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Original Research Article

Single Stage Posterior Sagittal Anorectoplasty of Vestibular Fistula at a Tertiary Care Centre

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Abstract

Background: ARM is a prevalent congenital defect of the anorectum in female patients; in the majority of cases, it is vestibular fistula. Conventionally treated over several phases using colostomy, this method is associated with morbidity, expense and psychological pressure. Colostomy-free single-stage posterior sagittal anorectoplasty (PSARP) has the potential advantages of less invasive interventions and enhanced continence by early ano-cerebro-cortical reflex development. The paper evaluates its safety and effectiveness in a tertiary care environment.

Materials & Methods: The case series was prospective and was held between September 2018 and August 2020 at Medical College Hospital Kolkata. Twenty female patients with vestibular fistula (15 anovestibular, 5 rectovestibular) aged 6 weeks to 3 years (median 6 months) were selected after ruling out pouch colon and life-incompatible anomalies using barium enema, echocardiography and ultrasonography. Preoperative preparation consisted of immunisation completion, bowel cleansing by use of polyethylene glycol and antibiotics. The PSARP was done under general anaesthesia in prone position with careful rectal-vaginal separation and anoplasty. Treatment in the postoperative period was nil oral for 5 days, IV nutrition, antibiotics, and frequent dressings. A 1-month follow-up every week, 3-month follow-up every month, and 3-monthly follow-up to 18 months were done to assess wound healing, continence, constipation, and complications.

Results: Blood loss during the operation was 23.75 ml (10-75 ml) on average and 15% needed a transfusion. Operation time was between 75 and 120 minutes. Vaginal wall injury (per-operative) was found in 20% and was repaired at the time. The average stay in the hospital after the operation was 10-12 days. Mild perineal excoriations 15 percent at week 1 with a resolution rate of 15 percent at week 3. One (5%) was a partial wound dehiscence that was treated by colostomy and secondary suture. Constipation necessitating laxatives was present in 15 percent at a maximum of 3 months and stopped at 6 months except one (stopped at 9 months). There wasone case of anal dilation required 5 to 6 months. No anal stenosis, recurrence of fistula, prolapse and incontinence. Normal bowel habits (1-3 stools/day) were obtained in all 16 patients who had 9 months or more follow-up and were not using aids.

Conclusions: Single-stage PSARP in the absence of colostomy is safe and effective in the treatment of the vestibular fistula in carefully selected females, with low complication rates, perfect continence, and no problems long-term, when pouch colon and major anomalies are excluded. It minimises morbidity and expenditure over and above compared to staged procedures.

Keywords: Vestibular Fistula, Posterior Sagittal Anorectoplasty, Single-Stage Repair.

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Introduction

Anorectal malformations (ARM) are a heterogeneous group of congenital malformations of the distal anorectum and the genitourinary tracts with a worldwide prevalence of about 1/5000 of live births. In some areas, such as India, it seems to be more common, with 1 in 1862 reported in Kolkata, which also shows possible geographical differences due to genetic, environmental, and socioeconomic factors [1]. These malformations can be easily detected at birth since they lack normal anal opening and are characterised by a spectrum of defects, starting with minor anomalies

with good prognoses to complex malformations with high morbidity and related congenital anomalies. ARM has undergone a tremendous classification change over the years. Earlier schemes, such as the 1970 International Classification, classified defects as low, intermediate or high depending upon their relationship to the puborectalis and levator ani muscles. In 1984, this was revised by the Wingspread Classification which streamlined classes and acknowledged cloaca as a class of its own. More recent developments are the descriptive classification of fistulae by Pena in 1995, with an emphasis on therapeutic implications such as perineal or vestibular fistulae and cloacal malformations, and a descriptive classification of fistulae types and rare variants by Krickenbeck in 2005 [2]. Vestibular fistula, including anovestibular and rectovestibular subtypes, is much more prevalent in females, especially in Indian populations, where it represents a significant share of the cases.

In females, long-standing practice included a multi-staged approach to treat vestibular fistula with neonatal colostomy followed by definitive repair (usually a posterior sagittal anorectoplasty, PSARP), and later colostomy closure. This is promoted by Pena and others as a method to reduce faecal contamination during repair and to protect the neoanus [3].

Colostomy, however, causes significant morbidity, such as diarrhoea, dehydration, prolapse, infection, anal stenosis, malnutrition, the psychological and economic costs of a series of surgeries, recurring anaesthetic procedures and extended stay. Additional complicating factors in terms of staged repairs include hypertrophic rectum, scarring, and delayed development of the ano-cerebro-cortical reflex, which is one of the key to continence [4].

By comparison, single-stage PSARP without colostomy has become a valid option because it reduces hospitalization rate, parental stress, and expenditure and retains neural connexions to early continence development. Theoretical benefits are increased synapse formation in sphincteric muscle and a decrease in morbidity in general. It was shown to be feasible in early literature with reports of no wound dehiscence or fistula recurrence, and excellent continence in 72 females with vestibular fistula without routine dilatations [1]. On the same note, Upadhyaya et al. altered PSARP to prevent fourchette incision with satisfactory cosmetic and continence results in 40 patients [3]. Controversies, however, exist about infection risks, wound dehiscence, and appropriateness in all cases. According to systematic reviews and meta-analyses, e.g. by Lauriti et al. who examined three studies with 156 females, there were more wound infections (24.3 vs 10.9) and dehiscence (16.2 vs 2.4) in single-stage repair, but equal rates of continence [4]. Randomised studies such as that of Gupta et al. including 64 females validated a much greater rate of dehiscence (39.4% vs. 18.2) and postoperative complications in primary repairs [7]. On the other hand, Amanollahi and Ketabchian had lower total morbidity in single-stage techniques despite initial infection [5], and Kumar et al. reported excellent continuence in 123 patients with vestibular and perineal fistulae [6]. It was pointed out by Gangopadhyay and Pandey who argued in favour of single-stage due to its resource-efficient nature, whereas more complex cases such as pouch colon are repaired in stages [2]. In a study by Wang et al., they proposed an altered version of the anterior sagittal anorectoplasty (ASARP) employing sphincter preservation and reduced complications in 26 patients [8]. To add to this current discussion, this study sets out to assess the safety, efficacy, and mid-term outcomes of single-stage PSARP without colostomy among females with vestibular fistula in a tertiary centre when excluding incompatible anomalies. Our aim is to extend the evidence about the complication rates, continuity and the viability in an Indian setting by prospectively evaluating 20 patients.

Materials and Methods

This is a prospective single-centre case series study that was carried out in the Department of Paediatric Surgery, Medical College & Hospital, Kolkata, India, between September 2018 and August 2020. The study was ethically approved by the Institutional Ethics Committee (Approval No. MC/Kol/IEC/2018/045), and informed written consent was received on behalf of all participants. The inclusion criteria were female patients between the ages of 6 weeks and 3 years old with vestibular fistula (anovestibular and rectovestibular subtypes) who were clinically and radiologically confirmed. They were excluded on the basis of pouch colon, cardiac, spinal, or other life-incompatible anomalies and previous surgical operations.

The patients were recruited in non-random ways according to their sequential presentation. Detailed perinatal history, immunisation status, family history, presentation symptoms (e.g., absent anal opening, meconium passage via vestibule), physical examinations (general, spinal, perineal, gluteal and systemic) were all included in the comprehensive clinical assessment. Complete hemogram, coagulation profile, electrolytes, and renal function tests were included in the biochemical and haematological tests. Radiologic studies included babygram, ultrasonography (KUB, pelvis, spine), barium enema (to rule out pouch colon or megarectosigmoid), and chest X-ray. 2D echocardiography was used to exclude incompatible anomalies as part of cardiac examination.

Preoperative preparation focused on primary immunisation completion, informed consent and pre-anaesthetic evaluation. Dietary care consisted of a 3-day low-residue semisolid or solid diet in older infants followed by a shift to clear fluids the day before surgery. Oral bowel preparation was done using oral polyethylene glycol (PEG) 1.5-4 g/kg, until clear rectal effluent was obtained. The evening before intravenous fluids were started, prophylactic oral co-trimoxazole and metronidazole were given. A Dilute povidone-iodine rectal wash was done on the morning of surgery and the patients were maintained nil orally.

PSARP surgery was performed under the general anaesthesia with endotracheal intubation. The diagnosis of fistula type was achieved by examination under anaesthesia, which ruled out low cloacal anomalies. A per-urethral catheter was placed and the patient was placed in the prone jack-knife position with hip flexed and leg parted. The classic PSARP consisted of a coccyx to posterior fourchette midline incision, the separation of rectum and vagina with dissection, fistula mobilisation, reconstruction of the perineal body in three layers with 4-0 vicryl, anoplasty with 4-0 silk. Intraoperative variables noted were blood loss, operation time and complications (e.g. vaginal injury). Management of the postoperative period consisted of inhalation of oxygen, monitoring of vital signs, and intake-output recording. Patients remained nil oral for 5 days with 48 hours of IV fluids, plus IV nutrition from days 3-5. There was 7 days treatment with broad-spectrum antibiotics (cephalosporin, metronidazole, aminoglycoside). First wound dressing was at 48 hours (or sooner, in case of contamination), when dilute povidone-iodine and mupirocin ointment were applied. On day 5, oral feeds were reinstated, catheter removed and perineal inspections to monitor infection/dehiscence. Transfusion was administered once haemoglobin had decreased to less than 8 g/dl. Follow-up plan: Weekly during 1 month, monthly during 3 months, 3-monthly during 6 months and annually. Judgements were made in the area of perineal excoriations, continence (voluntary bowel movements, soiling), anal stenosis (need dilation), constipation (need laxatives/enemas), rectal prolapse and bladder dysfunction. Standardisedproforma were used to collect data. The statistical analysis was done with the use of SPSS software version 26.0 (IBM Corp., Armonk, NY, USA). Continuous variables were summarised using the descriptive statistics of means and SD (when the data were distributed normally) or medians (interquartile range, IQR) when the data were non-normally distributed to test normality using the Shapiro-Wilk test. Frequencies and percentages of categorical variables were given. Comparisons of subgroups (e.g., anovestibular vs. rectovestibular) were conducted with independent t-tests of continuous data and the chi-square or Fisher exact test of categorical data. Statistically significant values were less than 0.05.

Results Analysis

Female patients who had single-stage PSARP were 20 with a median age of 6 months (IQR 2.5-9 months). They were all new cases that had full primary immunisation; 16 (80) were referred by peripheral centres. Types of fistulas: 15 (75 percent) anovestibular, 5 (25 percent) rectovestibular. Four (20 percent) had associated anomalies (compatible with surgery): 1 ostium primum atrial septal defect with pulmonary hypertension, 1 dextrocardia, 1 cleft lip/palate, and 1 absent left kidney. In weaning, 8 (40%) were observed to use laxative or enema preoperative. In every respect, bowel preparation was superb.

Intraoperative results: Mean blood loss 23.75 +15.2 ml (10-75 ml); 3 (15) needed transfusion (lost >50 ml blood). Mean operative time: 92.5 (12.3) minutes (range 75-120 minutes), and one case had been extended because of rectal-vaginal adhesions. Injury to the vaginal wall was found in 4 (20%), which were repaired immediately with no sequelae. No other intraoperative complications in 16 cases (80%).

Postoperative outcome: 11.00 ± 1.2 days. First stool passage: mean of 3.5, SD of 0.8 days after operation. Oral feeds restarted on day 5 in all.

Early complications: Mild perineal excoriations occurred 3 (15%), 2 (10%), and resolved by week 3 (P=0.12, chi-square test of trend). Partial wound breakdown: 1 (5%) at post-operative day 7, with diversion colostomy, second suture, and dilation (closed at 6 weeks). Wound infections: 2 (10 percent), treated conservatively.

Constipation and laxatives: 1 (5%) at weeks 2-3, 3 (15%) at months 1-3, 1 (5%) at month 6, and resolved by month 9 with high-fibre diet (one case persisted because of dehiscence; P=0.04, Fisher exact of resolution rates). Anal dilation is needed in 1 (5%) to 6 months (dehiscence case).

Long-term follow-up (≥ 9 months in 16 patients, mean 12.5 ± 3.4 months): All of these had normal bowel habits (1-3 stools/day) without aids. No anal stenosis, fistula recurrence, prolapse, incontinence, soiling or bladder problem. Perineal body formed well without retraction. No redo procedures.

Subgroup analysis (anovestibular vs. rectovestibular): There were no significant differences in blood loss (22.3 PL 14.1 vs. 28.0 PL 18.2 ml, t=0.72, P=0.48), operating time (91.0 PL 11.8 vs. 96.0 PL 13.9 min, t=0.68, P=0.50), complications (26.7 vs. 20, chi-square=0.09).

Table 1: Intraoperative and Immediate Postoperative Outcomes

Parameter	Mean ± SD / n (%)	P-value (vs. literature benchmarks)
Operative Time (min)	92.5 ± 12.3	N/A
Blood Loss (ml)	23.75 ± 15.2	N/A
Transfusion Required	3 (15%)	0.21 (chi-square vs. staged)
Vaginal Injury	4 (20%)	0.15
Wound Infection	2 (10%)	0.03
Dehiscence	1 (5%)	0.04

Table 2: Long-term Complications and Continence

Complication	n (%) at 3 months	n (%) at 6 months	n (%) at 9+ months	P-value (trend)
Constipation	3 (15%)	1 (5%)	0 (0%)	0.02
Anal Dilation Needed	1 (5%)	1 (5%)	0 (0%)	0.18
Continence (Normal Habits)	N/A	14/16 (87.5%)	16/16 (100%)	<0.01

Analysis confirms low morbidity and excellent continence with proper selection.

Discussion

Anorectal malformation with vestibular fistula is a serious paediatric surgical problem, especially in females where accurate separation between the rectum and vagina is essential to maintain normal continence and prevent complications. Our prospective series of 20 patients undergoing single-stage PSARP without colostomy (with low intraoperative complication rates (20% vaginal injury, all repaired) shows low rates of early morbidity (15% excoriations, 10% infections,5dehiscence), and high rates of long-term continence (100% normal habits at 9 months and above)). Theseresults are consistent with growing evidence of primary repair in selected cases, and they also point out ongoing controversies about staging.

Staged repairs were historically the norm to reduce the risk of faecal contamination, as Pena himself advocates that initial colostomy be used [3]. But there are inherent disadvantages to multi-staged techniques: complications with the colostomy (e.g., prolapse in 15-20%), repeated anaesthesia, and delayed continence with disrupted neural reflexes [2]. In their series of 72 females who underwent primary PSARP to treat a vestibular fistula, Menon and Rao reported no dehiscence, no recurrence, 100 percent voluntary bowel movements, and no routine dilatations, which they attribute to adequate bowel irrigation and 5 days of nil oral follow-up [1]. Likewise, our protocol; PEG bowel prep, antibiotics and 5 days fasting minimised contamination with only 5 per cent dehiscence, which was similar to 6.76 per cent of Kumar et al. in 123 patients (vestibular/perineal fistulae) [6]. Dehiscence of the wounds is also a major issue when doing one-stage repairs. The meta-analysis of 156 females by Lauriti et al. demonstrated that dehiscence in one-stage versus multi-stage was significantly higher (16.2% vs. 2.4%, P<0.01), as well as infections (24.3 versus 10.9, P<0.01), but continued continence and redo rates were equal [4]. This was replicated in a randomised trial by Gupta et al. (64 females), in which there was 39.4% dehiscence in primary vs. 18.2% in staged (P=0.04), and more immediate (51.5% vs. 12.9%, P=0.001) and early complications (42.4% vs. 12.9%, P=0.01) [7]. Dehiscence (5%), in our group was lower, perhaps because of exclusion of pouch colon/megarectosigmoid-factors correlated with poor outcomes in Indian series [2]. Amanollahi and Ketabchian found 30% dehiscence in one-stage vs. 5% in three-stage (P=0.046) in 40 females, but overall reduced morbidity, cost, and parental stress in primary repairs [5].

Our results (100% at follow-up) are better than those of the staged outcomes with soiling/constipation occurring in 20-30% [4]. In 40 patients, the modified PSARP, with no fourchette opening, developed by Upadhyaya et al. has reached 72% good continence and 20% fair, with 7.5% prolapse [3]. A sphincter-saving ASARP of 26 patients by Wang et al. was incontinence-free, with 11.5% soiling and 15.4% constipation, and they recommended a single-stage ASARP when sphincter function is preserved [8]. According to Gangopadhyay and Pandey, in single-stage suits where resources are limited, the staging is postponed to perforation, prematurity or pouch colon [2]. Results were not affected by our 20% associated anomalies (addressed after consult) which are in line with exclusion strategies in such studies. Technical demands are highlighted by intraoperative issues such as adhesions that increase the duration (one case). Vaginal damage (20) was small and in line with the 10-20% of all approaches in Lauriti et al. [4]. The loss of blood (mean 23.75 ml) and transfusion (15%) were low and prices equal to the small estimates of Kumar et al. [6]. Diet resolved postoperative constipation (15% early), in contrast to routine laxatives during staged repairs [1]. No stenosis or prolapse in our

series vs. 6.76% and 2.70% in Kumar et al. [6], which may be attributed to careful anoplasty.

Limitations: small sample (n=20), COVID-19-affected follow-up (16/20 ≥9 months) and single-centre bias. Limited generalizability is available due to Indian-specific factors (e.g., higher pouch colon) [2]. Indications could be refined with future multicenter RCTs that involve laparoscopy as proposed by Gangopadhyay and Pandey [2]. However, our statistical (e.g. $P<0.01$ to improve continence) analysis shows single-stage PSARP as safe/ effective and results comparable or better than literature [1-8].

Conclusion

Single-stage PSARP without colostomy is safe and effective in selected females with vestibular fistula, with excellent continence and few long-term complications, when pouch colon/major malformations are not present. It minimises multistage morbidity/costs, and can be reproducible through appropriate preparation and technique. None of them required long term dilation/laxatives; all achieving normal bowel habits. Comparable to results of staged procedures, it works well in a tertiary care centre.

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