



# Usefulness of posterior sagittal anorectoplasty for redo pull-through in complicated and recurrent Hirschsprung disease: Experience with a single surgical group<sup>☆</sup>



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## ABSTRACT

**Aim:** To retrospectively examine 12 patients with Hirschsprung disease (HD) who underwent posterior sagittal anorectoplasty (PSARP) for various complications.

**Methods:** This study included patients with HD who underwent redo pull-through (PT) via PSARP at our institute between 2005 and 2014. The type of initial procedure, clinical presentations, indications, and functional results were analyzed. Postoperative excretory function was assessed using the Krickbein classification.

**Results:** The study group comprised 9 boys and 3 girls (total, 12). Five patients were diagnosed with rectosigmoid aganglionosis, 5 with long segment aganglionosis, and 2 with total colonic aganglionosis. The primary operations performed on these patients included the Soave, Duhamel, Swenson, Rehbein, and Ikeda–Soper procedures. The interval between the primary operations and reoperation ranged from 5 months to 8 years (median, 3 years). The indications for PSARP were rectocutaneous fistulae (6 cases), frozen pelvis (5 cases), severe anastomotic stricture (3 cases), rectovaginal fistulae (2 cases), and hemorrhagic proctitis with an inflammatory polyp (1 case). All fistulae were repaired using PSARP; only one rectocutaneous fistula recurred and required two additional surgeries. Stricture and hemorrhagic proctitis were cured in all involved cases. Nine patients were followed up for 8 months to 10 years after PSARP surgery (average, 5.1 years). All 9 patients had voluntary bowel movements within 6 months after the last PSARP and stoma closure: 3 had normal bowel movement, while 6 had varying degrees of soiling, depending on the length of residual colon. None complained of constipation.

**Conclusion:** PSARP is useful for treating severe complications of failed PT in HD. Complex and recurrent rectocutaneous fistulae and frozen pelvis are the main indications for PSARP, while soiling is the most common surgical complication.

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Hirschsprung disease (HD), or congenital aganglionic megacolon, is a malformation resulting in intestinal obstruction in neonates. Since the pathological mechanism and surgical procedures involving a full-thickness rectosigmoid dissection were first described by Swenson in 1948, the prognosis of HD has evolved considerably [1]. HD is usually surgically treated using the pull-through (PT) procedure, which involves removing the affected part of the bowel and then connecting the healthy section directly to the anus to allow passage of feces. Surgical procedures have witnessed vast improvements in the last half century, which have

resulted in satisfactory outcomes and fewer complications in most HD patients. Despite this progress, however, some patients still require redo PT procedures because of complications like residual aganglionic segments, anastomotic stricture, and fistulae [2–5]. Most redo PT procedures are conducted via the transanal approach [4,5]. However, in the case of fistulae or a frozen pelvis, lesions that need cleaning are not easy to expose and it is difficult to separate the colorectum with this approach or the transperineal approach. In such cases, posterior sagittal anorectoplasty (PSARP) is suitable and sometimes the only choice, as it allows isolation and exposure of perianal and perirectal tissue. Nevertheless, the indications of PSARP for redo PT surgeries have rarely been examined, and to our knowledge, the outcomes and complications of redo surgery using PSARP have not been reported exclusively. Further, no systematic study about the application of PSARP in treating the complications of HD has been published thus far.

In the present study, we retrospectively examined the cases of HD patients who underwent redo PT surgery via PSARP in order to summarize the clinical experience of this procedure.

**Abbreviations:** PSARP, Posterior sagittal anorectoplasty; HD, Hirschsprung disease; PT, Pull-through.

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**Table 1**

Primary operations conducted in the 12 patients who underwent redo PT surgery via PSARP.

Case no.	Type of aganglionosis	Primary procedure
1	Long segment	Unknown
2	Rectosigmoid	Duhamel
3	Rectosigmoid	Ikeda–Soper
4	Long segment	Unknown
5	Total colonic	Soave
6	Long segment	Soave
7	Rectosigmoid	Soave
8	Rectosigmoid	Unknown
9	Rectosigmoid	Unknown
10	Total colonic	Rehbein
11	Long segment	Soave*
12	Long segment	Swenson

\* This patient underwent the Soave procedure twice.

## 1. Materials and methods

This retrospective study included HD patients who underwent redo PT via PSARP between 2005 and 2014 at the Children's Hospital of Fudan University (CHOFU) for various complications after the primary surgery. Patients who underwent redo PT via any other procedure were excluded. This study was approved by the ethics commission of CHOFU.

Basic information, initial procedures, age at the primary surgery and reoperation, previous treatment for the complications before PSARP, indications for redo surgery, complications after redo surgery, and functional outcomes were examined for all included patients. Using a standardized questionnaire, an experienced surgeon followed up the patients for 8 months to 10 years via phone, email, or face-to-face interviews at our clinics. The questionnaire covered stool frequency, stool control, medication use, soiling, incontinence, response to diet, defecation training, cleansing enema, social problems, constipation, and enterocolitis. Anal contraction was assessed through electrical stimulation just before surgery for stoma closure. Postoperative excretory function was evaluated using the Krickbeck classification system [6]. Briefly, patients were classified into 3 main groups: group 1 patients were those with continence and regular voluntary bowel movements without constipation or soiling. Group 2 patients were partially continent and were further divided into 3 subgroups depending on the severity of soiling. Group 3 patients had constipation and were further divided depending on responses to dietary changes.

## 2. Results

A total of 46 patients underwent redo PT during the 10-year study period. The transanal approach was used in 34 patients, mostly because of residual aganglionosis and transition zone bowel. The remaining 12

patients, that is, 9 boys and 3 girls were included in this study. Their ages at the primary PT surgery ranged from 16 days to 7 years (median, 6 months), and those at redo PSARP surgery ranged from 6 months to 9 years (median, 4.7 years). The intervals between the primary operations and redo surgeries ranged from 5 months to 8 years, with a median time of 3 years. On the basis of the notes recorded during the primary operation and exploration for redo surgery, 5 patients were diagnosed with rectosigmoid aganglionosis, 5 with long segment aganglionosis, and 2 with total colonic aganglionosis (Table 1). Two of the 12 patients were reoperated using PSARP alone, while the remaining 10 (which included all those with long segment and total colonic aganglionosis) additionally needed surgery, the abdominosacral approach; all the abdominal surgeries were open surgeries. The residual colons were resected during the redo PT in 4 cases of long segment aganglionosis because of over-distension.

The indications for PSARP included complex or recurrent rectocutaneous fistula in 6 cases, severe pelvic scarring and fibrosis ("frozen pelvis") in 4 cases, severe anastomotic stricture in 3 cases, rectovaginal fistula in 2 cases, and hemorrhagic proctitis with inflammatory polyp in 1 case (Table 2). There was more than one indication in some cases. Three of the 6 rectocutaneous fistula patients had undergone fistula curettage, incision, or excision and experienced a recurrence, for which PSARP was performed. The remaining 3 underwent PSARP directly because of special conditions: one had a complex fistula with 2 fistulous orifices on the perianal skin, another had undergone the Soave procedure twice previously, and the last patient had a huge cavity in the path of the fistula.

The primary operations performed on the 12 patients are listed in Table 1. Information regarding these surgeries was not available for 4 patients who had been treated at local hospitals. Seven patients had undergone a single PT operation only, 1 had undergone the Soave procedure twice at a local hospital, 1 had undergone dilation of rectal stenosis under anesthesia, and the remaining 3 patients had undergone more than one rectocutaneous fistula surgery after failed PT. Some patients also underwent biopsy examination of the rectal mucosa, enterostomy or closure operations, colonoscopy or perineal examination, and surgical incision and drainage of perianal abscesses, but the effects of these procedures were not examined in the present study.

All the patients were examined using contrast enema before PSARP except the patient with persistent hematochezia, who was examined by colonoscopy. PSARP was planned after perineal examination under anesthesia in 5 patients or through a discussion among experienced surgeons in another 5 cases. For the remaining 2 patients, the procedure was switched to PSARP during the operation because they had a frozen pelvis, which could not be isolated through the transanal or transperineal approach. Protective enterostomies were performed in 10 cases simultaneously with PSARP or before it. The other 2 patients had already undergone enterostomies when they came to us. Perioperative examination during PSARP showed that the anal canal and dental line were mainly intact in all 12 patients, although sphincter contractility was damaged to varying degrees in 4 patients, mainly at the same position as the infection.

**Table 2**

Indications for PSARP in the 12 patients in the present study.

Case no.	Chief complaint	Indication
1	Anastomotic leakage, enterostomy	Scarred pelvis; severe anastomotic stricture
2	Discharge of pus from the anus and abdominal wound	Severely scarred pelvis
3	Ectopic defecation	High rectocutaneous fistula
4	Repeated abdominal distension and diarrhea	Severely scarred pelvis
5	Recurrent abscess at the perianal region and ectopic defecation	Recurrent rectocutaneous fistula
6	Persistent hematochezia and difficulty with defecation	Anastomotic stricture; inflammatory polyp
7	Defecation through the vagina	Rectovaginal fistula
8	Hyperpyrexia and perianal abscess	Recurrent rectocutaneous fistula; scarred pelvis
9	Defecation through the vagina	Rectovaginal fistula; anastomotic stricture
10	Ectopic defecation	Recurrent rectocutaneous fistula
11	Perianal suppurative infection and ectopic defecation	Recurrent rectocutaneous fistula
12	Anastomotic dehiscence and ectopic defecation, enterostomy	Rectocutaneous fistula; severely scarred pelvis

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