Department of Surgery , Pediatric surgery unit
College of Health Sciences, Addis Ababa University

Master's Thesis

Outcomes of Outcomes of Anerior sagittal Ano-Rectoplasty in the Management of Rectoves Tibular Fistula at Two Referral Hospitals

Primary Investigator
Zinedin Beker , MD
Final Year Pediatric Surgery Resident, **Department of Surgery , Pediatric surgery unit**
College of Health Sciences, Addis Ababa University

Advisor
Dr Amezene Tadesse (MD,FCS-ECSA ,Associate Professor of Surgery
Consultant General and Pediatric Surgeon)

May, 2021

Declaration

I **Zinedin Beker** declare that this paper is a result of my independent research work on the topic entitled "Outcomes of Outcomes of Anerior sagittal Ano-Rectoplasty in the Management of Rectoves Tibular Fistula at Two Referral Hospitals" in partial fulfillment of the requirements for specialty certificate for General Pediatric Surgery at Addis Ababa University, College of Health Sciences, Department of Surgery, Pediatric Surgery Unit. This work has not been submitted for a degree to any other university. All the references are also acknowledged.

Dr. Zinedin Beker

Signature: _____

Date: ___/_____/_____

Confirmation

This is to certify that Zinedin Beker has carried out this research work on the topic entitled "Outcomes of Outcomes of Anerior sagittal Ano-Rectoplasty in the Management of Rectoves Tibular Fistula at Two Referral Hospitals" under my supervision. This work is original in nature and has not been presented for a degree in any University and it can be submitted for the partial fulfillment of the requirements for the award of the specialty certificate for General Pediatric Surgery.

Prof. Amezene Tadesse (Professor of Pediatric Surgery, FCS-ECSA,)

Signature: _____

Date: _____/_____/_____

Addis Ababa, Ethiopia

Abstract

Background

Over the years, a great deal of information has been acquired concerning the many variants of ARM. The current mortality for these anomalies for the most part has been very low and is related to associated anomalies and unfavourable chromosomal and genetic syndromes rather than the result of treatment of the ARM per se.

In general, agreement has been reached regarding some embryologic and anatomic considerations, diagnostic evaluation, and preoperative assessment. Most investigators would agree that results are better following repair of low defects when compared to intermediate and high anomalies and those with cloaca. There remain several areas of controversy regarding the choice and timing of the procedure and methodology used to assess results. Certain poor prognostic factors have been identified, including abnormal sacrum, deficient pelvic innervation, poor perineal musculature and disorders of colonic motility.

Objectives

To investigate the overall outcome ASARP in the management of recto vestibular fistula in Tikur Anebessa Hospital & Minilik Referral Hospital , Ethiopia from January 1, 2015 to December 30, 2020

Methods

A cross-sectional study design with convenience sampling technique was used on 74 patients who had ARM with RVF. Krickenbecks bowel function scoring system was used to assess the bowel functions score. Patients cards, follow-up charts and direct phone contacts with families were used to collect data from selected ARM patients who undergone ASARP in the two of the referral Hospitals from January 1,2015 to December 30, 2020.

Result : In this study we do not found statistically significant associations between the type of operations and the occurrence of Post-operative early wound complications or anal stricture But the overall frequency of early post-operative complications more in two staged ASARP (14.8%) and anal stenosis is observed in the Three staged ASARP group more frequently (21%) . This study also found lower rates of screening for associated anomalies

and higher rate of postoperative neo anal stricture (17.6%). Majority studied patient have an excellent bowel function (96.2%).

Keywords: Anorectal malformation, Anterior Sagittal Anorectoplasty, Recto-Vestibular Fistula

Acknowledgement

I would like to express my deepest gratitude to my advisors, Dr. Amezene Taddesse and paediatric surgery department of AAU for their incalculable support and critical and constructive comments.

Table of Contents

Abstract ii

Acknowledgement iv

List of tables vii

Abbreviations ix

Chapter one 1

Introduction 1

1.1 Background 1

1.3 Significance of the study 5

2. Chapter two 7

Objective of the study 7

2.1 General objective 7

2.2 Specific objective 7

3. Chapter three Materials and Methods 8

3.1 Study design 8

3.2 Setting 8

3.3 Study participants 8

3.5 Sampling procedure 8

3.6 Inclusion criteria 8

3.7 Exclusion criteria 9

3.8 Variables 9

3.8.1Dependant variable 9

3. 8.2 Independent variable 9

3.9 Data Quality control 9

3.10 Data collection 9

3.11 Instrument 10

3.12 Data analysis 10

3.13 Ethical clearance 10

3.14 Definition 10

3.15 Dissemination of the result: 12

4. Chapter four 13

4. Result 13

4.1 Demographic Data 13
4.2 Preoperative clinical status 15
4.3. Operative characteristics 19
5.1 Discussion 23
Reference list 31

List of tables

Table 1 Krickenbecks ARM classifications …….. **Error! Bookmark not defined.**7

Table 2 Table 2. The characteristics of some independent variable, Addis Ababa, 2013……..16

Table 3 Table 3, Birth weight……..17

Table. Distribution showing the presentation of symptoms, Addis Ababa,2013..... 17

Table 5 Frequency of screening workups and Positive findings ……19

Table 6 Types of associated congenital anomaly .....

Table 7 . The Management of major early post-operative complications ……21

Table 8. The frequency of early postoperative complications to the type of operations Cross tabulation ……22

Table 9 Distribution of the study subjects by the incidence if anal stenosis among the different stages, Addis Ababa, 2013…….22

Table 10 Crosstab between poor BFS with early or stoma associated complication……..24

List of Figures

Figure 1 … Figure 1, Overall diagnosis ……18

Figure 2 Pelvic/spinal x-ray ……...19

Figure 3 Mode of operations ……..……20

Figure 4 The type of operation done ……..……23

Abbreviations

AA Anterior Anus

ARM(s) Anorectal malformation(s)

AS Anal stenosis

ASARP Anterior sagittal anorectoplasty

PSARP Posterior sagittal anorectoplasty

BFS Bowel function score

CM Cloacal membrane

EUROCAT European Surveillance of Congenital Anomalies Registry

PF Perineal fistula

MRH Minilik referral hospital

RA Rectal atresia

RUF Rectourethral fistula

VF Vestibular fistula

TASH Tikur anebessa specialized hospital

Chapter one

Introduction

1.1 Background

Anorectal malformations (ARMs) comprise a spectrum of congenital anomalies that continue to present a challenge for pediatric surgeons.(1) ARMs affect around 1:2000-2500 births,(2-4) ranging in severity from mild anterior displacement of the anus to very complex malformations of the hindgut and urogenital tract.(3,4) Advances in modern surgical

techniques and neonatal care have greatly improved survival among ARM patients over the last decades, and early mortality is now unusual in the absence of fatal associated cardiac or chromosomal defects.(5) Accordingly, the focus of surgical care has shifted beyond initial survival of the patient towards ensuring that children treated for ARMs to grow up having bowel function that is compatible with a good quality of life.(6) For most, this means being able to actively participate in their social environment without significant limitations from bowel function, for which fecal continence is a major determinant.(7-9)

Posterior sagittal anorectoplasty (PSARP), first introduced in 1982 by De Vries and Peña (10) and followed later by its limited modification anterior sagittal anorectoplasty (ASARP),(11) represents the basis of the modern surgical approach to ARMs with

termination of the anal canal outside the voluntary sphincter complex. PSARP, entailing exposure of structures under direct vision and restoration of the normal anatomical relationships between structures has replaced earlier classical operations, including abdominoperineal or sacroabdomino- and sacroperineal pull through(12-15) as the ‘gold standard’ approach.(16) Other significant developments have included recommendations for centralization of surgery for ARMs to specialist tertiary units,(16) increasing understanding of their pathologic anatomy, and improved treatment of major functional complications such as constipation.(16)

The literature that has accumulated concerning the outcomes in childhood has been more optimistic than preceding the PSARP era, (17) but the results for severe ARMs in particular have varied widely, and there remains limited information on the

outcomes of modern treatments up to adulthood. Several studies addressing the management of ARM, especially the timing and single-stage or multistage approach, and various perioperative diagnostic studies have been emerging. Nevertheless, many surgeons prefer using the anterior sagittal anorectoplasty approach in dealing with congenital rectovaginal fistulas in females (4, 5, 6).

In Ethiopia colostomy is being performed for most ARM patients despite their sex or type of ARM either for the relief of obstruction or as the prerequisite for the definitive procedure. Most patients were managed by PSARP even for female ARMs with RVFs. (18).

1.2 Statement of the problem

Anorectal malformations (ARM) represent a complex group of congenital anomalies resulting from abnormal development of the hindgut, allantois and Mullerian duct, leading to incomplete or partial urorectal septal malformations .Anorectal malformations (ARM) present an incidence rate ranging from 1:1,500 to 1:5,000 live births, comprising approximately 0.2–0.3% births, they have been reported to comprise up to 1.2% of reported birth defects and have variable clinical presentations ranging from mild forms that might require only minor surgical interventions to more complicated cases that need to be managed with multi-staged operations (17,19,20).

Associated anomalies occur frequently and range from 36.4% in the EUROCAT study to 45.2% in another large series. Although these would appear to be higher in supralevator lesions as opposed to low lesions, it is of considerable interest that low vaginal anomalies in females appear to have as high an incidence of associated anomalies as high rectal lesions. (21, 22, 23, 24)

In general, the male: female ratio associated with ARM is almost equal, with a 56:44 ratio previously reported in large collective series as well as in the EUROCAT study of isolated ARM (male: female ratio = 1.06). Exceptions to this are anal atresia (male preponderance) and ectopic anus and congenital anal fistula (female preponderance) (21, 22 & 24).Vestibular fistula is the commonest anorectal malformation in the female child (25). Over the last 20–30 years, there have been significant advances in the management of children with anorectal malformations, both medical and surgical. Changes have also occurred in the terminology used to describe the various types of anorectal malformation. Previously, the terms “low,” “intermediate,” and “high” malformations were used; however, these terms are not as anatomically clear as those used in the Krickenbeck Classification, developed in 2005.

Table 1.

KRICKENBECK CLASSIFICATION

Common malformations	Rare malformations
Perineal fistula	Pouch colon atresia/stenosis
Rectourethral fistula (prostatic, bulbar, bladder neck)	Rectal atresia/stenosis
Rectovesical fistula	Rectovaginal fistula
Vestibular fistula	H-type fistula
Cloaca	Others (e.g., posterior cloaca variant, cloacal exstrophy, covered cloacal exstrophy)
No fistula	
Anal stenosis	

ARM form a significant load on the surgical services, particularly in developing countries, not only in the emergency situation but also in terms of long-term corrective procedures. Although there have been major advances in the management of these children during the last 15 years, these patients still represent a continuing challenge as a result of the significant reconstructive problems involved, as well as the fact that a significant number suffer from faecal and urinary incontinence, as well as the possibility of sexual inadequacy in later life.

Despite a better understanding of embryology, the anatomy of ARM and the physiology of continence, the management of children born with ARMs continues to be a surgical challenge and is still fraught with numerous complications and often leads to less than perfect qualitative results (26, 27 & 28). Different surgical techniques have been described for the correction of such anomalies including cutback procedure, anal transplantation, YV plasty and posterior sagittal anorectoplasty (PSARP) which was introduced by Alberto Pena and Devries in early 1980s(29). For so many years we were managing all cases of anorectal malformations by PSARP only, but from year 2010 onwards we included the anterior sagittal anorectoplasty (ASARP) as an optional technique for management of vestibular fistula.

ASARP is an optimal technique for intermediate ARMs and shows better cosmetic and functional outcome by reducing postoperative constipation compared to PSARP.(4,31)Similar to PSARP, the incision in ASARP is oriented along the sagittal plane. However, the primary advantage of ASARP is that, only the anterior aspect of the sphincter muscle complex is divided and continence mechanism is preserved (30,32). Moreover, as clearly alluded by Wakhlu et al “ASARP allows placement and anchoring of the mobilized rectum

within the muscle complex; the sphincteric muscle and the perineal body are accurately reconstituted and a normal perineum is reconstructed" (30).

Delay in presentation of patients with ARM leads to progression of neonatal intestinal obstruction, sepsis, aspiration pneumonia, intestinal perforation, and sometimes death (33). Adejuyigbe et al. in a retrospective review of 86 patients managed on account of ARM at Ile Ife, Nigeria reported that 74 (86.0%) presented after 24h of birth(34). In that series, nearly 60% of the patients presented with gross abdominal distension and over one-quarter had associated vomiting (34). Govender and Wiersma (36) in a retrospective study of 273 patients who presented to a tertiary hospital over a period of 8 years reported that 158 (57.9%) presented after 24h of birth i.e., delayed. Similarly, 63% of 78 children treated for ARM at a major referral center in southwest Uganda presented after 48h of birth (35). Delayed presentation is worse among female children because there is still some faecal discharge through the vestibular fistula in most instances unlike in males where abdominal distension occurs over a few days in most patients with a rectourethral fistula (33).

Reports indicate that a large proportion, ranging from 25 to 94.8% of childbirths in Africa take place outside the hospital. Delivery outside a hospital setting is significantly associated with delayed diagnosis of ARM. Presentation outside the neonatal period is widespread as 19– 85% of patients with ARM in Africa present outside the first 4 weeks of life (33). Delay in diagnosis has been correlated with poorer outcome and a higher mortality (34). So, this study is planned to assess burden RVF, type of operative modalities we use and the overall outcomes of ASARP in the management of RVF in our center.

1.3 Significance of the study

The aim of this study is to assess overall outcomes of ASARP in the management of RVF at two of referral hospitals in Addis Ababa. The study will help the care providers, to use the one which works best order to practice the safest and the most effective care and options of surgical management. Moreover, it might be used as a standard tool for management of Low ARMs some acquired causes of RVF in Ethiopia.

2. Chapter two

Objective of the study

2.1 General objective

The general objective of this study is investigate the overall outcome ASARP in the management of RVF in TASUH & MRH, Ethiopia from January 1 2015 to December 30, 2020..

2.2 Specific objective

1. To compare the incidence of immediate complications, that is, perineal body disruption and surgical site infection between primary, two and three staged ASARP.

2. To compare the incidence of neoanus stricture (stenosis) between primary, two and three staged ASARP

3. To assess bowel functions in patients who undergone ASARP

4. To assess post ASARP stoma (colostomy) associated complications

3. Chapter three Materials and Methods

3.1 Study design

Cross sectional retrospective descriptive study was conducted from January 1 2015 to December 30, 2020. Krickenbeck bowel function score was used to assess the post-operative bowel functional status for those with age above three.

.

3.2 Setting

The study was conducted in TASH, Addis Ababa, Ethiopia. TASH is a tertiary teaching referral hospital for the whole of the nation with a population of 100 million of which 43.47 % are under 15 years old. It has 40 pediatric surgery beds and neonatal unit annually it operates around 1000 pediatric surgery cases. The unit is served by 7 pediatric surgeon, pediatric surgery fellows, pediatrics & pediatric surgery residents on training, nurses, interns and other supportive staff.

3.3 Study participants

In this study the source population were all pediatric surgical patients who were admitted at TASH and MRH for surgical treatment during the stated period.

All pediatric patients with RVF who underwent ASARP either single, two or three staged were included in this study during the stated period were studied.

3.5 Sampling procedure

All patients who meeting exclusion and inclusion criteria during study period were included in the study.

3.6 Inclusion criteria

All patients with RVF (congenital) who underwent ASARP at TASH and MRH

3.7 Exclusion criteria

- Patients who undergo ASARP for other conditions
- Failure to get consent from parents, for the interview session in clinic for assessment on continence

3.8 Variables

3.8.1Dependant variable

. Type of surgical procedures used

- Major postoperative complication following each surgery

3. 8.2 Independent variable

Independent variables include non-modifiable variables such as gender, race, age (in days) at time of surgery, gestational age, birth weight, Dur, preoperative clinical condition , indication for surgery and presence of sepsis or other postoperative complications

3.9 Data Quality control

In order to assure data quality, high emphasis was given to minimize errors using the following strategies: the questionnaire was be pretested and subsequent correction and modification was made depending on the input from the adviser & experts. Data collection was done by the investigator on daily basis and the collected data was reviewed and checked for completeness before data entry.

3.10 Data collection

The questionnaire that was developed by the investigator based on the literature on the topic in less developed countries.

The supervisor was consulted about the structure and content of the questionnaire & it's checked.

The questionnaire has four sections in which extracted data was captured. The first section was capture of the demographic information, the second section was the capture information on preoperative clinical status, Complications during the perioperative period, and the fourth section was capture information regarding postoperative course.

Data was collected after the researcher attained consent from all required authorities. The researcher and pediatric surgery resident attaching to the unit will follow all patients who are admitted for surgical treatment throughout preoperative and postoperative stay. Information regarding operative detail was taken from operation note, anesthesia sheet.

Information regarding postoperative course of the patient and immediate causes of death, in addition to close follow up by investigator, was gathered from progress note and from attending pediatric surgery resident attaching to the unit for recent cases.

Once the information gathered and questionnaire filled & completed as soon as possible, completeness checked and collected & kept with investigator until data entry for analysis.

3.11 Instrument

3.11 Krickenbeck bowel function score is used to assess the postoperative bowel function score

3.12 Data analysis

Once collected, the data was computerized and analysed in the Statistical package for social science (SPSS) Version 20. Cross-tabulation was used to determine group differences. Afterward, the Chi-square test was performed to determine whether the variables are statistically independent or if they are associated.

3.13 Ethical clearance

Ethical approval was obtained from the Addis Ababa University College of health science, TASH Research Committee. The study used data gathered throughout stay of patient in the hospital and during follow-up clinic visits. Information was collective and no names or identifying information on the child's were used. The study ensured that all the information obtained remained confidential and were not disclosed to anyone other than the research supervisors. The identifiable details of the participant child was removed and codes were used instead to maintain anonymity.

3.14 Definition

Anterior sagittal anorectoplasty (ASARP) Devised by Okada et al1 for repair of rectovestibular or anovestibular fistula, the patient is operated on in a lithotomy position; a

median perineal skin incision is made and the sphincter muscles are cut from it. Rectum is then brought out through the external sphincter muscles centrally.

Rectovestibular fistula (RVF) The rectum terminates at or just below the pubococcygeal line, with an anterior fine fistula, 1–2 cm along the vestibule, coursing immediately adjacent to the posterior wall of the vagina. The terminal gut is supralevator, the fistula translevator. The vagina is often double. Diagnosis is made based on the following features:

1. Three orifices in vestibule, urethra, vagina, and a fine rectal fistula in fossa navicularis. Orifice totally surrounded by red vestibular mucosa.

2. Probe into fistula passed cranially only.

Posterior sagittal anorectoplasty (PSARP) PSARP is the most commonly used operation for the repair of anorectal anomaly (ARM)In this procedure, the patient is positioned prone. The contraction of the anal complex is predetermined by electrostimulation prior to making a midline incision. The layers of muscles are split in layers with careful preservation before delivering the rectum down, as exactly described by de Vries and Pena.

3.15 Dissemination of the result:

The result of this research will be disseminated to the Surgical Department, pediatric surgery unit department, Addis Ababa University, Tikure ambessa, and MIlilik Refferal hospital. It will be sent for publication on scientific journals in related fields.

4. Chapter four

4. Result

A total of 142 ASARP procedures were done at the Two referral hospitals of which 130 and 15 at TASUF and MRH respectively. The charts of 89 patients were retrieved of which 74 of them undergone ASARP for ARM with RVF and the rest under gone ASARP for ARM with RPF and short common channel cloaca.

4.1 Demographic Data

Out of 74 study participants 91.9% of them were from TASH followed by MH (6.8%). The majority (45.9%0 of the patients were from Oromia region followed by Addis Ababa (21.5%). About 81% of the patient`s mothers were in the age range of 20-34.Concerning the birth weight of the child, 40.8% were in the range of 2600-3500, while 27.1% of them were reported that they didn`t know the birth weight. When participants were asked about place of delivery the majority (71.6) were reported that they deliver in health facility out of the center followed by home delivery (27%). (table1).

Table 2. The characteristics of some independent variable, Addis Ababa, 2013.

Variable	Frequency	percentage
Name of hospitals		
MH	5	6.8
TASH	68	91.9
TSASH	1	1.4
Administrative region		
Amhara	7	9.5
Oromia	34	45.9
Somali	3	4.1

Benshangul Gumuze	1	1.4
SNNRP	6	8.1
Harari	5	6.8
Deridawa	2	2.7
Addis Ababa	16	21.5
Mother`s age at child`s birth		
20-34	60	81.1
35-49	14	18.9
Birth weight at birth		
2000-2500	20	27.1
2600-3500	30	40.8
3600-4500	5	5.5
Unknown	20	27.1
Place of delivery		
Home	20	27
Health facility in the same center(TAH)	1	1.4
Health facility out of the center	53	71.6
perinatal history		
Yes	37	50%
No	37	50%

Table 3, Birth weight

	Frequency	Percent
low birth weight	11	14.9
normal birth weight	42	56.8
Macrosomia	1	1.4
Total	54	73.0

About 37(50%) mothers had prenatal Ultrasound but no prenatal findings were identified.

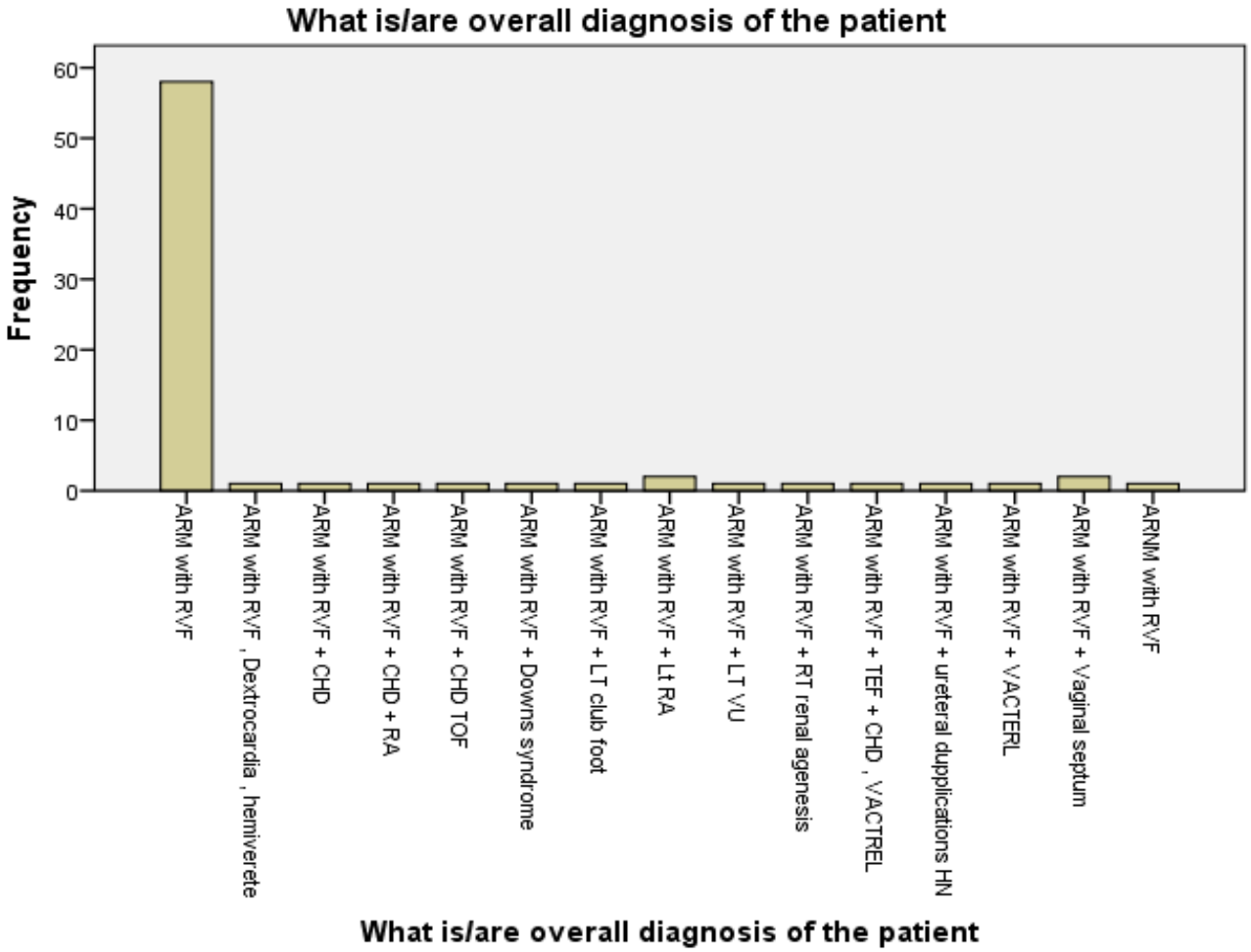
4.2 Preoperative clinical status

When we assess the presenting symptom, the majority (94.6%) of the study participants had experienced passage of fecal matter through abnormal route. While 95.9% of them didn`t experience Bilious vomiting and 85.1% of them also didn`t experience non bilious vomiting. Those participants who had abdominal distention were 35.1% and straining during defecation were seen in 25.7% (table3.)

Table4. Distribution showing the presentation of symptoms, Addis Ababa,2013

Variables	frequency	percentage
Passage of faecal matter through abnormal routes		
Yes	70	94.6
No	4	5.4
Bilious vomiting		
Yes	3	4.1
No	71	95.9
Non bilious vomiting		
Yes	11	14.9
No	63	85.1
Abdominal distention		
Yes	26	35.1
No	48	64.9
Straining during defecation		
Yes	19	25.7
No	55	74.3

All of the studied patients had ARM with RVF. None of the patients require any special support preoperatively nor did they have sepsis or shock 48 hours prior to surgery.When we see the overall diagnosis of the patient reported that ARM with RVF is the major diagnosis followed by ARM with RVF +Lt RA. All the others are in small amount (figure1).



Most of the patients had at least Ultrasonography. Table 3. Shows the distribution of the study participants by what they were screened, accordingly 67/74(90.5%) had abdominal U/S and overall 8.1 % patients screened had abnormal result. Common findings on ultrasound are renal agenesis, hydro nephrosis and ureteral duplications. While 23/74 (31.1 %) of them had pelvic /spinal x-ray, 2.7 % were abnormal common findings were Hemivertebra. Only 25.7% and 23% reported had **Echocardiogram**, and Spinal U/S respectively (table4). Common finding on echocardiography were VSD, ASD, PDA and TOF (8.1%). And there was one spinabifida case.

Characteristics	Frequency	Percentage
Associated Congenital malformations,		
Yes	68	91.9
No	6	8,1
Echocardiogram		
Yes	19	25.7
No	55	74.3
Spinal U/S		
Yes	17	23.0
No	57	77.0
Abdominal U/S		
Yes	67	90.5

Table 5.

No	7	9.5

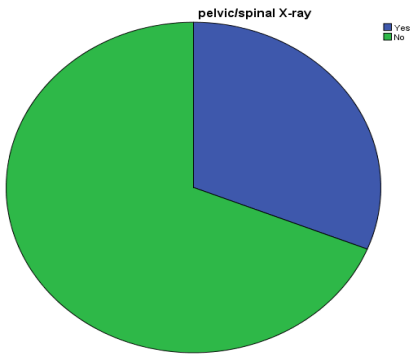


Figure 2. Pelvic/spinal x-ray

Other rarely ordered investigations and diagnostic procedures were cystoscopy , VCUG and IVP Associated congenital and chromosomal anomalies were found in 16/74 (21.6%) of which anomalies of genitourinary system accounts for the majority 56 %(9/16) followed by cardiac anomalies accounting for 31.25% (5/16). Table ….. Summarizes the idetifed anomalies and the specific types. Note that more than one associated anomalies are seen in single patient (18.5%)

Table 6. Types of associated congenital anomaly

System	Frequency	Types
Genitourinary malformations	9/16 (56.25%)	Renal agenesis 5
		Vaginal septum 2
		Hydro nephrosis 2
		Ureteral duplications 1
Cardiac anomalies	5/16 (31.2 %)	VSD 2
		ASD,TOF and Dextrocardia 3
Chest	1/16 (6.25%)	TEF 1
CNS anomalies	1/16 (6.25%)	Spina bifida 1
MSS anomalies	1/16 (6.25%)	Hemivertebre 1

Chromosomal anomalies	1/16 (6.25%)	Down syndrome 1

4.3. Operative characteristics

The majority of the operation was done on planned that is Elective 41/74 (55.4%) and there is also a significant number were in emergency level 33/74 (44.6%).

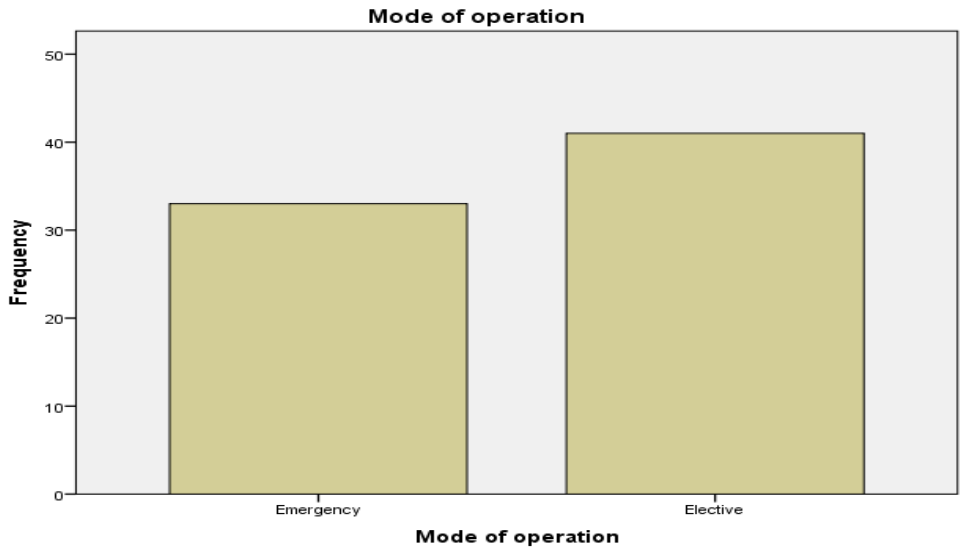


Figure 3

When we see the type of surgery patients undergone most 32/74(43.2%) three staged, 27/74 (36.5 %) two staged and 15/74 (20.3 %) single staged ASARP.

Colostomy was opened in 59 /74(79.7 %) patients of which 5/59(8.5%) had colostomy associated complications

Prolapse, requiring reoperation for refashioning 2/59 (3.3%) , complete wound dehiscence 1/59 (1.6 %) and parastomal herniation treated conservatively 3/59 (5 %)

Early perineal postoperative complications were seen in 9/74 (12.2 %) of which Perineal body breakdown, neoanus intact seen in 4/74 (5.6 %), perineal wound infections in 3/74 (4.1%) , Perineal body breakdown, neoanus retracted seen 2/74 (2.7%) . The major (Perineal body breakdown and Perineal body breakdown, neoanus retracted) complications were managed with early primary repair 4/6 (66.7%), With delayed repair with covering stoma 1/6(16.7 %) and local wound care 1/6(16.7 %).

Table 7.

	Frequency	Percent
. Early primary repair	4	5.4
Delayed repair with covering stoma	1	1.4
Treated conservatively with wound care	1	1.4
Total	6	8.1

The rate of early postoperative complications is similar for those who undergo two staged and three staged repair but with lesser rate of early complications were seen in single stage group.

Table 8. The frequency of early postoperative complications to the type of operations Cross tabulation

		Early postoperative complications		
		Yes	no	Total
Type of operation done	primary ASARP	1	14	15
	Two staged ASARP	4	23	27
	Three staged ASARP	4	28	32
Total		9	65	74

Most the patients 73/74 (98.6 %) were allowed to feeding within 48 hours while only 1/74 (1.4%) allowed to feed after 72 hours. Most 58.1 % received antibiotics for 3 to 5 days, 29.7 % for 1 to 2 days and 12.2 % received for more than 5 days.

When we see the durations anal dilatations overall and before closure of the colostomy 51/74 (68.9 %) undergone dilatations for more than 9 months, 13/74 (17.6) for 6 to 9 month and only 10/74(13.5 %) for 3 to 6 months.

The neo anus stricture rate in this study is found to be 17.6 % (13/74) of which 7 (53.8%) were managed with dilatations under General anaesthesia and 6(46.2 %) managed with redo anoplasty

From patients with neo-anus stenosis most 7/13 (53.8%) had three stage ASARP, 4/13(30.7%) had two staged ASARP and 2/13(15.4 %) had three staged ASARP. the proportions of neoanus stricture within specific type of operation were 13.3 %,14.8 %,and 21.9 % for single , two and three staged ASARP respectively .

Table 9.Distribution of the study subjects by the incidence if anal stenosis among the different stages, Addis Ababa, 2013

Variable	Type of operation done				
Anal stenosis	Primary ASARP	Two staged ASARP	Three staged ASARP	Chi square	
Yes	13.3%	14.8%	21.9%	.737	
No	86.7%	85.2%	78.1%		
Early postoperative complications					
Yes	6.7%	14.8%	12.5%		
No	93.3%	85.2%	87.5%	.605	

43 0f the 74 patients were above age of 3 i.e. above the age of toilet training of which 26 0f the participants responded to phone call for bowel function assessment by Krickenbeck bowel function tool and 25/26 (96.5%) responded to have voluntary bowel movement, 1/26(3.8 %) don't have voluntary bowel movement. 2/26 (7 %) had Grade 1 soiling and 1/26 (3.8%) have constipation managed with diet . Overall bowel function score is good in 96.2%.

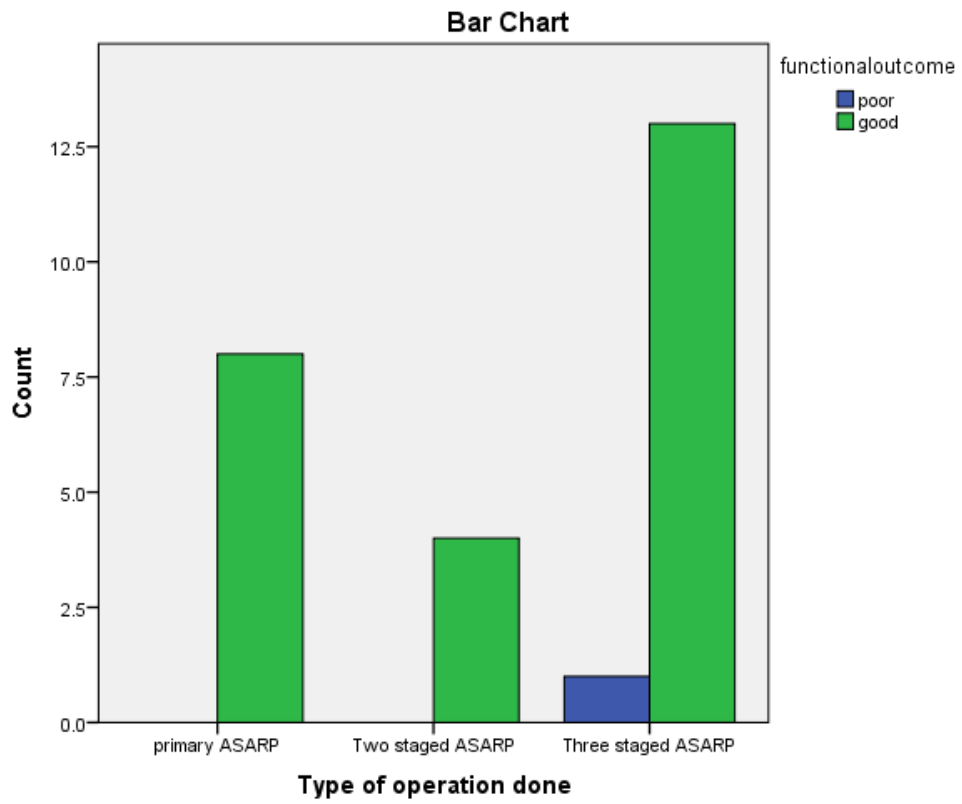


Figure 4

Table 10 Crosstab show the patient with poor BFS has no either early or stoma associated complications

Stoma associated complications			functional outcome	
			poor	Good
Yes	Early postoperative complications	Yes		1
		No		1
	Total			2
No	Early postoperative complications	no	1	15
	Total		1	15
Total	Early postoperative complications	yes	0	1
		no	1	16
	Total		1	17

5. Chapter five discussion and conclusion

5.1 Discussion

In this study 74 patients undergone ASARP for ARM with RVF of which majority (71%) of them were delivered at health facility and most them had Normal birth weight (77.8%) and the maternal age for most of them was between 20 to 35 years. Most of patients enrolled in the study reported passage of faecal matter through an abnormal route (94.6%).

Regarding screening in our study 67/74(90.5%) had abdominal U/S and While 23/74 (31.1 %) of them had pelvic /spinal x-ray (2.7 % were abnormal). Only 25.7% and 23% reported had Echocardiogram, and Spinal U/S respectively. Overall 8.1 % patients screened had abnormal result. Common findings on ultrasound are renal agenesis, hydronephrosis whereas spinal/pelvic X-ray revealed a case of hemivertebra. Among the patient who had echocardiography found to have VSD, ASD, PDA and TOF (8.1%). And there was one spinabifida case.

In Survey on the management of anorectal malformations (ARM) in European paediatric surgical centres of excellence, where 23 countries involved and experts were asked about preoperative screening and they answered Investigations routinely performed in the neonatal period Plain and/or lateral X-ray of the entire spinal column 11 (44 %) , Plain and/or lateral X-ray of lumbar and sacral spine 11 (44 %) Echocardiography 23 (92 %) Abdominal ultrasound, including the urinary tract 24 (96 %) Spinal ultrasound 22 (88 %) in contrary to our study in this survey significantly higher number of patients are screened . this might be because of a better availability of facilities and experts in the Europe . (35)

In contrary to our study in Beijing , china (2015) they have screened all patients with chest X-ray, spine X-ray, abdominal ultrasonography, echocardiography, and fistulogram prior to surgery. The spine MR was performed in suspected cases of vertebral defect. In comparison to this we screen lesser number of patients again this might because of a better availability of facilities and experts in this developed country. (37)

The large number of associated congenital malformation 16/74 (21.6%) seen in this study is similar with the studies conducted in other studies where genitourinary system accounts for the majority 56 % (9/16) followed by cardiac anomalies accounting for 31.25% (5/16)

In study done in Bangladesh on 23 with RVF they found 6/23(26%) patients had associated anomalies: two patients had renal anomaly (1 pelviureteric junction obstruction, 1 fused kidney), one had a cardiac anomaly (tetralogy of Fallot with atrial septal defect), one had askeletal anomaly (polydactyly) and one had incomplete cleft palate. (40) This result is in line with our study with regard to the types and frequency of associated anomalies. Almost a close result were found In the Chinese study were four patients had associated anomalies (4/26, 15.3 %), including

an urachal cyst (1/26, 3.8 %), a renal duplication (1/26, 3.8 %), a patent foramen ovale (1/26, 3.8 %), and a spinal abnormality (1/26, 3.8 %). (37)

In this study the majority of the operation were done on planned base Elective 41/74 (55.4%) and there is also a significant number were in emergency level 33/74 (44.6%). Most of the patients 32/74(43.2%) undergone three staged, 27/74 (36.5 %) two staged and 15/74 (20.3 %) single staged ASARP.

In contrary to our study in Alexandria, Egypt a 22 year retrospective single centre study was undertaken, were 594 patients included; About 526 patients had RVF. It showed all of these patients had been subjected to ASARP technique which had been done in one stage without proceeding colostomy in about 470 (79%) cases and in staged fashion with a protective colostomy in 124 (20 %) patients. This study revelled they have done more single stage repair than ours this might be because of an increased experience with Primary ASARP and a more promising results from previous studies. (38)

In support to the above findings in a survey of members of the Egyptian paediatric surgical association on a current management of anorectal malformation in Egypt were 91 paediatric surgeons were asked their surgical preference in the management of ARM with RVF, 9(10.98%) surgeons prefer to do primary neonatal ASARP, 20 (24.39%), from 1 to 3 months for after the third month for life by 40 (48.78%), whereas 13 (15.85%) did not perform single-stage repair. As regard to outcome of primary versus staged repair for rectovestibular fistula, 64.7% of participants stated that primary repair has similar outcomes compared with staged repair, 19.5% stated that staged repair has lesser complications and better outcome, and 15.8% of participants did not perform single stage repair. (42)

In the European survey in female patients a primary anorectal repair without a diverting enterostomy is routinely performed for perineal fistula by 23 participants (92 %), vestibular fistula by 15 (60 %), and in patients with no fistula with rectal gas below the level of the coccyx by 8 (32 %). (35)

In comparison to the above studies and surveys we do less single stage repairs and we had more stoma associated morbidity (8.5%).

Colostomy was opened in 59 /74(79.7 %) patients of which 5/59(8.5%) had colostomy associated complications including prolapse, requiring reoperations and refashioning 2/59(3.3%), complete wound dehiscence 1/59 (1.6 %) and parastomal herniation treated conservatively 3/59 (5 %)

In an overview of ARM management in Africa (2019) Colostomy for ARM may be associated with complications in 12.9–78.9% of African children. The most commonly reported complications following colostomy for ARM are prolapse, skin excoriations and haemorrhage. (33)

In Kenya among the 31 patients who had colostomy, four (10%) patients developed colostomy complications: 1(2.4%) had prolapse and three (7.4%) had retraction of the colostomy. (39) When compared to this in our study we have relatively low stoma associated complications this might be because of an increased experience and high number of patients operated in this centre.

In study done Malaysia , UKM Medical Centre, Kuala Lumpur, Three (3/25) 12 % patients developed complications two patients with a single complication, that is, one developed prolapsed stoma and another had retracted stoma, and the third patient had multiple complications, including, bleeding, intestinal obstruction requiring laparotomy, and parastomal herniation.

In the same study the overall interval from creation of stoma to its closure ranged from 4 to 34.2 months. Mean duration taken to complete all three stages of repair was 11.4 months, that is, almost 1 year which is almost similar to our study where 31/59 (52.5%) patients waited for their stoma to be closed for more than a year . (43)

In this study early perineal postoperative complications were seen in 9/74 (12.2 %) of which Perineal body breakdown, neoanus intact 4/74 (5.6 %), perineal wound infections 3/74 (4.1%), Perineal body breakdown, neoanus retracted 2/74 (2.7%). The major (Perineal body breakdown and Perineal body breakdown with retracted neoanus) complications were managed with early primary repair 4/6 (66.7%), With delayed repair with covering stoma 1/6(16.7 %) and local wound care 1/6(16.7 %).

Study done in Pakistan children's Hospital & the Institute of Child Health, , were 37 patients enrolled showed amongst the 30 patients of primary ASARP minimal surgical site wound infection occurred in 3 patients (10%) of primary ASARP which responded to conservative measures. Major wound infection with complete disruption requiring colostomy followed by redo ASARP occurred in 2 patients (5% of primary ASARP) Out of those 8 patients with previous colostomy only 2 patients had minimal wound infection. This paper result is almost comparable to our study. (36)

China Minor wound infection occurred in 3 patients (3/26, 11.5 %) in postoperative period (37)

In this study When we see the durations anal dilatations overall and before closure of the colostomy 51/74 (68.9 %) undergone dilatations for more than 9 month, 13/74 (17.6) for 6 to 9 month and only 10/74(13.5 %) for 3 to 6 month, The neo anus stricture rate in this study is found to be 17.6 % (13/74) of which 7 (53.8%) were managed with dilatations under General anaesthesia and 6(46.2 %) managed with redo anoplasty .

From patients with neo-anus stenosis most 7/13 (53.8%) had three stage ASARP, 4/13(30.7%) had two staged ASARP and 2/13(15.4 %) had single staged ASARP, the proportions of neoanus stricture within specific type of operation were 13.3 %,14.8 %,and 21.9 % for single , two and three staged ASARP respectively.

Pakistan: out of 37 ARM with RVF operated cases there were 2 (5%) cases of anal stenosis found in the period of follow up. (36)

In the Egyptian study 465/596 (95%) had normal caliber and only 30(5%) had neo anal stenosis of which 23 of them were successfully managed with regular anal dilatations and the rest 7 were managed with redo anoplasty. (38).

In Prospective study undertaken in India from 1992 -2017 On 157 pts of which 146 were VF 85%,undergone primary ASARP early comps 5.09% and Anal stenosis was only seen 5 patients (2 managed with anal dilatations, 3 with redo anoplasty) Bowel function good in all except one sever soiling and perineal excoriations (44)

Comparing our result with the above papers we do have relatively higher rates of neo anal stricture and high number of redo-anoplasty for the anal stricture cases

In our study Krickenbeck bowel function scoring tool was used for patients older than 3 years. It's considered it a good outcome when the patients were free from soiling or suffer from grade 1 soiling only, and a poor outcome for those presenting with grade 2 or 3 soiling. Accordingly of 26 participants responded to phone call for bowel function assessment and 25/26 (96.5%) responded to have voluntary bowel movement, 1/26(3.8 %) don't have voluntary bowel movement. 2/26 (7 %) had Grade 1 soiling and 1/26 (3.8%) have constipation managed with diet. Overall bowel function score is good in 96.2%.

Similarly in the Chinese study which have used similar scoring tool shows none of the patients was classified as a poor outcome. Three patients (3/26, 11.5 %) had soiling once or twice per week. Four patients (4/26, 15.4 %) had constipation which was managed by changes of diet. (37)

In Egypt were patient followed over a longer period of time and on the first post-operative year 41/596 (6.9%) developed constipations which managed with anal dilatations and laxatives. In this study they have found those with staged surgery had significantly higher rates of constipations (P=0.009) than those who undergone single staged ASARP. When compared to our paper we haven't identified statistically significant associations between the rate of constipations and the type of surgery patients undergone. (38)

In Japan Retrospective study undertaken to asses functional outcomes of patients with perineal and vestibular fistula treated by ASARP on 28 patients was undertaken and 6/28 had ARM with RVF all are assed using Krekenbecks bowel function tool and they found, Of the 6 pts 1 continent with G1 soiling, 1 G2 constipations And 2 G3 constipations. . (45) This paper showed relatively higher constipation rate than our study

In Indian study were 94 patients with RVF assessed, 54 were toilet trained of which 53/54(98.5%) were continent with voluntary bowel movement and 1/54(1.85%) was incontinent. This study shows almost similar result to our paper. (41)

5.2 Strength of the study

This study used standardized Krickenbeck classification, bowel function tool to assess the functional status and this might help researchers to see the gap where other research is needed.

5.3 Limitation of the study

The outcomes of this study cannot be generalized to all ARM with RVF patient outside of the study area since both study areas are referral tertiary hospitals with high burden of cases.

The sample size is small to run a logistic regression to test the relationship between categorical variables

5.4 Conclusion:

In this study we do not found statistically significant associations between the type of operations and the occurrence of Post-operative early wound complications or anal stricture But the overall frequency of early post-operative complications more in two staged ASARP (14.8%) and anal stenosis is observed in the Three staged ASARP group more frequently (21%) . This study also found lower rates of screening for associated anomalies and higher rate of postoperative neo anal stricture (17.6%). Majority studied patient have an excellent bowel function (96.2%).

5.5 Recommendations

We recommend further study with a lager sample size to run a logistic regression test to see if there is statistically significant associations between the types of surgeries patients undergone and the development of early post-operative complications and anal stenosis.

We recommend also separate study of the anal dilatation regimen and family adherence to the anal dilatation schedule.

Reference list

1. Peña A, Hong A. Advances in the management of anorectal malformations. Am J Surg 2000; 180: 370-376.
2. Rintala R, Lindahl H, Louhimo I. Anorectal malformations – results of treatment and long-term follow-up of 208 patients. Pediatr Surg Int 1991; 6: 36-41
3. Rintala RJ. Anorectal malformations – management and outcome. Semin Neonatol 1996; 1: 219-230.
4. Rintala RJ. Congenital anorectal malformations: anything new? J Pediatr Gastroenterol Nutr 2009; 48: S79-S82.
5. Rintala RJ, Pakarinen MP. Imperforate anus: long- and short-term outcome. Semin Pediatr Surg 2008; 17: 79-89.
6. Iwai N, Fumino S. Surgical treatment of anorectal malformations. Surg Today 2013; 43: 955-962.
7. Goyal A, Williams JM, Kenny SE, Lwin R, Baillie CT, Lamont GL, Turnock RR. Functional outcome and quality of life in anorectal malformations. J Pediatr Surg 2006; 41: 318-322.
8. Ditesheim JA, Templeton JM. Short-term v long-term quality of life in children following repair of high imperforate anus. J Pediatr Surg 2000; 35: 1218-1221.
9. Bai Y, Yuan Z, Wang W, Zhao Y, Wang H, Wang W. Quality of life for children with fecal incontinence after surgically corrected anorectal malformation. J Pediatr Surg 2000; 35: 462-464.
10. De Vries PA, Peña A. Posterior sagittal anorectoplasty. J Pediatr Surg 1982; 17: 638643.
11. Okada A, Tamada H, Tsuji H, Azuma T, Yagi M, Kubota A, Kamata S. Anterior sagittal anorectoplasty for rectovestibular and anovestibular fistula. J Pediatr Surg 1992; 27: 85-88.
12. Stephens FD. Imperforate rectum; a new surgical technique. Med J Aust 1953; 1: 202-203.
13. Rhoads JE, Pipes RL, Randall JP. A simultaneous abdominal and perineal approach in operations for imperforate anus with atresia of the rectum and rectosigmoid. Ann Surg 1948;127: 552.
14. Rehbein F. Operations der anal – und rectumatresie mit recto-urethrafistal. Chirurgie 1959; 30; 491-494.
15. Kieswetter WB, Turner CR. Continence after surgery for imperforate anus: a critical analysis and preliminary experience with the sacro-perineal pull-through. Ann Surg 1963; 158: 498-512.
16. Davies MC, Liao L-M, Wilcox DT, Woodhouse CRJ, Creighton SM. Anorectal malformations: what happens in adulthood? BJU International 2009; 106: 398-404.

17 Cuschieri A (2001) Descriptive epidemiology of isolated anal anomalies: a survey of 4.6 million births in Europe. Am J Med Genet 103:207–215
18 (Management of Anorectal Malformation: Experience from Ethiopia Maru Gama1, Amezene Tadesse2 1. Saint Paul Millennium Medical College 2. School of Medicine, Addis Ababa University
19 Skandalakis JE, Gray SW, Ricketts R (1994) The colon and rectum. In: Skandalakis JE, Gray SW (eds) Embryology for Surgeons, 2nd edn. Williams and Wilkins, Baltimore, pp. 242–281
20 Cuschieri A (2002) Anorectal anomalies associated with or as part of other anomalies. Am J Med Genet 110:122–130
21 Kulshrestha S, Kulshrestha M, Sing B, Sarkar B, Chandra M, Gangapadhyay AN. Anterior sagittal anorectoplasty for anovestibular fistula. Pediatr Surg Int 2007; 23:1191–1197.
22 Endo M, Hayashi A, Ishihara M, Maie M, Nagasaki A, Nishi T, Saeki M (1999) Analysis of 1,992 patients with anorectal malformations over the past two decades in Japan. Steering Committee of Japanese Study Group of Anorectal Anomalies. J Pediatr Surg 34:435–441Louw JH (1959) Malformations of the anus and rectum: a report on 85 consecutive cases. S Afr Med J 33:874–881
23 Smith ED, Saeki M (1988) Associated anomalies. Birth Defects Orig Artic Ser 24:501–549
24 Smith ED (1988) Incidence, frequency of types, and etiology of anorectal malformations. Birth Defects Orig Artic Ser 24:231–246
25 Chaterjee SK. Lesions in the wingspread list management in the neonatal period. In: Chaterjee SK, editor. Anorectal malformations-A surgeon experience chap 8. New Delhi, India, Oxford University; 1991. p. 48-64..)
26 Smith ED. The bath water needs changing, but don't throw out the baby: an overview of anorectal anomalies. J Pediatr Surg 1987;22: 335–48.
27 Yazbeck S, Lucks FI, St-vil D. Anterior perineal approach and threeflap anorectoplasty for imperforate anus: optimal reconstruction with minimal destruction. J Pediatr Surg 1992;27:190–5.
28 Sigalet DL, Laberge JM, Adolph VR, Guttman FM. The anterior sagittal approach for high imperforate anus: a simplification of the Mollard approach. J Pediatr Surg 1996;22:335–48)
29 Pena A, Devries PA (1982) Posterior sagittal anorectoplasty: important technical considerations and new applications. J Pediatr Surg 17(6):796–811 2.
30 Wakhlu A, Pandey A, Prasad A, et al. Anterior sagittal anorectoplasty for anorectal malformations and perineal trauma in the female child. J Pediatr Surg. 1996;31(9):1236–1240.
31 Shehata SM. Prospective long-term functional and cosmetic results of ASARP versus PSARP in treatment of intermediate anorectal malformations in girls. Pediatr Surg Int. 2009;25:863–888.
32 Rangel SJ, de Blaauw I. Advances in pediatric colorectal surgical techniques. Semin Pediatr Surg. 2010;19(2):86–95.
33 Overview of Anorectal Malformations in Africa Taiwo A. Lawal)

34 Adejuyigbe O, Abubakar AM, Sowande OA, Olayinka OS, Uba AF. Experience with anorectal malformations in Ile-Ife, Nigeria. Pediatr Surg Int. (2004) 20:855–8. doi: 10.1007/s00383-004-1297-1

35 Anna Morandi1 • Benno Ure2 • Ernesto Leva1 • Martin Lacher2, Survey on the management of anorectal malformations (ARM) in European pediatric surgical centers of excellence)

36 The Anterior Sagittal Anorectoplasty Technique (ASARP) for Treatment of Female Anorectal Malformations: Our Experience MUHAMMAD AFZAL, NABILA TALAT, ARSALAN RAZA WASTI, Sarfraz Ahmed, Muhammad Bilal Mirza, Muhammad Saleem -- Pak Pediatr J 2017; 41(3): 153-57)

37 Wang C, Li L, Liu S, Chen Z, Diao M, Li X, et al. The management of anorectal malformation with congenital vestibular fistula: a single-stage modified anterior sagittal anorectoplasty. Pediatr Surg Int 2015; 31:809–814.

38 Anterior sagittal anorectoplasty as a technique for the repair of female anorectal malformations: A twenty two-years-single-center experience Ahmed Elrouby* , Saber Waheeb, Ahmed Koraitim)

39 PATTERN OF ANORECTAL MALFORMATIONS AND EARLY OUTCOMES OF MANAGEMENT AT MOI TEACHING AND REFERRAL HOSPITAL ELDORET- KENYAP. KURADUSENGE, R. T. KUREMU, G. JUMBI and P. W. SAULA

40 Primary Anterior Sagittal Anorectoplasty for Rectovestibular Fistula: Md. Abdul Aziz, Tahmina Banu,1 Ramanandan Prasad and Abdur Rashed Khan, Department of Paediatric Surgery, Dhaka Shishu (Children) Hospital and Bangladesh Institute of Child Health, Dhaka, and 1Department of Paediatric Surgery, Chittagong Medical College and Hospital, Chittagong, Bangladesh: Asian J Surg 2006;29(1):22–4]

41 Single stage repair of vestibular and perineal fistula with out colostomy , Basant Kumar,Deepark kandp,Shyam B.Sharma Rajasthan ,India ,journal of peadiatric surgery 2008 43 1848-1852

42 Current management of anorectal malformation in Egypt: a survey of members of the Egyptian pediatric surgical association Nezar A. Abo Halawaa, Abdelghany E.M. Abdelgawadb and Essam A. Elhalabyb Ann Pediatr Surg 13:203–206 c 2017

43 Anorectal anomaly with rectovestibular fistula: a historical comparison of neonatal anterior sagittal anorectoplasty without covering colostomy and postoperative anal dilatation to the classical three-stage posterior sagittal anorectoplasty, Dayang Anita Abdul Aziz1 Ramamoorthy Velayutham2 Marjmin Osman1 Zarina Abdul Latiff3 Felicia SK Lim4 Mahmud Mohd Nor1 , Malaysia 2017

44 Ram babu goyal , Rahul Gupita ,Girish Prabhkar , Monika Bawa , journal of indian associatins of pediatric surgery 25(3),134 ,2020 ,ASARP experience Prospective study from 1992 -2017

45 Functional asst of pts with perineal and vestibular fistula treared by ASARP , AJPS 15(1),36,2018 Naoki hashizume , Kimio Asagiri ,Suguru fukahori, shinii,nobuvuki saikusa Retrospective study.

46