A prospective single-center cohort study on a modification of the posterior sagittal anorectoplasty technique for vestibular fistula in females

Original Article

Ahmed Kamal Sayed^a, Ibrahim Ali Ibrahim^b, Ahmed Hossney Ahmed^a, Mohamed Fathy^a and Alaa AE Moustafa^a

Department of Pediatric Surgery, ^aMinia University Hospitals, Minia University, Minia, ^bAssiut University Pediatric Hospital, Assiut, Egypt.

ABSTRACT

Background: The posterior sagittal anorectoplasty (PSARP) approach is used to repair the anorectal malformations (ARMs) with rectovestibular fistula. The incision extends from the coccyx posteriorly to the vestibule anteriorly cutting the perineal body (PB) skin. Dehiscence of the PB can have several long-term adverse events.

Aim: In this study, we modify the PSARP approach by leaving the PB intact and making the incision just limited to the anoplasty area.

Patients and Methods: We operated on 20 patients with a rectovestibular fistula using this modified technique in a single center between February 2023 and January 2024. The incision is limited to the anoplasty area with a final result of the skin incision is completely incorporated into the anoplasty. We assessed the mean operative time, intraoperative complications, postoperative complications, duration of hospital stay, scar cosmesis, and assessment of the continence for patients aged over 3 years old.

Results: Five patients were stooling via a colostomy and 15 patients via a fistula. Patients' ages ranged between 3 and 18 months. The mean operative time was 88.9 min. All cases healed with excellent cosmetic results. Intraoperatively, one case had a vaginal injury. Postoperatively, one case developed partial rectal mucosal prolapse. All patients were too young to be evaluated for bowel control.

Conclusion: This technique preserves all the benefits of the traditional PSARP and has the potential to eliminate PB dehiscence and its long-term complications. We believe that with a larger number of patients and longer follow-up periods, this technique will be a common approach.

Key Words: Anorectal malformations, perineal body, posterior sagittal anorectoplasty, PSARP, vestibular fistula.

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Corresponding Author: Ahmed Kamal Sayed, MSc, MBBCh, Department of Pediatric Surgery, Minia University Hospitals, Minia University, Minia, Egypt. **Tel.:** +201025979232, **E-mail:** a.kamal@s-mu.edu.eg

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INTRODUCTION

One in every 5000 live births is born with anorectal malformations (ARMs) which include a widespread spectrum of anatomic presentations^[1]. The diagnosis is commonly determined shortly after birth by routine physical examination and radiological investigations^[2]. ARMs are frequently associated with different congenital anomalies disturbing the short- and long-term outcomes^[3]. The etiology of this condition is supposed to be due to dysmorphogenesis of the urorectum and cloaca in early fetal development^[4]. The Wingspread classification in 1984 classified this spectrum of defects into high, intermediate, and low anomalies with different categories for males and females based on the relation of the rectal pouch with the levator ani muscle^[4].

In females, the most common ARM is an imperforate anus with a rectovestibular fistula^[5]. Management of this

problematic defect has significantly improved mainly after the introduction of posterior sagittal anorectoplasty (PSARP) by DeVries and Pena in the early 1980s^[6].

The perineal body (PB) is a complex fibromuscular structure found between the anal triangles and urogenital structures. This structure assists several vital functions, including preventing expansion of the urogenital hiatus, fixing the anorectum and vagina, preserving the orgasmic platform, providing a physical barrier between the rectum and vagina, and maintaining urinary and fecal continence^[7].

Steps of the traditional PSARP for rectovestibular fistula include doing a midline incision from the coccyx extending anteriorly through the anal sphincter complex and the PB to the fistula tract located within the vestibule. However, dissection of the PB affects the integrity of the region, making it susceptible to dehiscence postoperatively^[6].

After PSARP, several gynecologic complications have been illustrated, such as urethrovaginal fistula, rectovaginal fistula, acquired vaginal stricture, and insufficient or shortened PB. PB dehiscence is a common complication associated with the PSARP technique, which can have substantial implications for long-term functional outcomes^[8,9]. The morbidity from PB dehiscence can range from a superficial separation of the incision to affecting fecal continence^[7,10].

We aim in our study to evaluate a modification in the traditional PSARP technique by making the incision limited to the anoplasty area and leaving the PB intact to reduce the risk of postoperative PB dehiscence and related sequelae, and the effect of this modification on the outcomes and prognosis for vestibular fistula patients.

PATIENTS AND METHODS:

Our study is a prospective, single-center (at the Pediatric Surgery Unit, Minia University Hospital, Egypt), and single-arm clinical trial. After institutional review board approval (IRB Number: 644/2:2023), we included 20 female patients presented to our department and diagnosed with imperforate anus with rectovestibular fistula from February 2023 to January 2024. All patients' parents gave informed consent to participate in our study. Our trial was registered at PAN Africa Clinical Trials Registry (PACTR), Trial Registration Number: PACTR202407845732769; Date of Registration: 29 July 2024 'Retrospectively Registered'.

Patients were excluded if: (1) they were associated with major neurological, cardiac, syndromic, or skeletal anomalies; (2) they had other forms of ARMs; (3) they had a history of other anorectal operations; (4) they had recurrent rectovestibular fistula; (5) they were older than 14 years old; or (6) they were immunocompromised.

Preoperative preparation and positioning

All cases were checked for other elements of the VACTERL (Vertebral, Anal, Cardiac, Tracheoesophageal, Renal, and Limb anomalies) association before surgery. Patients stooling via fistula (noncolostomized) were admitted two days preoperatively for chemical and mechanical bowel preparation. Chemical preparation is a regimen of intravenous third-generation cephalosporin, Cefotaxime, at a dose of 50 mg/kg/6 h, plus Metronidazole at a dose of 15 mg/kg/day. Mechanical bowel irrigations with warmed saline 0.9% using a soft rectal tube (Foley catheter, size 14–18), three times per day at a dose of 25 ml/kg, which can be increased to 100–150 ml/kg for older females.

Antibiotic prophylaxis (Cefotaxime at a dose of 50 mg/kg) was administered, and a urinary Foley catheter was inserted for all cases after induction of general anesthesia.

Each patient was positioned prone, with a cushion placed below the hips to elevate the pelvis.

Operative steps

The anterior and posterior bounds of the sphincter complex were defined using electrical stimulation. A midline incision was then made using a cutting monopolar at the beginning of the posterior border of the muscle complex ending just before the PB (Fig. 1). The incision was made only in the middle of the sphincter ellipse, without any incision either anterior or posterior to the anoplasty. The dissection was then extended in the midline with the use of monopolar cautery, until the posterior rectal wall was reached. Retractors were placed with their pins positioned to expose the posterior rectal wall as well as the adjacent subcutaneous tissue (Fig. 2). The dissection continued, remaining right on the rectal wall; first, we defined the posterior and lateral walls before starting the circumferential dissection.

Full-thickness 4–0 vicryl traction stitches were attached to the rectal wall to mobilize the posterior rectum. The cautery was used to split the posterior rectal wall until we reached the rectal mucosa of the anterior wall, detecting the fistula from within (Fig. 3). The following step is to separate the rectum from the vagina. The rectum was cautiously elevated superiorly and outward to completely separate the common wall between the vagina and the rectum and reach the areolar tissue defining the two separate walls (Fig. 4). This complete dissection and separation of the rectal attachments allowed the rectum to be fully mobilized and achieve an appropriate length to reach the anoplasty without any tension.

When the rectum was adequately mobilized, the distal fistula was completely closed with continuous 4-0 Vicryl sutures, leaving the PB skin intact (Fig. 5). The parasagittal fibers, ischiorectal fat, and muscle behind the PB were closed with absorbable sutures. The site above the fistula was repaired using muscle.

The rectum was gently tractioned and sutured to the posterior limit of the sphincter complex. The anoplasty occupied the extent of the original incision and was performed with interrupted 4-0 Vicryl sutures, apposing the rectal mucosa to the skin. No posterior sagittal incision required repair. The ends of the sutures were left long and held in place with mosquito clamps to maintain exposure. After the anoplasty was completed, the long suture ends were cut to relieve tension in the area (Fig. 6).

Postoperative care and follow-up

After the patient recovered from anesthesia, feeding was started immediately, as the skin incision was entirely incorporated into the anoplasty, and there was no concern about the passage of stool through the rectum. Cases who are taking regular food or formula-fed were given laxatives to keep their stools thin and easy to pass.

The patients were scheduled for regular outpatient visits on a weekly basis for the first month, and then monthly visits thereafter. During each visit, the anoplasty was examined to ensure proper healing. Additionally, laxatives were continued to prevent constipation. A protocol of regular anal dilation was initiated two to three weeks postoperatively using Hegar dilators.

Intraoperative events

We assessed the operative time and intraoperative complications, including blood loss, vaginal injury, difficulty in identifying and separating the vagina from the rectum, and the number of cases converted to traditional PSARP.

Postoperative outcomes

We assessed the mean follow-up period, duration of hospital stay, and continence for patients over three years old using the 'International Krickenbeck Continence Score'^[11], while for patients younger than three years old, we used indicators (clinical assessment of anal tone, frequency of stools per day, and the presence of perianal excoriation beyond the first three months postoperatively) to predict continence. This was done through direct questioning of parents and inspection of the perianal area during the follow-up period. Postoperative complications included wound infection, wound dehiscence, anal stenosis, anal stricture, rectal mucosal prolapse, mislocated anus, constipation, remnant of the original fistula, and recurrent fistula. We evaluated cosmesis using 'The Scar Cosmesis Assessment and Rating Scale'^[12], which consists of two sections: clinician questions and patient questions^[12] which is formed of two sections; clinician questions and patient questions. We used only the clinician questions section because the patient section of the score included questions about itching and pain, while our patients were too young to answer those questions.

Statistical analysis

Descriptive statistics were completed using SPSS version 20.0. Alpha is set at the level of 5%. We present continuous variables as means±SD, and discrete variables as frequencies and ranges.



Fig. 1: Incision site limited to the center of the sphincter ellipse stopping just before the perineal body.



Fig. 2: Delivery of the posterior rectal wall with two full-thickness traction sutures aides in mobilization of the rectum.



Fig. 3: Detection of the fistula from within the rectum.



Fig. 4: Complete separation of the anterior rectal wall from the posterior vaginal wall.



Fig. 5: Closure of the fistula with continuous sutures with two full-thickness sutures to mark the edges of the fistula.



Fig. 6: The neoanus with no posterior sagittal incision and intact perineal body.

RESULTS:

Twenty patients with rectovestibular fistula underwent anoplasty by limiting the incision to the area of the sphincter complex and leaving the PB intact (modified PSARP). At the time of correction, five patients had a colostomy while 15 patients were stooling via a fistula. The age of the included patients ranged between 3 and 18 months. Preoperative bowel preparation was performed on patients stooling via a fistula. One of the patients had a large ventricular septal defect that was corrected surgically. Another patient had a common urogenital sinus, which was evaluated by cystoscopy and surgically corrected using partial urogenital mobilization. The baseline characteristics of the patients are shown in (Table 1).

The mean operative time was 88.9 min with minimal estimated blood loss. A vaginal wall injury occurred in one patient, for which a primary repair was performed, and no fistula developed during follow-up. There were no other intraoperative complications. None of the cases were converted to the traditional PSARP (Table 2).

The mean follow-up period was 7.2 months. The duration of the hospital stay ranged between 1 and 2 days for the colostomized patients and between 2–3 days for noncolostomized patients. No dehiscence of the anoplasty or other postoperative complications occurred, except for one case that developed a mild rectal mucosal prolapse, which will be corrected at the age of two years. All patients had excellent cosmetic results for their anus and perineum, with the Scar Cosmesis Assessment and Rating Scale scores of 0–1 for colostomized patients and 1–3 for noncolostomized patients, respectively (Table 3).

All patients were followed up in our outpatient clinic every week during the first month and then every month (Fig. 7). Two cases had a remnant dimple in the vestibule, with no fistula or sinus, as evidenced by the contrast enema. Additionally, there was no fecal discharge from the vestibule during follow-up in noncolostomized patients.

The five colostomized patients were returned for elective colostomy closure after reaching an adequate anoplasty size for their age. All of the included patients were younger than three years old and could not be evaluated using the International Krickenbeck Continence Score. During follow-up, all cases had good anal tone, passed 2–3 stools per day, and none of the cases had persistent perianal excoriation beyond the first three postoperative months.

Table 1: Baseline characteristics data of the included patients

Age at correction (months)*	3–18
Weight (Kg)*	4–12
Stooling (n)**	
Colostomy	5
Via Fistula	15
Preoperative Bowel Preparation (n)**	15

Variables are expressed as range*, frequency (n)**.

Table 2: Intraoperative events

Operative Time (min)*	88.9±14.58
Blood loss (ml)**	10-25
Intraoperative complications (n)	
Vaginal injury	1
Difficulty in identifying and separating the vagina from the rectum	1
Cases converted to ordinary PSARP (n)	0

Variables are expressed as mean \pm SD *, range **, frequency (n)||. PSARP, posterior sagittal anorectoplasty.

Table 3: Postoperative outcomes

Follow-up period (months)*	7.2±1.8
SCARS ^b	
Colostomized cases	0–1
Non-colostomized cases	1–3
Postoperative complications (n)**	
Wound infection	0
Wound dehiscence	0
Anal stenosis	0
Anal stricture	0
Rectal mucosal prolapse	1
Mislocated anus	0
Constipation	0
Remnant of the original fistula	0

Recurrent fistula	0
Duration of hospital stay (days)	
Colostomized cases	1–2
Noncolostomized cases	2–3

Variables are expressed as mean \pm SD *, frequency (n)**, range||. SCARS, scar cosmesis assessment and rating scale.



Fig. 7: Shape of the neoanus and the perineum at the follow-up (a) after 1 week, (b) after 1 month, (c) after 3 months, (d) after 6 months.

DISCUSSION

The traditional PSARP procedure involves a thorough understanding of sphincter muscle anatomy and precise surgical techniques, achieved through excellent exposure obtained by making an incision from the coccyx down to the fistula, including the PB^[6,13,14]. However, the literature reports a wide variation in the rate of wound and PB dehiscence after a single-stage PSARP, ranging from 0 to 48%^[2,15-18]. Several short-term negative impacts result from PB dehiscence, including infection, healing of the wound by secondary intention with poor cosmetic results, and prolonged hospital stay. Even more problematic are the long-term implications: increased fibrosis and scarring of the PB after dehiscence may cause anterior migration of the neoanus outside the muscle complex, potentially affecting fecal continence^[15,16].

Furthermore, scarring of the PB may cause narrowing of the vestibule with no sufficient length or strength of the PB to maintain its integrity during intercourse and vaginal delivery^[19–21]. Vilanova-Sanchez *et al.* found introital stenosis in 26.6% of their patients^[22]. At a large referral center for colorectal surgery, Vilanova-Sanchez *et al.* found that the most common reason for redo perineoplasty was an insufficient PB^[22]. Moreover, in adult women, a shortened or scarring PB is associated with significant dyspareunia and an increased risk of third to fourthdegree vaginal lacerations. Accordingly, Vilanova-Sanchez *et al.* concluded that cesarean delivery is favored for women with abnormal PB after surgical correction of ARMs^[23]. To decrease the frequency of wound dehiscence, several management practices have been used. These include mechanical bowel preparation, protective colostomies, delay of oral intake following the operation, loperamide administration, and antibiotics^[2,15,18,24]. Most recently, Martynov *et al.* reported the application of a perineal wound vacuum as a tool to counteract wound dehiscence^[25].

In our study, we evaluate a modified anoplasty technique for the surgical correction of patients with rectovestibular fistula by eliminating the division of the PB and making the incision limited to the area of the sphincter muscle complex instead of the long incision of the traditional PSARP.

Twenty cases with rectovestibular fistula were included in our study. Our mean operative time, which was calculated from the electrical marking of the sphincter complex to the last stitch, was shorter than that in the published literature by Badillo et al. 2023 and Ostertag-Hill et al. 2023 who operated on their cases by a similar technique^[26,27]. No intraoperative complications were observed, except for one case in which a vaginal injury occurred during dissection and was repaired primarily during the same procedure. No postoperative complications were observed during the follow-up period except for one case that developed a mild rectal mucosal prolapse which is comparable to the results published by Pena et al. that overall incidence is 3.8% in 833 patients with different types of ARMs^[28]. Our patients experienced frequent soiling and perianal dermatitis during the first three to four weeks postoperatively, which was treated with local agents and improved. The observed perianal dermatitis is in accordance with the published data by Hakalmaz et al. and Divarci et al.^[29,30]. By limiting the incision to the anoplasty area, patients can benefit from a shorter hospital stay and less restriction of their activity with no fear of PB dehiscence. All patients had good cosmetic results for their anus and perineum. Our results are comparable to those published in the literature^[26,27].

We could not assess fecal continence in our patients because they were younger than three years old. However, we expect good results based on the type of malformation addressed in our study, as reported by Minneci *et al.*^[31], and none of the cases had postoperative anatomic complications or persistent severe perianal dermatitis or soiling. All cases had good anal tone and pass stools 2–3 times per day.

Our study has a few limitations; (1) a relatively small sample size. However, this could be explained by the incidence of ARMs in the population and the nature of our study which is prospective. (2) Short follow-up period with no ability to assess for fecal continence, we will follow our patients in the future for good and accurate assessment of the long-term data.

CONCLUSION

In conclusion, we demonstrate that performing PSARP on vestibular fistula patients with a more limited incision to the anoplasty area is both safe and effective, while preserving all the benefits of the traditional PSARP. Furthermore, it has the potential to eliminate PB dehiscence and its long-term complications. This technique has less operative time, minimal estimated blood loss, insignificant intraoperative and postoperative complications, and excellent cosmetic results. We believe that with a larger number of cases and long-term follow-up periods, this technique will be commonly used.

CONFLICT OF INTEREST

There are no conflicts of interest.

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