zone: (1) performing the study when the patient does not have colitis, (2) presence of a confirmed histological diagnosis of Hirschsprung disease, (3) and using a proper radiological technique.^{8–10}

The certainty of the diagnosis of Hirschsprung disease and determination as accurately as possible of the length of the aganglionosis is mandatory before operating. It prevents unpleasant surprises faced by the surgeons when they are confronted with unexpected intraoperative situations, e.g., a total colonic aganglionosis during a planned primary transanal pull-through and overstretch of the bowel creating a tense anastomosis with an ischemic bowel resulting in postoperative dehiscence, stenosis, or fistulas.

When the surgeon does not have results from an intraoperative histological study, “the contrast enema for Hirschsprung disease” becomes an indispensable tool of great value for planning the type of operation.

A safety plan for performing a contrast enema for Hirschsprung disease

All patients with clinical or radiological evidence indicating the possibility of Hirschsprung disease need rectal irrigations to treat the intestinal obstruction and colitis. If the rectal irrigations resolve these problems, a rectal biopsy is performed to confirm the diagnosis. While waiting for the biopsy report, irrigations and oral feeding are continued for the patient.

Because of all patients with Hirschsprung disease born with intestinal obstruction in different degree, a spectrum of colitis exists if not appropriately treated. Do not perform the contrast enema in a patient suffering from colitis (Fig. 7). How do you know when the time is right for the contrast enema? The perfect timing is when the patient on irrigations is gaining weight, eating well, and forming normal yellow seedy feces. To perform the contrast enema, mild distension of the proximal ganglionated bowel is necessary to render the transition zone visible. Therefore, the parents are instructed to stop one irrigation before the contrast enema. For example, if the patient requires rectal irrigations every 6 h, the caregiver is told to hold the irrigation 6 h before the study.

Technique for the “contrast enema for Hirschsprung disease.”

Confirm the presence of a dilated segment of the colon on the scout image. Insert a small catheter (e.g., an 8-French catheter) 1 cm into the rectum. If a Foley catheter is used, do not inflate the balloon. The contrast agent (e.g., Iopamidol, do not use barium) is slowly hand injected and monitored under fluoroscopy. Because approximately 80% of the patients with Hirschsprung disease have aganglionosis of the rectum or rectosigmoid, the initial images should be obtained in the lateral position. The transition zone may be missed if the study begins with anteroposterior images. Continue injecting the contrast until the dilated segment is confirmed. Once the dilated bowel is observed, the study is completed



Fig. 7. Contrast enema in a 1-day-old boy with low intestinal obstruction. The study shows inconclusive data for diagnosis. The patient improved with rectal irrigations and underwent a rectal biopsy. See Fig. 2 showing the same patient.

(Fig. 8). Different factors do not enable demonstration of the transition zone, with the most common factor being the presence of colitis. In this situation, the spastic colon does not permit the formation of the transition zone, and a spiculated mucosa is observed (Fig. 9). Furthermore, using a catheter with an inflated balloon, inserting a long segment of the catheter, injecting the contrast agent at a high pressure, and rapid filling of the colon do not enable demonstration of the transition zone.

Error trap #4. Intraoperative histopathological findings concerning the transition zone in the colon for the anastomosis

The goal of performing intraoperative biopsies is to confirm the definitive presence of ganglion cells in the bowel for the anastomosis. However, a “histological transition zone” anastomosed to the rectum has been proposed as a cause of postoperative obstruction. As a consequence, surgeons nowadays avoid performing the colorectal anastomosis in this zone. The histological findings of this segment are the presence of rare ganglion cells or an adequate number of ganglion cells coexisting with hypertrophic nerves. To guarantee normal plexuses during the pull-through, an intraoperative biopsy is required. Because the length of the transition zone is variable, the surgeons may perform an extensive resection of healthy ganglionated bowel when the intraoperative biopsy concerns the transition zone.

A safety plan to avoid an extensive bowel resection facing the histological transition zone

Different studies have confirmed the length of the transition zone to be ≤ 5 cm.^{11–13} Consequently, to avoid an extensive and unnecessary bowel resection, if ganglion cells are present in the intraoperative biopsy sample, but it concerns a transition zone; a safety plan is to perform the anastomosis 5 cm proximal to the site where the biopsy sample was obtained.

Error trap #5. Invasion of and damage to the anal canal produce fecal incontinence

The importance in preserving the anal canal to assure fecal continence is that this area possesses great sensitivity, thus allowing the patient to differentiate between solid, liquid, gas, pain, pressure, heat, cold, longitudinal, or rotatory movement.¹⁴ Thus, damage to the anal canal during the pull-through is associated with postoperative fecal incontinence.^{15–19} Because the pull-through requires partial or total resection of the rectosigmoid (natural fecal reservoir), which is another component necessary for fecal continence, preservation of the anal canal is mandatory.

Surgeons need to protect the anal canal and 1–2 cm of the rectum during the pull-through. The transanal approach changed the direction to perform the mucosectomy (Soave procedure) or the proctectomy (Swenson procedure). Before 1995, these operations were performed through laparotomy. Therefore, mucosectomy and proctectomy began proximal to the peritoneal reflection. These procedures required prolapse or eversion of the rectum, facilitating a clear identification of the pectinate area and distal rectum (Fig. 10). The transanal approach does not require prolapse of the rectum. Consequently, identification of the anal canal is hindered.

Consequently, the risks involved with this approach are starting the operation too close to the anal canal, thus, causing damage to this area and overstretching the anal canal and anal sphincter. Even when the traction sutures are placed above (proximal to) the pectinate area, circumferential incision and placement of the sutures during the anastomosis shorten the initially proposed length between the rectum and anal canal. As a result, the anastomosis could be completed in a lower (distal) location, damaging the anal canal. Another source of damage to the anal canal is inadequate use of colorectal retractors or use of traction sutures on the anal canal to expose the rectum.

A safety plan to preserve the anal canal

Adequate use of a colorectal retractor and convenient location of the traction sutures

To place the colorectal retractor, first set the hooks at the limit between the anal skin and anoderm. When fixing the hooks to the ring, avoid using too much traction. Then, observe the anal canal and identify the rectal mucosa that is slightly pinker. To ensure an adequate length of the traction sutures from the anal canal, use of a ruler is recommended. Then, place the traction suture 1–2 cm proximal (above) to the pectinate area. While placing the traction sutures, take care at all times to maintain a proper distance between the traction sutures and the pectinate area, and once the traction sutures are placed the entire circumference of the rectum, then, reposition the hooks proximally (above) over the pectinate area, setting the hooks on the rectal mucosa. This maneuver protects the anal canal during the operation (video <https://youtu.be/YCcLET7pm28>).

Another way to protect the anal canal is setting the hooks on the rectum covering the anal canal before starting the placement of the traction sutures. This option may lead to some sutures

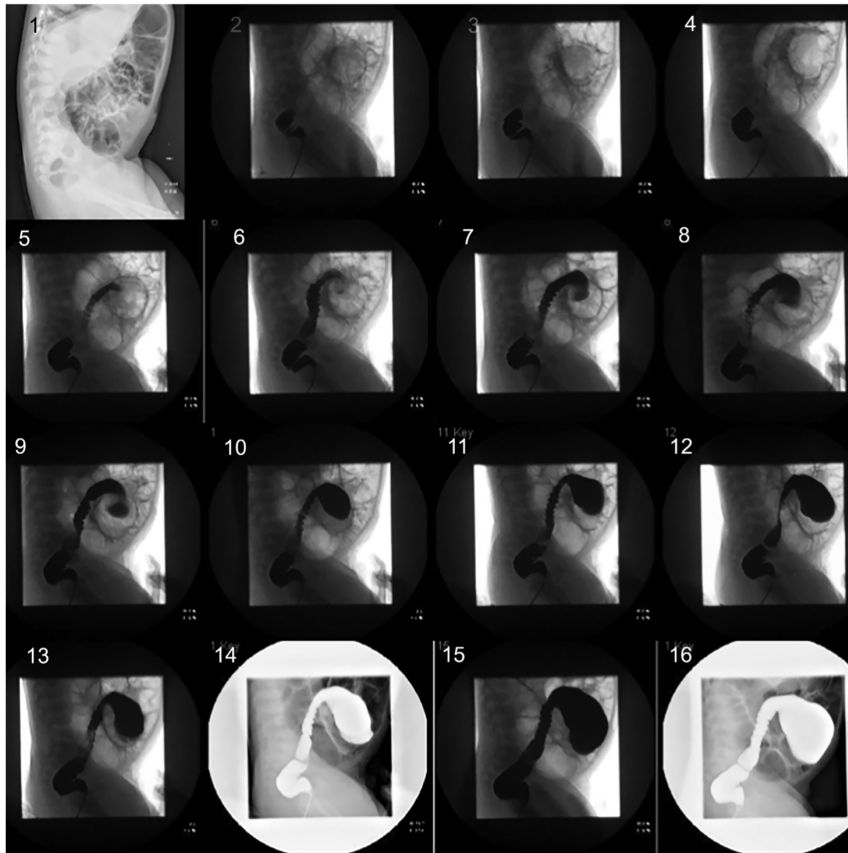


Fig. 8. A sequence of images of a contrast enema in a 5-week-old boy. At 3 days of life, the patient underwent a rectal biopsy, confirming Hirschsprung disease diagnosis. Rectal irrigations every 8 h were established and stopped 8 h before the study. Notice the distention of the colon and the slow injection of the contrast with low pressure to avoid dilation of the rectum. In images 7–9, the dye looks diluted while filling the proximal dilated segment. The subsequent images show the transition zone.

being placed nearer to the anal canal and others farther away. This asymmetric circumference possesses the risk for damage to the anal canal.

Error trap #6. Mapping of the colon and subsequent failure of enterostomy closure because of unsuspected rectal aganglionosis (short segment Hirschsprung)

Neonatal low intestinal obstruction should lead to the suspicion of Hirschsprung disease, which always (100% of times) affects the rectum. Intestinal obstruction localized below the peritoneal reflection or distal sigmoid can create an impressive colon dilation, and the transition zone is not evident. This situation encompasses 80%–85% of Hirschsprung cases. This scenario is not uncommon when a newborn undergoes an “urgent” exploratory laparotomy because of

a low intestinal obstruction, and the only intraoperative finding is a dilated colon. Then, the surgeon creates an enterostomy (colostomy or ileostomy) and obtains multiple biopsy samples from the colon (e.g., ascending, transverse, descending, proximal, and distal stoma) searching for Hirschsprung disease. After the intestinal diversion, the patient improves, and pathology report reads “biopsies with ganglion cells”. Therefore, the surgeon feels confident in closing the ostomy because all biopsy samples show ganglion cells, ruling out the diagnosis of Hirschsprung disease. However, the postoperative outcome is again an intestinal obstruction or dehiscence of the anastomosis, and the surgeon does not realize the reason for this outcome. Then, the surgeon again needs to create another enterostomy. Rectal aganglionosis must be excluded in this scenario. Mapping of the colon is not to confirm the diagnosis of Hirschsprung disease.

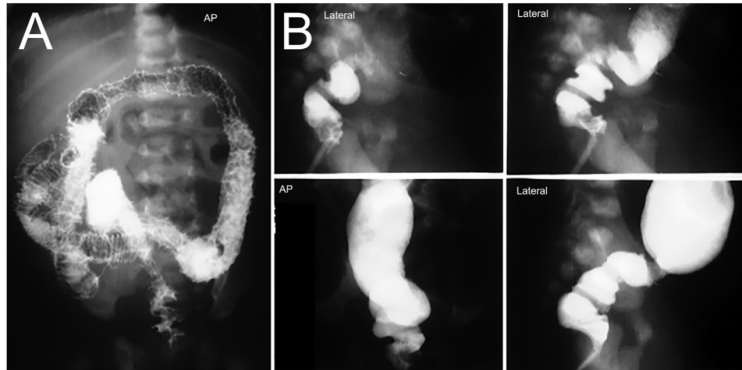


Fig. 9. (A) Contrast enema in a 2-day-old boy with intestinal obstruction. Notice the spastic colon and the spiculated mucosa. It is not possible to identify a transition zone. The patient was referred the same day to our Hospital. We started rectal irrigations and metronidazole treatment. A rectal biopsy on day 10 confirmed the diagnosis of Hirschsprung disease. (B) At the age of 2 months, the contrast enema demonstrated the transition zone in the rectum in lateral view. The transition is not visible in the anteroposterior image.

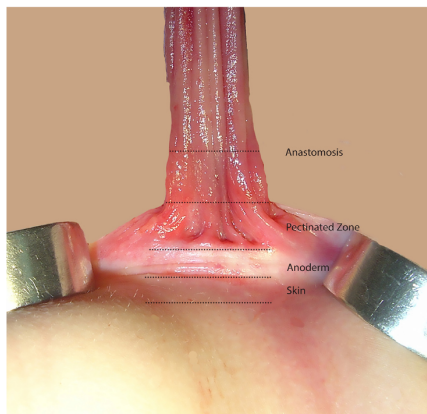


Fig. 10. Rectal prolapse during an open Soave procedure showing the limits of the anal canal consisting of the anoderm and the pectinate zone. Additionally, the prolapse shows the limits of the skin and anoderm and the proposed line of the anastomosis.

A safety plan for performing an enterostomy closure created because of neonatal intestinal obstruction

In the scenario of a non-diagnostic laparotomy in a newborn with low intestinal distal obstruction and intraoperative dilation of the colon, it is appropriate to perform an enterostomy (colostomy or ileostomy) at the discretion of the surgeon and his/her circumstances. However, the surgeon must perform rectal biopsy and confirm the presence of ganglion cells before closing the stoma. The gold standard for exclusion of Hirschsprung disease is a rectal biopsy sample with definitive ganglions cells. The presence of

ganglion cells in several biopsy samples (mapping) from the colon does not exclude the diagnosis of Hirschsprung disease.

References

- Qualman SJ, Jaffe R, Bove KE, Monteforte-Munoz H. Diagnosis of Hirschsprung disease using the rectal biopsy: Multi-institutional survey. *Pediatr Dev Pathol.* 1999;2(6):588–596.
- Schmitt LA, Jaffe R. Optimal handling and processing techniques of biopsies for the diagnosis and treatment of Hirschsprung's disease. *J Histotechnol.* 2004;27:277–280. doi:10.1179/his.2004.27.4.277.
- Kapur R. Pathology of enteric neuromuscular disorders. In: Faure C, Thapar N, Di Lorenzo C, eds. *Pediatric Neurogastroenterology*. Switzerland: Springer; 2017:191–209.
- Kapur R, Reed RC, Finn LS, Patterson K. Calretinin immunohistochemistry versus acetylcholinesterase histochemistry in the evaluation of suction rectal biopsies for Hirschsprung disease. *Pediatr Dev Pathol.* 2009;12:6–15. doi:10.2350/08-02-0424.1.
- Kapur R, Raess PW, Hwang S, Winter C. Choline Transporter immunohistochemistry: an effective substitute for acetylcholinesterase histochemistry to diagnose Hirschsprung disease with formalin-fixed paraffin-embedded rectal biopsies. *Pediatr Dev Pathol.* 2017;20:308–320. doi:10.1177/1093526617697060.
- Ridaura C. Problemas en el diagnóstico histopatológico de la enfermedad de Hirschsprung. *Acta Pediatr Mex.* 2003;24:166–171.
- Osler W. On dilatation of the colon in young children. *Arch Pediatr.* 1893. <https://archive.org/details/101314111.nlm.nih.gov>.
- De la Torre-Mondragón L. Enfermedad de Hirschsprung. Mitos y realidades a 120 años de su descripción. *Acta Pediatr Mex.* 2008;29:139–146.
- Frangia G, Günther P, Schenk JP, Strube K, Kessler M, et al. Contrast enema for Hirschsprung disease investigation: diagnostic accuracy and validity for subsequent diagnostic and surgical planning. *Eur J Pediatr Surg.* 2016;26:207–214. doi:10.1055/s-0035-1546755.
- Zhu T, Sun X, Wei M, Yi B, Zhao X, et al. Optimal time for single-stage pull-through colectomy in infants with short-segment Hirschsprung disease. *Int J Colorectal Dis.* 2019;34(2):255–259. doi:10.1007/s00384-018-3179-3.
- Kapur RP. Practical pathology and genetics of Hirschsprung's disease. *Semin Pediatr Surg.* 2009;18:212–223. doi:10.1053/j.sempedsurg.2009.07.003.
- Kapur RP, Kennedy AJ. Histopathologic delineation of the transition zone in short-segment Hirschsprung disease. *Pediatr Dev Pathol.* 2013;16(4):252–266. Epub 2013 Mar 15. doi:10.2350/12-12-1282-OA.1.
- Kapur RP. Histology of the transition zone in Hirschsprung disease. *Am J Surg Pathol.* 2016;40:1637–1646. doi:10.1097/PAS.0000000000000711.
- Duthie HL, Cairns FW. Sensory nerve-endings and sensation in the anal region of man. *Br J Surg.* 1960;47:585–595.
- Levitt MA, Martin CA, Olesovich M, et al. Hirschsprung disease and fecal incontinence: diagnostic and management strategies. *J Pediatr Surg.* 2009;44(1):271–277.
- Levitt MA, Dickie B, Peña A. Evaluation and treatment of the patient with Hirschsprung disease who is not doing well after a pull-through procedure. *Semin Pediatr Surg.* 2010;19:146–153.

17. Levitt MA, Dickie B, Peña A. The Hirschsprung patient who is soiling after what was considered a "successful" pull-through. *Semin Pediatr Surg*. 2012;21:344–353.
18. Bischoff A, Frischer J, Knod JL, et al. Damaged anal canal as a cause of fecal incontinence after surgical repair for Hirschsprung disease—a preventable and underreported complication. *J Pediatr Surg*. 2017;52:549–553. doi:10.1016/j.jpedsurg.2016.08.027.
19. De la Torre L, Cogley K, Santos K, Morales O, Calisto J. The anal canal is the fine line between "fecal incontinence and colitis" after a pull-through for Hirschsprung disease. *J Pediatr Surg*. 2017;52:2011–2017. doi:10.1016/j.jpedsurg.2017.08.040.