MAR Pediatrics (2026) 7:2

Research Article

Management and Outcome of Vestibular Anus in Sudan

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Received: 04 February 2026

Published: 01 March 2026

DOI: https://doi.org/10.5281/zenodo.18616359

Background

In female children, the most prevalent types of anomalies are vestibular fistula and ectopic anus. Despite advancements in anatomy, physiology, and embryology (3), surgical challenges remain regarding post-operative cosmetic results and continence outcomes.

Anorectal malformations, which are birth defects characterized by the absence or deformity of the anus, occur equally in both males and females, with an incidence of 1 in 5000 births. ARMs encompass a spectrum of congenital anomalies ranging from minor lesions to complex defects. Different surgeons may use varying terminologies to describe the types of ARMs. The reality is that there exists a broad spectrum of defects, making any classification attempt somewhat arbitrary and imprecise. The traditional classification into 'high', 'intermediate', and 'low' defects often leads to ambiguous or uncertain results. However, studies indicate that a low version is present 90% of the time in females and 50% of the time in males. Typically, ARM necessitates immediate surgical intervention to create a passage for feces, unless a fistula is available or until corrective surgery can be performed. Treatment varies based on the severity of the anomaly, with options including perineal anoplasty alone or a two-stage approach involving colostomy followed by definitive repair (1).

The most frequently observed anomaly in newborn girls is rectovestibular fistula. A perineal examination typically reveals a normal urethra, a normal vagina, and an additional opening behind the vaginal orifice, which is the rectal fistula located in the vestibule. For newborns exhibiting clinical signs of a rectovestibular fistula, a diverting colostomy is considered the safest approach for surgeons lacking extensive experience with anorectal anomalies. While performing a colostomy prior to the main repair helps prevent infection complications, the creation of a colostomy in neonates must be approached with caution.

Materials and Methods

A descriptive prospective, multicenter hospital study design was employed.

Study area and settings:

The research was carried out at Elribat University Hospital, Khartoum Teaching Hospital, Khartoum North Teaching Hospital, Soba University Hospital, and Madani Teaching Hospital in Sudan.

Study population:

The study included all patients diagnosed with vestibular fistula in the Department of Pediatric Surgery at Elribat University Hospital, Khartoum Teaching Hospital, Khartoum North Teaching Hospital, Soba University Hospital, and Madani Teaching Hospital in Sudan during the period from March 2018 to March 2019.

Sampling:

Sample frame:

All patients diagnosed with imperforate anus with recto-vestibular fistula.

Inclusion criteria

All patients with RVF who underwent surgical repair.

Exclusion Criteria

Patients with imperforate anus without fistula, urogenital sinus, cloaca, or absent vaginal opening were excluded from the study.

Sample size:

All patients diagnosed with vestibular fistula at the time of the study.

Sample type:

Quota methodology (i.e., all patients with RVF who underwent surgical repair).

Data collection methods:

Research data was gathered through a questionnaire and individual examinations of each patient. Patients were selected to participate in this study and were interviewed face-to-face on various aspects using a structured questionnaire.

Data management and analysis:

Data was managed and analyzed using a computer with the Statistical Package for Social Sciences (SPSS) program, version 25. Tables, pie charts, and bar charts were utilized for data presentation. Chi-squared analysis, correlation, and regression tests were employed to compare the groups, with the significance level established at $P<0.05$.

Quality control

All research assistants were trained on research procedures, including the consent process and data collection, one week prior to the study. Additionally, monthly study meetings were held to review challenges encountered and to address them. The questionnaires were also piloted. The Principal Investigator checked the data daily.

Results

Patients were gathered from various pediatric surgery centers: Algazira Pediatric National Centre 17 (25.8%), Soba University Hospital 16 (24.2%), Khartoum Teaching Hospital 15 (22.7%), Ribat University Hospital 10 (15.2%), and Khartoum North Teaching Hospital 8 (12.1%).

The majority of patients were aged between 1-3 years 30 (45.5%), while those older than 3 years numbered 27 (40.9%), and the age of patients ranged from the first day to 1 year 9 (13.6%) (Figure 1).

The predominant ages at which vestibular anus was observed included: since birth 48 (72.7%), 2-4 months 9 (13.6%), first month 8 (12.1%), and 5-12 months 1 (1.5%) (Table 1).

Regarding the age of presentation, the distribution was as follows: 2-4 months 20 (30.3%), first month 17 (25.8%), since birth 17 (25.8%), and 5-12 months 12 (18.2%) (Figure 2).

Patients born at home accounted for 42 (63.6%), while those born in a hospital were 24 (36.4%) (Figure 3).

The majority of patients did not have associated abnormalities 60 (90.9%), with cardiac abnormalities present in 5 (7.6%) and cardiac issues combined with cleft lip in 1 (1.5%) (Table 2).

A total of 62 (93.9%) experienced abdominal distension, while 4 (6.1%) did not (Figure 4).

52 (78.8%) had constipation, whereas 14 (21.2%) did not (Table 3).

Among those who underwent preoperative dilatation, 40 (60.6%) were not dilated preoperatively, and 26 (39.4%) had preoperative dilatation (Figure 5).

59 (89.4%) had colostomy, while 7 (10.6%) did not (Table 4).

27 (46%) had colostomy at the age of 2-6 months, 20 (34%) at the first month, 9 (15%) at 7-12 months, and 3 (5%) at over one year (n=59) (Figure 6).

59 (89.4%) underwent repair with colostomy, while 7 (10.6%) had repair without colostomy (Table 5).

The most common type of repair was limited PSARP 32 (48.5%), ASARP 25 (37.9%), and anal transfer 9 (13.6%) (Figure 7).

43 (65.2%) experienced complications, while 23 (34.8%) did not (Figure 8).

The most frequent complications included anal stenosis 25 (58.1%), minimal wound infection 8 (18.6%), perineal body disruption 5 (11.6%), rectovaginal fistula 3 (6.9%), and combined perineal body disruption with anal stenosis 1 (2.3%) (n=43) (Table 6).

46 (69.7%) of patients did not have perineal body contraction and 20.

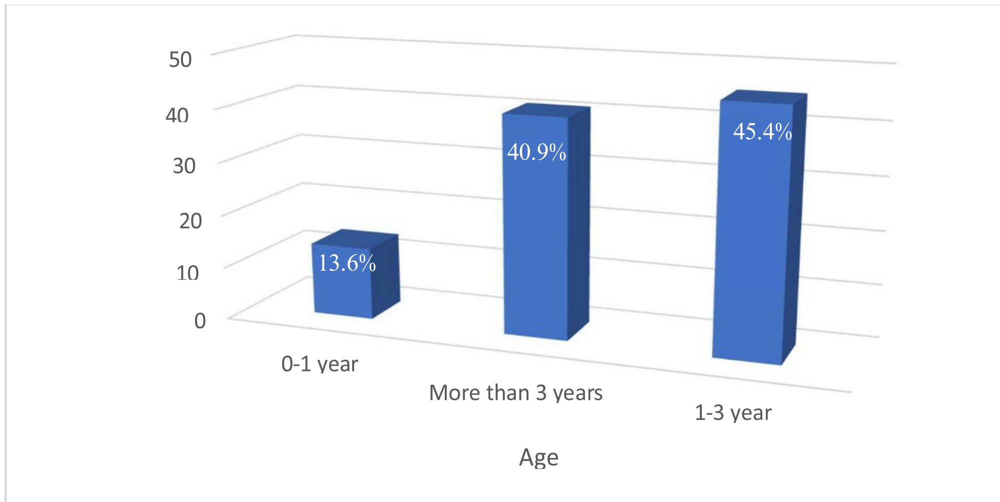

Figure 1 Shows Distribution of Cases According to Age Group

Noticed age	Frequency	Percent
5-12 months	1	1.5
First month	8	12.1
2-4 months	9	13.6
since birth	48	72.7
Total	66	100.0

Table 1 Shows distribution of cases according to age

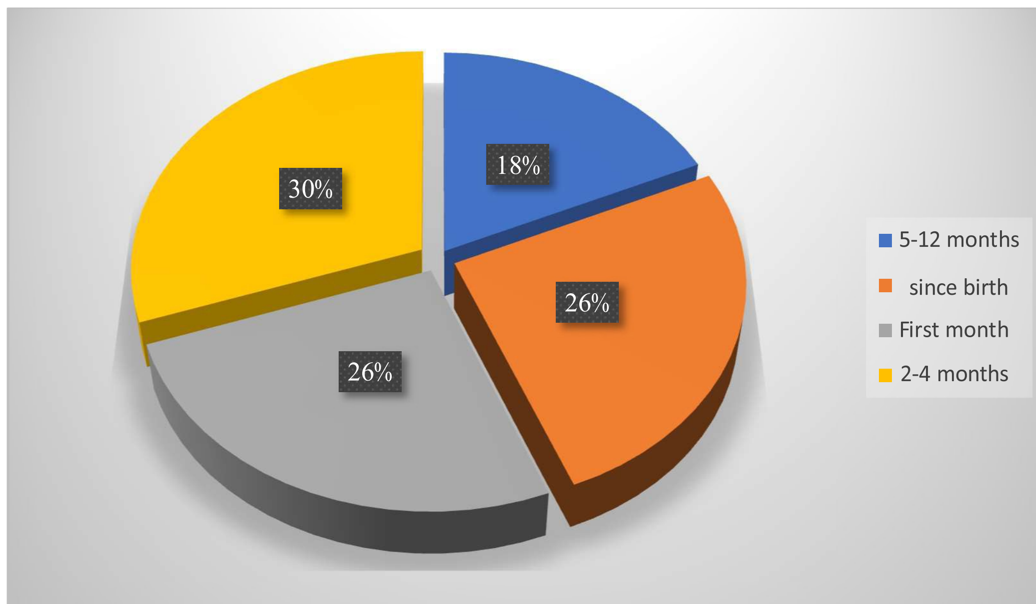

Figure 2 Shows distribution of cases according to presentation of age

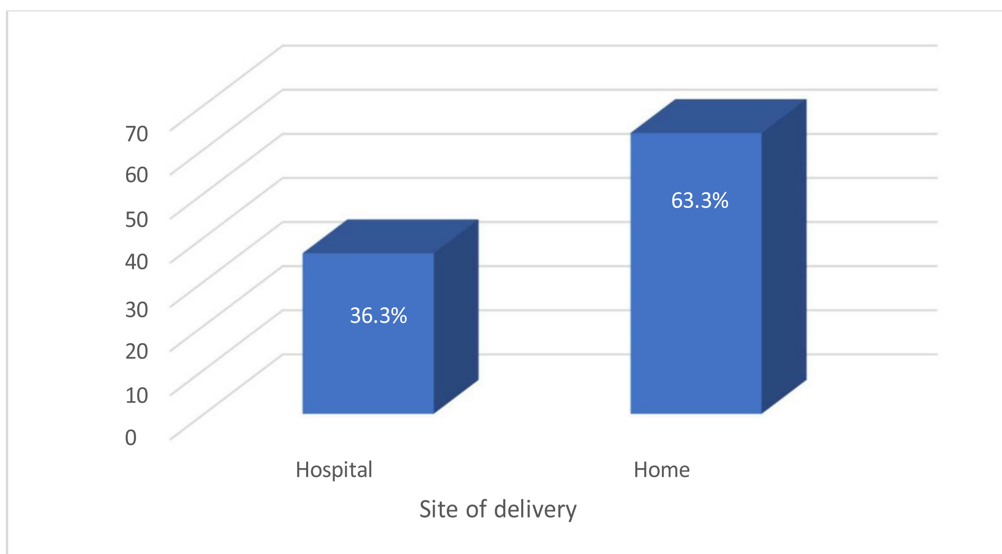

Figure 3 Shows distribution of cases according to site of delivery

Associated abnormalities	Frequency	Percent
Cardiac + cleft lip	1	1.5
Cardiac	5	7.6
No associated abnormalities	60	90.9
Total	66	100.0

Table 2 shows distribution of cases according to associated abnormalities

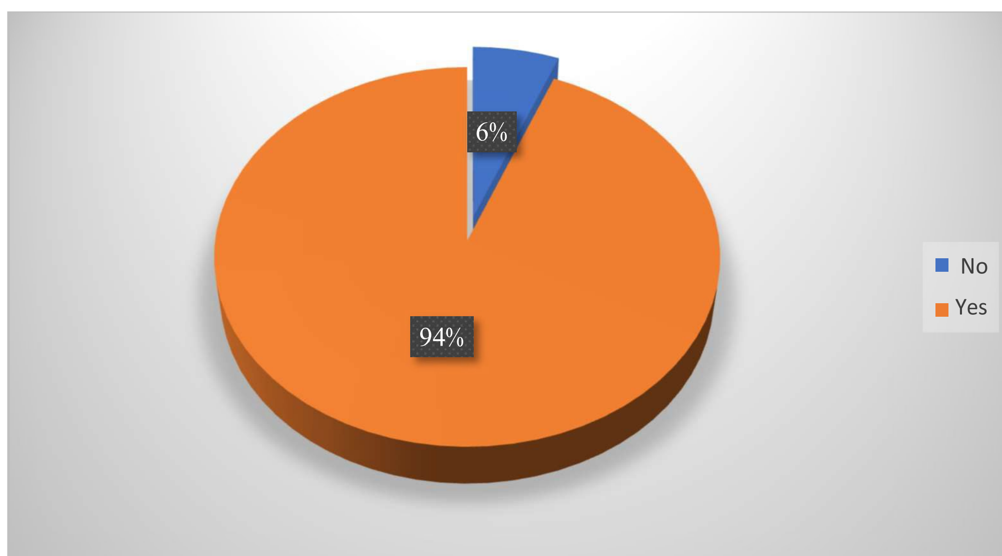

Figure 4 Shows distribution of cases according to abdominal distension

Constipation	Frequency	Percent
No	14	21.2
Yes	52	78.8
Total	66	100.0

Table 3 Shows distribution of cases according to constipation

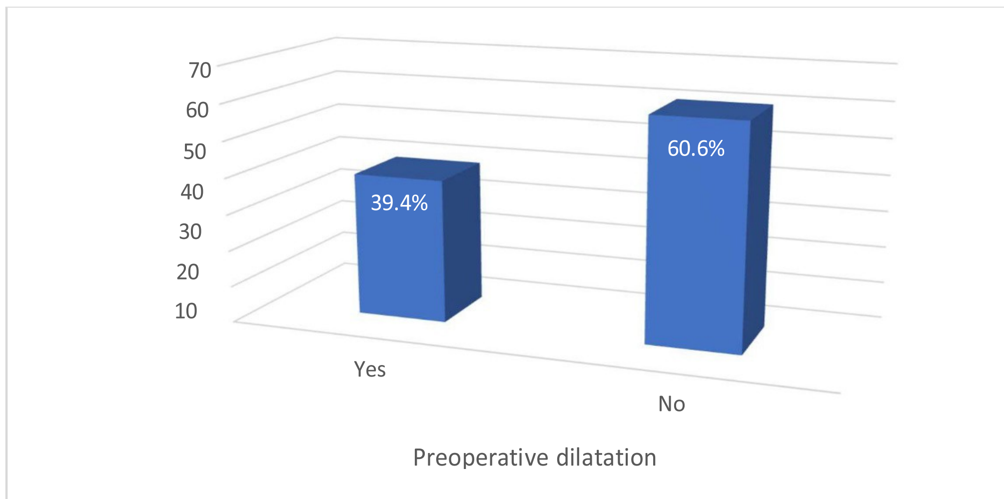

Figure 5 Shows distribution of cases according to preoperative dilatation

colostomy	Frequency	Percent
No	7	10.6
Yes	59	89.4
Total	66	100.0

Table 4 shows distribution of cases according to colostomy

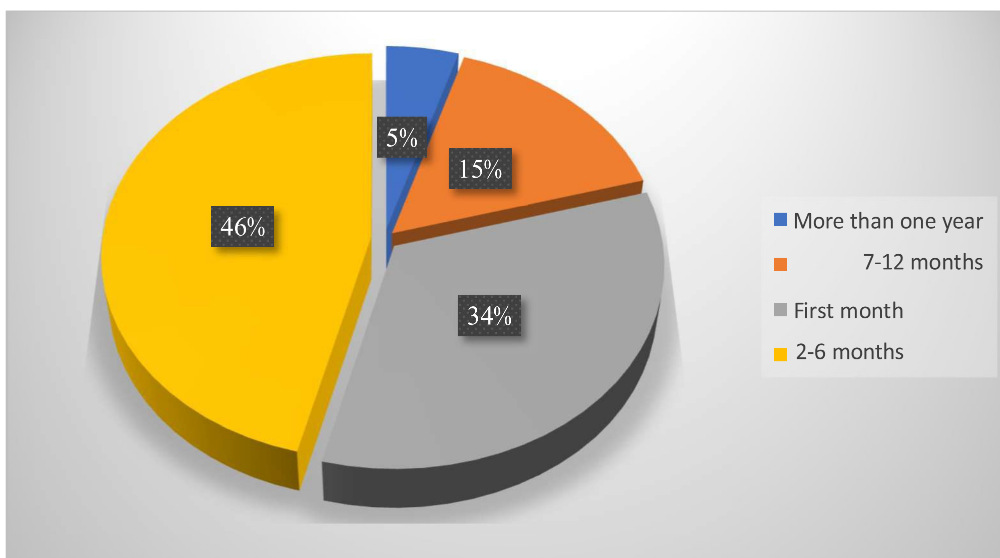

Figure 6 shows distribution of cases according to age of colostomy

Repair	Frequency	Percent
Without colostomy	7	10.6
With colostomy	59	89.4
Total	66	100.0

Table 5 Shows distribution of cases according to repair

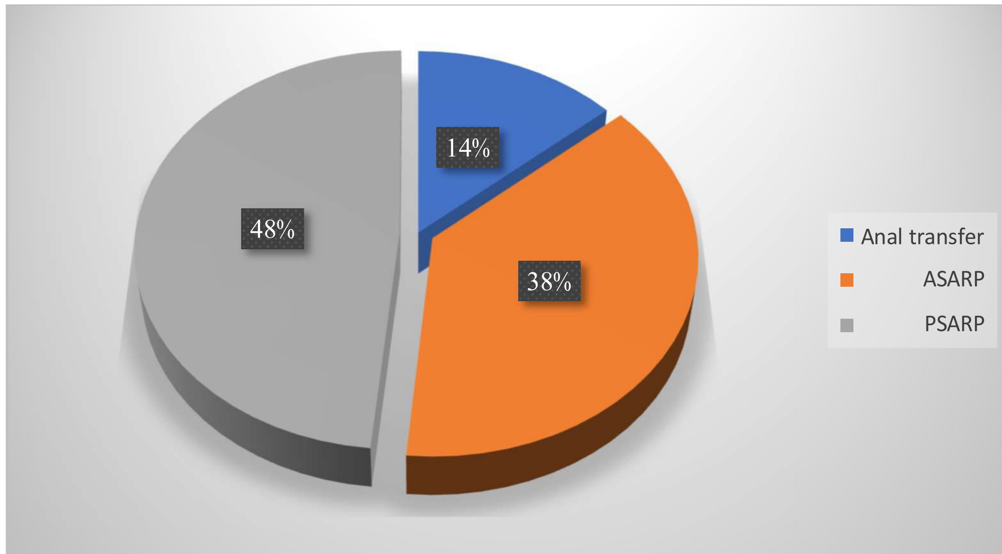

Figure 7 Shows distribution of cases according to type of repair

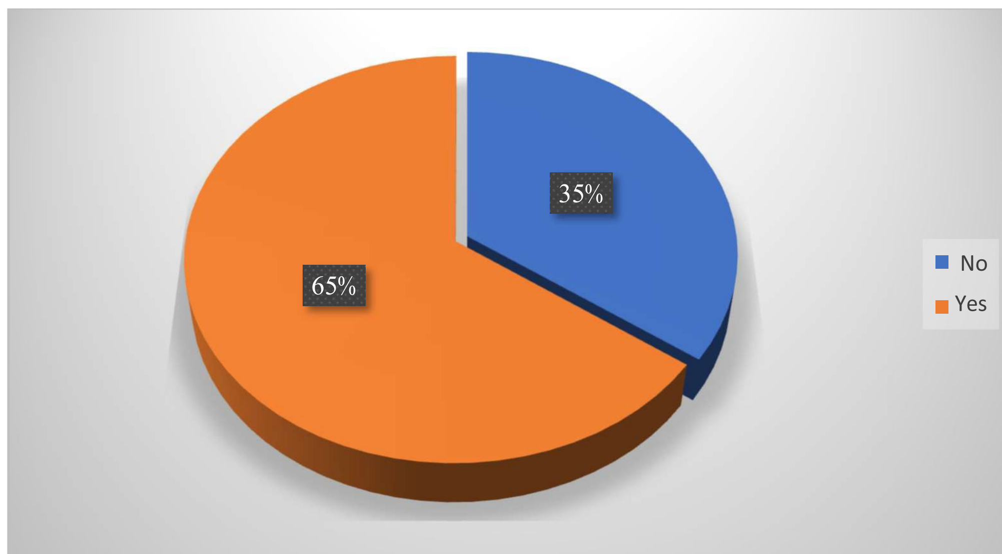

Figure 8 shows distribution of cases according to complications

Complications	Frequency	Percent
Perineal body disruption + Anal stenosis	1	2.4
Rectovaginal fistula	3	7.1
Perineal body disruption	5	11.9
Minimal wound infection	8	19.1
Anal stenosis	25	59.5
Total	42	100.0

Table 6 Shows distribution of cases according to type of complications

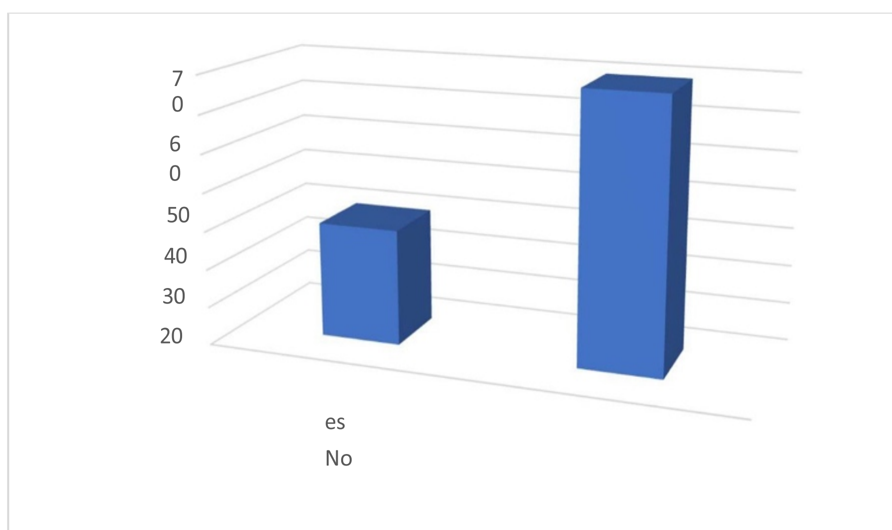

Figure 9 shows distribution of cases according to contraction of perineal body

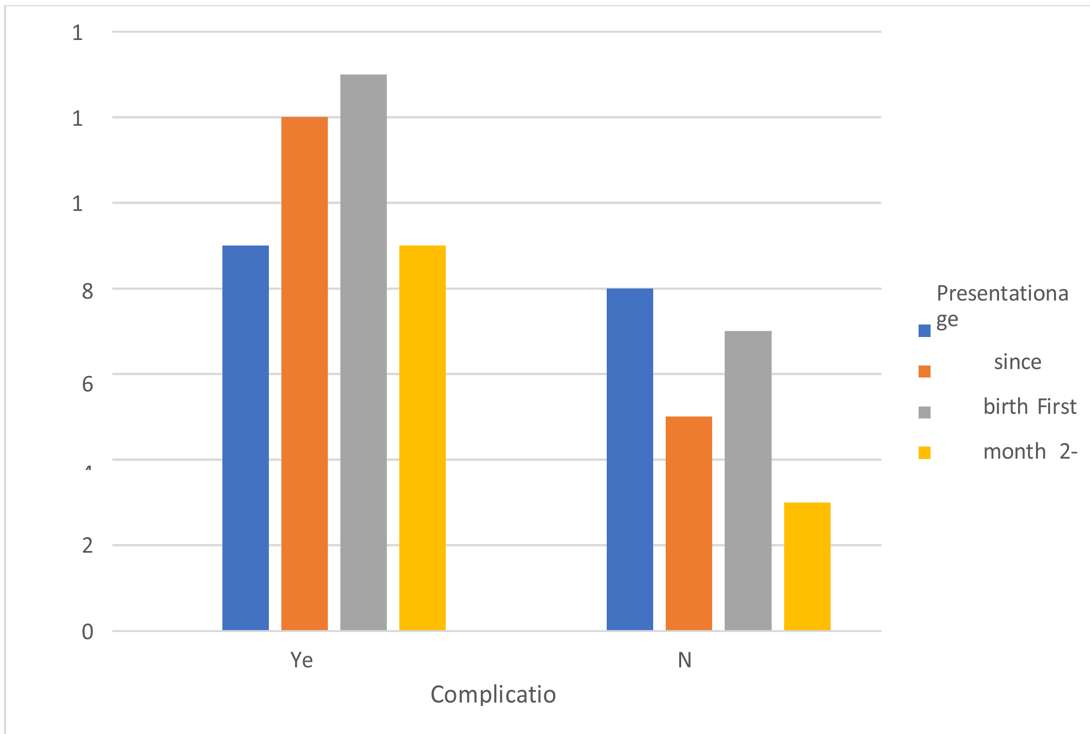

Figure 10 shows distribution of cases according to relation between age at presentation and complication

		Presentation age				Total
		since birth	First month	2-4 months	5-12 months	
Associated abnormalities	Cardiac	2	1	0	2	5
	No associated abnormalities	14	16	20	10	60
	cardiac + cleft lip	1	0	0	0	1
Total		17	17	20	12	66

Table 7 Shows distribution of cases according to relation between age at presentation and associated abnormalities

		Complications					Total
		Minimal wound infection	Perineal body disruption	Anal stenosis	Rectovaginal fistula	Perineal body disruption + Anal stenosis	
Type of repair	Anal transfer	1	1	6	0	0	8
	ASARP	3	3	7	1	1	15
	PSARP	4	1	12	2	0	19
Total		8	5	25	3	1	42

Table 8 shows distribution of cases according to relation between type of repair and complication

		Repair		Total
		With colostomy	Without colostomy	
Complications	Yes	37	6	43
	No	22	1	23
Total		59	7	66

Table 9 shows distribution of cases according to relation between with and without colostomy andcomplication

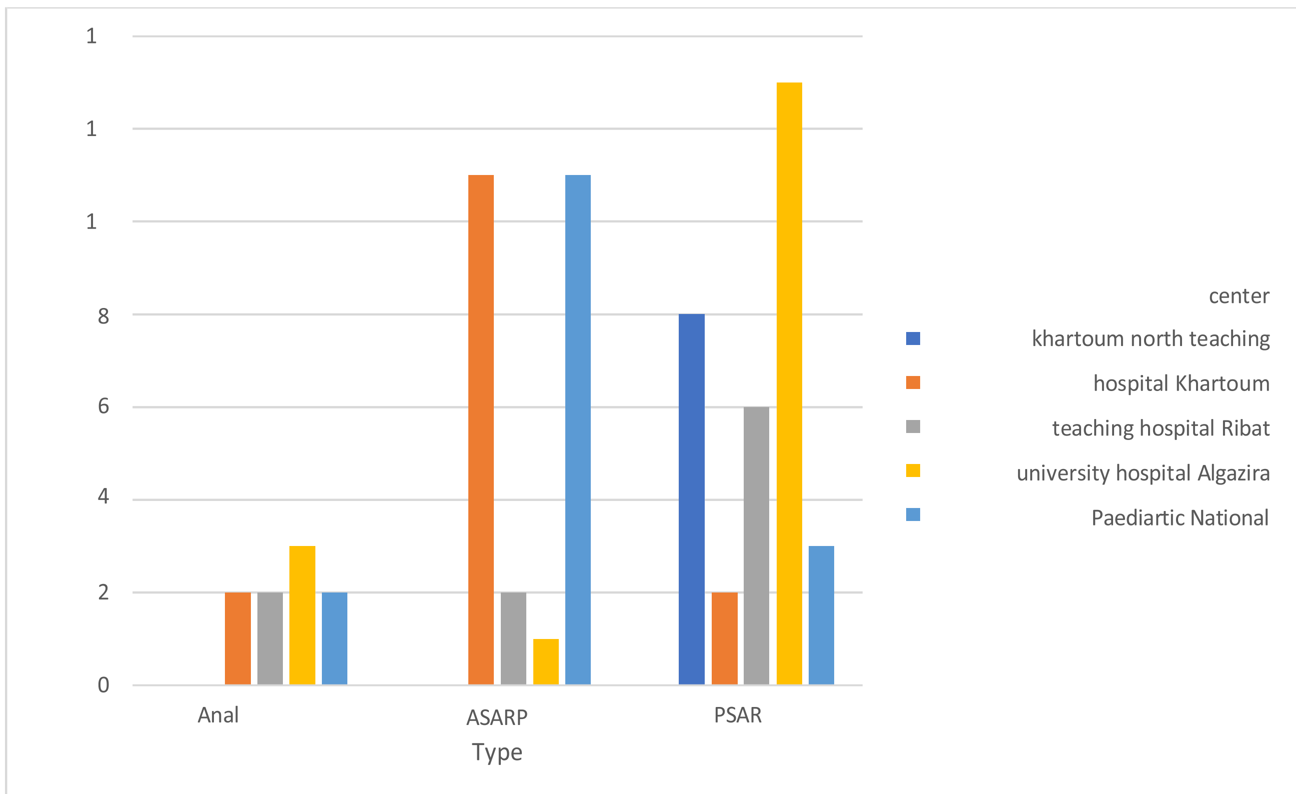

Figure 11 shows distribution of cases according to relation between center of pediatrics surgery and type of repair

		center					
		Khartoum north teaching	Khartoum teaching hospital	Ribat university hospital	Algazira Paediartic National	Soba university hospital	
Repair	With colostomy	8	12	10	15	14	59
	Without colostomy	0	3	0	2	2	7
Total		8	15	10	17	16	66

Table 10 shows distribution of cases according to relation between center of pediatrics surgery and repair

Discussion

In this descriptive prospective, multicenter, hospital-based study, we examined all patients diagnosed with vestibular fistula within the pediatric surgery department. The aim was to evaluate the feasibility, safety, benefits, and follow-up of anterior or posterior sagittal anorectoplasty in low-type ARMF (rectovestibular) cases, as well as to compare the outcomes and complications associated with one-stage and three-stage repairs in females with vestibular fistula. A total of 66 cases were recorded from March 2018 to March 2019.

The ages of the patients ranged from the first day of life to 1 year and 9 months (13.6%). The majority of patients were aged between 1 and 3 years (30 cases, 45.5%), while 27 patients (40.9%) were older than 3 years. This finding aligns with a similar study conducted in Egypt (9), where ninety-two percent of patients were aged ≤1 year, with a mean age of 7 months and a median age of 6 months. Most surgeries were performed on patients aged between 3 and 7 months (64%). Early identification of vestibular fistula significantly improved surgical outcomes and reduced complications.

Most patients did not have associated abnormalities (60 cases, 90.9%), while 5 patients (7.6%) had cardiac abnormalities, and 1 patient (1.5%) had both cardiac issues and a cleft lip. This is consistent with findings from an Indian study (10), where 16 patients (38%) presented with associated congenital anomalies, including atrial septal defect, ventricular septal defect, and patent ductus arteriosus as the cardiac anomalies.

Out of the total, 62 patients (93.9%) experienced abdominal distension, while 4 patients (6.1%) did not. Additionally, 52 patients (78.8%) suffered from constipation, whereas 14 patients (21.2%) did not experience this issue. This observation is in line with the Egyptian study (9), where constipation was reported in 15 patients (50%). Various factors have been explored to account for the persistence of constipation following the repair of anorectal anomalies, including a narrow neoanus requiring dilatation, hypomotility of a significantly dilated rectosigmoid colon, neurogenic causes, dyssynergic defecation, and the potential presence

of aganglionosis. No single factor could fully elucidate the pathogenesis of constipation in these patients, indicating a multifactorial etiology. Nevertheless, to the best of our knowledge.. Among those who underwent preoperative dilatation, 40 (60.6%) were not dilated prior to surgery, while 26 (39.4%) were. Over the last ten years, numerous authors have asserted that simple dilatation of the fistula is adequate in most instances. Out of 59 patients, 59 (89.4%) underwent colostomy, whereas 7 (10.6%) did not. Specifically, 27 (46%) had colostomy performed between the ages of 2 to 6 months, 20 (34%) during the first month, 9 (15%) between 7 to 12 months, and 3 (5%) after one year (n=59). Of those, 59 (89.4%) were repaired with colostomy, while 7 (10.6%) were repaired without it. This finding is consistent with the results of the Waheeb study conducted in Egypt (12). Traditionally, the surgical procedure is executed in three stages, which include a colostomy, posterior sagittal anorectoplasty (PSARP), and subsequent closure of the colostomy. However, various studies indicate that PSARP can be performed in a single stage without colostomy, yielding comparably favorable outcomes. The ASARP technique, with or without colostomy, is being utilized as an alternative to PSARP in certain medical centers, offering improved cosmetic and functional results by minimizing postoperative constipation in comparison to PSARP. The conventional surgical correction involves a diverting colostomy, typically performed during the neonatal period, followed by posterior sagittal anorectoplasty around the age of one year, and closure of the colostomy several months thereafter. In many developing nations, neonatal surgery remains in its early stages. Performing a diverting colostomy on a female neonate with a fistula unnecessarily exposes the patient to risk, as the gastrointestinal tract is already self-decompressing. Furthermore, up to 61% of these children present with associated anomalies that may render neonatal general anesthesia significantly hazardous. Therefore, unnecessary neonatal surgeries should be avoided. The predominant types of repair included limited PSARP at 32 (48.5%), ASARP at 25 (37.9%), and anal transfer at 9 (13.6%). ASARP, initially described for various conditions such as postoperative fecal incontinence, vestibular anus, rectal prolapse, and perineal trauma, has been adopted as an alternative approach to PSARP, yielding comparably positive outcomes. Although not statistically significant, ASARP demonstrates marginally superior results compared to PSARP regarding postoperative outcomes.

Complications in anal transfer were observed in 8 patients (18.6%), ASARP in 15 patients (34.9%), and PSARP in 20 patients (46.5%). Out of 43 patients (65.2%), complications were noted, while 23 patients (34.8%) experienced no complications. The majority of complications were due to anal stenosis in 25 patients (58.1%), minimal wound infection in 8 patients (18.6%), perineal body disruption in 5 patients (11.6%), rectovaginal fistula in 3 patients (6.9%), and combined perineal body disruption with anal stenosis in 1 patient (2.3%) (n=43). This aligns with the findings of an Indian study (10), which reported that superficial wound infections were twice as common in the PSARP group compared to the ASARP group. The infection rate for

ASARP in this series was nearly 10%, which is comparable to previous studies. Nevertheless, all patients were managed conservatively with local wound care. ASARP offers several advantages in treating vestibular fistula compared to PSARP, including comparable post-operative complications, favorable cosmetic outcomes, excellent continence, and a reduced need for laxatives.

Among the patients, 46 (69.7%) did not exhibit perineal body contraction, while 20 (30.3%) did. This finding is consistent with the results of the Waheeb study conducted in Egypt (12). The perineal body and posterior fourchette were meticulously closed from within outwards, resulting in an aesthetically pleasing appearance of the perineum. Another benefit was the elimination of the proximal pelvic colostomy, which is traditionally associated with the PSARP technique, thereby reducing the risk of wound sepsis. Overall, there were minimal complications, with no instances of fecal impaction or anal stenosis. Additionally, a shorter hospital stay was noted as another advantage.

Conclusion

- The risk of wound dehiscence is associated with limited PSARP and ASARP procedures performed without colostomy.
- In our locality, a two-stage correction of RVF is considered safer and more advantageous compared to a one-stage procedure.
- The early identification of vestibular fistula has led to improved surgical outcomes and a reduction in complications.
- Pre-operative dilatation does not significantly contribute to the reduction of complications in the early stages.
- The significance of performing Limited PSARP, ASARP, or anal transfer is limited and largely depends on the surgeon's experience.

Recommendation

- It is recommended to train midwives to conduct genital examinations on every neonate for the early detection of RVF.
- A three-stage repair is deemed safer and associated with fewer complications.
- A combined clinic involving pediatric surgeons, gynecologists, social workers, and psychologists should be established for all patients with ARMF.
- This study should be followed up after two years to evaluate the mechanisms of continence, the necessity for laxatives, and the need for dilatation.

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