Stricture rate in patients after the repair of anorectal malformation following a standardized dilation protocol

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Abstract
Purpose The aim of this study was to determine and analyze the stricture rate in patients who underwent a PSARP or PSARVUP and followed a postoperative protocol of anal dilation (Fig. 1).
Methods A retrospective review of patients with anorectal malformation (ARM) who underwent a primary PSARP or PSARVUP from February 2016 to October 2021 was performed. Data collected included patients’ demographics, type of ARM, age at the time of operation, postoperative complications, with emphasis on whether there were any strictures or any difficulties during dilations, and on follow-up. During the surgical repair, emphasis was placed on preserving the blood supply of the bowel and performing a tension-free bowel-to-skin anastomosis.
Results Eighty-four patients met the inclusion criteria. Forty-four patients were females: 21 recto-perineal fistula, 12 cloaca, 9 recto-vestibular fistula, one imperforate anus without fistula, and one patient had a complex anorectal and vaginal malformation with an anal stricture and a rectovaginal fistula. Forty patients were males: 14 recto-perineal fistula, 11 recto-urethral bulbar fistula, 6 recto-urethral prostatic fistula, 6 imperforate anus without fistula, and 2 bladder neck fistula. One patient had an anal stenosis with sacral agenesis, without a presacral mass. Patient ages ranged from 0 to 79 months (mean 7.5 months, median 5 months) at the time of surgery. Follow-up time ranged from 7 to 73 months (mean 38 months, median 35 months). No patient suffered of a postoperative anal stricture. Six patients suffered of a rectal prolapse that required a surgical repair.
Conclusion Post-operative anal stricture after PSARP and PSARVUP can be avoided with proper surgical technique and postoperative care. Namely, by preserving adequate blood supply of the bowel and avoiding tension at the anoplasty, and by adhering to a structured protocol of anal dilations.

Keywords PSARP · Anal dilation · Dilation protocol · Anal stricture · Imperforate Anus · Anorectal Malformation

Introduction
Postoperative anal dilations are standard treatment since the early introduction of the posterior sagittal anorectoplasty procedure (PSARP) for children born with an anorectal malformation (ARM) [1]. Postoperative anal dilations are performed by over 80% of the pediatric surgeons [2] following a variety of protocols based on their personal preferences. In our practice, parents are taught how to perform anal dilations on their children 14 days after surgery. We start with a small Hegar dilator (8–9 mm), and advise the parents to increase the diameter every week, until they reach a predetermined diameter based on the patient’s age, when parents will then start the tapering phase of the protocol (Fig. 1). Recently, we were surprised to read reports in the literature about stricture rates as high as 26% following a PSARP, with some 22% of cases requiring further surgical intervention for stricture repair, [3]; with or without following a postoperative protocol of anal dilations. We aimed to analyze our stricture rate after PSARP and PSARVUP following our standardized protocol of anal dilations, in part because some surgeons believe that anal dilations are not necessary and that dilations produce severe pain and may result in significant psychological sequelae [4].
Methods

A retrospective review of all patients with anorectal malformation (ARM) who underwent a primary PSARP or PSARVUP at our institution from February 2016 to October 2021 was conducted, with patient follow-ups until April 2022 included in the review. All included patients followed our postoperative anal dilation protocol (Fig. 1). Patients were evaluated for presence of anal stricture at the time they reached the final Hegar size number (predetermined based on the patient’s age), and at the one-year follow-up clinical visit. Colorado Multiple Institutional Review Board (COMIRB) approval was obtained (COMIRB #22–0428). Data collected included patients’ demographics, type of ARM, age at the time of operation, postoperative complications, with an emphasis on the presence or absence of stricture, difficulties noted during dilations, and follow-up. During the surgical repair, emphasis was placed on preserving the blood supply of the bowel and on performing a tension-free bowel-to-skin anastomosis.

Results

Eighty-four patients underwent a PSARP or PSARVUP due to anorectal malformation at our institution and followed our postoperative anal dilation protocol. Patient ages ranged from 0 to 79 months (mean 7.5 months, median 5 months) at the time of surgery. Follow-up time ranged from 7 to 73 months (mean 38 months, median 35 months).
Forty-four (52%) patients were females. Twenty-one of them had a recto-perineal fistula. Twelve had a cloacal anomaly (8 had a common channel < 2.5 cm and 4 had a common channel > 3.5 cm). Nine had a recto-vestibular fistula and one with trisomy 21 had an imperforate anus without fistula. One patient had a complex anorectal and vaginal malformation with anal stenosis and a rectovaginal fistula.

Forty patients were males (48%). Fourteen had an ARM with a recto-perineal fistula, 11 had a recto-urethral bulbar fistula, and 6 had a recto-urethral prostatic fistula. Six had an imperforate anus without fistula (3 of which had trisomy 21), and two had an ARM with a recto-bladder neck fistula. One patient had an anal stenosis with sacral agenesis, without a presacral mass.

When asked about pain, suffering or discomfort experienced during dilations, parents described that most children manifested some discomfort but no real pain. Most parents were anxious prior to the first dilation.

None of the 84 patients suffered from postoperative anal stricture, and all patients reached the final Hegar size, as determined by their ages, without any problems. Eighty-one patients had no stricture at the 1-year follow-up. The remaining three patients have not yet had their 1-year follow-up as of the time of this submission.

Six patients, two female and four male patients, suffered from rectal prolapse postoperatively and required surgical intervention (Table 1). Rectal prolapse repair was performed within 2–12 months after the PSARP procedure.

**Discussion**

The need for anal dilations after a PSARP has been debated recently in the literature [3, 5–7]. We speculate that when anoplasties are performed without respecting the limits of the sphincter, or in patients without a sphincter, then the result is a large, patulous anal orifice, somewhat resembling a colostomy. These particular patients do not need dilations as they will never develop strictures (Fig. 2).

On the other hand, when the limits of the sphincter are respected, the anus is visibly closed at the end of the

| Patient | Gender | Diagnosis | Sacral Ratio | Spinal cord | Time after PSARP (months) | Repair technique^a |
|---------|--------|-----------|--------------|-------------|---------------------------|-------------------|
| 1       | F      | ARM with recto-perineal fistula | 0.8          | Normal      | 8                         | Plication         |
| 2       | F      | Complex cloacal malformation, 4 cm of common channel, bilateral hydrocolpos, 2x hemivagina, hemiuteri | 0.9          | Normal      | 12                        | Full thickness    |
| 3       | M      | ARM with recto-urethral bulbar fistula | 0.5          | Normal      | 8                         | Plication         |
| 4       | M      | ARM with recto-urethral bulbar fistula | 1            | Normal      | 2                         | Full thickness    |
| 5       | M      | ARM with high recto-urethral prostatic fistula | 0.35        | Low conus at L3 | 4 | Full thickness |
| 6       | M      | ARM with recto-bladder neck fistula | 0.8          | Asymmetric sacrum, low conus at L3, hemi-vertebra L5/L6 | 4 | Full thickness 1st attempt plication 2nd operation |

^aRepair techniques: full thickness resection or mucosal resection and muscular plication
In this cohort, most patients were younger than 6 months of age at the time of their PSARP. We acknowledge that trying to perform anal dilations in toddlers or older children should be discouraged, and this is one of the reasons why we prefer to do the PSARP at a young age.

The rate of rectal prolapse requiring surgical intervention in this study population was 7% and in the literature its occurrence ranges from 3.8% up to 27% [3, 8–10]. The incidence of rectal prolapse is important to mention when evaluating skin-level strictures after a PSARP and when debating the need for postoperative anal dilations. When rectal mucosal tissue is left exposed and is visible immediately after the conclusion of the PSARP, then the patient will have no risk for stricture at the muco-cutaneous junction. However, we advise against this practice as a means of preventing strictures because prolapsed mucosa has other inconveniences.

We acknowledge the limitations of this study due to it being both retrospective and non-randomized. Because of this, we cannot isolate the impact of the surgical technique versus the impact of the postoperative anal dilations in the incidence of postoperative strictures reported in this series.

**Conclusion**

Postoperative anal strictures after PSARP and PSARVUP can be avoided with a technically correct operation that emphasizes the preservation of adequate blood supply to the bowel and an anoplasty fashioned without tension, and a follow-up plan consisting of a structured (standardized) protocol of anal dilations.

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**Declarations**

**Competing interests** The authors declare no competing interests.

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