



Colorectal Conditions

Primary Posterior Sagittal Anorectoplasty Outcomes for Rectovestibular and Perineal Fistulas Using an Accelerated Pathway: a Single Institution Study

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ABSTRACT

Background: Posterior sagittal anorectoplasty (PSARP) is the most common surgical treatment for patients with anorectal malformations. Such patients are often subjected to prolonged *nil per os* (NPO), antibiotic use, and use of parenteral nutrition. Our aim was to review our institutional experience with patients undergoing PSARP using an accelerated standardized postoperative pathway.

Methods: Our hospital database was queried from 2004 to 2016 for patients diagnosed with imperforate anus who underwent a surgical procedure. Short term outcomes, hospital length of stay (LOS), and cost were evaluated.

Results: Sixty-three patients were identified during the study period. Of these patients, 34 (54.0%) had a fistula to the urogenital tract or had no demonstrable fistula and one cloaca and 29 (46.0%) had a fistula in the perineum. Approximately half of patients underwent primary PSARP, including 8 patients with fistulas located in the vestibule and vagina in girls and two with no apparent fistulas (12.7% of total cohort). Only two postoperative complications occurred: one superficial surgical site infection and one perineal wound dehiscence. Among the whole cohort, median LOS was 3 days. Median time to PO intake was 2 days, and median cost was \$11,532. No complications occurred among the subset of 8 patients undergoing primary PSARP.

Conclusion: Patients undergoing PSARP experienced similar outcomes compared to historical series, suggesting that the accelerated pathway for early refeeding and reduced use of antibiotics may be beneficial in appropriately selected patients.

Type of study: Case series with no comparison group

Level of evidence

Level IV.

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Anorectal malformations or imperforate anus represents a wide spectrum of anomalies that affect the development of the hindgut. Both males and females are affected, with males exhibiting a slightly higher prevalence. The diagnosis is usually determined shortly after birth by routine physical examination. Physical exam findings include absent anus, intestinal obstruction, stool emptying from the perineum, introitus, or vaginal orifice, or fecaluria in a male. Radiologic investigations include distal colostogram to identify site of fistula, voiding cystourethrogram, plain abdominal radiographs, and Ultrasounds of

the spine, kidneys and perineum. De Vries and Pena described the anatomy of the Posterior Sagittal anorectoplasty (PSARP) procedure in 1982 for children and infants with anal atresia [1]. This procedure standardized the surgical care of children born with anorectal malformations and led to improved outcomes for bowel continence. When the procedure was initially described a protective colostomy was performed in patients with lesions that did not have a perineal fistula or those that involved the urogenital system to reduce the risks of postoperative complications. Customarily, patients with perineal fistulas do not require colostomy [2]. Short term complications following PSARP included anastomotic disruption, perineal or pelvic sepsis, superficial surgical site infections, urologic complications, and rectal prolapse [3]. In the past decade, many have published results for primary PSARP for rectovestibular lesions with good outcomes. In these reports, many of these patients have undergone a bowel prep, prolonged course of antibiotics and nothing per os (NPO), often requiring parenteral nutrition [4].

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While PSARP without protective colostomy has shown to be an effective and safe surgery, there has been little work done on surgical outcomes with an accelerated standardization of postoperative care. The aim of our study was to assess our short-term outcomes using an accelerated postoperative protocol for the care of our patients undergoing PSARP. All patients undergoing a PSARP with or without protective colostomy were included in the study. We evaluated the safety and costs with respect to overall postoperative hospitalization stay, duration of antibiotics and time to oral intake.

1. Methods and methods

This study was approved by our Institutional Review Board (Study #00005111). Data for this study were obtained from our Institutionally-based data repository of PSARP patients. Patients who underwent PSARP at our institution from 2004 to 2016 were identified within this prospectively maintained data repository. Individual charts were analyzed for the data points of interest. Patients were followed up for 30 days. Operating surgeons all adhered to the same pre- and postoperative care of children with anorectal malformations. No patients underwent mechanical bowel preparation prior to surgery. All patients received perioperative antibiotics in the operating room, which were continued for a maximum of 48 h postoperatively. All patients without a protective stoma were kept NPO for less than 48 h postoperatively, when a diet was initiated, thereby avoiding the need for central venous access and parenteral nutrition. All patients had a urinary drainage catheter in place for 48 h.

Outcomes of interest for our cohort of patients undergoing PSARP included postoperative complications, postoperative length of stay (LOS), time to oral feedings, duration of antibiotics, and hospitalization costs. These outcomes were also assessed for a particular subgroup of patients, using the accelerated pathway, who would have traditionally been treated according to more conservative pathways: patients presenting with vestibular and vaginal fistulas and no fistula who underwent primary PSARP without the use of a diverting colostomy.

2. Results

Sixty-three patients were identified during the defined study time period that underwent a PSARP. Approximately half of the study cohort underwent primary PSARP, including 8 patients, 2 of which with vestibular fistulas, 4 with rectovaginal fistulas, and two patients with no fistula; the remaining patients undergoing primary PSARP had rectoperineal fistulas. The remainder of the study cohort underwent a 3-stage procedure, defined as an initial diverting colostomy, followed by the PSARP procedure, with a later colostomy closure. The most frequent type of fistula among patients undergoing 3-stage PSARP was a rectourethral fistula (43.7%), of which 3 patients had a rectobladderneck fistula, 2 patients had a rectobulbar fistula, and 4 had a rectoprostatic fistula. The patients with rectobladderneck fistulas required a combined abdominal and perineal approach. One patient in the 3-stage PSARP cohort had cloaca. The two patients with no fistula that underwent primary PSARP had both an invertogram and perineal US demonstrating a 1 cm distance from bowel gas to the marked location on the skin where the anus should be located.

Characteristics of the entire study cohort, dividing by approach, are shown in Table 1. Of note, a significantly higher proportion of patients in the primary PSARP cohort underwent PSARP within the newborn period (19 of 31 patients [61.3%] less than 30 days of age at surgery) versus those undergoing 3-stage PSARP within the newborn period (4 of 32 patients [12.5%], $p < 0.001$). Of the patients undergoing primary PSARP outside of the newborn period ($n = 12$), the ages at time of PSARP were 2 months ($n = 2$), 3 months ($n = 3$), 5 months ($n = 3$), 6 months ($n = 2$), 8 months ($n = 1$), and 9 months ($n = 1$); no postoperative complications occurred among this subset of 12 patients.

Table 1
Characteristics of PSARP patients.

Variable	Primary PSARP (n = 31) 6 days (2 days–3 months)	3- Stage PSARP (n = 32) 4 months (2.5–7)
Age (median, IQR)		
Sex		
Male	29.0%	68.7%
Female	71.0%	31.3%
Location of anomaly		
No fistula	6.4%	25.0%
Perineal fistula	74.2%	18.8%
Rectourethral	0.0%	43.7%
Vestibular fistula	19.4%	9.4%
Cloaca	0.0%	3.1%
Comorbid conditions		
Tethered cord	12.9%	25.0%
Vesicoureteral reflux	16.1%	9.4%
Tracheoesophageal fistula	0.0%	12.5%
Trisomy 21	0.0%	0.0%
Myelomeningocele	0.0%	6.3%

Among the whole cohort, the median LOS for the PSARP procedure hospitalization was 3 days (IQR 2–5 days) with median postoperative LOS of 3 days (IQR 2–4 days). The median length of time to PO intake was 2 days (IQR 1–2 days), and the median total cost was \$11,532 (IQR \$7306–\$17,136).

Postoperative complications were categorized into major and minor subsets (Table 2). A minor complication occurred in 1.6% of the total study cohort, which was a superficial surgical site infection in the perineal wound occurring in a patient with a rectourethral fistula who did have a descending loop colostomy. PSARP was performed at 6 months of age. The distal colon was clear of stool as demonstrated by the distal colostogram and intraoperative distal colonic lavage with dilute betadine solution. This healed with antibiotics and local care without apparent problems. The patient has been followed for 8 years for management of his constipation with stimulant laxatives. There was also 1 major complication (1.6% of cohort), which was comprised of a partial anastomotic dehiscence of the perineal wound that required a return to trip to operating room for a diverting loop colostomy on postoperative day 3. This complication occurred in a patient with a rectoperineal fistula who underwent primary “limited PSARP” on day of life 4 without prior diverting stoma. The patient has been followed for 6 years for management of his constipation with stool softeners. The patient has no problems with soiling or incontinence. The perineum has healed without evidence of prolapse or stricture. Notably, no complications occurred among the subset of 8 patients with primary PSARP.

3. Discussion

Posterior sagittal anorectoplasty is the most common surgical procedure performed for the management for anorectal malformations. In the last decade, primary PSARPs are being increasingly performed for vestibular fistulas because of the risks and complications associated with colostomies [3]. Multiple studies support primary PSARP not only for rectoperineal fistulas but also for anomalies in selected patient populations involving the vestibule [5–11]. Kuijper et al. assessed the feasibility, safety, advantages, and follow-up of anterior or posterior

Table 2
Complication profiles of cohorts.

Type	Primary PSARP (n = 31)	3- Stage PSARP (n = 32)
No complications	30 (96.8%)	31 (96.9%)
Surgical site infection	0 (0%)	1 (3.1%)
Perineal wound dehiscence	1 (3.2%)	0 (0%)
Early bowel obstruction	0 (0%)	0 (0%)
Central venous line sepsis	0 (0%)	0 (0%)

anorectoplasty performed without colostomy in 35 children and showed that anorectoplasty without colostomy is safe and exhibits low morbidity with good outcomes [6]. Additional benefits proposed for primary PSARP were noted in a prospective study by Adeniran et al., showing that after posterior sagittal anorectoplasty patients without colostomy were able to pass stool without need of stool softeners or enemas [10]. Key benefits of these approaches include relieving the alimentary tract obstruction at birth, eliminating urinary tract contamination, establishing anorectal continuity thus giving maximal potential for normal defecation reflexes at birth, and early development of the cerebral cortical fibers via sensations of rectal fullness allowing continence to develop.

In the present study, both newborn and nonnewborn patients with a variety of anorectal malformations received accelerated postoperative care, defined as shorter time to oral feedings of 2 days or less, avoidance of parenteral nutrition with central venous access, and with an average postoperative length of stay of 3–4 days and short course of antibiotics of 48 h. Traditionally, PSARP for rectoperineal fistulas done outside of the newborn period without a protective stoma as well as those fistulas involving the vestibule or vagina undergoes a preoperative bowel prep 24 h prior to the surgical procedure, placement of a central venous catheter for total parenteral nutrition (TPN), and patients are kept nothing per os (NPO) for 7 days [4]. Our series contained a wide range of patient ages and anorectal malformations, as well as both patients who underwent a primary PSARP and those who underwent a 3-stage procedure, and our outcomes from this accelerated postoperative care pathway appear to be safe and well tolerated. This was particularly true in the selected subgroup of our patients who underwent primary PSARP with vestibular, vaginal and no fistulas, which traditionally would have been treated with the more conservative pathway described above.

Overall, in our series of patients who underwent an accelerated pathway, low complication rates were observed, with one patient having an anastomotic dehiscence requiring diverting colostomy and one patient with a minor surgical site infection. We had no patients with rectal prolapse or urologic injury. Our short term complication rates were comparable with those reported in the literature: specifically, rectal prolapse 1.5%, partial wound or anal dehiscence and or infection of 0.6%, and urologic injuries ranging from 0.2% to 0.4% [4].

Other aspects of PSARP procedures that are highly variable across series and remain debated among pediatric surgeons are the appropriate duration of antibiotic therapy, as well as the length of time remaining NPO postoperatively. Compared to our study, higher postoperative infectious complication rates have been reported to be 33% to 41% when either perioperative to 24 h of antibiotics was used as compared to 11% to 26% when antibiotics were extended to 2 to 5 days, in patients who remained NPO for 3 to 5 days [6]. Clinical practice guidelines for antimicrobial prophylaxis in general surgical procedures recommend preoperative-dose time within 60 min before surgical incision, with 120 min allowed for some agents such as fluoroquinolones and vancomycin. The duration of prophylaxis postoperatively recommends a single dose or continuation for less than 24 h. This recommendation includes the presence of indwelling drains and intravascular catheters [14]. Despite a robust guideline for adults, pediatric-specific data are sparse. However, antibiotic selection and recommendations for the pediatric patients mirror those of adults.

Kumar et al. retrospectively evaluated 123 patients with vestibular and perineal fistulas that underwent a repair without a colostomy. Nonnewborn patients received mechanical bowel prep 24 h prior to surgery. Patients received postoperative antibiotics for 7 days, with a range of 5 to 12 days, and remained NPO for 3 to 6 days. Early complications included one missed rectal injury and two total wound dehiscences all requiring redo surgery; minor wound complications such as minor wound infections and superficial wound dehiscence had an occurrence rate of 10.6% [5]. In another series, Kulshrestha et al. published a similar group of patients who underwent primary ASARP for

rectovestibular fistula, all of whom received preoperative bowel preparation. They then stratified a group of patients to early (clear liquid diet initiated postoperative day 2) versus late (NPO for 9–10 days with partial parenteral nutrition) feeding. One patient from the early group had a major wound disruption requiring a sigmoid colostomy and later a redo repair, three patients had a subcutaneous leak which required partial opening of the perineal incision and healing by secondary intent, with 2 patients requiring a revision, and an additional seven patients had dehiscence of the mucocutaneous or skin sutures that did not require further treatment; the delayed group did not experience significant immediate postoperative complications [9]. Findings from both of the series contrast our own, in which only one patient who underwent a primary PSARP for a rectoperineal fistula suffered a major wound dehiscence requiring colostomy and redo anoplasty, and only one patient undergoing a three-stage procedure for a rectourethral fistula experienced a surgical site infection.

Aziz and colleagues prospectively evaluated outcomes for 23 patients with rectovestibular fistulae repaired via an anterior approach without a protective ostomy. Their patients did not have preoperative mechanical bowel prep but did receive preoperative antibiotics 3 days prior to surgery, and oral feeds were initiated on postoperative day 4; in their series, 3 patients (13.0%) developed a partial wound disruption [7]. Kuijper and Aronson assessed duration of antibiotics, time to oral intake and use of TPN following 35 patients undergoing either an anterior or posterior sagittal anorectoplasty procedure. Patients received either 24 h of antibiotics or antibiotics for 2 to 5 days, and remained NPO for either 3 or 5 days. Patients kept NPO for 5 days also received TPN. In the group of patients receiving 24 h of antibiotics, 43% has superficial wound infections, with one patient requiring colostomy and a second patient required a delayed redo anoplasty. The group receiving 2 to 5 days of antibiotics had an 11% rate of minor wound complications; these findings led the authors to conclude that at least 2 to 5 days of therapeutic antibiotics is indicated to reduce infectious complications [6]. In our series, infectious complication rates remained consistently low with a maximum course of 48 h of antibiotic therapy within our accelerated standardized care pathway.

The belief that a protective stoma will mitigate anastomotic complications or infectious complications has not been borne out [5–11]. The potential for ostomy complications and need for an additional operative procedure to close the ostomy, when compared to postoperative infectious complications reported infectious complication rates in the literature, must be weighed when making this decision [12,13]. Currently, the trend for many surgeons is to perform a one-staged procedure, for high anomalies. The cost benefits of a one-stage versus a three-staged procedure are significant, especially in the setting of comparable results [5].

4. Conclusion

To our knowledge, this is the only study reporting a defined surgical pathway for patients undergoing either a primary or 3-stage PSARP. The aim of this retrospective study was to evaluate the safety of early postoperative feeds and reduced antibiotic use in the treatment of ARMS. We conclude that patients undergoing PSARP who are started on early postoperative feeds (thus obviating the need for central venous access and parenteral nutrition) and decreased use of antibiotics experienced good outcomes, even among a subgroup of patients undergoing a primary PSARP with vestibular, vaginal and no fistulas. When we compare our complication rates to those in the reported literature, our rates are comparable or less compared to others. The results do suggest that in certain patients, regardless of receiving protective colostomy, an accelerated pathway may be beneficial to these patients. Thus far, there are no randomized prospective trials to direct length of antibiotic use and time to oral feeding following primary posterior sagittal anorectoplasty. As such, we are left with compiling data from published series and formulating a best practice guideline for the care of these often medically complex patients.

This study has several limitations. The data represented includes a small population. A large multi-institutional study will need to be conducted to validate these preliminary results.

References

- [1] deVries PA, Pena A. Posterior sagittal anorectoplasty. *J Pediatr Surg* 1982;17: 638–43.
- [2] Wilkins S, Pena A. The role of colostomy in the management of anorectal malformations. *Pediatr Surg Int* 1988;3:105–9.
- [3] Pena A, Migotto-Krieger M, Levitt MA. Colostomy in anorectal malformations: a procedure with serious but preventable complications. *J Pediatr Surg* 2006;41: 748–56.
- [4] Coran AG, Adzick NS, Krummel TM, et al. *Pediatric surgery*, 7th ed. St. Louis, MO: Mosby; 2012: 1289–1311.
- [5] Kumar B, Kandpal D, Sharma SB, et al. Single-stage repair of vestibular and perineal fistulae without colostomy. *J Pediatr Surg* 2008;43:1848–52.
- [6] Kuipper C, Aronson D. Anterior or posterior sagittal anorectoplasty without colostomy for low-type anorectal malformations: how to get a better outcome? *J Pediatr Surg* 2010;45:1505–8.
- [7] Aziz A, Banu T, Prasad R, et al. Primary anterior sagittal anorectoplasty for rectovestibular fistula. *Asian J of Surg* 2006;29:22–4.
- [8] Pakarinen M, Rintala R. Management and outcome of low anorectal malformations. *Pediatr Surg Int* 2010;26:1057–63.
- [9] Kulshrestha S, Kulshrestha M, Singh B, et al. Anterior sagittal anorectoplasty for anovestibular fistula. *Pediatr Surg Int* 2007;23:1191–7.
- [10] Adeniran JO. One-stage correction of imperforate anus and rectovestibular fistula in girls: preliminary results. *J Pediatr Surg* 2002;37:16–9.
- [11] Liu G, Yuan J, Geng J, et al. The treatment of high and intermediate anorectal malformations: one stage or three procedures? *J Pediatr Surg* 2004;39:1466–71.
- [12] Patwardhan N, Kiely EM, Drake DP, et al. Colostomy for anorectal anomalies: high incidence of complications. *J Pediatr Surg* 2001;36:795–8.
- [13] Nour S, Beck J, Stringer MD. Colostomy complications in infants and children. *Ann R Coll Surg Engl* 1996;78:526–30.
- [14] Bratzler DW, Dellinger EP, Olsen KM, et al. Clinical practice guidelines for antimicrobial prophylaxis in surgery. *Am J Health-Syst Pharm* 2013;195–283.