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Anal endosonography and bowel function in patients undergoing different types of endorectal pull-through procedures for Hirschsprung disease☆

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ABSTRACT

Background: The reasons for fecal incontinence after surgery for Hirschsprung disease (HD) remain unclear. The aim of this study was to examine the anal sphincters by anal endosonography and manometry after transanal endorectal pull-through, with or without laparotomy or laparoscopy, in HD patients. Furthermore, we aimed to correlate these findings to bowel function.

Patients and methods: Fifty-two HD patients were followed after endorectal pull-through. Anal endosonography and manometry were performed without sedation at the age of 3 to 16 years.

Results: Endosonographic internal anal sphincter (IAS) defects were found in 24/50 patients, more frequently after transanal than transabdominal procedures (69 vs. 19%, $p = 0.001$). In a multiple variable logistic regression model, operative approach was the only significant predictor for IAS defects. Anal resting pressure (median 40 mm Hg, range 15–120) was not correlated to presence of IAS defects. Daily fecal incontinence occurred more often in patients with IAS defects (54 vs. 25%, $p = 0.03$).

Conclusions: Postoperative IAS defects were frequently detected and were associated with daily fecal incontinence. IAS defects occurred more often after solely transanal procedures. We propose that these surgical approaches are compared in a randomized controlled trial before solely transanal endorectal pull-through is performed as a routine procedure.

Hirschsprung disease (HD) is a congenital disorder with disturbed migration of enteric nerve cells to the distal intestine, and the absence of ganglionic cells leads to functional obstruction in the affected bowel segment. Different surgical techniques may be used to treat the obstructive symptoms caused by the aganglionic segment. Most of these procedures include resection of the aganglionic bowel and an anastomosis between ganglionic bowel and the anal canal. Unfortunately, a significant number of HD patients experience postoperative problems with persistent constipation and/or fecal incontinence. Particularly, fecal incontinence is a serious complication because it is difficult to treat satisfactorily and has a profound negative impact on social life and mental health [1].

During the transanal endorectal pull-through operation (TEPT), a popular modification of the Soave operation, the colonic mobilization and resection are performed transanally without laparotomy or laparoscopy [2,3]. Theoretically, the risk of anal sphincter damage may be higher after TEPT than after other surgical procedures, where only a minor part of the operation is performed through the anal opening [4–6]. So far, few prospective studies have evaluated anorectal function after introduction of TEPT, and the data are inconclusive [2,4,5,7]. Importantly, there are no agreed-on methods for assessing functional outcome after HD operations, and this makes comparison of outcome after different surgical techniques difficult. In addition, objective postoperative examinations of the anal sphincters in HD patients are almost absent in the literature [6,8–11]. Postoperative evaluation of the anal sphincters by anal endosonography (AES) and manometry may add important information about how surgery affects the anal sphincters and make it possible to compare different surgical approaches with respect to anal sphincter integrity and function.

The aim of this study was to examine the anal sphincters by AES and anal manometry in HD patients operated with TEPT or endorectal pull-through assisted by laparotomy or laparoscopy. Furthermore, we aimed to correlate the endosonography and manometry findings to bowel function.

1. Patients and methods

This prospective cohort study was conducted in a tertiary pediatric surgery center. The study was approved by the regional ethics

☆ Contribution of each author: This study was initiated and supervised by KB and RE. Each author participated in preparing the study protocol, patient inclusion and examination, interpretation of the results and final conclusions. KJS was responsible for data extraction, data entry and analysis and preparation of the final manuscript with the close collaboration of the other named authors.

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committee (IRB0006244), and written informed consent from the parents was obtained.

1.1. Patients

All children operated with an endorectal pull-through procedure for HD from 1998 to 2011 were eligible for this study. Background characteristics were recorded at the time of operation. Patients with aganglionosis oral to the sigmoid colon were excluded. The patients were prospectively followed according to our research protocol for HD [2]. Since 2008, anal manometry and AES without sedation have been offered as a supplement in the clinical follow-up in HD patients older than 3 years. Only the patients in whom manometry and/or AES were performed were included in this study.

1.2. Surgical technique

Since 1998, one stage modified Soave-like endorectal pull-through with a short muscle cuff has been the standard operation for rectosigmoid HD. From 1998 to 2001, five surgeons performed laparotomy assisted endorectal pull-through [3]. Shortly, mobilization of the aganglionic colon and rectum was performed through a laparotomy. Frozen sections were taken to identify the transition zone and ganglion cells. The anal canal was exposed using stay sutures around the anal verge, assisted by two handheld Langenbeck's retractors when necessary. The anorectal mucosa was incised circumferentially 5–10 mm oral to the dentate line. Endorectal dissection was performed transanally, leaving a 2–3 cm long muscle cuff which we did not split. A hand-sewn circular end-to-end anastomosis was then performed.

TEPT, as described by De la Torre-Mondragón et al. and Langer et al. [12,13], was introduced in 2001. Four surgeons performed TEPT, and two of these had experience from laparotomy assisted endorectal pull-through. Stay sutures were placed around the anal verge to expose the anal canal and the distal rectal mucosa. A handheld nasal speculum or two Langenbeck's retractors were used for further exposure when necessary. The transanal endorectal dissection and the length of the muscle cuff were identical to the open procedure. The transanal dissection was continued outside the bowel wall to mobilize the aganglionic segment by division of the mesocolic blood vessels close to the bowel wall. Biopsies to verify ganglionic bowel were obtained after mobilization of the transition zone through the anus or through a small umbilical incision.

Laparoscopy assisted endorectal pull-through [14] was introduced in our department in 2008 as an alternative to TEPT for patients with difficult colonic mobilization or at the surgeon's preference. Three of the surgeons familiar with TEPT performed laparoscopy assisted endorectal pull-through. In the statistical analyses, we divided the children into two groups according to the intended operative approach for colonic dissection: the TEPT group included those who had a completely transanal procedure or a transanal procedure converted to laparoscopy or laparotomy because of difficult transanal dissection. The transabdominal approach group (TAA) included the children operated by planned laparoscopy or laparotomy assisted endorectal pull-through.

1.3. Anal endosonography

AES was performed without anesthesia in the left lateral position, using a Hitachi EUB 6500 HV system and a rectal probe (EUP-R54AW-19) (diameter 12 mm) with a 10 MHz rotating transducer covered by a latex balloon filled with degassed water. The ultrasound probe was covered by a latex condom, inserted into the rectum, and slowly withdrawn throughout the length of the anal canal. Serial axial images through the anal canal were captured on a computer and de-identified. The images were assessed, first independently by two of the authors (KJS & KB) to evaluate interrater reliability, and then by the three authors in common if there was disagreement.

The thickness of the anal sphincters was measured in the mid anal canal at the 3 and 9 o'clock positions, using image-processing software, Image-Pro Express (Media Cybernetics, Inc, Bethesda, MD- USA). Mid anal canal was defined as the most cranial level at which the external anal sphincter (EAS) formed a complete ring anteriorly. The internal anal sphincter (IAS) thickness was defined as the width of the hyporeflective band surrounding the subepithelium (Fig. 1a). IAS is surrounded by a broader zone of moderate reflectivity representing two muscle layers, innermost the longitudinal smooth muscle layer

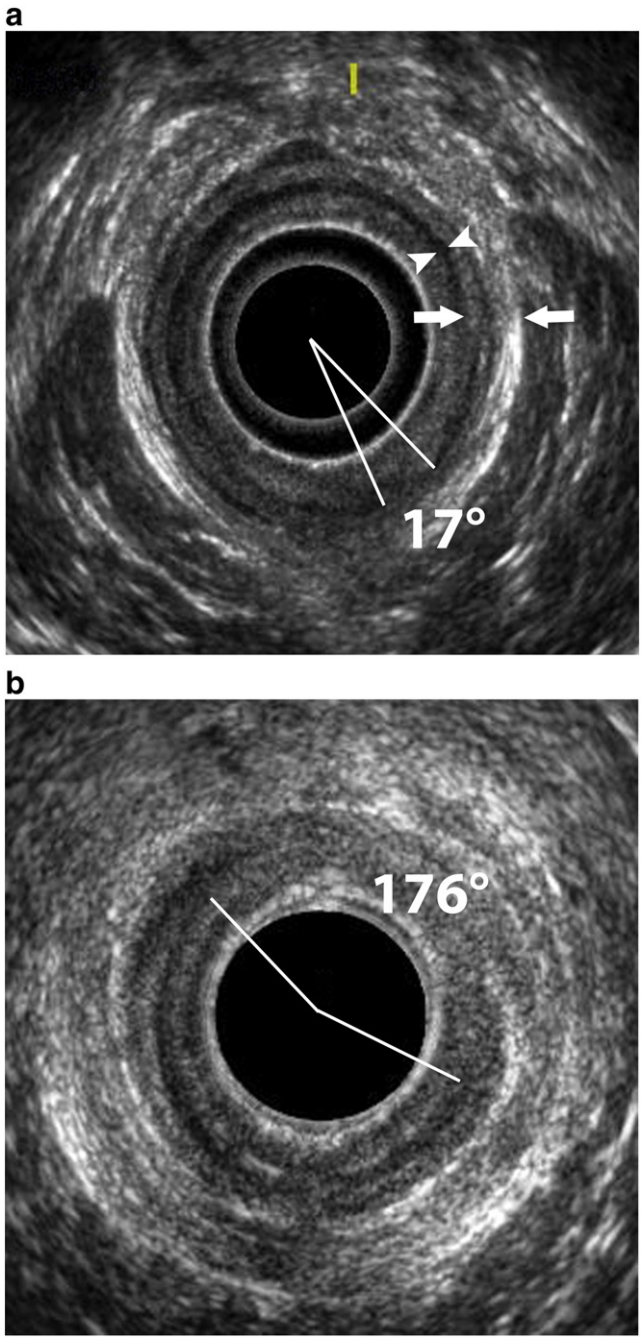

Fig. 1. a, Axial image from the mid anal canal in a 7 year old boy (anterior is uppermost). The IAS can be seen as a hyporeflective band (arrowheads) surrounding the subepithelium. A small, relatively hyperreflective defect (17°) can be seen in the posterior part. The surrounding broader zone of moderate reflectivity (arrows), representing the longitudinal smooth muscle layer and the EAS, is intact. b, In this 8 year old boy, nearly half of the IAS is absent while the EAS is intact.