Defecation Disorders after Surgery for Hirschsprung's Disease in Children; an Iranian Experience

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Abstract: Hirschsprung's disease (HD) is a congenital disease of the intestinal nervous system characterized by absence of ganglionic cells in distal colon followed by functional obstruction. After corrective surgery, the majority of children with HD develop defecation disorders such as soiling, constipation, fecal incontinence, and/or enterocolitis. The aim of this investigation was to determine the prevalence, diagnoses, therapies, and 6-month clinical outcomes in children with HD after corrective surgery. In this cross-sectional study performed at pediatric surgery ward in Tabriz Children's Hospital, prevalence, diagnosis and treatment of defecation disorders after surgery for HD were studied. First, defecation pattern was determined in 230 HD patients who had undergone surgery within previous ten years. Later, thirty patients with severe defecation disorders were recruited. Diagnostic and therapeutic procedures were performed. Clinical outcome was evaluated after six months. Defecation pattern was normal in 65% of the patients. In 21% of the cases, defecation disorders were mild and negligible, with no need of treatment. In 13% (30 patients), the pattern was impaired. Soiling, constipation, fecal incontinence and enterocolitis were the postoperative disorders. Of 30 patients with defecation disorders, 18 children (60%) and 12 patients (40%) had undergone multi-stage and TOSEPT surgical procedures, respectively. Defecation disorder was developed in 25.3% and 7.7% of the patients underwent multi-stage and transanal one-stage endorectal pull-through (TOSEPT) surgical procedures, respectively. Twelve patients and 10 children were treated with reoperation and medical therapy, respectively. Clinical outcome was excellent in five patients (16.7%), good in 15 patients (50%), fair in 8 patients (26.7%), and poor in 2 patients (6.6%). In conclusion, majority of the children with HD and postoperative defecation disorders have a favorable long-term clinical outcome when treated with minimally invasive surgical methods such as TOSEPT. Moreover, postoperative defecation disorders can be successfully treated using surgical procedures, medical therapy, as well as teaching both parents and their children.
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1. Introduction

Hirschsprung's disease (HD) is a congenital disease of the intestinal nervous system characterized by absence of ganglionic cells in distal colon followed by functional obstruction (Kenny et al., 2010). Common surgical methods for HD are multi stage (Swenson, Duhamel and Soave methods). Transanal one-stage endorectal pull-through (TOSEPT) is the newest minimally invasive restorative surgery procedure for HD patients (Aslanabadi et al., 2008; Gunnarsdóttir and Wester, 2011). One of the post-operative complications following surgery for HD is defecation disorders. Some studies reported up to 60% incidence rate of these disorders in children with HD after operation (Zhang et al., 2005; Fujiwara et al., 2007; Huang et al., 2008; Stensrud et al., 2010; Chumpitazi and Nurko, 2011; Zakaria, 2012). These disorders not only pose many problems for the patients and parents, but also have harmful psychological and physical effects. As some parents do not consider these disorders abnormal, they attribute these disorders to the nature of the disease and therefore try to get along with them.

With regard to high prevalence of the defecation disorders in HD patients, particularly in multi stage surgical procedures, a precise diagnostic and treatment strategy should be planned. Therefore, we aimed at evaluating all HD patients underwent surgical procedures for their disease during previous

ten years to determine the prevalence of defecation disorders and manage these complications.

2. Material and Methods

In a descriptive-cross sectional study at Pediatric Surgery Ward in Tabriz Children's Hospital, all 288 HD patients underwent surgery during previous ten years (from March 2000 to March 2009) were studied. Through telephone call, all parents were asked about their children's defecation patterns including number of defecations, fecal soiling, fecal continence, diarrhea, constipation and anal stenosis. Of 230 patients answered telephone calls, 30 patients suffering from defecation disorders were recruited for further investigation and treatment. Medical history was taken and clinical examinations were performed completely. Clinical and paraclinical data including age, gender, length on the involved piece (short segment of rectum, rectosigmoid, long segment of colon, or extended long segment of colon), and type of surgical procedure (multi-stage or TOSEPT) were recorded in questionnaires.

Paraclinical diagnoses such as barium enema or rectal biopsy were performed if required. In cases of transition zone report in barium enema indicating residual aganglionosis, reoperation was performed. Patients with normal barium enema were categorized in two groups: patients with non-retentive fecal incontinence and those with obstructive symptoms, soiling or enterocolitis. In the first group, after ruling out other diagnoses, treatment was performed through teaching both parents and children bowel self-control and food diet adjustment. In the second group, non-surgical interventions including specific food diet, treatment with laxative, teaching chronic constipation preventive measures. Patients were followed up every two months for the period of six months. At sixth month, treatment results were recorded and compared with the previous findings. The recorded complications included constipation, fecal incontinence, fecal soiling, enterocolitis and anal stenosis. These complications were diagnosed and treated based on the previous investigations (Azarfarin et al., 2013; Keshtgar et al., 2003; Levitt et al., 2009; Menezes et al., 2006; Dasgupta and Langer, 2008).

Based on the severity of symptoms before and after the treatment interventions as well as patients and their parents' satisfaction, clinical outcome was categorized as following: (1) poor, no improvement or worsening symptoms during 6-month follow up requiring continuous treatment or other treatment methods; (2) fair, partial improvement in symptoms, reduction in bowel movements in cases with fecal incontinence and increased frequency of defecation in constipation requiring long-term medical or supportive treatment; (3) good, satisfactory improvement in symptoms and signs requiring short-term treatments during first three months of follow up; and (4) excellent, total improvement of symptoms requiring no further treatments.

Data were presented as mean ± standard deviation (SD) and frequency (%). The statistical analysis was performed using SPSS for windows version 19.0 using Chi-square test and multi-variant test, whenever appropriate. A P value <0.05 was considered statistically significant.

3. Results

Two hundred and thirty HD patients underwent surgery during previous ten years were studied. Defecation pattern was normal in 65% of the patients. In 30 patients (13%), 23 boys and 7 girls, defecation patter was abnormal. Other cases include mild defecation disorder requiring no treatment. In all studied patients, rectosigmoid was the involved segment. Of 230 patients, 74 children and 156 patients had undergone classic (multi-stage) and TOSEPT surgical procedures, respectively. Of 30 patients with defecation disorders, 18 children (60%) and 12 patients (40%) had undergone multi-stage and TOSEPT surgical procedures, respectively.

Defecation disorder was developed in 25.3% and 7.7% of the patients underwent multi-stage and TOSEPT surgical procedures, respectively. All enterocolitis cases were seen in patients underwent multi-stage surgical procedures. Fecal incontinence was detected in three patients underwent multi-stage surgical procedures (Swenson, Duhamel, and Soave methods: each 1 patient). Two patients underwent TOSEPT had fecal incontinence.

Twelve patients and 10 children were treated with reoperation and medical therapy, respectively. In 24 cases, parents were taught appropriate toileting and proper diet. Performed operations included sphincteroplasty (2 patients), resection of the residual aganglionic section and re-pull-through (2 patients), surgical treatment of anal stenosis (4 patients), colostomy or ileostomy (3 patients), and sigmoid stricturoplasty (1 patient). Broad spectrum antibiotic and rectal washing were administered in two patients with severe enterocolitis. Maintenance therapy with stool softener or laxatives was used in treatment of constipation and soiling.

Six-month follow-up of the patients showed persistent defecation disorder symptoms in 8 patients (26.6%). Clinical outcome was excellent in five patients (16.7%), good in 15 patients (50%), fair in 8 patients (26.7%), and poor in 2 patients (6.6%).

4. Discussion

The present study revealed that defecation pattern was normal in 65% of HD patients underwent surgery. Moreover, normal defecation pattern was more seen in children treated with TOSEPT method rather than the multi-stage methods. This finding is similar to that of the investigations by Huang et al (2012) and Teitelbaum et al (1997) indicating more favorable results of TOSEPT method regarding control over defecation and normal defecation pattern compared to other HD surgical treatment procedures.

In our study, soiling (50%) followed by constipation (33%) was the most frequent defecation disorder after surgery for HD. Soiling was still present in five patients following the required treatments. It seems that soiling is a persistent complication attributed to the highly invasive surgical methods. However, soiling has not been considered as a severe defecation disorder in some studies and therefore it has not been a target of treatment. Similarly, Lu and Chen (1998) found that 74% of their surgically treated HD patients had soiling. Nonetheless, Hyman (2005) and Menezes and Puri (2005) reported constipation as the most frequent post-operative defecation disorder in HD patients.

In our study, clinical outcome was excellent in 16.7% and good in 50%, while only two patients (6.6%) had no response to the treatment. In the study by Chumpitazi and Nurko (2011), following treatments such as reoperation, medical therapy, and teaching parents and children special defecation methods, the clinical outcome was excellent, good, and poor in 28%, 38.6%, and 31.6%, respectively. Comparing results shows more favorable outcome in our study. This might be due to more use of minimally invasive surgical methods, i.e. TOSEPT, leading to decrease in the number of patients with constipation and fecal incontinence. Furthermore, continuous use of bougienage after TOSEPT results in reduction of postoperative anal stenosis incidence and enterocolitis.

In conclusion, majority of the children with HD and postoperative defecation disorders have a favorable long-term clinical outcome when treated with minimally invasive surgical methods such as TOSEPT. Moreover, postoperative defecation disorders can be successfully treated using surgical procedures, medical therapy, as well as teaching both parents and their children.

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