Comparison of outcomes of two methods of surgery (J-pouch and ileorectal anastomosis) in children with total colonic aganglionosis, Hirschsprung’s disease.

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Abstract

Background and objectives: Hirschsprung disease is commonly diagnosed in early childhood by aganglion areas in rectal biopsy. Diagnosed infants usually undergo a primary procedure and the definitive surgical treatment is usually performed several months later. Different surgical methods have been proposed for its treatment, but the detailed outcome of each method should be further investigated. Thus, we aimed to retrospectively assess the outcomes between two surgical methods for consistency of intestinal continuity, including J-Pouch and ileorectal anastomosis, in total colectomy procedure, in patients with total colon aganglionosis in our center.

Materials and methods: In this study, we retrospectively assessed all children undergoing total colectomy after primary ileostomy in Children’s Medical Center Hospital, from 1994 to 2016. In this center, intestinal continuity was provided by J-pouch procedure from 1994 to 2003, and by ileorectal anastomosis from 2003 to 2016. In the second method, 0.2 mg/kg loperamide was started after the first surgery (ileostomy) and was increased until the skin around ileostomy was just like the intact skin around colostomy with no significant inflammation. Data including demographic characteristics, need for re-ileostomy, duration of hospitalization, duration of NPO after surgery, and amount of loperamide were recorded, and compared between the two groups. Postoperative short-term complications were also recorded and compared. During the three-year follow-up period, all patients were assessed for soiling and fecal continence.

Results: Among 37 patients undergoing total colectomy due to total colon aganglionosis, 48.6% underwent J-pouch procedure (group 1) and 51.4% ileorectal anastomosis (group 2). In general, 54.1% were female and 45.9% were male. Mean hospitalization time was significantly lower in the second group (P=0.000). Mean NPO time was 7.06 ± 2.55 days in the first group and 3.63 ± 0.49 days in the second group (P=0.000). The rate of enterocolitis and mean surgical duration were significantly higher in the first group (P=0.001, and 0.000). None of the patients reported any fecal incontinence or constipation after surgery in both groups. Other postoperative complications had no statistically difference regarding leak, peri-anal inflammation, number of defeactions, soiling, anastomosis stricture, need for re-ileostomy, pelvic abscess, peritonitis, and adhesion band.

Conclusion: As the results of the present study indicated, the surgical method of ileorectal anastomosis has significant advantages to J-pouch procedure, including less hospitalization time, surgical duration and NPO duration, no cases of enterocolitis, fecal incontinence or constipation, which, in general, indicates that ileorectal anastomosis is a better method than J-pouch.

Keywords: Hirschsprung disease, Surgical procedures, Operative procedures.
including J-Pouch and ileorectal anastomosis, in total colectomy, in patients with total colon aganglionosis, in our center.

Materials and Methods

Study design

In this study, we retrospectively assessed all children undergoing total colectomy due to total colon aganglionosis, in Children’s Medical Center Hospital, from 1994 to 2016. Diagnosis of the disease was made by contrast enema and biopsy, then all patients underwent a primary ileostomy and after several months, when the child could tolerate a gross surgery, they underwent total colectomy. In this center, intestinal continuity during this procedure was provided by J-pouch procedure from 1994 to 2003, and by ileorectal anastomosis from 2003 to 2016. The classic J-pouch procedure was performed through the first group and in the second method; ileum was directly attached above dentate line by direct anastomosis. In order to reduce the number of defecations, increase the absorption time, and reduce inflammation, 0.2 mg/kg loperamide was started after the first surgery (ileostomy) and the dose was increased until the skin around ileostomy was just like the intact skin around colostomy with no significant inflammation.

All children with established Hirschsprung disease, who underwent total colectomy due to total colon aganglionosis, were enrolled into the study and patients who had other accompanying diseases, such as syndromic patients were excluded from the study. In addition, any patient with involvement of ileum or proximal parts of the intestine was not included in the study.

Data including gender, age at ileostomy and at definitive surgery, type and duration of the definitive surgery, need for re-ileostomy, duration of hospitalization, duration of NPO after surgery, and amount of loperamide were recorded, and then compared between groups. Postoperative short-term complications were recorded as well, including anastomosis leak and stricture, peri-anal inflammation, pelvic abscess, peritonitis, constipation, enterocolitis, adhesive bands, and long-term complications, included soiling, manometric results, and fecal incontinence.

Statistical analysis

Continuous variables are presented as mean ± standard deviation and qualitative variables are reported as frequencies (percentage). Comparison of variables between the two groups was performed by chi-square and one-way ANOVA tests. Statistical analysis was performed using SPSS 20.0 software (SPSS Inc., Chicago, IL, USA). P-values less than 0.05 were considered statistically significant in all tests.

Results

A total of 37 patients underwent total colectomy during the study period, among which 48.6% underwent J-pouch procedure (Group 1) and 51.4% underwent ileorectal anastomosis (Group 2). In general, 54.1% were female and 45.9% were male patients. The sex distribution of patients was not significantly different between the two groups (p=0.56). Mean age of patients at ileostomy was 19.83 ± 29.14 months in the first group and 21.74 ± 47.76 months in the second group (P=0.885). Mean age of patients at definitive surgery was 13.17 ± 3.2 months in the first group and 17.47 ± 3.7 months in the second group (P=0.001).

Mean hospitalization time was significantly lower in the second group compared with the first group (4.95 ± 0.7 vs. 9.11 ± 3.37 days, P=0.000). Besides, mean NPO time was remarkably lower in the second group as well (3.63 ± 0.49 vs. 7.06 ± 2.55 days, P=0.000). Mean amount of loperamide, which was only administered in the second group, was 2.68 ± 0.885 mg.

Postoperative complications are summarized in Table 1, which indicates no statistically difference regarding leak, peri-anal inflammation, number of defecations, soiling, anastomosis stricture, need for re-ileostomy, pelvic abscess, peritonitis, and adhesion band. Surprisingly, although 8 patients in the first group experienced enterocolitis, no case of enterocolitis was recorded in the second group which was statistically significant (P=0.001) Mean surgical duration was 227.78 ± 26.69 minutes in the first group and 88.42 ± 13.54 minutes in the second group (P=0.000) (Figures 1 and 2).

<table>
<thead>
<tr>
<th>Variables</th>
<th>Group 1</th>
<th>Group 2</th>
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<tr>
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</tr>
<tr>
<td></td>
<td>Negative</td>
<td>15</td>
<td>19</td>
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<tr>
<td>peri-anal inflammation</td>
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<td>1</td>
<td>4</td>
</tr>
<tr>
<td></td>
<td>Negative</td>
<td>17</td>
<td>15</td>
</tr>
<tr>
<td>Number of defecations</td>
<td>Once</td>
<td>4</td>
<td>3</td>
</tr>
<tr>
<td></td>
<td>Twice</td>
<td>4</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Three</td>
<td>3</td>
<td>8</td>
</tr>
<tr>
<td></td>
<td>Four</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
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</tr>
<tr>
<td></td>
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<td>17</td>
</tr>
<tr>
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<tr>
<td></td>
<td>Negative</td>
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<td>15</td>
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<tr>
<td>Need for re-ileostomy</td>
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<td>0</td>
</tr>
<tr>
<td></td>
<td>Negative</td>
<td>16</td>
<td>19</td>
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<tr>
<td>Pelvic abscess</td>
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</tr>
<tr>
<td></td>
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<td>Adhesion band</td>
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<tr>
<td></td>
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Figure 1. Intraoperative photo of total colectomy and ileorectal anastomosis in total colon aganglionosis.
reported a 10% prevalence of postoperative enterocolitis, which
Criado et al. evaluated 73 patients undergoing transanal
complication as it causes morbidity and mortality [13]. Martínez-
Hirschsprung-related enterocolitis is an important postsurgical
incontinence or constipation.

Accordingly, the three-year follow-up period in the present
study might not be sufficient enough for evaluation of fecal
complications after surgery and the rest of complications was not statistically
different between the groups.

Importantly, the proposed ileorectal anastomosis procedure is
associated with higher patients’ comfort as the result of lower
hospitalization time, NPO duration and surgical duration (P=0.000), and lower
rate of enterocolitis and (P=0.001). None of the patients in
both groups reported any fecal incontinence or constipation after surgery and the rest of complications was not statistically
different between the groups.

Several studies reported postsurgical complications after surgery
for Hirschsprung. Levitt et al. considered fecal incontinence a
serious problem after operative management of Hirschsprung
disease and attributed it to the surgical technique [11]. In
addition, a very recent systematic review indicated higher rate
of postoperative complications between two procedures, in order to give
physicians and researchers a broad overview on different
postsurgical complications. However, the limitations of current
study included small sample size and short follow-up, which
was due to the low incidence of the disease. Therefore, it is
suggested that multicentric studies provide larger sample
sizes. In addition, future randomized controlled trials are
recommended in order to provide a powerful evidence on the
efficacy of each therapeutic procedure.

Hirschsprung-related enterocolitis is an important postsurgical
complication as it causes morbidity and mortality [13]. Martínez-
Criado et al. evaluated 73 patients undergoing transanal
endorectal descent procedure for treatment of Hirschsprung and
reported a 10% prevalence of postoperative enterocolitis, which
was not associated with length of aganglionic segment [14].
Elhalaby and colleagues also reported 21.4% postoperative
enterocolitis [15] and Hackam et al. reported a prevalence of
32% with no cases of mortality, which was associated with
mechanical factors related to anastomotic complications and
intestinal obstruction [16]. In the study by Marty et al., all
cases of Hirschsprung-related death were due to postoperative
enterocolitis [8], which indicates the significance of this
postoperative complication. Carneiro et al. have also reported
less postoperative enterocolitis after Duhamel procedure,
compared to pull-through procedure and mentioned that it was
associated with patient’s sex and family history of Hirschsprung
disease [17]. Yet, the results of the present study indicated no
cases of postoperative enterocolitis in ileorectal anastomosis,
compared to 44.4% in J-pouch procedure, which was not
associated with patient’s sex. Although different procedures
have reported different rates of postoperative enterocolitis, none
of the studies have reported zero cases for this complication,
which was reported by the present study in the ileorectal
anastomosis method (Table 1).

Marty and colleagues reported severe soiling as a common
problem (12.6%) after surgical procedures, including
endorectal pull-through, modified Duhamel and long side-to-
side anastomosis for total colonic aganglionosis [8], which was
5-10% and statistically not different between the two procedures
of the current study. A meta-analysis of studies compared the
outcomes between laparoscopically-assisted and totally trans
anal endorectal pull-through procedures in 405 patients, which
documented no mortality rate with no statistically significant
difference in fecal incontinence, constipation, and enterocolitis
between the procedures [18].

The strengths of present study included comparing various
complications between two procedures, in order to give
physicians and researchers a broad overview on different
postsurgical complications. However, the limitations of current
study included small sample size and short follow-up, which
was due to the low incidence of the disease. Therefore, it is
suggested that multicentric studies provide larger sample
sizes. In addition, future randomized controlled trials are
recommended in order to provide a powerful evidence on the
efficacy of each therapeutic procedure.

In conclusion, as the results of current study indicated, the
surgical method of ileorectal anastomosis had priorities to
J-pouch procedure, in patients with total colon aganglionosis,
including less hospitalization time, surgical duration and
NPO duration, no cases of enterocolitis, fecal incontinence
or constipation, which, in general, proposes that ileorectal
anastomosis is a more efficient method than J-pouch.

None of the patients in both groups reported any fecal
incontinence or constipation after surgery.

Discussion

As the results of the present study indicated, the surgical
methods of ileorectal anastomosis (group 2) had some priorities
to J-pouch procedure (group 1), including less hospitalization
time, NPO duration and surgical duration (P=0.000), and lower
rate of enterocolitis and (P=0.001). None of the patients in
both groups reported any fecal incontinence or constipation after surgery and the rest of complications was not statistically
different between the groups.

Figure 2. Barium enema of an infant with total colon aganglionosis.


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