Mangment of Late post-operative complications in patients with Hirschsprung's disease

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Abstract: Background: Postoperative complications following Hirschsprung's disease surgery is a challenging health problem. Its proper timing and disgnosis of such complicaitons is of utmost importance. **Objective:** to evaluate the post-operative complications in patients with Hirschsprung's Disease after unsuccessful initial surgery and its management. **Patients and methods:** the study included 30 children from dameitta and Al-Hussin Unviersity hospitals (Al-Azhar University). All were reviewed for history taking, clinical examination and surgical intervention, complications and its treatment. **Results:** The most common initial clinical presentations were constipation and abdominal distension in all studied children. The most common surgical procedure was Soave (50.0%), then trans-anal-endo-rectal pull through (20.0%), myomectomy (16.7%), LAEPT (6.7%) and each of TAAPT and Swenson (3.3%). Late postoperative complications were in the form of constipation (26.7%), anastomic stricture (20.0%), enterocolitis, fecal incontinence and increased stool freuquncy, each (13.3%), rectal prolapse (10.0%) and fecal fistula (6.7%). Treatment before redo operation, (46.7%) underwent anal dilation, (40.0%) bowel management and (13.3%) medical treatment. Redo procedures were in the form of Soave (20.0%), strictureroplasty (6.7%), Duhamel (3.3%); Swenson (10.0%), resection anastomosis (10.0%) and each of trans-anal pull through and myomectomy, (3.3%). **Conclusion:** postoperatrive complications after HD surgery still represent a surgical problem. Proper diagnosis and selection of initial surgical intervention and timing of this intervention may reduce the occurrence of postoperative complicaitons.

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1. Introduction

Hirschsprung's disease is an inherited condition that causes newborn intestinal obstruction. Children with the disease suffer dangerous intestinal blockages because they lack key nerves that drive the muscle contractions needed to move digested material. The disease occurs more often in males than in females, and sometimes is associated with Down syndrome and other inherited conditions **(Marc et al., 2012).**

Surgical treatments for Hirschsprung's disease have evolved over the decades. Surgery involves "pull-through" techniques to remove non-functioning portions of the bowel. Most surgeries are successful. However, there is little published about the outcomes for patients who do not fare well after initial treatment **(Elhalaby et al., 2008).** Various methods have been developed for the surgical treatment of Hirschsprung disease (HD). The traditional pull- through techniques described by Swenson, Soave, and Duhamel have been modified in attempts to reduce the size of the surgical wound, minimize injury to surrounding structures during intra-abdominal dissection, and improve bowel function. Laparoscopic surgery has been adopted in the management of HD **(Hussam et al., 2008).**

Patients who have undergone surgery for Hirschsprung disease can suffer from severe constipation and incontinence. Management of patients with these surgical problems has greatly informed care of patients with idiopathic constipation **(Elhalaby et al., 2008).** There various reasons explaining why some patients with Hirschsprung's disease may continue to suffer from obstructive symptoms or recurrent enterocolitis after the definitive management that necessitates medical treatment or even redo surgery. The causes of these unfavorable postoperative functional results can be grouped into two categories: the first one is related to incomplete resection of the aganglionic and/or the hypo-ganglionic transition zone, and the second is due to local anatomical problems including anastomotic strictures, rigidity of the ano-rectal cuff, and fistulas **(Langer, 2004).** Additionally, there are patients with marked dilatation of the rectosigmoid colon secondary to years of constipation, and this bowelsegment is unable to regain its muscular tone **(Coran and Teitelbaum, 2000).**

Three main diagnostic procedures are routinely used to reach a rapid and an accurate decision: 1. the ano-rectal palpation by the most experienced surgeon, general anesthesia may be needed in patients with stricture. 2. Radiological contrast studies to show the gross anatomical pathology, the length of the remaining colon, the severity of any existing stricture, and possibly the length of the transition zone. 3.

Biopsies and histopathological examinations by an experienced pathologist. Using a well-defined algorithm based on the clinical status as well as precise diagnostic workup can put an end to the long suffering of patients with postoperative complications **(Langer, 2004).**

The decision to redo surgery is not always easy and usually represents a major psychological trauma to the patients and/or their families. A conservative approach using medications such as laxatives, colonic wash out, enemata, metronidazole for entero-colitis or less invasive procedures such as dilatations should be tried initially. However, these conservative methods or limited surgical procedure such as myectomy, longitudinal division of the anorectal cuff, V-Y-plasty did not work in the presence of residual aganglionosis. The decision to redo pull-through should not be delayed once a residual aganglionic segment is confirmed. In other cases with a confirmed existence of ganglion cells at the pulled through colon, such a decision was made only after all other measures of medical therapy, anal dilatation, and other less technically challenging surgical procedures have been attempt Choosing the redo pull-through technique there is no consensus regarding the ideal technique for redo pull-through. Endorectal pull-through was the preferred and the safest one **(Schweizera et al., 2007).**

Aim of the work

Evaluation and management of post-operative complications in patients with Hirschsprung's Disease after unsuccessful initial surgery.

2. Patients & methods

This study was designed as a randomized, unblended controlled study that was approved from ethics committee of faculty of medicine Al-Azhar university, in the periode between (2013- 2016). The study was conducted on 30 cases with late post-operative complications of HD. Conservative approach using medications was applied at first. If no response, minimal invasive procedures was applied if they fail re-do surgery was the only solution.

Inclusion criteria:

Patients with the following criteria were included in the present study: 1) both sexes, 2) age < 18 years, 3) postoperative complications which were happen after one month from the operation, and 4) the management was done in Al-Azhar university hospitals however the initial operation done in or out our hospital.

Exclusion criteria:

Patients with one or more of the following were excluded from the study: 1) If the patients exceed the age of 18 yers at time of study, 2) Pre-operative, intra-operative and immediate post- operative complications.

Preoperative preparation:

After plain abdominal radiology, washouts were undertaken at 4-6-h intervals until the child was decompressed and stable. A suction rectal biopsy was taken to establish the diagnosis. In most cases a barium enema was performed to determine the length of the aganglionosis. Washouts were then resumed and operation scheduled for the next list. During this period the child was kept fasting and total parenteral nutrition was started to provide nourishment and fluids. Antibiotics were commenced 4 h before operation.

Operative technique: was done as described by **Cass (1990)**

Post-operativ care:

The urinary catheter is removed on day 3 and the transanastomotic silastic drain on day 5. Nutrition is maintained with parenteral feeding and oral intake commenced on day 4. Antibiotics are continued for 5 days post-operatively. If there are any unexplained fevers or redness of the wound, flucoxacillin is added. Anal dilatations are not routinely undertaken in the first 3 months and have not been found to be necessary. However, long designated sutures can be left in place so that should there be signs of an obstruction and calibration with dilatation warranted, there would be clear identification of the position of the lumen. Once established on full oral feeds, the child can be discharged from hospital, usually on the 6^{th} or 7^{th} post-operative day. Three months post-operatively the anal anastomosis is calibrated and the parents shown how to perform a digital examination. These are not routinely performed, but reserved for situations where there is any distension, loose stools, or failure to defecate for 48 h.

Any late complications were ducomented and treated according to the clinical situations.

Redo pull through

All redo procedureswere performed in the presence of one senior surgeon. The workup protocol for patients with Hirschsprung's disease not doing well (obstructive or incontinent) always consisted of a contrast enema and inspection under anesthesia with biopsy. The time to wait for conservative management before deciding to perform redo surgery depended on the indication for redo surgery. The surgical technique used in a TERPT procedure was done as by **Dickie et al. (2014)**. The surgical procedure is performed in prone position with the placements of lone star pins just above the dentate line. Next a circular incision is made 1–2 cm above the dentate line and the full thickness plane of dissection is found with a consequent full-thickness pull-through. Any possible muscular sleeve (in case of a previous Soave procedure), or Duhamel pouch is removed. Once a redo Soave was performed, in that case the muscular

cuff (which was not obstructing) was only additionally cut over the full length. If necessary, an additional laparoscopy or laparotomy is performed for themobilization of the proximal colon (proximal to the splenic flexure). During surgery biopsies were taken to verify the presence of ganglion cells or aganglionosis and pathological nerve fibers. Final specimen was always sent to pathology for definitive diagnosis.

Statistical Methods:

Statistics in this study were performed by compute using Epi Info Software version 6.04, a word processing, database, and statistics program. Categorical variable were presented as relative frequency and percent distribiton, while quanititative data were presened as mean and standard deviation as measurements of central tendency and dispersion respectively.

3. Results

This study included 30 patients with Hirschsprung's disease. All these patients were subjected to functional colostomy before pull-through operation for Hirschsprung's disease, and their data were retrieved by checking their files and focusing on the findings in the following points: history taking, radiological examination, rectal biopsy, clinical manifestations, surgical treatment and postoperative follow-up. The study was performed at El Hussin and Damitta, Al azhar universty hospitals (Department of pediatric surgery).

The present study included 30 children, 24 of htem (80.0%) were males and 6 were females (20.0%) with male to female ratio of 4: 1. Age ranged from 20 days to 144 months; the mean age of the studied group was 41.45±39.18 months. One child (3.3%) was below 1 month of age; 9 children (30.0%) were up to one year of age; 10 children (33.3%) were from 1 – 3 years of age and 10 children (33.3%) were above 3 years of age. The most common initial clinical presentations were constipation and abdominal distension in all studied children, then delayed meconium passage in 27 children (80.0%), vomiting in 10 children (33.3%), enterocolitis in 8 children (26.7%) and fecal soiling in 2 children (6.7%) (Table 1).

The rectal biopsy was diagnostic in all patients, and barium enema was only diagnostic in 24 patients. As regard to seurgical procedure in studied children, the most common was Soave, done for 15 children (50.0%), tehn trans-anal-endotrectal pull through in 6 children (20.0%), myomectomy in 5 children (16.7%), LAEPT in 2 children (6.7%) and each of TAAPT and Swenson in one child (3.3%). Late postoperative complications were defined as those which occurred after 30 days of pull through operation for Hirschsprung's disease. These complications were in the form of constipation in 8 children (26.7%), anastomic stricture in 6 children (20.0%), enterocolitis, fecal incontinence and increased stool freuquncy, each in 4 children (13.3%), rectal prolapse in 3 children (10.0%) and fecal fistula in 2 children (6.7%). Treatment before redo operation was as the following; 14 children (46.7%) underwent anal dilation, 12 (40.0%) bowel management and 4 children (13.3%) medical treatment. The redo procedure was in the form of Soave in 6 children (20.0%); strictureroplasty in 2 children (6.7%), Duhamel in one child (3.3%); Swenson in 3 children (10.0%), resection anastomosis in 3 children (10.0%) and each of trans-anal pull through and myomectomy, each in one child (3.3%) (Table 2).

Table (1): Sex, age and initial clinical presentation in studied children

		n	%
Sex	Male	24	80.0
	Female	6	20.0
Age group	Neonate (<4weeks)	1	3.3
	Babies (up to 1 year)	9	30.0
	Toddlers (1-3 years)	10	33.3
	Above 3 years	10	33.3
	mean±SD; Min. - Max. (months)	41.45±39.18; 0.67-144	
Clinical presentation	Delayed meconium passage > 48h	27(80%)	80.0
	Constipation	30	100.0
	Abdominal distension	30	100.0
	Enterocolitis	8	26.7
	Vomiting	10	33.3
	Fecal soiling	2	6.7

Table (2): Surgical procedure in studied children

		n	%
Surgical Intervention	Soave	15	50.0%
	Trans anal endo-rectal pull through	6	20.0%
	Myomectomy	5	16.7%
	LAEPT	2	6.7%
	TAAPT	1	3.3%
	Swenson	1	3.3%
Late PO Complications	Constipation	8	26.7
	Anastomotic stricture	6	20
	Enerocolitis	4	13.3
	Fecal incontinence	4	13.3
	Increased stool frequency	4	13.3
	Rectal prolapse	3	10.0
	Fecal fistula	2	6.7
Treatment before Redo operation	Anal dilatation	14	46.7
	Bowel Managment	12	40.0
	Medical Treatment	4	13.3
Redo pull through Procedures	Soave	6	20.0
	Strictueroplasty	2	6.7
	Duhamel	1	3.3
	Swenson	3	10.0
	Trans anal pull through	1	3.3
	Myomectomy	1	3.3
	Resection anastmosis	3	10.0

4. Discussion

Hirschsprung disease (HD) is a chronic condition with long-term effects on the physical and psychosocial health of a patient **(Thakkar et al., 2017).** Hirschsprung's disease (HD) is a disease for which several surgical procedures have been developed and modified over the years. In the late 90s both laparoscopy and transanal (endorectal) pull-through (TERPT) techniques were introduced as less invasive procedures. Both laparoscopy and transanal pull-through procedures are increasingly becoming surgery of choice **(Dingemans et al., 2016).**

Although surgery is effective in most HD patients, up to 32% of these patients continue to have symptoms and complaints **(Dasgupta and Langer, 2008).** These persisting symptoms and complaints include fecal incontinence, constipation and persistent stooling problems, and Hirschsprung's diseaseassociated enterocolitis (HAEC) **(Ralls et al., 2012).**

Additional laxative treatment is generally needed and a very small fraction of the patients with obstructive symptoms needs secondary surgery. There are several indications when considering redo surgery for HD. Indications are obstructive symptoms caused by the primary disease with residual aganglionosis or transition zone pull-through. Other causes for obstructive symptoms are torsion of anastomosed colon, stenosis or fibrosis of the anastomosis, or residual cuff tissue after Soave-like surgery. In some cases the indication for redo surgery is a dysfunctional Duhamel pouch with subsequent obstructive symptoms, fecal incontinence or abdominal complaints **(Gobran et al., 2007).**

The present study was designed to evaluate the post-operative complications in patients with Hirschsprung's Disease after unsuccessful initial surgery and its management. It included 30 children from dameitta and Al-Hussin Unviersity hospitals (Al-Azhar University). All their sheets were reviewed for history taking, clinical examination and surgical intervention, complications and its treatment. The present study included 30 children, 24 of them (80.0%) were males and 6 were females (20.0%) with male to female ratio of 4: 1. These results are comparable to those reported by **Thakkar et al. (2017)** who reported that, 72% of included children were males and 28% were females. In addition, **Menezes et al. (2006)** reported that, Of the 259 patients with HD, 200 were males (77.2%) and 59 females (22.8%). Also, **Wang et al. (2004)** reported that, they analyzed 147 patients (125 boys and 22 girls) who received surgical treatment for HD. These results are comparable to the present work.

In the present work, the most common initial clinical presentations were constipation and abdominal distension in all studied children, then delayed meconium passage in 27 children (80.0%), vomiting in 10 children (33.3%), enterocolitis in 8 children (26.7%) and fecal soiling in 2 children (6.7%). These results are comparable to those reported by **Vu et al. (2010)** who reported that, presenting symptoms included abdominal distention (100%), emesis (70.5%), and lethargy (5.8%). Meconium was passed within 24 h in 13.7%, between 24 and 48 h in 41.1%, and greater than 48 h in 45.2%. In addition, results of the present study was comparable to those reported by **Mabula et al. (2014)** who reported that, at presentation, sixty-four (58.2%) patients presented with complete intestinal obstruction while 42 (38.2%) presented with chronic intestinal obstruction and 4 (3.6%) patients had intestinal perforation. There was no patient with enterocolitis. Constipation and abdominal distention were the most common presenting complaints in 94.5% and 92.7% of cases, respectively.

In the present study, the rectal biopsy was diagnostic in all patients, and barium enema was only diagnostic in 24 patients (80.0%). These results are comparable to those found by **Vu et al. (2010)** who reported that, a contrast enema was diagnostic in 49 of the 51 patients who underwent the study (96%). Confirmation of the diagnosis of Hirschsprung disease was made by a rectal biopsy in all patients pre-operatively. In addition, **Adiguzel et al. (2016)** reported that, HD was diagnosed in 38 patients based on barium enema and anorectalmanometry findings. In 11 (22 %) patients, HD was diagnosed before TEPT with rectal biopsy, and in one patient, HD was diagnosed based on laparotomy with biopsy. Preoperative barium enema and anorectal manometry were performed in 49 and 47 patients, respectively. Barium enema showed a transitional zone in 43 (86 %) patients. The transition zone was present in the rectosigmoid in 38 patients, rectum in 3, and proximal to the sigmoid colon in 2 patients. Barium retention for more than 24 h was found in 45 (90 %) patients. Anorectal manometry revealed lack of the rectoanal inhibitory reflex in 39 (78 %) patients. In all the patients, the diagnosis was confirmed using perioperative frozen section biopsy examination. Unfortunately we did not use rectal manometery in the present work. In addition, it was reported that, three tests are typically used for the initial workup of HD. The first one is barium enema, and the typical finding of this test is the transition zone. The second is anorectal manometry: in this test, the absence of the rectoanal inhibitory reflex is indicative of HD. The third diagnostic test is rectal biopsy, which remains the gold standard for the diagnosis of HD **(Adiguzel et al., 2009).** A rectal biopsy is usually a simple and incident-free procedure, but it can result in bleeding,

perforation, perirectal fibrosis, and sepsis, and is associated with a failure rate of 13–22 %. The first test was usually performed for the workup of HD is anorectal manometry, which has demonstrated good sensitivity and specificity (91 and 93 %, respectively) in a systematic review **(de Lorijn et al., 2006).**

As regard to surgical procedure in studied children, the most common was Soave, done for 15 children (50.0%), then trans-anal-endo-rectal pull through in 6 children (20.0%), myomectomy in 5 children (16.7%), LAEPT in 2 children (6.7%) and each of TAAPT and Swenson in one child (3.3%). In their work, **Dingemans et al. (2016)** reported that, the initial surgical procedur in included children was in the form of Rehbein in 3 children (19.0%), Duhamel in 3 children (19.0%), TERPT (Soave-like) in 8 children (50.0%) and TERPT (Swenson-like) in 2 children (13.0%). These results are comparable to those of the present study. On the other hand, **Mabula et al. (2014)** reported that, the definitive pull-through was performed in 94 (85.5%) patients. Out of 94 patients who had definitive pullthrough, 76 (80.9%) underwent Swenson's pull-through and the remaining twelve (12.8%) and four (4.3%) patients underwent Duhamel's and Soave's pull-through procedures, respectively. In one (1.1%) patient who had an ultra-short segment of the disease, Lynn's dorsal myectomy was done because of the ease of application. Ileostomy was performed after total colectomy in one (1.1%) patient who had total colonic involvement.

As regard to late postoperative complications, these were in the form of constipation in 8 children (26.7%), anastomic stricture in 6 children (20.0%), enterocolitis, fecal incontinence and increased stool freuquncy, each in 4 children (13.3%), rectal prolapse in 3 children (10.0%) and fecal fistula in 2 children (6.7%). Our results are in agreement with **Thakkar et al. (2017)** who reported that, therewere 15 episodes of enterocolitis in 11 (15%) patients. This incidence lies also within the range reported by **Demehri et al. (2013)**. They reported enterocolitis to occur in 5 – 42% after HD surgerical intervention. The etiology of enterocolitis is multifactorial and includes defective mucosal immunological tolerance, partial obstruction, infection as well as genetic causes. The latter has particularly been implicated when considering the increased risk of enterocolitis in patients with Trisomy 21. In addition, it was reported that, EC is considered as one of the main complications in patients with HD. The rates of postoperative EC vary from 0 to 66.6 % in various published series **(Ekema et al., 2003; Langer, 2004).**

The rate of fecal incontinence in the present work (13.3%) lies with the range of previous literature, where it was reported that, rates of fecal incontinence post pull-through for HD range from 3 to 53% **(Bai et al., 2002). Menezes et al. (2006)** reported that, at long-term follow up, 10.3% of patients had fecal incontinence. These results are comparable to the present work. Howevr, some authors noted that patients with severe incontinence showed no improvement over time, whereas others noted a marked degree of improvement in the rates of continence with time **(Saleh et al., 2004).**

In the present work, constipation was reported in 28.6% post HD surgery. Constipation following a pull-through for HD can be attributed to prolonged colonic transit time, post-operative stricture or retained aganglionic segment. Hence, the value of a good history and thorough physical examination supported by the necessary diagnostic investigations cannot be undermined. The typical rate of constipation post pullthrough ranges from 6 to 34%. Some series have noted the Swenson procedure to have a high degree of constipation **(Saleh et al., 2004).**

In the present work, fecal incontinence and increased stool freuquncy, each in 4 children (13.3%). These results run in percetanges of previous literature, where it was reported that, patients with total colonic aganglionosis would generally have a poorer outcome with respect to their bowel function, when compared to patients with long segment and rectosigmoid disease. They would tend to have a higher stooling frequency and greater incidence of soiling **(Menezes et al., 2006).** However some series have described the stooling frequency to have decreased steadily over time and even the rate of incontinence to have improved **(Wildhaber et al., 2005).**

In the present work, anastomitic stricture was reported in 20% postoperatively. **Rouzrokh et al. (2010)** reported that, the most common postoperative complication of HD surgery was anastomotic stricture. Anal stenosis is one of the most serious and disabling complications of anorectal surgery. Several studies have indicated it in 5–10% of anorectal surgery **(Brisinda et al., 2009; Rouzrokh et al., 2010)**.

As regard to treatment before redo operation; 14 children (46.7%) underwent anal dilation, 12(40.0%) bowel management and 4 children (13.3%) medical treatment. In their work, **Ralls et al. (2016)** reported that, twenty-eight percent of patients at their institution underwent intervention in an attempt to alleviate obstructive symptoms prior to RedoPT as per published algorithm. These included anal dilations (two patients), posterior myotomy or myectomy (seven patients), or Botox injection into the anal sphincters. The indications for dilations were stricture or tight cuff.

As regard to redo procedures, it was in the form of Soave in 6 children (20.0%); strictureroplasty in 2 children (6.7%), Duhamel in one child (3.3%); Swenson in 3 children (10.0%), resection anastomosis

in 3 children (10.0%) and each of trans-anal pull through and myomectomy, each in one child (3.3%). These results are more or less comparable to those reported by **Sheng et al. (2012)** who reported that, the re-operation ranged from posterior sagittal approach combined with laparotomy in seven patients, Soave (conventional and transanal) procedure in seven patients, Duhamel procedure in one patient, Rehbein operation in three patients, simply re-using the stapling device in five patients after initial Duhamel operation, and repairing the rectovaginal fistula via laparotomy in one patient after initial Duhamel operation. They added, the indications for re-operation can be classified into three groups: (1) Anatomical problems, such as anastomotic stricture, twisted pull-through, retained dilated segment, obstructing Duhamel pouch, and obstructing Soave cuff; (2) pathological problems, for instance, residual aganglionosis, retained transition zone bowel; (3) other problems, for example, HD-associated enterocolitis, fistulae (rectocutaneous, recto-urethral and rectovaginal), etc.

Performing a Redo is difficult in many respects. This is true both in the operating room but also in the initial workup and decision tree. Only experienced pediatric surgeons following an appropriate algorithm should take on these cases. Determination that redo is necessary is the first step. However, the redo operation carries risks far greater than the original surgery and each patient should be approached on an individual basis and an operative intervention should be planned according to its presentation, type of complication, underlying pathology, and previous surgical history. Because of this, operative approach can vary widely and is heavily dependent on these factors as well as surgeon preference as to the type of repair. An open ERPT was the most commonly performed RedoPT procedure (38%). This was followed by a Swenson (25%), Duhamel (13%), and transanal ERPT (7%) **(Ralls et al., 2016).**

Conclusion:

Postoperatrive complications after HD surgery still represent a surgical problem. Proper diagnosis and selection of initial surgical intervention and timing of this intervention may reduce the occurrence of postoperative complicaitons.

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