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ORIGINAL ARTICLE

Results of rectoanal manometry after a novel laparoscopic technique: laparoscope-assisted heart-shaped anastomosis for Hirschsprung's disease

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Abstract

Purpose The present research utilizes a mid-term follow-up study to assess the results of anorectal manometry after laparoscope-assisted heart-shaped anastomosis (LHSA) for Hirschsprung's disease (HSCR), and compares it to a more generally applied approach, the laparoscope-assisted Soave procedure (LSP).

Methods Retrospectively, patients from January 2015 to June 2017 who received LHSA or LSP were included in this study. After surgery, anorectal manometry was performed by the outpatient department. Anal sphincter resting pressure, anal canal length, amplitude of anal contraction, and frequency of anal contraction pre- and postoperatively were recorded. Additionally, mid-term complications were also monitored.

Results Preoperative manometry showed no statistically significant difference between the LHSA and LSP groups. Postoperatively, anal sphincter resting pressure was lower in the LHSA group (60.64 ± 9.33 vs. 68.84 ± 11.80 mmHg, $p = 0.001$). Furthermore, anal canal length of the LHSA group was shorter than that of the LSP group (1.41 ± 0.18 vs. 1.53 ± 0.25 cm, $p = 0.015$). Frequency of anal contraction also showed a statistically significant difference between the LHSA and LSP groups (13.53 ± 2.17 vs. 12.50 ± 2.03 per minute, $p = 0.032$). The complication rates showed no significant difference and were as follows: incidence of enterocolitis was 13.89% in the LHSA group and 20.45% in the LSP group, incidence of constipation was 11.11% after LHSA and 27.27% after LSP, and incidence of soiling was 13.89% after LHSA and 25.00% after LSP.

Conclusions Manometric results of this study show satisfactory outcomes after LHSA. LHSA is an advanced surgical technique to make intestinal anastomosis easy and ensure a good prognosis.

Keywords Hirschsprung's disease · Anorectal manometry · Heart-shaped anastomosis · Soave

Background

Hirschsprung's disease (HSCR) is one of the most common digestive tract malformations, with an incidence of 1/5000 [1, 2]. Aganglionosis of the distal bowel is the main pathological feature. HSCR clinically manifests as digestive problems, such as constipation, abdominal distention, and vomiting. Common surgical methods of treatment include Swenson, Soave, Rehbein, Duhamel, and their modifications [3–7]. After removal of the dysfunctional bowel, the normal bowel is pulled out and anastomosed to the residual rectum. But after this radical operation, Hirschsprung's associated enterocolitis, soiling, and constipation are still common [8, 9]. Our medical center has designed a new anastomosis method called heart-shaped anastomosis for HSCR [10], and previous articles have reported satisfactory outcomes [11, 12]. Minimally invasive laparoscope-assisted heart-shaped

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anastomosis (LHSA) is studied in the present study. Heart-shaped anastomosis is characterized by a higher anterior anastomosis and a lower posterior anastomosis after the split of the posterior rectum wall. Previous long-term follow-up studies reported good prognoses for LHSA [10, 13]. In this study, anorectal manometry is used to collect more detailed follow-up information. Because of the aganglionosis of the distal bowel, manometry is an important preoperative examination and will typically show lack of rectoanal inhibitory reflex in the bowels of HSCR patients [14]. In the present study, anal sphincter resting pressure, anal canal length, amplitude of anal contraction, and frequency of anal contraction were used to evaluate the postoperative anorectal condition in mid-term follow-up after the HSCR operations. Furthermore, complications were also included to assess the mid-term postoperative side effects. Results of LHSA were then compared to another surgical procedure employed by our medical center—laparoscope-assisted Soave procedure (LSP).

Absence of ganglion cells results in the establishment of an abnormal nerve reflex loop in the rectum and anal canal, which affects the contraction of the aganglionic bowel. Preoperative manometry typically shows an absence of the rectoanal inhibitory reflex. Furthermore, anal contraction loses its normal pattern, and the anal sphincter resting pressure may rise because of the spasmodic and narrow bowel lacking in ganglion cells. Contraction of the anal internal sphincter is the primary mechanism to maintain resting pressure. Persistent cramps of the aganglionic bowel lead to an abnormal resting pressure and contraction rhythm. Herein, the anal sphincter resting pressure, anal canal length, amplitude of anal contraction, and frequency of anal contraction measured by manometry enabled a comprehensive comparison of postoperative outcomes between LSP and LHSA.

Patients and methods

Patients

This retrospective study included 80 HSCR patients that were operated on in our medical center from January 2015 to June 2017. Postoperative pathological examinations were used to ensure a definitive diagnosis. None of these patients had a previous diagnosis of Down syndrome, hypothyroidism, anorectal malformation, or a total colonic aganglionosis. None of the participating patients required staging operations or reoperations. Among the participating patients, 44 underwent LSP, and 36 underwent LHSA. Detailed patient demographics are shown in Table 1.

Prior to surgery, both the purpose and the associated risks of the surgical procedure were fully explained to the legal guardian of the patient who then signed an informed consent for the operation. The surgical method was selected randomly, and a single experienced surgeon finished all the procedures for the patient. Additionally, all patients completed the entire therapeutic course with one pediatric gastrointestinal surgery group at the Pediatric Surgery Department of Tongji Hospital of Tongji Medical College of Huazhong University of Science and Technology (Wuhan, China). This study was approved by the ethics committee of the Tongji Hospital and adhered to the tenets of the Declaration of Helsinki. The purpose of this study was reviewed with all participating patients prior to their inclusion, and signed informed consent was obtained.

Table 1 Characteristics of patients

	LSP ($n=44$)	LHSA ($n=36$)	p value
Sex			
Male	32	26	0.960
Female	12	10	
Age at operation (month)			
Mean ± SD	18.97 ± 27.03	21.35 ± 24.19	0.683
Follow-up time (month)			
Mean ± SD	12.27 ± 0.64	12.17 ± 0.71	0.517
Length of removed bowel			
Subtotal colectomy	20	16	0.928
Left colectomy	24	20	

LHSA laparoscope-assisted heart-shaped anastomosis, *LSP* laparoscope-assisted Soave procedure, *SD* standard deviation

Prior to the operation, the three main examinations used to diagnose and guide the therapeutic strategy were the histochemical acetylcholinesterase reaction in the rectal mucosa, anorectal manometry, and barium enema [14]. Patients who qualified for the operation were given colonic irrigation with normal saline for 7 days and oral metronidazole for 3 days to prepare the colon. To confirm the diagnosis, postoperative hematoxylin–eosin staining of paraffin sections was used. Patients were then asked to return to the outpatient clinic for reexamination approximately 12 months after the operation.

Follow-up data were collected through medical records, outpatient visits, detailed telephone interviews, and mail correspondence with the patients. The demographic data, examination information, operation procedures, and postoperative outcomes, including Hirschsprung's associated enterocolitis, constipation, and soiling, were recorded comprehensively. Hirschsprung's associated enterocolitis was manifested as postoperative fever, abdominal distension, and acute watery diarrhea. Constipation meant reduced voluntary bowel movements. Soiling was defined as an involuntary bowel movement between two voluntary bowel movements. A specially designed Excel spreadsheet was used to record the information for this study.

Surgical techniques and anorectal manometry

Surgical techniques

Our previous articles have described the establishment of laparoscopic access [15]. Intraoperative multi-place biopsy of the seromuscular layer of the colon was carried out to determine whether the enteral nervous system was normal or not. According to this result, confirmation of the resection length was made based on the intraoperative biopsy and preoperative examinations. Then, the affected colon that was 5 cm proximal to the most distal bowel with normal nerve plexus to the peritoneal reflection of the rectum was mobilized. Soave anastomosis was done using the classical procedure. The heart-shaped anastomosis was a modified procedure for HSCR that has been widely used in many Chinese medical centers since the 1990s. Previous articles have reported the details of the procedure [10]. First, with the assistance of a laparoscope, sponge forceps grasped the rectum through the anus and pulled out the bowel. Then, the rectum was cut open. After that, the affected bowel, as determined by the intraoperative biopsy, proceeded to be pulled through. Next, a longitudinal split was made in the posterior anorectum approximately 0.5 cm above the dentate line. Finally, the seromuscular layer of the remaining rectum and the normal colon was anastomosed. Notably, the anastomosis was heart-shaped because the anterior anastomosis was higher (about 4 cm) above the anal verge and the posterior anastomosis was lower (about 1.5 cm) above the anal verge.

Anorectal manometry

A six-channel probe anorectal manometer (MMS UPS2020, The Netherlands) was used to evaluate the condition of the rectum and anal canal in HSCR patients [14]. Anorectal manometry was performed with the same manometer before and after the operation. Half one hour before the examination, patients took chloral hydrate at 6% orally with dose of 1 ml/kg. During the examination, the child was in the left lateral position and was detected by the high-resolution gastrointestinal motility monitoring system. The pressure catheter has an outer diameter of 3.5 mm and an inner diameter of 0.8 mm, and is equipped with a 5 cm long low compliance airbag at the top. There are four channels, and each channel opening is 1 cm apart with a difference of 90 degrees, with an aperture of 0.5 mm. The water infusion rate was 0.3 mL/min. During the test, the catheter and air bag were coated with lubricating oil and inserted into the anus for about 15–20 cm. While injecting air into the air bag, the catheter was pulled outward to make the air bag fully extended. After the child was quiet, the catheter was pulled out 0.5 cm at a time by slow fixed-point pulling method, and the pressure changes in each channel were continuously recorded by the computer. According to the pressure curve, the positions of the anal sphincter and external sphincter were determined. Then, the doctor inserted the catheter again and pulled it outward according to the above method. When the side holes of each channel reached the position of the internal and external sphincters, air was injected into the air bag and the changes in the pressure were recorded. A typical rectoanal inhibitory reflex was defined as a relaxation wave with a $\geq 70\%$ decrease of pressure in response to a 10–30 mL increase in the stimulation volume. If the rectoanal inhibitory reflex was not induced by stimulation with the maximum volume after three repeated attempts, it was considered negative. Additionally, anal resting pressure, anal canal length, amplitude of rectum contraction and frequency of rectum contraction were recorded.

Statistical analysis

SPSS Version 18.0 was used to analyze all variables included in our statistical analysis. Dichotomous variables were analyzed by χ^2 test. Student's t test was used to analyze continuous parameters. All statistical tests described above were two-sided. A p value of < 0.05 was considered to be statistically significant.

Results

In total, 44 patients underwent LSP and 36 patients underwent LHSA from January 2015 to June 2017. The LSP group consisted of 32 males and 12 females. The LHSA group included 26 males and 10 females. The operation age of the LSP group was 18.97 ± 27.03 months and that of the LHSA group was 21.35 ± 24.19 months. Follow-up time for the LSP and LHSA groups was 12.27 ± 0.64 and 12.17 ± 0.71 months. These results showed no statistically significant differences. The length of the removed bowel between the two groups also showed no significant difference. Detailed statistics are shown in Table 1.

During follow-up, the incidence of Hirschsprung's associated enterocolitis was 20.45% in the LSP group and 13.89% in the LHSA group. Constipation happened in 27.27% of patients with LSP and 11.11% of patients with LHSA. Additionally, 25.00% of patients with LSP and 13.89% of patients with LHSA displayed soiling. Findings of postoperative complications showed no statistically significant difference and are shown in Table 2.

Anorectal manometry detected the anal sphincter resting pressure, anal canal length, amplitude of anal contraction, and frequency of anal contraction. In preoperative examinations, these indicators showed no significant differences (Table 3). In postoperative examinations, anal sphincter resting pressure of the LSP group was 68.84 ± 11.80 mmHg, and

Table 2 Complications after HD operations

	LSP ($n=44$)	LHSA ($n=36$)	p value
Enterocolitis, n (%)	9 (20.45)	5 (13.89)	0.559
Constipation, n (%)	12 (27.27)	4 (11.11)	0.0946
Soiling, n (%)	11 (25.00)	5 (13.89)	0.2684

HD Hirschsprung's disease, *LSP* laparoscope-assisted Soave procedure, *LHSA* laparoscope-assisted heart-shaped anastomosis

that of the LHSA group was 60.64 ± 9.33 mmHg ($p=0.001$). Anal canal length of the LSP group and the LHSA group was 1.53 ± 0.25 and 1.41 ± 0.18 cm, respectively ($p=0.015$). Frequency of anal contraction of the LSP group was 12.50 ± 2.03 per minute and that of the LHSA group was 13.53 ± 2.17 per minute ($p=0.032$). However, amplitude of anal contraction showed no statistically significant difference (Table 3).

Discussion

HSCR is a congenital digestive disease that has been studied since the eighteenth century. Gene defects, such as *ret*, *ednrb*, and *sox10*, can cause abnormal migration and colonization of enteric crest cells during embryonic development [16–19]. Thus, these children will manifest constipation, delayed passage of meconium, and malnutrition because of bowel dysfunction caused by aganglionosis. Excision of the affected bowel can be performed using many procedures, such as Swenson, Soave, Duhamel, and Rehbein procedures. Our medical center has developed a novel procedure, which is characterized by a heart-shaped anastomosis that we have combined with a minimally invasive laparoscopic technique [11]. This article analyzes its outcome through the use of anorectal manometry.

Anorectal manometry is an important examination for patients with HSCR because it helps doctors evaluate the function of the anorectal sphincters and the enteric nervous system [20, 21]. When evaluating bowel function, manometry can help doctors get results based on pressure change. This is relatively safer compared to X-ray or biopsy. It is valuable to evaluate anal sphincter resting pressure, anal canal length, frequency of anal contraction, and amplitude of anal contraction to decide the innervation condition of intestinal smooth muscle besides rectoanal inhibitory reflex. Normal enteric nervous system controls regular movements of intestinal muscles to form pressure and contraction with rhythm. Previous article developed a scoring system to diagnose HSCR that is composed of the histochemical acetylcholinesterase reaction in the rectal mucosa, anorectal manometry, and barium enema [14].

Results showed that the anal sphincter resting pressure after LSP was higher than that after LHSA. Moreover, the anal canal length after LSP was longer than that after LHSA. The anal sphincter resting pressure after LHSA was 60.64 ± 9.33 mmHg, which indicates a relatively normal status of the rectum and anal canal. Higher pressure is caused by excessively powerful contraction of the internal sphincter and nearby muscles. When defecation occurs, the anal sphincter resting pressure decreases. Excessive contraction hampers this process. The results of anal canal lengths also suggest that it is related to the length of the anal high-pressure zone. Frequencies of anal contraction after LSP and LHSA both increased when compared to preoperative examinations. Frequency after LHSA was higher than that after LSP, which suggests that the postoperative nerve reflex loop of LHSA recovers better. LHSA creates a wide anastomosis and preserves the internal sphincter to the greatest extent. By avoiding a complex operation in the pelvic cavity, nerves and vessels are less harmed. With these advantages of LHSA, relatively

Table 3 Results of anorectal manometry of patients pre- and post-HD operations

	LSP	LHSA	p value
Preoperation			
Anal sphincter resting pressure (mmHg), mean ± SD	66.77 ± 12.68	67.81 ± 14.19	0.732
Anal canal length (cm), mean ± SD	1.58 ± 0.29	1.63 ± 0.25	0.505
Amplitude anal contraction (mmHg), Mean ± SD	15.86 ± 5.20	15.64 ± 3.93	0.831
Frequency anal contraction (per minute), mean ± SD	8.66 ± 1.74	8.19 ± 1.77	0.242
Postoperation			
Anal sphincter resting pressure (mmHg), mean ± SD	68.84 ± 11.80	60.64 ± 9.33	0.001*
Anal canal length (cm), Mean ± SD	1.53 ± 0.25	1.41 ± 0.18	0.015*
Amplitude anal contraction (mmHg), mean ± SD	17.64 ± 4.47	16.64 ± 4.06	0.304
Frequency anal contraction (per minute), mean ± SD	12.50 ± 2.03	13.53 ± 2.17	0.032*

HD Hirschsprung's disease, *LSP* laparoscope-assisted Soave procedure, *LHSA* laparoscope-assisted heart-shaped anastomosis, *SD* standard deviation

*$p<0.05$

normal function of rectum and anal canal can be achieved, which was shown by the better manometric results. The rectoanal inhibitory reflex is difficult to recover because the abnormal nervous system of the residual rectum and anal canal remains. We speculate that multiple pathophysiological triggers lead to the results. First, operation points are different between LHSA and LSP: LSP reserves rectal muscle layer, but LHSA tries to form a wide anastomosis and resects most diseased rectum with enough sphincter to control defecation by the heart-shaped anastomosis. Then, after LHSA, resistance is smaller and the nerves nearby may be more possible to grow into the aganglionic residual rectum than that after LSP. Second, the pathology of HSCR still affects the manometric results post-radical operation. Neurons cannot be regenerated to normal in the residual rectum, so rectoanal inhibitory reflex cannot reappear in most HSCR children. But reconstructed rectum can have nearly normal pressure and contraction because of the anastomosis of residual rectum and bowel with normal enteric nervous system. Complications after the two procedures showed no significant difference in the mid-term follow-up. But in our previous article, LHSA showed a better prognosis in long-term follow-up [11]. This may be attributed to the sample and different follow-up times of the present article. However, the ratio of complications of the LHSA group was lower than that of the LSP group (Table 2).

Because of the better manometric results and easily managed procedure, LHSA may be a good choice for HSCR treatment. Heart-shaped anastomosis has been widely used in Chinese medical centers. Furthermore, it is also used for the treatment of Hirschsprung's disease. In the future, a prospective study should be conducted to provide more convincing evidence.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval The Institutional Review Board of Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology approved the protocol of the study (Permit Number 2010-HP0761, Wuhan, China). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Written informed consent was obtained from the guardians of the patients included in this study.

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