


ORIGINAL RESEARCH

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Comparative study between one-stage versus three-stage repair of anorectal malformation with recto-vestibular fistula

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Abstract

Background: This is a retrospective comparative study that aimed to compare the short-term and intermediate-term outcomes of the one-stage and three-stage repair in the treatment of female neonates with a recto-vestibular fistula. Female patients who were presented with recto-vestibular fistula between 2017 and 2020 have been included in the study, and they were divided into two groups. *Group A* is the group of patients that underwent one-stage repair, and *Group B* is the group of patients that underwent three-stage repair. Short- and intermediate-term outcomes were recorded in both groups and the results were compared.

Results: Thirty-six female infants with recto-vestibular fistula were included in our study divided into two groups. *Group A* included 20 cases for whom we performed one-stage posterior sagittal anorectoplasty, and *Group B* also included 16 cases for whom the three-stage repair was done. The results showed that the three-stage repair had lower hospital stay (8 days) than the one-stage repair (15 days), and the three-stage repair has lower rates of wound infection, wound dehiscence, and recurrence than the one-stage repair.

Conclusion: The three-stage repair of anorectal malformations in females with recto-vestibular fistula is a safer technique, has lower rates of complications, and achieves comparable functional results to the one-stage repair.

Keywords: Vestibular fistula, Anorectal malformation, Repair

Background

Anorectal malformations (ARMs) comprise a wide spectrum of diseases, affecting both boys and girls, involving the distal anus and rectum as well as the urinary and genital tracts [1]. In females, the vestibular fistula represents the most common type of anorectal defect with associated malformations [2, 3].

The vestibular fistula was described as a fistulous tract between the rectum and the low female genital tract by Bryndorf and Madsen in 1960 [4]. In these cases, perineal

inspection shows a normal urethra, normal vagina, and another orifice, which is the rectal fistula in the vestibule.

The traditional surgical correction of high and intermediate ARMs in newborns is staged over several weeks or months. This usually entails a divided colostomy, posterior sagittal anorectoplasty (PSARP), and then colostomy closure as the third stage [5].

One-stage repair has many advantages; first, the potential colostomy complications are avoided. Especially in developing countries, the social and parental inconvenience is also eliminated, and the morbidity and mortality of 2 more general anesthetics and 2 more operations are also averted. Saving the cost related to these extra operations in addition to the cost of colostomy appliances,

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stoma care medications, and outpatient clinic follow-up visits is another advantage [6].

Taking this into consideration, we decided to compare the repair of ARMs in females with recto-vestibular fistula in one stage without colostomy using the posterior sagittal approach, with the traditional three-stage repair.

Methods

A comparative retrospective study was done to compare the results of repairing ARMs in females with recto-vestibular fistula in one stage without colostomy using the posterior sagittal approach (PSARP), with the results of the three-stage repair.

All female patients who presented with ARMs with a recto-vestibular fistula between January 2017 and January 2021 were included in the study.

The study was conducted in 2 groups:

Group (A): Female infants who had ARMs with recto-vestibular fistula and underwent one-stage repair (PSARP). They were kept postoperative *nil-per-os*, for a period of 7 days, and a wide pore peripheral line or central venous line was inserted for total parenteral nutrition and antibiotic infusion.

Group (B): Female infants who had ARMs with recto-vestibular fistula and underwent three-stage repair (colostomy, PSARP, and closure of colostomy).

Exclusion criteria included other female ARMs such as Cloaca, ARMs without fistula and recto-vaginal fistula, association with major anomalies such as major cardiac anomalies, caudal regression anomalies, and renal anomalies.

Patients of both groups were assessed after 2 weeks, 1 month, 3 months, and 6 months postoperatively in the outpatient clinic for follow-up of anal size and complications such as anal stenosis, soiling, constipation, or recurrence.

Results

The statistical analysis was done by Statistical Package for Social Sciences (SPSS) version 16. Correlations between different parameters had been evaluated using Spearman rank correlation: P values < 0.05 are considered significant. Descriptive analysis tools will be used for single variables such as range, mean, and standard deviation.

In *Group A*, the median age was a 12-month range (5–30), while in *Group B*, the median age was a 12-month range (6–32).

Regarding hospital stay: *Group B* had a lower hospital stay with an average of 8 days (range 4–30) than *Group A* recording 15 days (range 7–35) that were statistically significant with a P value of 0.03. *Group B* had lower rates of

recurrence (0%), while in *Group A*, 4 (20%) patients had a recurrence, with a significant difference P value = 0.03.

The results of our study were summarized in Table 1.

Discussion

We conducted this study in order to compare the repair of ARMs in females with recto-vestibular fistula in one stage without colostomy using the posterior sagittal approach, with the three-stage repair.

Thus, to achieve this goal, thirty-six female infants with recto-vestibular fistula were included in our study and divided into two groups, *Group A* included 20 female infants with recto-vestibular fistula that underwent one-stage repair, and *Group B* included 16 cases that underwent three-stage repair (colostomy, PSARP, closure of the colostomy).

In our study, all infants included in the study had the repairing approach at the age of 3–18 months without any significant differences.

While in the study by J.O. Adeniran, there were 4 female infants ranging from 3 days to 1.5 years old [7]; however, in the study by A. Mirshemirani et al., all cases were operated on at age of 1 to 20 days of life [8].

The presenting symptoms of our cases were constipation and abdominal distension.

The complications of the surgery were compared between 2 groups of the presenting study; the most common complication was wound infection as in the one-stage operation, and there were 10 patients (50%) versus 6 patients (37.5%) in the 3-stage operation with no significant difference ($p=0.1$). In J.O. Adeniran's study, only one patient who had PSARP had a superficial wound infection. While in A. Mirshemirani et al. study, 3 cases had postoperative wound infections [7, 8].

Wound dehiscence occurred in 6 cases (30%) versus 4 cases (25%) in the one-stage and 3-stage operations, respectively. This was in contrast with what was found in Mirshemirani et al study, as no cases developed wound dehiscence or stricture formation [8].

Table 1 Results of our study

Complication	Group A (n= 20)	Group B (n=16)	P values
Average hospital stay	15 (range 7–35)	8 (range 4–30)	0.03
Wound infection	10 (50%)	6 (37.5%)	0.1
Wound dehiscence	6 (30%)	4 (25%)	0.08
Recurrence	4 (20%)	0 (0%)	0.03
Stricture	0 (0%)	0 (0%)	-
Constipation	3 (15%)	0 (0%)	0.1
Soiling	3 (15%)	3 (18.8%)	0.2
Vaginal wall injury	0	0	-

Soiling occurred in 3 cases (15%) in one-stage repair versus 3 cases (18.8%) in the three-stage operation, while only one case had soiling in the study by Mirshemirani et al. [8]

Recurrence occurred in 4 cases (20%) in one-stage repair versus no cases (0%) in three-stage repair, and this was in contrast to what was revealed by Mirshemirani et al. who found that recurrent fistula did not develop in any patients [8].

In our study, we did not encounter vaginal wall injuries in both groups, yet, it is a serious complication that may render recurrence, meticulous dissection, and use of magnification during the operation are among the strategies to avoid such a complication.

All complications that had happened to all cases in our study showed no significant differences between both surgical techniques except for the hospital stay and the recurrence rate.

Colostomy complications are very common, even fatal for many, especially in developing countries. Sowande et al. [9] reviewed 67 patients who had colostomies at Obafemi Awolowo University, Nigeria, and reported a total of 50 complications in 32 patients (47.8%). Only 30 patients (44.8%) had definitive surgery and eventual closure of colostomy. The 37 patients lost to follow-up could not have been carrying stomas around for so long and probably died. In their 10-year review of anorectal anomalies at the University of Ilorin, Nigeria, only 10 boys out of 26 (38.5%) and 9 girls out of 19 (47.4%) lived to have definitive operations after a preliminary colostomy [7–9].

Thus, less than 50% of patients who had colostomies eventually had definitive surgery and colostomy closure. This indicates an intolerably high waste rate.

Although Pena in 1997 recognized that colostomy represents a significant source of morbidity, he still proposed the formation of a colostomy for recto-vestibular anomalies. Fernando Heinen in 1997 added some weight to this argument. In his review of a total of 227 patients, 97 girls had recto-vestibular fistulas. All had preliminary colostomy before PSARP [10, 11].

However, other surgeons have argued for a definitive repair without colostomy, even for boys, and very recently, as early as the neonatal period. The advantages of bypassing the colostomy stage are many. First, colostomy complications are eliminated completely. This is even more important in developing countries where:

- Colostomy is socially unacceptable
- Colostomy bags are difficult to be afforded
- Most of the parents are illiterate and cannot manage colostomies (which these unfortunate patients have for 6 to 8 months)
- There are no stoma-care nurses

The traditional operations are of immense disadvantage to the baby, the parents, and the entire household.

In our study (group B of patients), two patients had prolapsed stoma (12.5%) without the functional obstruction of the stoma and 10 patients had peristomal skin maceration (62.5%).

Three operations mean three admissions, which means the mother is separated from the rest of the family, and nobody may be able to take proper care of the rest of the other children. The cost also is significant. It is quite difficult for many of these parents to pay for one operation, and it is more difficult when there are 3 major operations within 6 to 8 months. It would therefore be an advantage if the patient can have only one admission, no colostomy is done, and the parents pay only once for an operation.

Conclusion

The three-stage repair of anorectal malformations in females with recto-vestibular fistula is a safer technique, has lower rates of complications, and achieves comparable functional results to the one-stage repair, but more costly in time and money than the one-stage repair.

Abbreviations

ARMs: Anorectal malformations; PSARP: Posterior sagittal ano-recto plasty.

Acknowledgements

Not applicable.

Authors' contributions

Concepts: MG, KA, SK; Design: MG; Definition of intellectual content: MF, ME, AA, MR; Literature search: MG, OA, MR; Clinical studies: MG, KA; Experimental studies: KA, AA; Data acquisition: MF, OA, AA, MR; Data analysis: MF, KA, MR; Statistical analysis: MG, ME; Manuscript preparation: MG, OA; Manuscript editing: MG, OA, AA, MR; Manuscript review: ME, SK, MR; Guarantor: OA, SK. The author(s) read and approved the final manuscript.

Funding

The authors declare there are no funding resources for this research.

Availability of data and materials

Yes, available. Please contact the author for data requests.

Declarations

Ethics approval and consent to participate

The ethics approval was obtained from the institution's research ethics and scientific committee, and consent to participate is obtained from all guardians prior to enrollment in the study.

Consent for publication

Obtained and included in the informed consent.

Competing interests

The authors declare that they have no competing interests.

Received: 12 July 2022 Accepted: 4 November 2022

Published online: 24 November 2022

References

1. Pena A, Levitt M. Anorectal malformations. In: Stringer M, Oldham K, Mouriquand PDE, editors. *Pediatric Surgery and Urology: Long term outcomes*. Cambridge: Cambridge University Press; 2007.
2. Forrester MB, Merz RD. Risk of selected birth defects with prenatal illicit drug use, Hawaii, 1986-2002. *J Toxicol Environ Health A*. 2007;70:7–18.
3. Zwink N, EkkehartJenetzky and Hermann Brenner B. Parental risk factors and anorectal malformations: systematic review and meta-analysis. *Orphanet J Rare Dis*. 2011;6:25.
4. Bokhari I, Ali SU, Farooq AR, Khan A. Late presentation of a patient with an anorectal malformation (ARM). *J College Phys Surg Pak*. 2010;20(12):825–7.
5. Pena A, Hong AR, Midulla P, Levitt M. Reoperative surgery for anorectal malformations. *Semin Pediatr Surg*. 2003;12:118–23.
6. Elbatarny A, Elafifi M, Ismail K, Metwally M, Hashish A, Elmahalway M, et al. Evaluation of one-stage posterior sagittal anorectoplasty for the repair of high and intermediate anorectal malformations. *Ann Pediatr Surg*. 2009;5(1):36–45.
7. Adeniran J. Anorectal malformations in Ilorin, a 10-yr review. Presented at the Nigerian Surgical Research Society Conference in Ilorin, Nigeria; 2000.
8. Mirshemirani A, Kouranloo J, Rouzrokh M, Sadeghiyan MN, Khaleghnejad A. Primary posterior sagittal anorectoplasty without colostomy in neonate with high imperforate anus. *Acta Med Iran*. 2007;45(2):121–5.
9. Sowande O, Adejuyigbe O, Ogundoyin O. Colostomy complications in infants and children. *Nig J Surg*. 1999;6:19–22.
10. Pena A. Advances in anorectal malformations. *Semin Pediatr Surg*. 1997;6:165–9.
11. Heinen F. The surgical treatment of low anal defects and vestibular fistulas. *Semin Pediatr Surg*. 1997;6:204–16.

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