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SOAVE TECHNIQUE AND HIRSCHSPRUNG DISEASE

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Abstract :

Background and objectives: *Soave a single stage trans-anal procedure most commonly done early in life with good outcome. We used this technique in our patients with Hirschsprung disease (HD) presented in our hospital. Our research's main objective is to outline Soave technical aspects, outcome and rate of success in our center.*

Methods: *This is a retrospective review study series of HD patients in our center who managed by Soave trans-anal approach, to look for postoperative adverse event including: stricture, anastomotic leakage, enterocolitis, and bowel functions.*

Results: *In this study we operated 17 patients with HD, 14 of them primarily went for Soave trans-anal resection, except these three patients: 2 underwent a levelling colostomy, 1 with ileostomy, at neonatal period, follow by Soave. The length of resection was 20±10.5 cm. The follow-up period was 12.2 months (range 5–22 months). The patients ages between 3-14 years old. All patients on follow up bowel movements were voluntary, no fecal incontinence and no constipation which required the use of laxative.*

Conclusions: *Our research strongly approve the fact that a single stage Soave approach without transabdominal dissection is a good technique for late HD patients with a good outcome. A multicenter prospective study and large number of patients is required to validate our results.*

Keywords: *Hirschsprung disease; Soave; Trans-anal*

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INTRODUCTION:

Hirschsprung's disease (HD) is the most common congenital bowel motility disorder; affected infants usually present briefly after birth; HD is characterized by the absence of ganglion cells (aganglionosis) in a different length of the distal colon patient mainly present with symptoms and signs of distal intestinal obstruction that are ultimately fatal if not treated. 60 years ago, Dr. Ovar Swenson illustrate the first description of the cause of HD [1]. After that multiple techniques have been developed for elimination of the aganglionic distal colon. Removal of the aganglionic bowel, pulling thru ganglionic bowel, the principles of surgical repairs maintaining the anal canal and sphincter mechanism regardless of the technique.

The operative management of HD has developed dramatically, from full-thickness rectosigmoid dissection (Swenson) [2], endorectal dissection (Soave) [3], retro-rectal pouch procedure (Duhamel) [4,5], low anterior resection (Rehbein) [6] and most recently a primary repair [7,8] can be done trans-anally [9,10], and using

Laparoscopic techniques [11] but which technique provides the best short and long-term results still there is a debate.

Nowadays one of the laparoscopic operations is trans-anal Soave. Soave is the communist used technique with good results in early age group. In the late 1990s; de la Torre and Ortega- Salgado and Langer et al published that animal models and the initial series of children with HD; Trans-anal resection of the rectum was shown to be possible [34, 35]. the principal benefit of the trans-anal approach is avoiding the need for intra-abdominal mobilization of the rectum thru either laparotomy or laparoscopy [27].

Our aim here is to describe our experience with Soave technique cohort study of HD and its outcomes in the late presenters' groups.

SUBJECTS AND METHODS:

This is a retrospective study done in the Pediatric Surgery department of Alhada Military Hospital, Taif, Saudi Arabia; by reviewing our pediatric patients' (age from 3 to 14 years) electronic records; after applying the inclusion and exclusion criteria in HD confirmed diagnosis who underwent surgical repair in the period between (January 2016 and July 2018). The study was approved by Research Ethical Committee.

Our exclusion criteria:

1. Patients who didn't underwent surgery with confirmed diagnosis of HD
2. Histopathological result from rectal biopsy which shows other than HD.

All the files of our patients were reviewed for:

1.Demographic data: age, sex

2.Medical history: age of presentation, presenting symptoms, any medical illness.

3.Surgical history: for previous operations other than the primary repair of HD.

4.Confirmation of HD:

A.All patients were assessed for the transition zone by contrast imaging

B.Histopathology of rectal biopsy.

5.Preoperative data: preparation of the bowel preoperative and the use of intravenous antibiotics.

6.Operative data: efficiency of bowel preparation, age at the time of surgical correction, surgical technique used for repair, performance of intraoperative full thickness frozen section biopsy, length of each resected specimen.

7.Hospital course and Postoperative follow up: data on post-operative early and late complications, long term outcomes were evaluated especially fecal incontinence, constipation, rates of stricture formation, pelvic abscess, and enterocolitis.

Data were coded, and statistical analysis was done using Statistical Package for Social Sciences software program (SPSS) Version 20. Means±standard deviation were used for quantitative data analysis. Although qualitative data were expressed as percentages. A Chi-square and Fisher exact tests were applied for bivariate data analysis to test the statistical significance of associations, the level of significance was $P \leq 0.05$.

RESULTS:

Eighteen patients fulfilled the diagnosis of HD, but one female patient was excluded as she was above 14 years old and referred to adult surgeon service. The remaining 17 patients fulfilled all the study inclusion criteria were enrolled. Age at the time of operation ranged from 3 to 14 years with mean age of 7.9 ± 5.8 years. There were 13 male and 4 female patients with a ratio of 3.2:1.

Preoperative evaluation with contrast enema was done in all patients which revealed a transition zone in the rectosigmoid in 12 patients (70.5%), left colon in 4 (23.5%) and transverse colon in 1 (5.8%) as shown in figure 1. HD was confirmed by preoperative rectal biopsy histopathologic picture of aganglionosis in all patients. All patients underwent mechanical bowel preparation along with rectal irrigation prior to their definitive operation aiming for best operative and post-operative outcomes. Six patients had fecolotomy

on presentation and three necessitated irrigation under anesthesia to irrigate well.

Preoperative intravenous antibiotics with third generation cephalosporin and metronidazole were given to all patients with provisions made for allergies. One surgical technique was used in the studied cohort with trans-anal rectal dissection, short sleeve, Soave approach with careful preservation of the dentate line. The three patients with fecal diversion prior to definitive repair underwent trans-anal resection in the neonatal period. The transverse colostomy in two of them was left untouched for later reversal as a third stage; the ileostomy in the third patient required a stoma reversal in a next stage to pull-through procedure.

In the remaining 14 patients who had a well-defined sigmoid or lower left colon transition zone on contrast enema [figure 2], a purely Soave trans-anal resection was performed without preoperative diversion. The trans-anal dissection was performed using a lithotomy position. Sutures were placed initially just inside the anal canal at the level of the mucocutaneous junction for retraction, in this way the distal 1.5 cm of anal canal was preserved. Interrupted sutures were placed 1.0 cm proximal to the dentate line in a circumferential fashion to provide uniform traction.

Monopolar electrocautery was used to perform submucosal dissection leaving 3cm cuff and then a full-thickness dissection along the traction line of sutures. The dissection was carried into the peritoneal cavity and the colon was pulled-through to the appropriate level. The anastomosis was created in two layers, 1.0 cm proximal to the dentate line.

Full thickness frozen section from all the patients was taken and intra-operative biopsies were checked in the operating theatre to assure that the pull-through segment had ganglionic cells and the muscularis and submucosa had normal sized nerve trunks.

Only in 3 patients who had previous levelling stomas laparoscopy or laparotomy were used to assist the dissection but in all other patients with disease we didn't use it even in patient with disease that extended above the mid-sigmoid.

The average length of resected specimen was 22.6 ± 10.9 cm with no statistically significant difference, figure 2. The time consumed during the operation was slightly but didn't reach a statistical significance. None of the patients required perioperative blood transfusions, none had urinary retention, wound infection, enterocolitis, anastomotic leak or strictures, or intraabdominal abscesses.

All patients were followed up in the pediatric surgery outpatient clinic at 2 weeks, 2 months and 6 months. At two weeks, rectal examination using Hagar dilators was done to assess the need for rectal dilations. Two patients needed anal dilation in the first two clinic visits and improved after dilation. One patient had constipation requiring intermittent laxative usage. All patients had multidisciplinary care follow-up plane with pediatric surgery, pediatric gastroenterology and clinical nutrition departments. The patients were assessed for fecal and bowel functions on follow up where voluntary bowel movements and spontaneous voiding with good control was reported in 100% of patients.

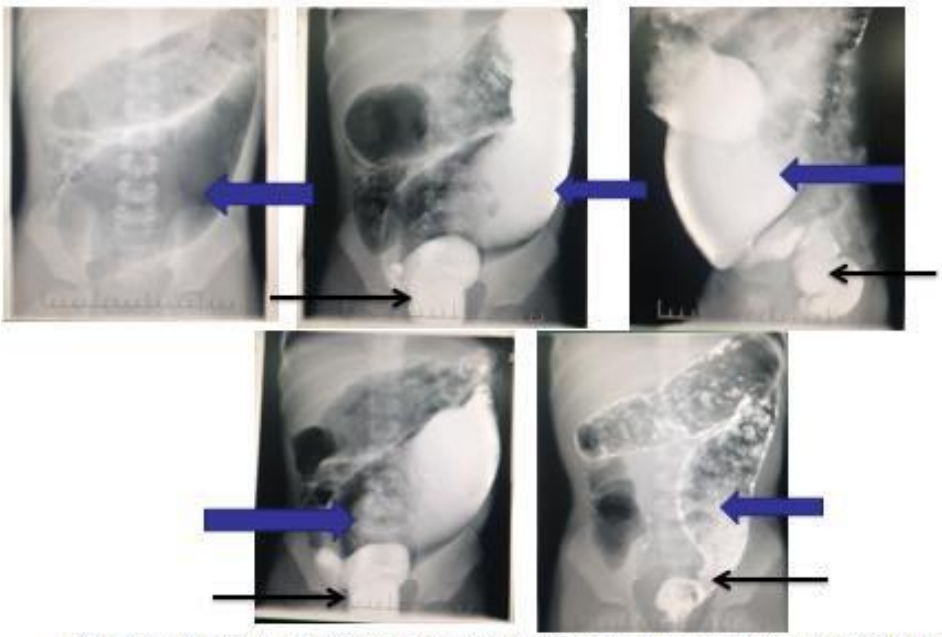

Fig. 1 contrast study showing dilated proximal colon (blue arrow) and distal portion (black arrow) with clear transition zone.

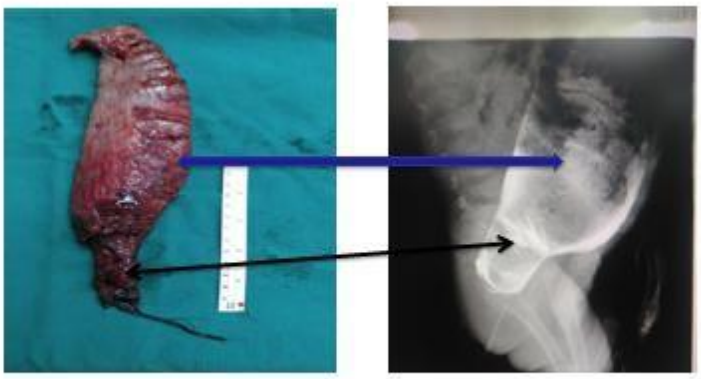

Fig. 2 well excised specimen to demonstrate with contrast study (arrows).

DISCUSSION AND CONCLUSIONS:

HD is the commonest congenital gut motility diseases seen in pediatric surgery; removing of bowel segment without ganglionic cells and pulling thru the ganglionic bowel with anal sphincter preserving is the majority of treatment. Many published HD series managed with either the laparoscopic or trans-anal pull-through techniques had developed with most of it with very good results. [16, 17, 18,33,28].

Few studies compared the laparoscopic to the trans-anal approach [19]. The laparoscopic approach was adopted by Miyano and his colleagues [27], while trans-anal pull-through was highlighted by Pratap and his colleagues; as it can be done by any pediatric surgeon, including those without laparoscopic skills [36]

We used Soave trans-anal technique in our center in all the patients irrespective of the age at presentation and age at surgical intervention. None experienced anastomotic leaks, intraabdominal abscesses or strictures, this compares favorably with literature which quotes leak rates of 5–7%, abscess formation in 2–6%and stricture rates of 5–24%[20,21,23-25] which might be explained by the fact that we performed Soave procedure without opening abdomen or any stoma creation as compared

to diversion before the pull through as done by others. The complications described by them after the neonatal age were due to the stoma as it done before Soave not the Soave itself [31]. Pediatric surgeons in those series preferred in older children preliminary dysfunction stoma for resolution of the dilatation and thickening, so that a subsequent trans-anal approach can be safely used, however in our series, we used good colonic irrigations to decompress the dilated colon like Onishi and his group. We didn't encounter any difficulty during trans-anal dissection to pull down this dilated segment of the rectum or colon [figure 2] and a single stage Soave procedure was performed without any appreciable complications in follow up.

Frozen biopsies full thickness sections intraoperative was done in all the patients in our cohort. We adopted our protocol where we didn't only depend on confirmation of presence of ganglion cells in intraoperative biopsies studies prior to performing an anastomosis, but also to have pathologic assurance of normal appearing nerves without hypertrophied nerve trunks in a circumferential biopsy sample which includes muscularis and submucosa; based on the report of Muller et al 2012[22]. These protocols explain the slightly prolonged operative time, which was attributed to the wait time for the results of the intraoperative biopsies.

Only one patient had constipation with intermittent laxative use, and none had bowel dysfunction as excessive pull of anal canal and external sphincter were avoided.

In concordance with Marc and his group [26], we com pletely removed the aganglionic bowel apart from the preserved 1 cm above the dentate line, without leaving behind a long cuff as we only leave around 3 cm cuff. Although we have a limitation of having a small cohort, but we can conclude that the Soave approach is a safe, reproducible and elegant technique if properly planed irrespective of the patient's age. A larger multicenter prospective study is warranted to solidify our conclusion.

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