Ghana College of Physicians and Surgeons

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TYPES, PRESENTATION AND CHALLENGES WITH THE MANAGEMENT OF ANORECTAL MALFORMATIONS AT KOMFO ANOKYE TEACHING HOSPITAL, KUMASI, GHANA.

A Dissertation presented to the Faculty of Surgery (Paediatric) in partial fulfilment of the requirement for the conferment of a Fellow of the Ghana College of Surgeons (FGCS)

BY

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DECLARATION

I declare that this work was written independently of any unauthorized help, and the use of secondary material has been duly acknowledged. I confirm that this dissertation is wholly mine and has not been part of any presentation for any other qualification in its present form or a similar version.

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ATTESTATION BY SUPERVISORS

We, the undersigned, attest that this dissertation titled "TYPES, PRESENTATION AND CHALLENGES WITH THE MANAGEMENT OF ANORECTAL MALFORMATION AT KOMFO ANOKYE TEACHING HOSPITAL, KUMASI, GHANA" was carried out by Dr. Fareeda Agyei under our supervision.

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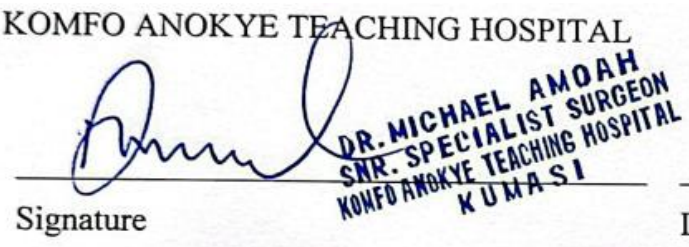

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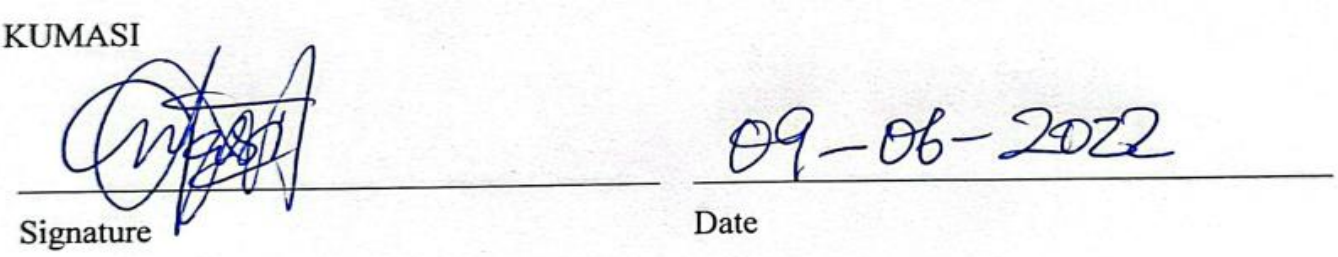

DEDICATION

This study is dedicated to Almighty God, the builder and lifter of men, as well as my husband and children.

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ABSTRACT

Introduction: Anorectal malformations remain a significant challenge to the child and the paediatric surgeon. Clinical outcomes depend on the surgeon's skill, the type of anorectal malformation, associated anomalies and the availability of perioperative and rehabilitation facilities. Anorectal malformation is rare, occurring in about 2-6 per 10000 live births. However, its rarity becomes relative in Sub-Saharan Africa, where few surgeons are paediatric surgeons, and only a handful of these few surgeons handle these cases. The burden of treating these patients falls on the few paediatric surgeons available in the region.

The paediatric surgeon is faced with challenging clinical scenarios; some children present with complications, either as a result of late presentation or from the initial surgery, done mainly by a local surgeon as a lifesaving procedure.

Understanding the challenges the patient, the parents and caregivers, and the paediatric surgeon face in this resource-limited setting is essential.

Aim: The aim of the study was to assess the types, modes of presentation, the challenges carers of patients with anorectal malformation encounter during management and the cost implication in a resource-challenged tertiary teaching hospital, Komfo Anokye Teaching (KATH), in Kumasi, Ghana.

Methods: The study was a cross-sectional descriptive study carried out on Anorectal malformations at the Paediatric Surgery Unit of Komfo Anokye Teaching Hospital (KATH) over a period of eight months. It employed both qualitative and quantitative study design which was taken at the same time period; a concurrent mixed methodology. Data on all children with anorectal malformation being managed by the Paediatric Surgery Unit of the Department of Surgery, KATH, were obtained. The ARM types, presentation, associated anomalies, surgical interventions, peri operative challenges; challenges with managing and taking care of a child with anorectal condition, and the cost of managing the condition were captured using a structured questionnaire (quantitative study). In-depth interviews of guardians of the children with anorectal malformations who were being managed were also conducted (qualitative study).

Results: A total of 61 patients (male: female ratio 1:0.96) were included in the study. A majority, 35 (57.38%), of the patients with anorectal malformations were observed between days 2 and 5. The average birth weight was 3.12kg (SD =0.51). Most of the patients, 88.52% reported late (> 24hrs). Majority of the patients 55.74% presented in a stable state at the emergency department. Acute intestinal obstruction accounted for 32.79% of presentations. All the 61 patients with anorectal malformation underwent a staged procedure. The majority, 60 (98.63%) out of the 61 patients,

underwent a three-staged procedure; an initial colostomy, definitive repair, and colostomy closure. Only 1 patient underwent a two-stage procedure in which colostomy + PSARP was done on day 1 of presentation. Closure of colostomy was done 14 months later. The definitive repair for all the children who underwent the three-staged procedure was PSARP. For this study, 45 (75%) had had PSARP, and 29 (48.33%) had closure of colostomy done at the end of the study. Overall complication after all surgeries was 39.3%. There was no significant association between, birth weight (p-value = 0.596), the presence of a fistula (p-value = 0.061), when the malformation was detected (p-value = 0.349) and time of presentation to KATH, (p-value = 0.306).

Overall Complications at the initial colostomy, PSARP and colostomy closure were 21.6%, 26.7%, and 27.59%, respectively. The mean duration between a colostomy and PSARP was 7.02 +/- 2.05 months, while between PSARP and colostomy closure was eight months. The mortality rate at the end of the study was 3.3%. The length of stay in the hospital was 11.89 +/- 8.93 days, 29.95 +/- 9.26 days, and 17.51+/- 7.0 days for the initial colostomy, PSARP and the closure of colostomy respectively.

The mean cost for the complete three-staged procedure was GHC11604.93 ($2053.90) (1$ = GHC5.65), with out-of-pocket payments accounting for 61.91% of the total cost. 44.3% of the caregivers rated the cost as moderate.

Findings from the qualitative study indicated that most caregivers lacked adequate knowledge regarding anorectal malformations. Most respondents highlighted the stigma associated with having a child with ARM and having a colostomy. Rescheduling surgeries, financial constraints, increased out-of-pocket payments, and lack of psychological support were some challenges caregivers had with managing anorectal malformations.

Conclusion: Anorectal malformation is challenging to the paediatric surgeon, the patient and the carer. There is a general lack of awareness about the condition among health professionals and carers. The stigma attached to having a child with colostomy is rife and it is associated with a lot of psychological trauma to the carers and family members. The cost of managing anorectal malformation is high and steps must be taken to help carers pay for the cost. Improving public awareness of the condition and our referral system, providing psychological support, training more stoma therapists, increasing the partial amount the NHIS pays for this condition will all go a long way to improve the management and the lives of children born with this congenital abnormality.

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LIST OF ABBREVIATIONS

ARM	Anorectal Malformation
ICU	Intensive Care Unit
KATH	Komfo Anokye Teaching Hospital
KUB	Kidney, Ureter Bladder
MBU	Mother and Baby Unit
NHIS	National Health Insurance Scheme
NICU	Neonatal Intensive Care Unit
PEU	Paediatric Emergency Unit
PICU	Paediatric Intensive Care Unit
PSARP	Posterior Sagittal Anorectoplasty

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CHAPTER ONE

INTRODUCTION

1.1 Background to the Study

Anorectal malformations (ARM) constitute a spectrum of rare but significant congenital gastrointestinal defects. They occur in 2-6 per 10000 live births.[1] It consists of defects involving children's anus, rectum and the urogenital system. Anorectal malformations may present as an isolated anomaly, associated with other congenital malformations, or as part of a syndrome. There are variable presentations. It could be an incidental finding on a routine inspection of neonates at birth. Others may present with associated sepsis or intestinal obstruction. Notably, ARM is one of the African children's primary causes of intestinal obstruction.[23] Although ARM is considered rare, its management constitutes a significant medical, socio-economic and psychological challenge for the child and the guardians.[3,4,5] ARM management requires a multidisciplinary approach. Highly specialised surgical and nursing care, as well as prolonged rehabilitation is needed for most patients.

1.2 Problem Statement

As much as the presentation is variable, the management outcome is also variable, depending not only on the type of ARM and associated anomalies but also on the availability of functional perioperative and operative facilities. Late presentation has been noted in Sub-Saharan Africa. (6,7)[7] Observably, Adejuyibe et al.[6] noted that 86% of children presented after 24hrs, with 60% presenting with abdominal distension. Reports have attributed this to the fact that 94.8% of children's births occur outside the hospital in Africa.[5] Delivery outside the hospital has been significantly linked to delayed presentation and diagnosis of ARM.[5,7]

Surgical management usually depends on the type of anomaly, associated anomalies and availability of skilled surgeons for such surgeries. In Low-and-middle-income countries (LMIC), paediatric surgeons face the enormous task of managing these challenging clinical scenarios with the few resources available.[3] This often leads to adopting modified strategies to initial workup and surgical correction of ARM.

1.3 Justification of the Study

Like other Sub-Saharan countries, most of the healthcare centres in Ghana have limited resources and few surgeons with the expertise and knowledge in the Management of ARM. KATH is the only tertiary hospital serving the whole Northern sector of Ghana, receiving most of the referrals of ARM.

In addition to this apparent lack of skilled personnel at the peripheral hospitals, some cultural and traditional beliefs often delay or prevent parents from seeking specialised management at the tertiary facilities where limited paediatric surgeons are found. Some of them present with complications such as intestinal obstruction and sepsis.[8]

The type and associated anomalies affect the surgical intervention, prognosis, and quality of life. Even with surgical interventions, the sequelae of ARM such as faecal incontinence, sexual dysfunction and constipation may continue into adulthood. Therefore, it is imperative to have a holistic picture of the presentation and Management of ARM. Studies[4,9,10] have intimated that parents and children who have ARM have to deal with psychological stress, especially with their colostomies and faecal incontinence. This study is warranted with the paucity of data on types, presentation, and initial ARM management in Ghana. This will help fill data gaps and may help review the approach to the management of the children with ARM.

1.4 Aims and Objectives

1.4.1 Main objectives

The aim of the study was to assess the types, modes of presentation, challenges carers and mothers encounter during the management of anorectal malformation, and the cost of managing the condition in a resource-challenged tertiary teaching hospital such as Komfo Anokye Teaching (KATH) in Kumasi, Ghana.

The study also aimed at identifying the factors contributing to the delay in presentation in patients with anorectal malformation.

1.4.2 Specific Objectives

The general objective was achieved by addressing the following objectives:

1. To determine the average presentation time of an ARM at KATH and the cause of the delay in the presentation.
2. To determine complications associated with creating colostomies.
3. To identify the unique challenges the carers face in managing a child with anorectal malformation.
4. To determine the length of time patients live with their colostomies and the factors contributing to the delay in reversing colostomies.
5. To determine the cost of managing a patient with ARM at KATH

1.4.3 Research Questions

The following research questions were answered to achieve the specific objectives

1. What is the average presentation time of an ARM at KATH and what are the causes of the delay in the presentation?
2. What challenges are encountered when colostomies are created for patients with an anorectal malformation?
3. How long do patients with anorectal malformation live with their colostomies. What factors contribute to the delay in reversing colostomies?
4. What is the cost of management of a patient with ARM at KATH?

CHAPTER TWO

REVIEW OF LITERATURE

2.1 Introduction

Anorectal malformations (ARM) are rare congenital birth defects. It affects the gastrointestinal and urological systems of children. The incidence is around 2-6 per 10,000 live births worldwide.[1] they range from mild forms which require minor surgery to complex forms which require multi-staged approaches.

Variations exist in geographical incidence and apparent ethnic disparity of ARM prevalence, associated anomalies and types.[11,12] Louw et al[11] reported a higher incidence of 5.5/10,000 live births in South Africa. This prevalence is higher than the prevalence worldwide. Anorectal malformations are more common in boys than in girls, with males tending to have more severe malformations than females.[11]

There is a spectrum of presentation, ranging from low anomalies with relatively simple management to higher anomalies requiring complex surgical management. It has been postulated that abnormal embryonic development of the distal end of the digestive tract between the sixth- and tenth weeks of gestation results in anorectal malformations.

The distal enteric component may end blindly or form a fistula between the rectum and the urinary tract, genital tract or perineum.[13] these children are born with an absent anal opening, an abnormal opening in the perineum or a fistulous tract connected to the genito-urinary system.

2.2 Aetiology

The aetiology of ARM remains unclear. Multiple factors have been suggested to play different roles in its development. Genetic and non-genetic factors have been found to contribute to the formation of anorectal malformations in animal models. In humans, first-degree children of parents with anorectal anomalies and patients with chromosomal anomalies have a high risk of developing the condition**.**[14,15,16] Anorectal malformations may be inherited as autosomal recessive or autosomal dominant. Familial cases are also reported.[17] A mother and her children all having anorectal malformation have been reported in some series.[18]The mother and her children all had a normal karyotype and the author found microdeletions that affected family members.[18]

About half of children with anorectal malformations have associated congenital anomalies.[19,14] Isolated anorectal malformations have been associated with CDX1 mRNA gene expression.[20] Genetic studies show down regulation of CDX1 gene expression in anorectal malformation compared to the control group. Other genes implicated are ‘Wnt, BMP4, GLi2 and Hox[21–23]. In patients with VACTERL associations, Hondel et al.[24] concluded that genetic disorders were twice as much as

isolated anorectal malformations. Mutation in the EFNB-2 gene has been suggested.[25] Neonates with chromosomal abnormalities like Down syndrome, trisomy 18, and Fragile X syndromes tend to have an ARM component.[26–28] Down syndrome constitutes about 40% of chromosomal anomalies. (28) non-chromosomal syndromes such as FG syndrome[29] and Towne Brookes syndrome[30] have been associated.

There is little evidence of the effect of drugs on the development of anorectal malformations. Some reports implicate thalidomide[31]. The induction of trans-retinoic acid has been suggested to cause anorectal malformations by interfering with normal caudal migration.[32] In Yu et al.[32] review, 94% of animal models had anorectal malformation after administering retinoic acid. Pre-conception folic acid intake has been weakly linked to the reduction in congenital malformation because of its role in the reduction of retinoic acid.[33] Significant statistical association has been demonstrated between benzodiazepines and anal atresia in mothers exposed to them.[34]

Infectious agents' effect on the development of anorectal malformation is equivocal. Cytomegalovirus is reported as a possible aetiology.[35]

Prenatal exposures of mothers in the first trimester to tobacco and alcohol, as well as maternal obesity and diabetes mellitus, have been suggested to affect the development of anorectal malformations.[35] Irradiation and electromagnetic exposures have been challenging to prove to affect the embryo in first trimester.[16]

2.3 Embryology

The embryology of the development of ARM has been speculative among investigators and paediatric surgeons**.** It was generally accepted that abnormal septal development of the cloaca resulted in ARM.[36] However, there was no consensus on the development of this septum. Bill and Johnson[37] postulated that it was due to an ectopic anal opening regarding the formation of fistulas in ARM. This theory suggested the migration of the rectum from a higher position to the average area of the anal opening.

However, in 1995, Kluth et al.[38], using scanning electron microscopy in rat embryos, could not observe this phenomenon. They observed a region of the future anal orifice in the dorsal part of the cloacal membrane. A recent study of human and animal embryos with a high incidence of ARM has shown that abnormal development of the dorsal cloaca resulted in ARM.[39] Smaller defects in the dorsal cloaca lead to distal defects, any cutaneous fistulas and a covered anus. The larger defects are associated with significant malformations and higher anomalies[40]

2.4 Incidence and frequency of Anorectal malformation

There is significant geographical variation in the incidence and prevalence of ARM. No significant association has been identified between ethnicity and ARM, although the disparity exists.[41] In addition to ethnic and geographical variations, inter-registry variations have been reported between regions (between 1.14-5.96 per 10,000).[11,41]True incidences are difficult to obtain in Sub-Saharan Africa as most countries have no formal birth registries. Estimates are made from hospital-based records. Moore et al.[12] noted a higher clinical load of ARM in Africa.

A wide diversity of associated anomalies has been associated with ARM. 2% of children with trisomy 21 have an ARM (between 2-8%).[41,42] other syndromes associated with ARM include Goldenhar and Di-George syndromes, Frazer's syndrome and Pfeiffer's syndrome and VACTERL (Vertebral, Anorectal, Cardiac, Tracheo-oesophageal, Renal and Limb) group of anomalies. 14% of ARM have vertebral anomalies, oesophageal anomalies (13%) and cardiac anomalies (13%) in Moore's[12] review of ARM in selected countries in Africa. Genitourinary anomalies occur between 20-54% of patients[43,44]. Higher ARM present with more associated anomalies and have a poorer prognosis.[45]

2.5 Classification of anorectal malformations

The classification of ARM has evolved throughout the centuries. Various systems and protocols have been developed to help with successful management. Stephens F.D developed a high and low classification in 1963. It recognized the importance of the pubococcygeal (PC) line and associated muscles in maintaining continence. Lesions above the PC line were designated high and those below as low. In 1970, the international classification was proposed based on Stephen and Smith's early work on ARM. It was later refined in the early 1980s at the Wingspread conference.[46] Lesions were reclassified into high, intermediate and low anomalies as shown in Table 1.

Table 2.1 Wingspread Conference classification of anorectal malformation

Level of anomaly	Male	Female
High	1. Anorectal agenesis a. Rectovesical fistula b. Without fistula 2. Rectal atresia	1. Anorectal agenesis a. Rectovaginal fistula b. Without fistula 2. Rectal atresia
Intermediate	1. Rectobulbar urethral fistula 2. Anal agenesis without fistula	1. Rectovestibular fistula 2. Rectovaginal fistula 3. Anal agenesis without fistula
Low	1. Anocutaneous(perineal) fistula 2. Anal stenosis	1. Anovestibular fistula 2. Anocutaneous (perineal) fistula 3. Anal stenosis
Miscellaneous	Rare malformations	Persistent cloacal anomaly Rare malformations

Source: Hutson JM, Holschneider AM. Anorectal Malformations in Children: Embryology, Diagnosis, Surgical Treatment, Follow-up. Berlin: Springer; 2006. p.178.

After the advent and adoption of posterior sagittal anorectoplasty (PSARP), Pena proposed a new classification considering the fistula's presence and position. (47) This is shown in Table 2.

In 2005, the Krickenbeck classification was introduced. It incorporated both Pena and Wingspread classification systems. In addition, a category was made for documenting functional outcome criteria. It gave equal emphasis to both constipation and soiling.[48] Table 3 show the classification after the Krickenbeck conference.

Table 2.2 Pena Classification of anorectal malformations

NON-SYNDROMIC ARM		
A non-syndromic ARM with fistula	Recto-perineal malformations	
	Imperforate anus with recto-urethral Fistula	Recto-urethral bulbar fistula Recto-urethral prostatic fistula Bladder neck fistula
	Imperforate anus in female	Recto-vestibular fistula Recto-vaginal fistula Cloacal malformation
Non-syndromic ARM without Fistula	Imperforate anus without fistula	Cloacal malformations with a short common channel (< 3 cm) Cloacal malformations with a long common channel (> 3 cm) H-shaped fistula (rectovaginal) Rectal duplication
SYNDROMIC ARM		
VACTERL	Axial mesodermal dysplasia	Klippel-Feil syndrome
MURCS (Mullerian duct aplasia, renal aplasia, -and cervicothoracic somite dysplasia)	OEIS (Omphalocele, exstrophy, imperforate anus, and spinal defects)	Sirenomelia-caudal regression
Trisomy 21,13,18	Pallister-Killian syndrome	Currarino syndrome
MIDAS syndrome	Cat-eye syndrome	Pallister-Hall syndrome
Christian syndrome	Parental unidisomy 16	Ulnar-mammary syndrome
Deletion 22q11 syndrome (del22q11.2)	Townes-Brock syndrome	Johanson-Blizzard syndrome
Okihiro syndrome	Hirschsprung disease	Heterotaxia
Rieger syndrome	Feingold syndrome	FG syndrome
Thanatophoric dwarfism	Kabuki syndrome	Short rib – polydactyly syndrome
Opitz BBB/G syndrome	Spondylocostal dysostosis	Baller-Gerald syndrome
Ciliopathies	Fraser syndrome	Lowe syndrome
X-linked mental retardation		

Source: Levitt MA, Peña A. Anorectal malformations. Orphanet J Rare Dis. 2007; 2:33.

Table 2. 3 Standards for Diagnostic Procedures: International Classification (Krickenbeck) for anorectal malformations.

Major clinical groups	a. Perineal (cutaneous) fistula b. Rectourethral fistula i. Bulbar ii. Prostatic c. Rectovesical fistula d. Vestibular fistula e. Cloaca f. No fistula g. Anal stenosis
Rare/regional variants	a. Pouch Colon b. Rectal atresia/stenosis c. Rectovaginal fistula d. H type fistula e. Others

Source: Hutson JM, Holschneider AM. Anorectal Malformations in Children: Embryology, Diagnosis, Surgical Treatment, Follow-up. Berlin: Springer; 2006. p. 181.

2.6Associated Anomalies

Associated anomalies may present individually or as a component of the VACTERL association. Genito-urinary associations are seen in 20-80% of anorectal malformations.[10,49] Unilateral agenesis and hydro nephrosis have been reported.[49]

Cardiac anomalies can be identified on routine examination. 32 of the 86 children in a study by Nour et al.[50] had cardiac anomalies. They found that 40% had significant heart defects, and 28% had minor heart anomalies. These anomalies alter the course of management of these patients. Cardiac anomalies are relatively low in Africa.[51] This is explained by the low screening done for anomalies. The most common heart anomalies are atrial septal defect, patent ductus arteriosus, and ventricular septal defects.[52]

Associated gastrointestinal anomalies like intestinal atresia are relatively uncommon.[17] Oesophageal atresia and tracheoesophageal fistula, a component of VACTERL association is fairly common and occasionally , gastrointestinal duplications[53] and Hirschsprung disease can occur.[54]

Spinal cord anomalies have been reported in up to 46% of children with anorectal malformations.[17]Ultrasonography raises the suspicion of an abnormality an MRI can be used to substantiate or confirm the diagnosis. Diagnosis occurs when the conus medullaris is located below L2-L3 or when there are other spinal anomalies.[55] Spinal cord anomalies affect the functional outcomes after definitive repair[56]. Low cases are reported in Africa. [7] Vertebral anomalies occur in 60-90%.[57,58] Bony vertebrae are usually screened in studies. The Common bony and spinal cord anomalies are tethered cord and sacral bone anomalies. Tethered cord is present in 9-42% of children with anorectal malformation.[59–63] Tethered cord is more common in complex anorectal malformations. Samuk et al. noted tethered cord in 100% of vaginal fistulas, 83.3% of cloacal exstrophy, 27.1% of bulbar fistula and 21.1% of perineal fistulas. Tethered cord, if left unresolved, may lead to tethered cord syndrome with subsequent neurological deterioration and bladder dysfunction.[59,64]

More than half of children having VACTERL associations in ARM have tracheoesophageal anomalies.[65,66] some of the anomalies can be detected during antenatal screening. MRI is highly predictive but not cost-effective.[67].

2.7 Identification and Presentation of Anorectal Malformation

2.7.1 Prenatal screening

Prenatal screening of congenital malformations has gained attention in recent times. Ultrasonography is the most common non-invasive screening modality for detecting pathologies.[68]

Despite its extensive usage, anorectal malformations are mostly diagnosed at birth, with only 16% diagnosed by ultrasound.[52,69] Signs that suggest anorectal malformation can be demonstrated around the 23rd week of pregnancy.[70–73] MRI can be used as a complementary imaging modality.[74] The foremost ultra-sonographic finding suggests ARM is the absence of target around 23 week's gestation.[65,71] Other indirect signs include detecting other malformations such as rectal or bowel distension.[74,75]

Accuracy rates are low for isolated anorectal malformation compared to high anorectal malformation or anorectal malformation with other associated anomalies. Rohrer et al. observed that only 3% of a low type of anorectal malformation were detected via prenatal screening compared to 39% of intermediate to the high type of anorectal malformations.[68] In contrast, Lee et al.[76] had diagnostic accuracy rates of 91% in high and intermediate types of ARM in their series.

2.7.2. Identification of anorectal malformation

Most anorectal malformations are identified clinically.[77] Identification remains crucial as it has been associated with early reportage for management.[77] Not uncommonly, defects are missed at birth, and

the baby presents days or weeks after birth[78]. The mother may take the baby home, and the child may develop an intestinal obstruction, or in some cases develop sepsis, because of translocation of bacteria.[9,79] Reasons for non-identification of anorectal malformation include lack of awareness, false assurance of the presence of structure that allows passage of meconium, and poor skills of midwives in examining babies at birth.[77,80] Lawal et al.[77] observed that only 19.5% of mothers were aware of anorectal malformation as a disease. Late Identification is not only limited to developing countries.[78,80]

2.7.3 Presentation: Neonatal age group

In the neonatal age group, prompt diagnosis and management are encouraged to achieve good outcomes and prevent complications. ARM is the leading cause of neonatal intestinal obstruction causing 57-67% of cases.[13,81] Neonatal intestinal obstruction accounts for 24-64% of African neonatal admissions.[81,82]

The late presentation of neonatal intestinal obstruction due to ARM is a unique challenge. This often leads to patients presenting with sepsis and aspiration pneumonia. In a one-year study of 273 patients with ARM who presented at a tertiary teaching hospital, Govender et al noted that 57.9 % presented 24 hours after birth[5]. In a similar study in Uganda,[83] 63% of patients with ARM presented 48 hours after birth. The delayed presentation was more common in females compared in the male neonates.

Deliveries outside the hospitals are associated with delayed presentation and diagnosis of anorectal malformations. It is therefore up to the most experienced caregiver to identify this anomaly when neonates present with it to the hospital. This is also challenging as mothers or caregivers may not know about this disease[77]. In a study in Nigeria by Lawal et al.[77] only 1 out of 5 mothers could identify anorectal malformations. In addition, much misinformation is given to parents, contributing to delayed presentations.[77]

2.7.4 Presentations outside the neonatal age group

Delay in diagnosis and referrals significantly increase mortality.[5,12] Delay in presentation beyond 24-48 hours in developed countries is unusual, as most deliveries are supervised, and there is strict adherence to neonatal assessment at birth. Deliveries outside the hospital significantly increase the delay in the presentation of ARM.[5] Extreme delay beyond the neonatal period is uncommon except in resource-challenged settings, with some children presenting during the adolescent period.

A case study by Ogundoyin et al.[84] reported a 25-year-old Nigerian woman with ARM and a rectovestibular fistula. She underwent a successful repair after an initial colostomy. The causes of delayed presentation include delayed detection, wrong advice to caregivers, cultural and traditional beliefs, lack of funds, and absence of health professionals trained to handle such cases.[77]

Even in African centres where patients report early, it may take a while before surgeries are performed because of the inadequate systemic support for paediatric surgical services, which often need a multidisciplinary team.

2.8 Diagnosis

A thorough examination of the perineum should be done at birth. Clinical features such as absence or presence of anus, presence of urethral and vaginal opening, presence and size of anal dimple, and presence and type of fistulas should be noted. Accurate determination of the exact type and anomaly level is crucial in the initial management as it guides the decision to do a primary perineal repair or perform a colostomy, deferring definite surgical management. The emphasis on physical examination varies between sexes as there are different clinical types.[3]

Prenatal diagnosis of ARM remains rare. It is even more unlikely in third-world, resource-challenged countries. Prenatal ultrasonography has a low sensitivity and specificity for detecting ARM.[85]

Diagnosis can be suspected when gross foetal anomalies are detected in-utero. Images that can raise visualization, tethered cord and dilated or calcified bowel.[85] Imaging support is also crucial after suspicion include absent kidneys, vertebral anomalies, omphalocele in the absence of bladder birth. Cross-table lateral radiograph should be taken to classify the malformation if there is no fistula. Abdomino-pelvic ultrasound scans should be done to evaluate associated genito-urinary malformations such as renal agenesis, hydronephrosis, fused kidneys and ectopic kidneys. Augmented pressure distal colostogram should be done to delineate the anatomy when a colostomy has been created for the patient. An echocardiogram should be done to evaluate for congenital heart diseases.[85,86]A cloacogram to determine the genito-urinary system and its confluence with the gastrointestinal system in a patient born with anorectal malformation with a persistent cloaca. Radiographs to prognosticate ARM include ultrasound scans , plain X –rays and magnetic resonance imaging to define the anatomy, determine any renal abnormalities, screen for vertebral anomalies, tethered cord and evaluate for other associated malformations.[87]

The absence of a visible fistula or no meconium from the urinary tract after 24 hours necessitates the performance of a cross-table, lateral x-ray. Radiological imaging could be misinterpreted within the first 24 hours as the rectum collapses, and results could be wrongly interpreted as high anomalies.[88] the prone, cross-table, lateral x-ray should be delayed for at least 18-24 hours to allow gas to reach the distal rectum. The gas bubble to the pubococcygeal line defines whether the blind pouch is above or below the attachment of the levator ani to the pelvic wall.[88] Associated anomalies should be identified before undertaking surgical interventions. Echocardiography should be performed to screen for cardiac anomalies. A nasogastric tube should be passed to rule out oesophageal atresia.

2.9 Initial Management

Children born with ARM and intestinal obstruction will ideally be admitted to a neonatal intensive care unit (NICU) or a high care unit for observation to rule out or treat intestinal obstruction, sepsis or aspiration if present and offer general support for patients who come with other associated anomalies e.g. tracheo oesophageal fistula. This will ensure optimal control of fluid and electrolyte losses, hypothermia, the institution of respiratory support if indicated and if need be - parenteral nutrition. This NICU support is mainly unavailable in resource-limited settings. Only 51% of the 37 surveyed hospitals and healthcare facilities in Sub-Saharan Africa have NICU or ICU.[89] The situation becomes dire for those infants who are extremely premature infants and those who have other systemic abnormalities in addition to their malformation.

The paediatric surgeon trains to ensure that the patients born with correctable anomalies survive. However, very few hospitals in West and East Africa have the complement of health care professionals to form a multidisciplinary team comprising of a neonatologists, paediatric intensivists, paediatric surgeons and anaesthesiologists and stoma therapists that is needed to treat and manage patients with ARM. The paediatric surgeon is made to play many different roles. The multidisciplinary approach gives the most optimal care to patients.

Timing of surgery is dependent on factors like the type of ARM, presence or absence of associated congenital abnormalities, the expertise of the surgeon, available skilled personnel to give neonatal anaesthesia, availability of imaging facilities, availability of related specialists, support facilities and the availability of parenteral feeds and NICU.[85,89,90]

In non-emergent cases, the decision about colostomy or primary operation should also be deferred after 24hours of life because significant pressure is required for meconium to be forced through a fistula orifice.[91] The presence of gas above the coccyx, meconium in urine, a flat bottom, gross sacral abnormalities, and significantly associated malformation requires initial colostomy with the postponement of definitive repair for subsequent operation. [91]

2.10 Primary Repair versus Staged Repair

Primary (definitive) repair and staged repair approaches have been described in ARM management. Primary repair is usually done for low types of ARM without the need for a protective diverting colostomy.[92] The main argument for primary stage repair is to avoid the complications of colostomy.[93]

2.11 Repair of Anorectal Malformation

The recommended treatment of high and intermediate types of ARM has been a three-staged procedure involving colostomy after birth followed by definitive surgery, usually a posterior sagittal anorectoplasty (PSARP) and then finally reversal of the colostomy as the last stage.[94]

PSARP is done at approximately 3-6 months of age, and if the child appears in an excellent nutritional status evidenced by a weight of a 5-8 kg.[92] PSARP involves Identification of the distal rectum, separation and ligation of any fistula, mobilization of the rectum, and placing it within the muscle complex and parasagittal fibres of the external sphincter. Improper localization of the distal rectum may lead to faecal incontinence and the need for a re-operation.[47]

Other approaches to definitive surgery include abdominal sacro peritoneal pull-through and anterior sagittal anorectoplasty (ASARP). The objective of ASARP is to preserve the internal sphincter, and it's sometimes used for females with rectovestibular fistulas.[95] Laparoscopy-assisted anorectal pull-through (LAARP) can be carried out but is primarily done in more advanced centres. [96,97,98]

The initial colostomy is done for colon decompression to relieve an intestinal obstruction or eminent intestinal obstruction, control sepsis and reduce the frequency of breakdown of definitive repair.[99] Some surgeons have, however, argued that a single-stage corrective procedure for selected high and intermediate ARM types is technically feasible, safe, and relatively cost-effective and avoids the complications related to colostomies. Primary PSARP was performed by Osifo et al on five selected patients. These patients had rectovestibular fistula, rectourethral fistula and ARM without fistula. All five patients were operated on between the second and seventh days of life and were discharged home eight and ten days after surgery. Elhalaby[100] in Egypt and Adeniran et al.[101] in Nigeria have demonstrated the feasibility of primary PSARP without colostomy in carefully well-selected males with "intermediate" types of malformations. Primary anorectoplasty without colostomy may be performed for the perineal fistula.[94] Primary repair may be done for rectovestibular fistula, depending on the surgeon's confidence and expertise[102,103]

Even though feasible, primary repair in intermediate and high ARM types is a common complication source.[93] Even with experienced surgeons practising in hospitals with good neonatal perioperative support, the wound infection rate may be as high as 26.3%.[104] This is often challenging to manage in most hospitals. These single staged repairs were done in facilities with a full complement of experienced staff. These include paediatric anesthesiologists, neonatal intensivists, neonatal nurses, and stoma therapists.

Diverting colostomy is the most effective way of protecting the pull-through.[90] The type of colostomy chosen depends on the surgeons' training, preference and healthcare resources.[105] A divided distal colostomy was recommended by Pena and has been generally seen as the preferred method.[90,106] the advantage is that it ensures adequate faecal diversion and allows enough pressure to make a high-pressure distal colostogram easier in the future. Reported complications include prolapse, retraction, skin excoriations, parastomal hernia, skin dehiscence and bleeding.[105,107,108]

There is a debate between the use of loop colostomy and divided colostomy as the ideal type of colostomy for ARM.[106,109] Surgeons who argued against loop colostomies noticed higher rates of

stoma prolapse in loop colostomies.[106,109] However, Van Den Hondel[109] and Mullassery et al.[110] in their review of colostomies in ARM noticed no significant difference between the two methods concerning stoma prolapse. There was no statistically significant difference between loop colostomy and divided colostomy. Complications like retraction, parastomal hernia, post anorectoplasty, need for revision, and post-stomal wound infection was compared.[105,109]

The prolapsed colostomies in these reports were located in the transverse colon. Stoma prolapse is dependent on the location rather than the type of colostomy.[106] Colostomy done in a mobile part of the colon is most likely to prolapse regardless of its type.[106] Placing the stoma at the distal end of the descending colon at the proximal end of the sigmoid colon would circumvent the problem of stoma prolapse.[110] Prolapse of loop colostomy can also be reduced if a skin bridge is performed.[111] The risk of prolapse was reported to be higher when there was a longer interval between the creation of stoma and the closing of a loop colostomy.[112]

After the colostomy closure, there is also a significant reported incidence of complications. Significant incidence of wound infection, incisional hernia, and adhesive intestinal obstruction has been reported after the reversal of colostomies. [113] Serial anal dilation programs follow after surgical repair.

Anal dilation is essential in preventing anal strictures and stenosis after a definite procedure. Pena et al.[94] provided a standard guideline for dilatation. However, differences exist in its application by different practitioners.[4] According to Pena et al., dilation should be started 2 weeks after the definite procedure using a dilator that fits snugly into the anus and dilation is done twice a day. Dilator size is increased by 1mm weekly until the desired size is reached. Once there is no resistance, the frequency of dilatation is reduced to once a day for one month, every third day for one month, twice a week for one month, once weekly for one month and then once a month for three months. When the desired size is reached, the colostomy can then be closed.[48]

Some surgeons do not follow this protocol and have developed an internal institutionalized protocol for dilation. Surveys in European centres showed that anal dilations are carried out with protocols other than that suggested by Pena et al.[114], with 39% not following any specific protocol. Dilation is individualized. Morandi et al.'s international survey showed that the outpatient clinic was the preferred place for starting dilation (83%). Both plastic and metal dilators can be used. In most reviews, metal dilators dominate.[4] Dilators can be used with lubricants. Corticosteroids have been applied in some cases where the anus was stiff.[4] Gloved fingers as dilators have been reported.[4]

Follow-up after the first dilatation is inconsistent in the literature. The frequency of dilation also differs. In Morandi et al. survey, mothers were asked to dilate twice a day following the first dilations. Temple et al.[115] suggested that weekly dilation would achieve the same results.

Dilation can be associated with rectal prolapse and wound dehiscence.[116,117] These findings are variable and unique to each patient. Redo surgery of definitive procedure. It is reported in less than 5% of cases. They are done mostly because of anal stenosis or rectal prolapse.[116,117]

2.12 Complications

Delayed ARM presentation leads to gross intestinal obstruction, sepsis, aspiration pneumonia and sometimes death.[8] Adejuyigbe et al.[6] reported that nearly 60% of patients had gross abdominal distension during the presentation. The overall mortality in children with ARM ranges from 4.3% to 31.0%.[7,95] Complications associated with diverting colostomy for anorectal malformation range from 18.4–78.9% in African children.[7,118,119]The most commonly reported complications following colostomy for ARM are prolapse, skin excoriations and haemorrhage.[7,118]

Despite advancements in surgical techniques, complications requiring secondary procedures are still common. Complications in the perioperative period are encountered. Though uncommon with PSARP, wound infection frequently occurs when a primary repair is done without colostomy. Complications have also been documented when a loop colostomy is used to divert faeces. A loop colostomy does not entirely divert faecal matter as in divided colostomy.[120] Rectal problems like dehiscence, retraction and acquired atresia have been described after pull-through procedures.[120]

Rectal strictures can develop due to non-compliance with the anal dilation. Though rectal strictures can be present in some forms of ARM, it can be acquired during operative management. A recto-urinary fistula can also develop following PSARP.[121]Rectal prolapse following PSARP has an incidence of three per cent (3%).[53] It is common in higher anomalies.

Long-term complications include faecal incontinence and constipation. Holschneider et al.[122] review of ARM noted that PSARP had better outcomes with regards to continence than previous outdated methods. However, there was a significant clinical burden. 11.8% of children were incontinent, and 41.3% were partially continent.[122]

Constipation is the most common sequelae in children after correction of ARM. Huang et al[123] noted a high prevalence of 64.5% in the low anomalies group and 78.6% with faecal incontinence in the high anomalies group.

2.13 Financial Burden of Anorectal Malformation

Health is expensive, and it is so worldwide. Over a year, an estimated $45.5 million has been spent on managing patients with an anorectal malformation in the United States of America.[124] This money is spent in a system where health care is streamlined, and insurance policies work well. In low-income countries like Ghana, the National Health Insurance Scheme (NHIS) will pay a percentage of the

overall health cost for the patients with the insurance, and the rest of the healthcare cost will be borne by the caregivers.

The average cost of managing patients with anorectal malformations is not known in Ghana, Sub-Saharan Africa and Africa, as there is a paucity of data on the cost burden of managing this malformation by the primary caregivers. In KATH financial records, an average of GHC8000 ($1415; $1 = 5.65) is spent on patients with an anorectal malformation during surgeries and hospital stays. This is high for the average Ghanaian whose daily wage is around cedis 11.82 cedis ($2.09). The NHIS bears part of this cost. Medications, laboratory and radiological investigations that are not covered by the health insurance are borne by the caregiver. There are no findings in literature on the exact cost of ARM at KATH.

In the United States, the average hospital stay for patients with ARM who underwent primary repair was about 7.3 days, with no statistical difference over 3 years. The amount of money spent increased from $52,104 to $72,631.[125] The amount of money spent doubled or tripled when there were associated co-morbidities. The higher the number of co-morbidities, the more expensive the cost of treatment was. Patients with anorectal malformation and no other identified co-morbidity had an average hospital length of stay of 4 days and paid $35 029 for services. Most of these payments were by Medicaid and Blue Cross, Blue Shield Health Insurance Scheme, covering about 90%of the cases. Self-payment cases were about 6%[124], and other insurance paid for 2% of the cases. (124) Timely diagnosis and surgical care in the neonatal period has a favourable cost-effectiveness ratio reducing the overall healthcare burden.[125]

CHAPTER THREE

METHODOLOGY

3.1 Study Type and Design

The study was a descriptive, cross-sectional study of patients using a mixed-method approach in collecting data on children with ARM who were being managed at Komfo Anokye Teaching Hospital. The children recruited were at different levels of their management for anorectal malformation. The concurrent mixed-method approach was used, where the quantitative and qualitative arms of the study are done separately and then compared. The study was done over eight months, from November 2020 to June 2021.

3.2 Study Setting

The study was conducted at the Komfo Anokye Teaching Hospital (KATH); the second-largest teaching hospital in Ghana, located in Kumasi in the Ashanti Region.

The Ashanti Region; which is the third-largest region, occupies 10.2% of the total land area in Ghana, and it is the most populated region in the country. It accounts for 19.4% of Ghana's total population, according to the 2010 census.

Figure 3.1 Map Showing the Regions of Ghana

Source: https://www.ghanamissionun.org/map-regions-in-ghana/

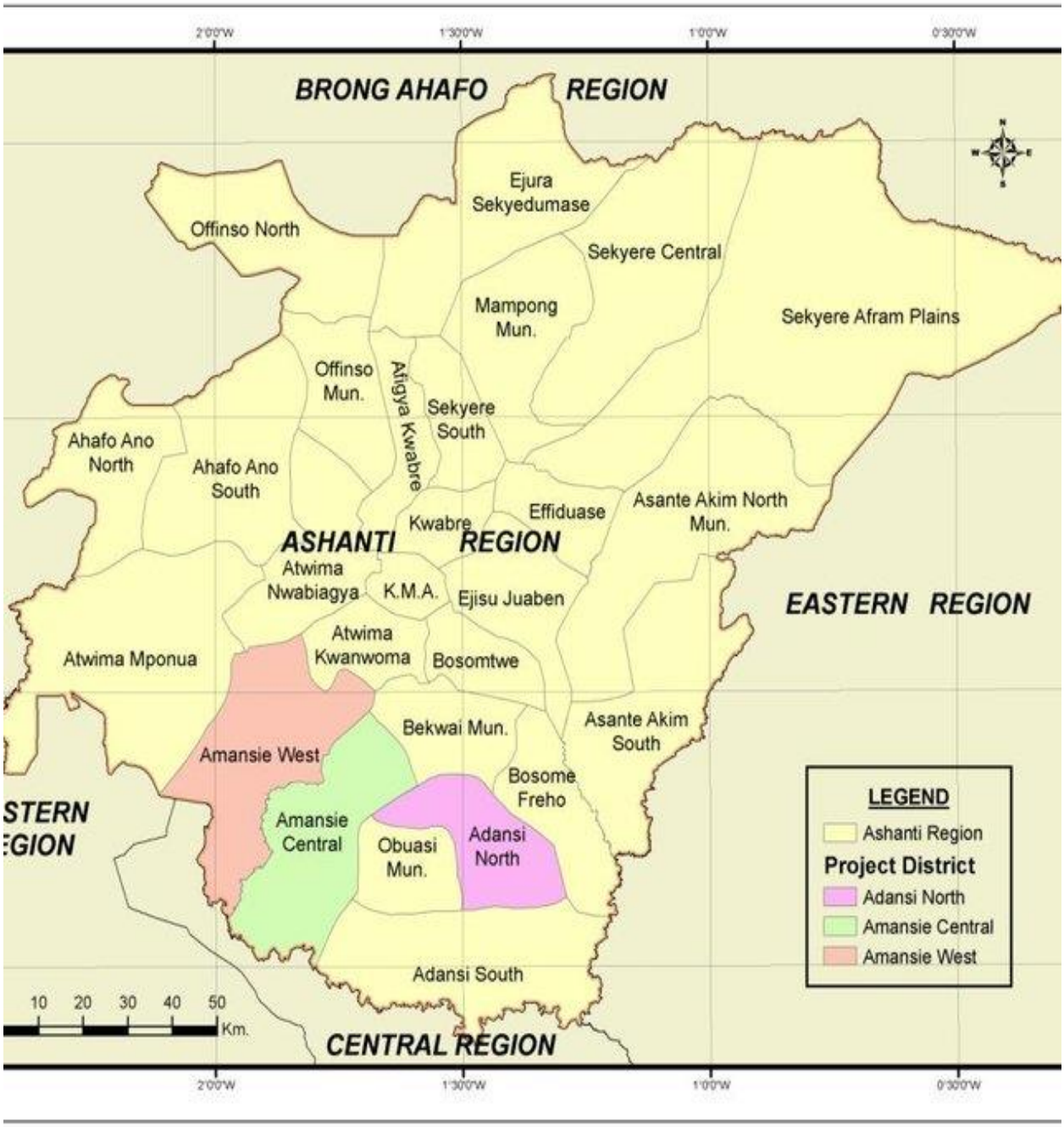

Figure 3.2 Map of Ashanti Region:

Source: https://www.researchgate.net/figure/Study-districts-in-Ashanti-Region_fig6_296701853

Kumasi, the capital city of the Ashanti Region of Ghana, is a metropolis located between latitude 6.35 º North and 6.40 º South, Longitude 1.30º West and 1.35 º East, with an elevation of 250- 300 meters above sea level. The land area of Kumasi is approximately 254km^2. The land area makes up about 0.9% of the total land area and accommodates 36% of the population in the region. Kumasi Metropolis shares its boundary to the north with Kwabre East and Afigya Kwabre Districts; bounded in the East

by Ejisu-Juaben and Asokore- Mampong Districts. In the south, it shares its boundaries with Bosomtwe and Atwima Kwamwoma. Kumasi is bounded by Atwima Nwabiagya in the west.

The population and Housing Census population of Kumasi in 2019 stood at 1,468,609. As of 2020, it had a growth rate of 2.13% annually, according to the national statistical services. Children from 0-15 years in the metropolis make up 33% of the population, with an overall sex ratio of 91.4 males to 100 females.

The Ghana Health Service's three-tier levels operate the health system within the metropolis, most of which are managed by the Regional Health Directorate. There are, in all, 136 health facilities providing services to the residents in Kumasi. Out of this number, private people own 115 hospitals (84.56%). Only 15.44% (21 out of the 136) regional hospitals are government-owned. The distribution of these facilities comprises the regional, quasi-government, mission, health centres, and teaching hospitals, i.e. the Komfo Anokye Teaching Hospital, KATH, where the study was conducted).

KATH is a tertiary teaching hospital, the largest in the region, with 1200 beds. It is affiliated with Kwame Nkrumah University of Science and Technology in Kumasi. It provides health care for about a third of Ghana's population, making it the second-largest hospital in the country.

The Surgery Department at KATH was officially established in 1975 and provided 24-hour surgical services for patients. The Department is well equipped and staffed with experienced doctors and nurses specialising in various surgical disciplines. The Surgery Department has been divided into distinct wards for the specialities. These sub specialties include paediatric surgery, general surgery, urology, trauma and orthopaedics, plastic surgery, neurosurgery and cardiothoracic surgery. The Paediatric Surgery unit has 30 beds and 18 cots on ward B3 and 15 floating cots at the Mother and Baby Unit (MBU) of KATH.

The unit has three Consultant Paediatric Surgeons and three general Specialists (senior residents), several Junior Residents (pre-membership level), and House Officers. The latter two rotate in and out of the unit.

The unit runs two outpatient clinics weekly and two weekly elective surgery days. An emergency team sees all emergency cases referred to the unit daily. From the theatre records, the unit performs, on average, ten emergencies every week. The unit can handle all emergencies referred and deal with any complications that may develop.

Patients were recruited from the Paediatric Emergency Unit, where all paediatric emergency cases are admitted and management started. Patients were also recruited from the Mother and Baby Unit (MBU) of Komfo Anokye Teaching Hospital, where all new-born babies are admitted and cared for up to two months.

3.3 Sampling and Sample Size

3.3.1 Quantitative study

The minimum sample for the study was determined using Yamane formula (1967)

$$n = \frac{N}{1+N(e)^2}$$

Where:

N = Population size

e = confidence level = 95% (0.05)

n = estimated sample size

An average of 50 cases of ARM presented half-yearly (6 monthly) at KATH for the past two years.

$$n = \frac{50}{1+50(0.05)^2} = 44.4$$

Thirty (30) percent of the sampling frame was added to allow for attrition. The high value was estimated because of the COVID-19 pandemic, which was likely to increase the attrition. Thus, a total of 57 participants were estimated for the study.

Consecutive sampling was done until a minimum of 57 participants were recruited.

3.3.2 Qualitative study

A purposive sampling technique was applied in recruiting willing parents of children with ARM who meet the inclusion criteria for the qualitative arm of the study. Fifteen (15) parents were sampled for interview. Interviews were conducted in person. However, saturation (a point where no new themes emerge from a qualitative interview) was reached at the 10th interview.

3.4. Study Population

The study population consisted of guardians and children with anorectal malformations at KATH.

3.5 Inclusion Criteria

1. All children diagnosed with anorectal malformation are managed at Komfo Anokye Teaching Hospital. Children recruited were at different stages of their management.

3.6 Exclusion Criteria

1. Patients who have had part of their surgeries done in another hospital other than KATH.

3.7 Data Collection

3.7.1 Quantitative data

A structured questionnaire was designed to capture information on all children presenting to the Mother and Baby Unit, Paediatric Surgery ward, Paediatric Emergency Unit and consulting room with anorectal malformation who meet the inclusion criteria. Consent was sought from the patient's parents or legal guardians before data was taken.

Data included demography of the child, such as age, current weight, birth weight and demography of both parents, such as age, occupation and educational level. Pregnancy details such as gestational age at booking of pregnancy, routine use of antenatal medications, mode of delivery and gestational age at birth were also captured. A detailed history was taken to note any delays in presentation and how the malformation was detected. Paediatric surgeons on duty and the primary researcher performed examinations of patients before surgery.

Details of the surgery were taken either from surgery or from patients' folders when the primary researcher was not part of the surgery. Complications related to the surgery were noted in the structured questionnaire. Wound complications after colostomy, PSARP, and reversal were noted. All the children recruited were followed up on the ward while on admission. After discharge, children were followed up in the Paediatric Surgery consulting room. Guardians were reached through mobile phones when patients did not turn up for reviews. The average cost of surgery was ascertained from the Accounts Department of KATH.

All data were collected by the primary researcher with the aid of a research assistant. Chi-square test was used to test for significance of association between independent and dependent variables. A P-value of < 0.05 was considered significant.

3.7.2 Qualitative data

A conducive environment was created for parents to express their thoughts on the issues raised freely. In-person, in-depth interviews of guardians were done using a semi-structured interview guide. Interviews lasted about 30minutes and were audio-taped. Notes were also taken with pen and paper/notebooks. All audios were stored in a password protected zip folder on the researcher's personal computer.

3.8 Data Processing and Analysis

Quantitative data were analysed using Statistical Package for Social Sciences (SPSS) version 22.0. Values were presented as proportions, means, and median where appropriate.

Ten (10) qualitative in-depth interviews of parents were transcribed verbatim into text in preparation for analysis. A deductive way using the thematic approach was employed. Data was checked several times for accuracy. The Research Assistant reviewed the transcribed work. Coding of the text was done manually to generate themes and subthemes.

3.9 Quality Assurance and Pretesting.

Pretesting of questionnaires was done using five children who had ARM. This was done before the final data for the study was collected. The interview's authenticity was ensured by religiously keeping records of interviews conducted and transcribing interviews verbatim. Where interviews were conducted in the local language, translation was done and then reviewed. Participants were not coerced to provide data. All procedures for analysis were made known to the research assistants. Translated and transcribed data were cross-checked and independently reviewed by research assistants.

3.10 Ethical and Legal Considerations

Written permission was obtained from the Paediatric Surgery Unit, Directorate of Surgery, Komfo Anokye Teaching Hospital Research and Development Unit to conduct the research.

Ethical clearance was obtained from the Committee on Human Research, Publications and Ethics of Komfo Anokye Teaching Hospital (KATH), Kumasi. (CHRPE/AP/370/20). The document is attached at the appendix iii.

Participation in the study was entirely voluntary, and guardians of children were allowed to withdraw from the study. The decision to withdraw did not affect the management of the child nor incurred any cost to the guardian.

Parents, guardians or informants signed a written consent form for participation in the study. Privacy of the patients and confidentiality of the information provided were ensured. There was no compensation given for participation in the study.

The author of this study did not have any competing interest in carrying out this study. The entire study was self-funded.

CHAPTER FOUR

RESULTS

This chapter presents the study results. The chapter has been divided in two main sections. The first section presents the analysis of the quantitative findings while the second section presents the qualitative findings.

4.1 Quantitative Study

A total of sixty-one (61) children; who were being managed for anorectal malformation at different management stages were included in the study. The presentation was made in sections.

The guardians' socio-demographic characteristics, pregnancy history of mothers, identification and presentation of anorectal malformation were captured. Treatment and cost analysis of the management of anorectal malformation were also captured. Findings are presented in tables and figures, preceded by narrations.

4.1.1 Socio-demographic characteristics of guardians

Twenty-two percent (22%) of the mothers were below 25 years. A majority (36.07%) were between the] ages of 30-35 years. About a quarter of the mothers, 15 (24.59 %), had no formal education, 27 (44. 26%) were junior high school graduates, 10 (16.39%) were senior high school graduates, and 4 (11.48 %) were graduates of tertiary education.

Most (78.6%) of mothers were employed with trading as the primary occupation, 39.34%, followed by civil service (18.03%). The minor group were artisans (8) 20%.

Findings on the socio-demographic characteristics of guardians are summarised in Table 4.1

Table 4.1 Socio-Demographic Characteristics of guardians

Variable	**Frequency (N=61)**	**Percentage (%)**
Age of the mother		
below 25 years	14	22.95
25 – 29years	18	29.51
30 – 34 years	22	36.07
35 years and above	7	11.48
Age of the father		
Below 25 years	8	13.11
25 – 29 years	10	16.39

30 – 34 years	21	34.43
35 years and above	22	36.07
Mother's occupation		
Unemployed	13	21.31
Artisan	5	8.20
Civil servant	8	13.11
Farming	11	18.03
Trader	24	39.34
Father's Occupation		
Artisan	6	9.84
Civil servant	25	40.98
Farming	11	18.00
Security personnel	3	4.92
Trader	16	26.23
The educational level of the mother		
Uneducated	15	24.59
Primary	2	3.28
Junior high School	27	44.26
Senior High School	10	16.39
Tertiary Education	7	11.48

4.1.2 Mothers' awareness of ARM

Fifty –six, 56 (91.8%) of mothers were not aware of anorectal malformation. Among the mothers who were aware of anorectal malformation, four (4) heard of it through the media, while one was taught the condition as part of the school curriculum.

4.1.3 Pregnancy history

None of the mothers took folic acid pre-conception. A greater proportion (70.49%) took folic acid during pregnancy. The majority (80.33%) of the mothers had their first visit to the antenatal clinic before eight weeks. 19.67% visited after eight weeks. The majority (88.52%) had no pregnancy-related condition. Table 4.2 summarises the findings.

Table 4.2 Pregnancy History

Variable	Frequency	Percentage
Did mother take Folic Acid		
No	43	70.49
Yes	18	29.51
Booking time		
After eight weeks	12	19.67
Before eight weeks	49	80.33
Routine intake of all antenatal medication		
No	16	26.23
Yes	45	73.77
Pregnancy-related condition		
No	55	88.52
Yes	6	9.84

4.1.4 Demographic characteristics of children

Of the 61 children, 31 (50.82%) were males, and 30 (49.18%) were females.

The birth weight of the children ranged from 1.5 kg to 4.1kg, with a mean of 3.12kg (SD = 0.51). Majority, 53 (86. 89%) of the patients had birth weight 2.5kg and above while 8 (13.11%) had birth weight below 2.5kg.

Majority were term babies 53 (86.89 %), 2 (3.28%) were post-term and 6 (9.84%) were pre-term. Term babies are children born between 37weeks and 42 weeks. Pre-term babies are less than 37weeks, while post-term babies are children born after 42 weeks. The demographic characteristics of the children are presented in Table 4.3

Table 4.3 Demographic characteristics of children

Characteristics	Frequency	Percentage
Weight		
>2.5kg	53	86.89
<2.5kg	8	13.11
Gestational age at birth		
Pre-term	2	3.28
Term	53	86.89
Post-term	6	9.83

4.1.5 Identification and presentation of anorectal malformation

The majority, 35 (57.38%), of the anorectal malformation was detected between day two and day five, 27.87% (17) were seen on day one, and (6) 9.83% were seen between day 6 and day 30. 4.92 (3) was noticed after one month.

Table 4.4 Identification and Presentation of Anorectal Malformation

Variable	Frequency (n= 61)	Percentage
When malformation was detected		
Day 1	17	+27.87
2-5days	35	57.38
6-30days	6	9.83
>30days	3	4.92
When malformation was brought to the hospital		
Within 24hours	7	11.48
24hrs - 48hrs	11	18.03
up to 72hrs	9	14.75
72hrs - 7 days	19	31.15
more than 7 days	15	24.59
Who detected malformation		
Health personnel	18	29.5
Mother	28	45.9
Relatives	15	24.6

After detection of the anorectal malformation, more than half of the children, 7 (11.48%) were brought to KATH within 24hrs, 11 (18.03%) within 48hours. 9 (14.75%) were brought in within 72hrs. 19 (31.15%) arrived in KATH between the 3rd and the 7th day while 15(24.59%) arrived after 7 days. After seven days. A greater proportion, 28(45.9 %), of the anorectal malformation were first detected by the mothers. 18 (29.5) % were detected by health professionals and 15 (24.6%) by relatives. The findings are summarised in Table 4.4.

4.1.6 Mode of presentation

Twenty 20 (32.79%) presented with intestinal obstruction, and 4 (6.56%) presented with sepsis at the emergency unit. Others (3) presented because of a noticed syndrome (Down syndrome). 34 (55.74%) were admitted through the emergency in a stable state. Figure 4.1 shows the state in which children with ARM presented to KATH

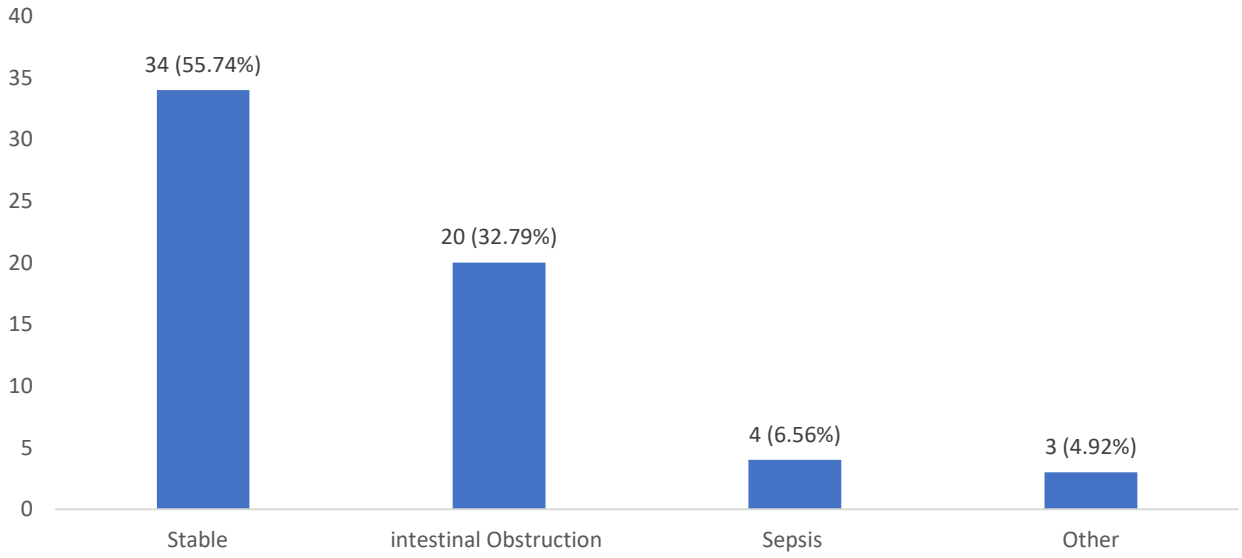

Figure 4.1 State in which Children with ARM presented to KATH

4.1.7 Type of anorectal malformation

The type of anorectal malformation varies by gender. Of the 31 males, 9 (29.03%) presented without a fistula, while 22 (70.97%) presented with a fistula. 11(35.48%) had recto-prostatic urethral fistula, 3 (9.67%) had recto perineal fistula, and 2 had a rectovesical fistula. The remaining 6 had anorectal malformation with a recto bulbar fistula. Figure 4.2 illustrates these findings.

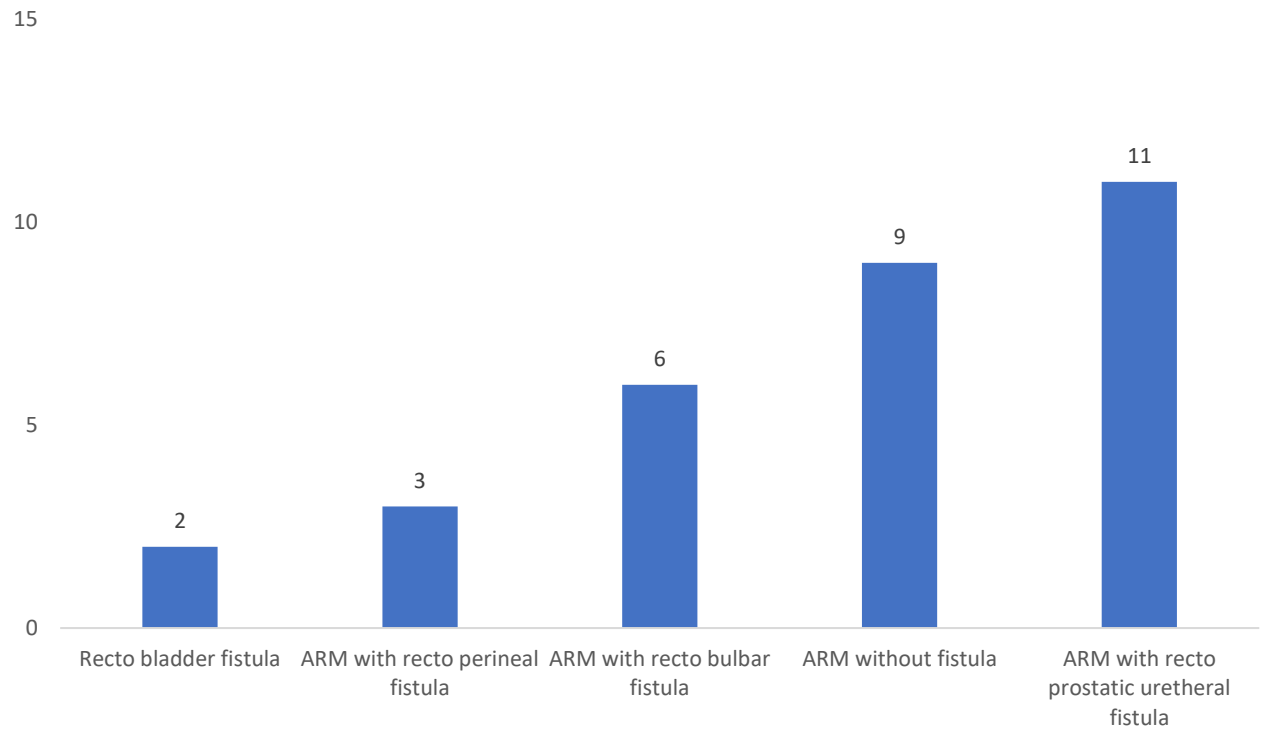

Figure 4.2 Distribution of Anorectal Malformation in Males

In all, 90% of the females presented with fistula. 10% (3) of the female had no fistula. 14 (46.67%) had recto vestibular fistula; five children each had a recto perineal fistula, respectively. Others were persistent cloaca 6.67% (2) and recto bladder fistula 3.33% (1). Figure 4.3 shows the distribution.

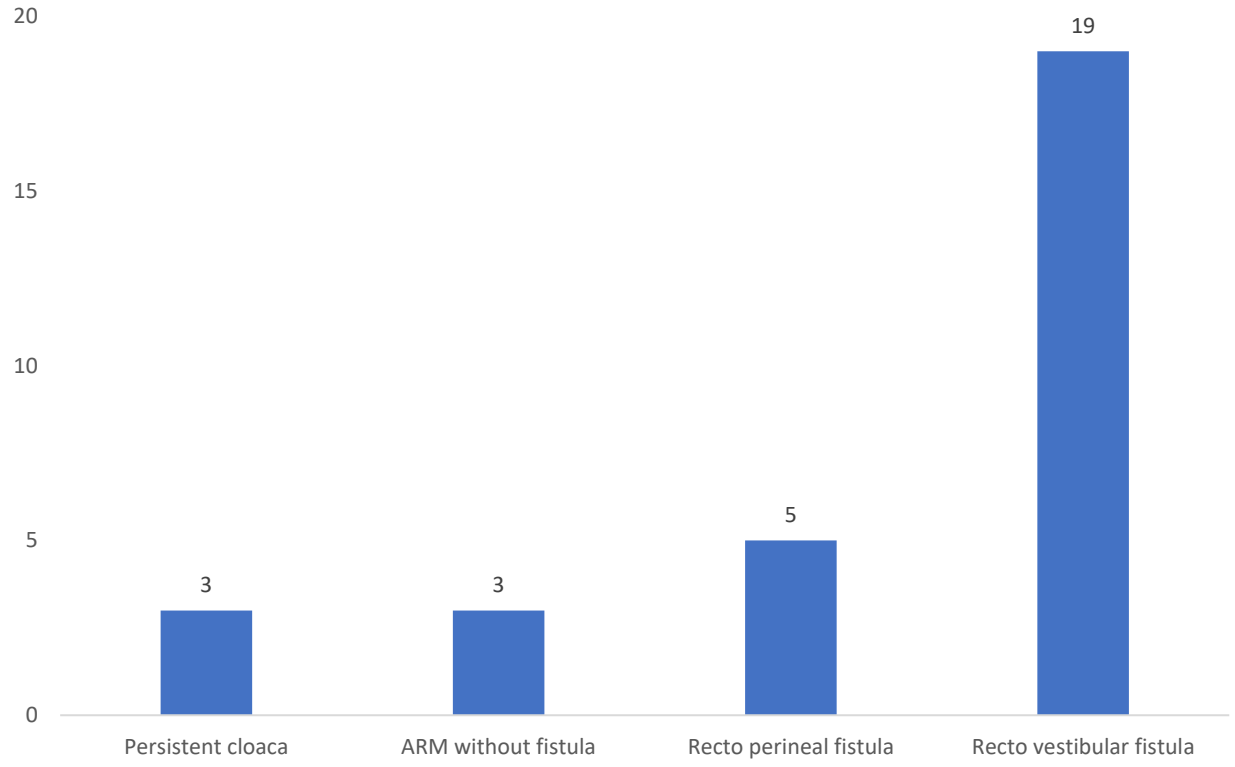

Figure 4.3 Distribution of Anorectal Malformation in Females

4.1.8 Associated anomalies

Seven congenital anomalies were detected. VACTERL anomalies were noted in 4 (57%) of the children. 1 spina bifida, and 3 Equino varus. Three syndromes were noted. 2 Down syndromes and an undiagnosed syndrome. It is shown in Table 4.5

Table 4.5 Distribution of Congenital Anomalies

Variable	Frequency (n = 7)	Percentage (%)
VACTERL associated anomalies		
Spina Bifida	1	14
Equino varus	3	43
Other syndromes		
Down syndrome	2	29
Undiagnosed syndrome	1	14

4.1.9 VACTERL screening

Complete VACTERL screening was not done for 33 children out of the 61 patients recruited for the study, making 54.1%. Of the remaining 28 patients, some form of screening for VACTERL malformation was done.

Investigations that were not done and why they were omitted are shown in Figure 4.4 and Figure 4.5 respectively. 72.1% had an echocardiogram (20), 68.9% had a spine x-ray (19) and 57.4% had a KUB ultrasound (16). An abdominal x-ray was performed on one child (3.6%).

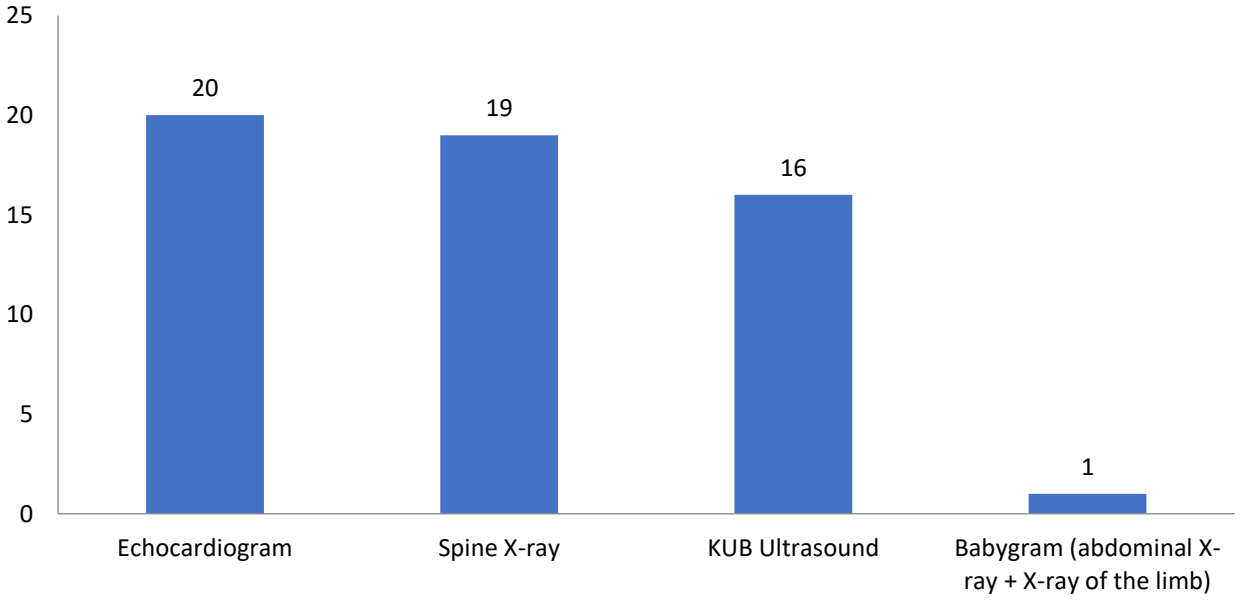

Figure 4.4 Frequency Distribution of VACTERL Investigation Done

Physicians' discretion to do a particular screening or investigation at a later date and deal with the emergent situation at hand, example creation of a colostomy for a patient with anorectal malformation with intestinal obstruction, was the most frequent reason for not doing a full VACTERL workup by the close of this study. These patients all have their VACTERL workup pending (to done at a later date). This constituted 24(72.72%) of all the reasons. Others are financial constraints 7(21.21%) and logistic challenges 2(6.06%). Challenges with logistics were faulty machines, and lack of personnel for investigations.

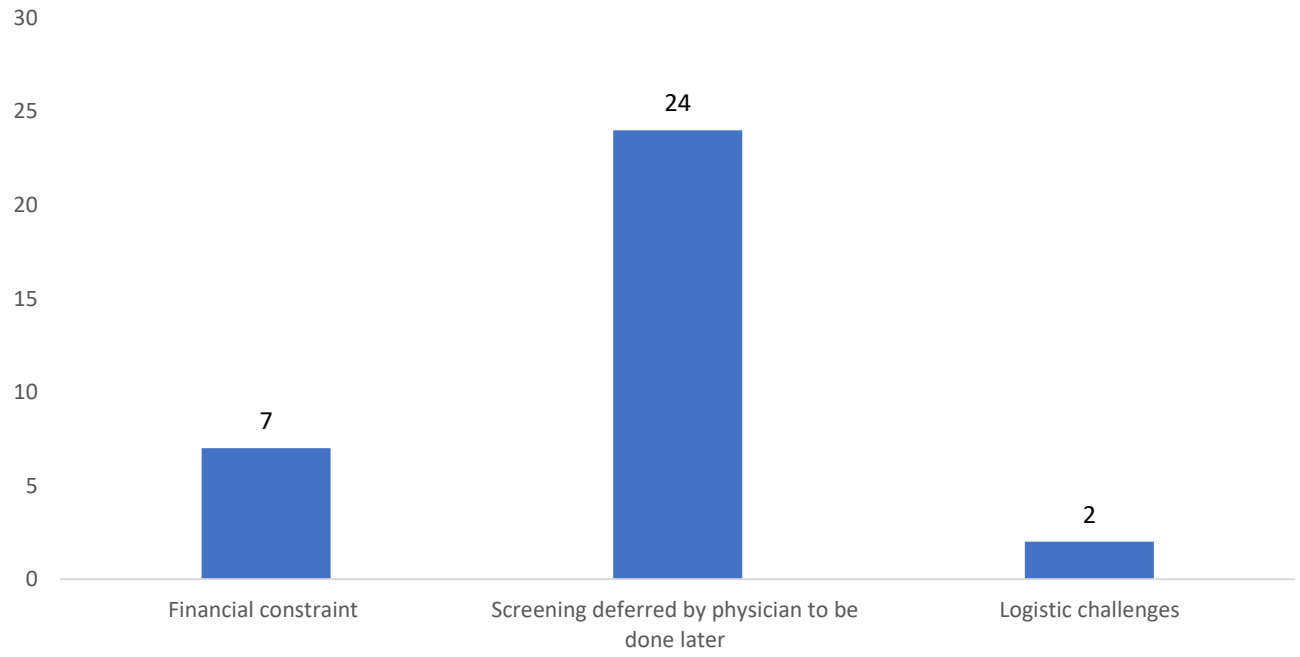

Figure 4.5 Reasons why Full VACTERL Investigation was not carried out.

4.1.10 Surgical management and complications

Two approaches were utilised to manage anorectal malformation in children: two-staged and three-staged procedures.

Two- staged procedure

Only one child (1.6%) underwent a two-staged procedure this patient had a colostomy and PSARP done on day one of presentation at KATH. Patient was a 3-day old male neonate with an anorectal malformation with a perineal fistula. He was discharged home 3 weeks after the repair. He had closure of his colostomy 14 months later. The late presentation for the closure of colostomy was because of financial constraints.

There was no anaesthetic or surgical complications post colostomy + PSARP for the two-staged repair. Colostomy was reversed in 14 months.

Three-staged procedure

The remaining (60) patients out of the 61 patients recruited underwent a three-stage procedure.

The three-staged procedure consisted of creation colostomy, posterior sagittal anorectoplasty (PSARP), and colostomy closure.

Initial colostomy:

Senior residents in Paediatric surgery performed the majority, 54 (90%) of the initial colostomy. The remaining 7 (10%) were performed by consultants. Figure 4.6 shows a divided sigmoid colostomy on 7-month-old patient with anorectal malformation with a divided sigmoid colostomy. The patient had mild excoriations around the stoma site.

Figure 4.6 A child with a divided sigmoid colostomy with mild excoriations around the stoma site.

General anaesthesia was administered in all cases. Nurse anaesthetists administered a more significant percentage of anaesthesia. No anaesthetic complication occurred. Findings are shown in Table 4.6

Table 4.6 Personnel Involved in Initial Colostomy

Variable	Frequency	Percentage
Category of surgeon		
Senior resident	54	90
Fellow	6	10
Category of Anaesthetist		
Doctor Anaesthetist	18	30

Nurse Anaesthetist	42	70

4.1.11 Complications post-colostomy

A total of thirteen complications were noticed post colostomy. 38.86% were superficial surgical site infections. 15.38% were mucocutaneous breakdown, and 1 (7.7%) wound dehiscence. There were redo-surgeries in 3 of the cases. The 3 cases were stenosis and complete closure of the distal stoma. Mortality occurred in one child, who died from heart failure he had very a large ventricular septal defect and an atrial septal defect as well.

Table 4.7 Complications Post-Colostomy

Variable	Frequency	Percentage of total cases done
Excoriations	5	8.20
Mucocutaneous breakdown	2	3.28
Wound dehiscence	1	1.64
Stenosis/ Complete closure of mucous fistula	3	4.29
TOTAL	11	18.03

The mean length of stay was 11.89 +/- 8.93 days. Most of the children (31), 51.67% were discharged in 10 days.

4.1.12 PSARP

Patients recruited for the study were at different levels of their management; all 61 patients had a colostomy. Forty –five (45) children out of the 61 had had a colostomy, and a PSARP and 29 children out of the 61 had colostomy, PSARP and a closure of colostomy done.

Thirty -five; 35 (78%) of PSARP was performed by residents, while consultants performed 22% of the surgeries. The average time between an initial colostomy and PSARP was 7.02 +/- 2.05 months. The maximum duration was 12 months.

Figure 4.7 shows an augmented pressure distal pressure colostogram of a patient with an anorectal malformation with a recto prostatic fistula, while Figure 4.8 shows PSARP done on a female patient with anorectal malformation with a rectovestibular fistula. The patient also has a prominent sacral dimple.

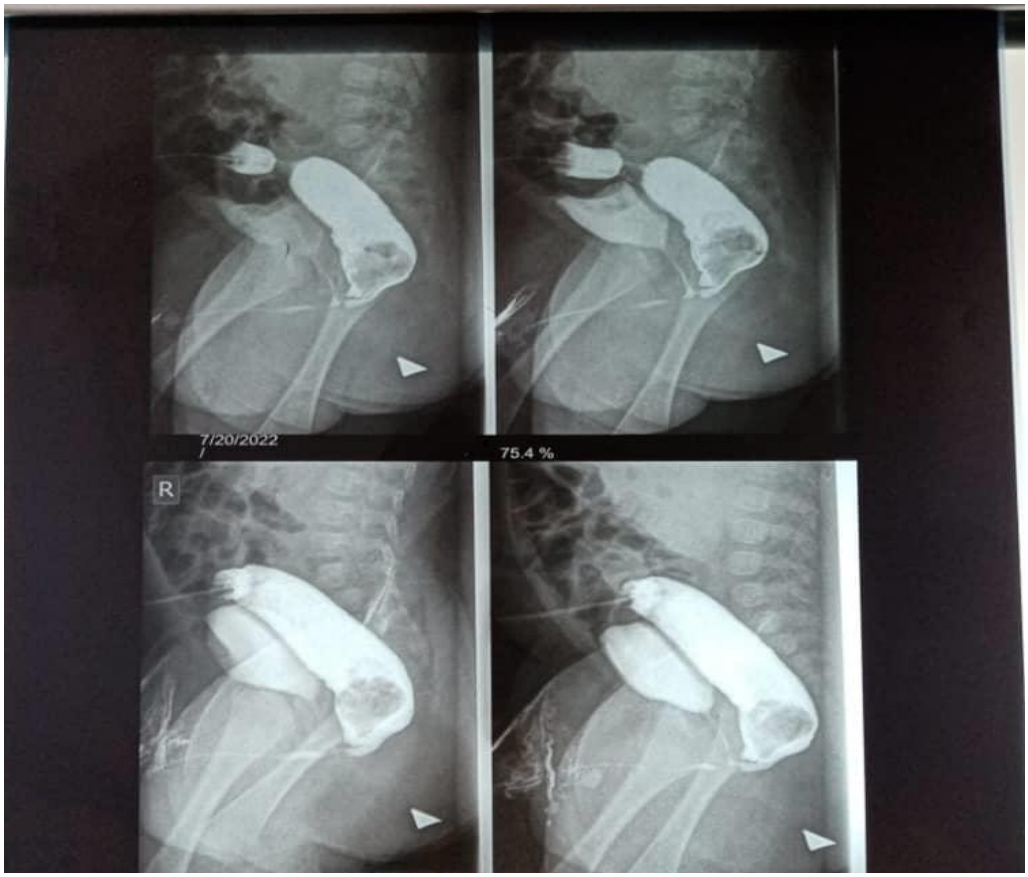

Figure 4.7 An augmented pressure distal pressure colostogram of an anorectal malformation with a rectoprostatic fistula.

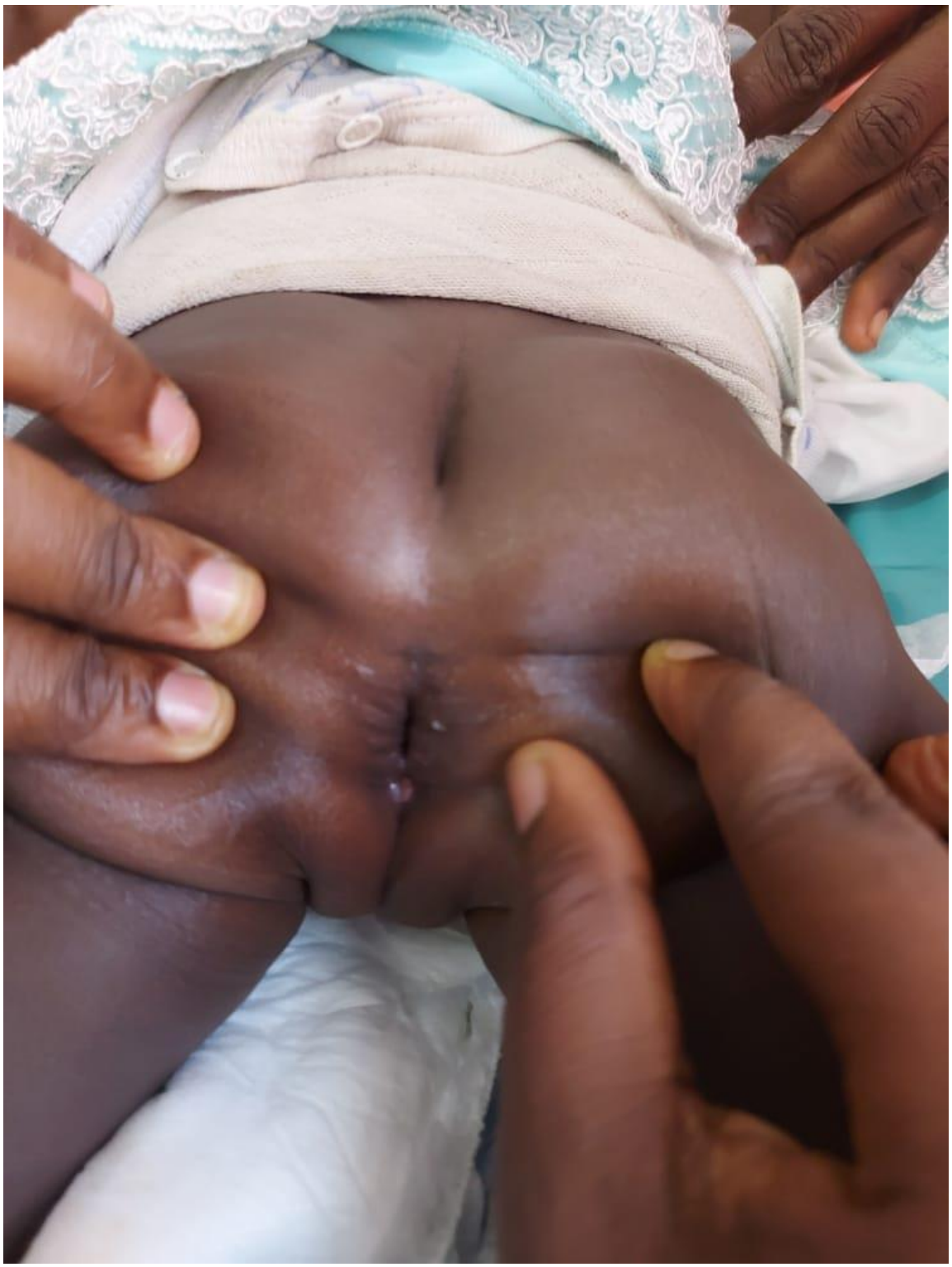

Figure 4.8 Post PSARP of a female patient with anorectal malformation with rectovestibular fistula. Patient also has a sacral dimple.

4.1.13 Complications and Incidents Post- PSARP

Six, 6 (13.33%) out of the 45 PSARPs done developed complications or had an incident. Gaped wounds accounted for 50% of the complications. Dislodged urethral catheter was 33.33%. There was also malpositioning of the neo-anus for one (1) child accounting for 16.67%. The gaped wounds were re sutured and patients discharged home on day 30 post surgery, no leakage of urine was observed in the patients with dislodged urethral catheters and they were discharged home on day 26. The malpositioned neo anus had a re do psarp done and discharged home on day 35.

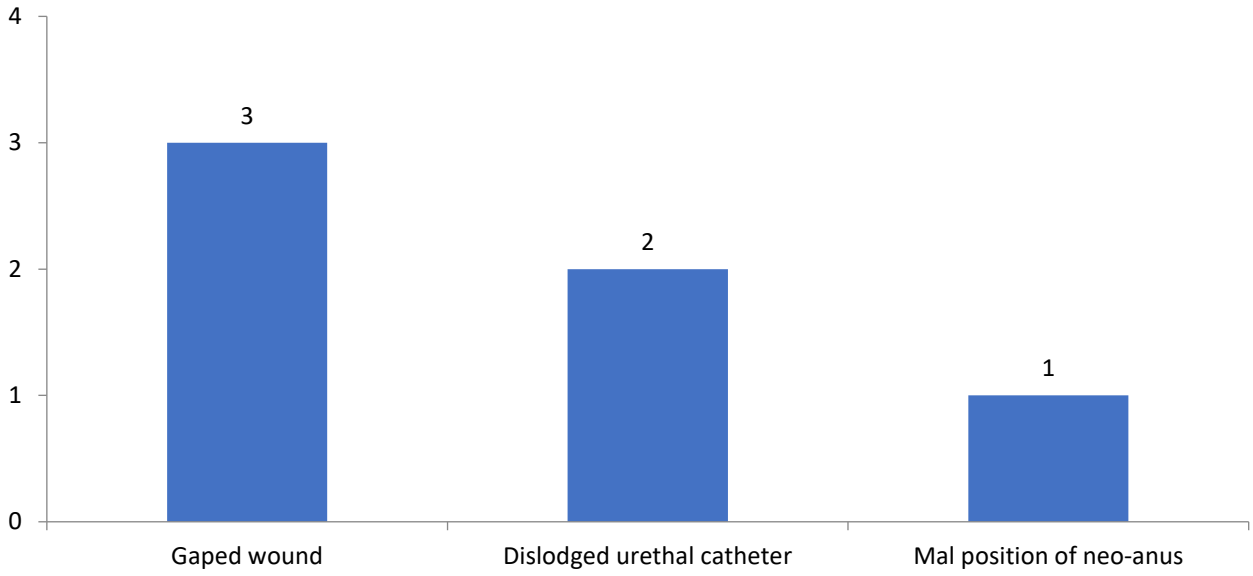

Figure 4.9 : Complications Post-PSARP

Length of stay prior to be being discharged

27 (60%) of the children were discharged within 30 days of PSARP. 13(28.89%) were discharged between thirty and forty days while 5(11.1%) were discharged after forty (40) days. The average length of stay post-PSARP was 29.95 +/- 9.26 days. Figure 4.10 shows the distribution.

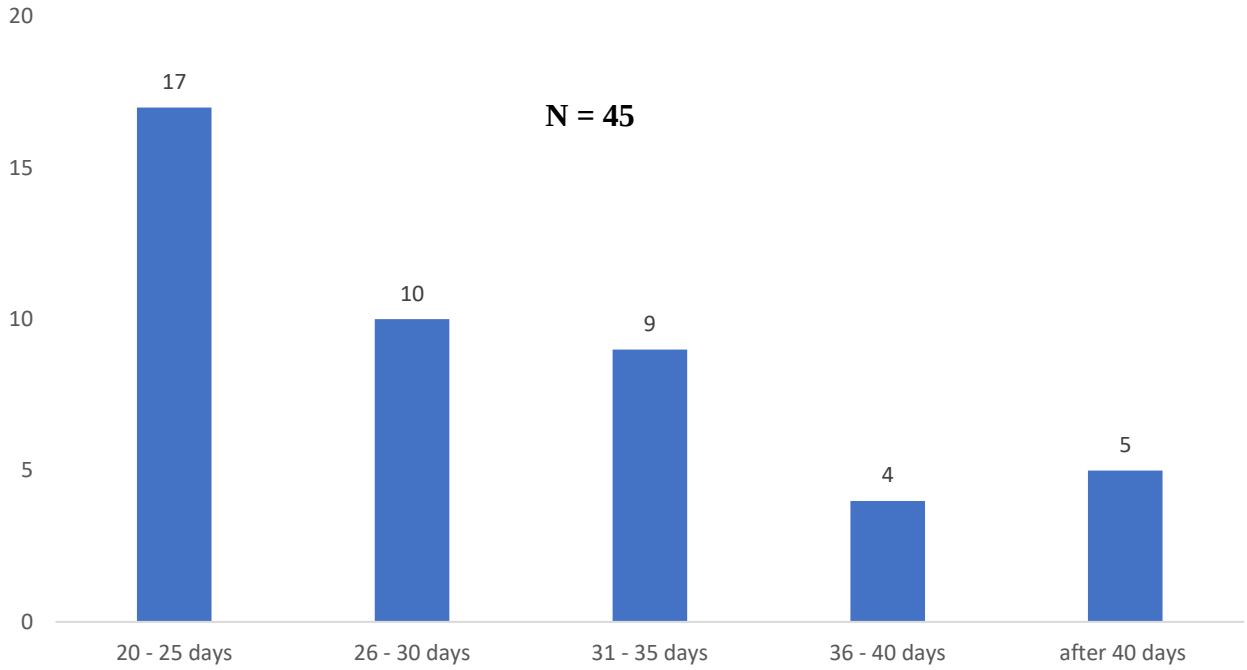

Figure 4.10 Length of Hospital Stay prior to being discharged

4.1.14 Reversal of colostomy

Twenty-nine of the children had a reversal of colostomy during this study.

A proportion of colostomy reversal was done by fellows (14) and senior residents (15).

The median time between PSARP and colostomy reversal was eight months, with a range of 4 -24 months for 28 children, the last child(an outlier) in the study had colostomy reversal 8 years and 4 months after PSARP . Of participants who had the reversal done between the 24 months, 50% had a reversal of colostomy done within six months.

Figure 4.11 shows a child with colostomy reversal performed without complications.

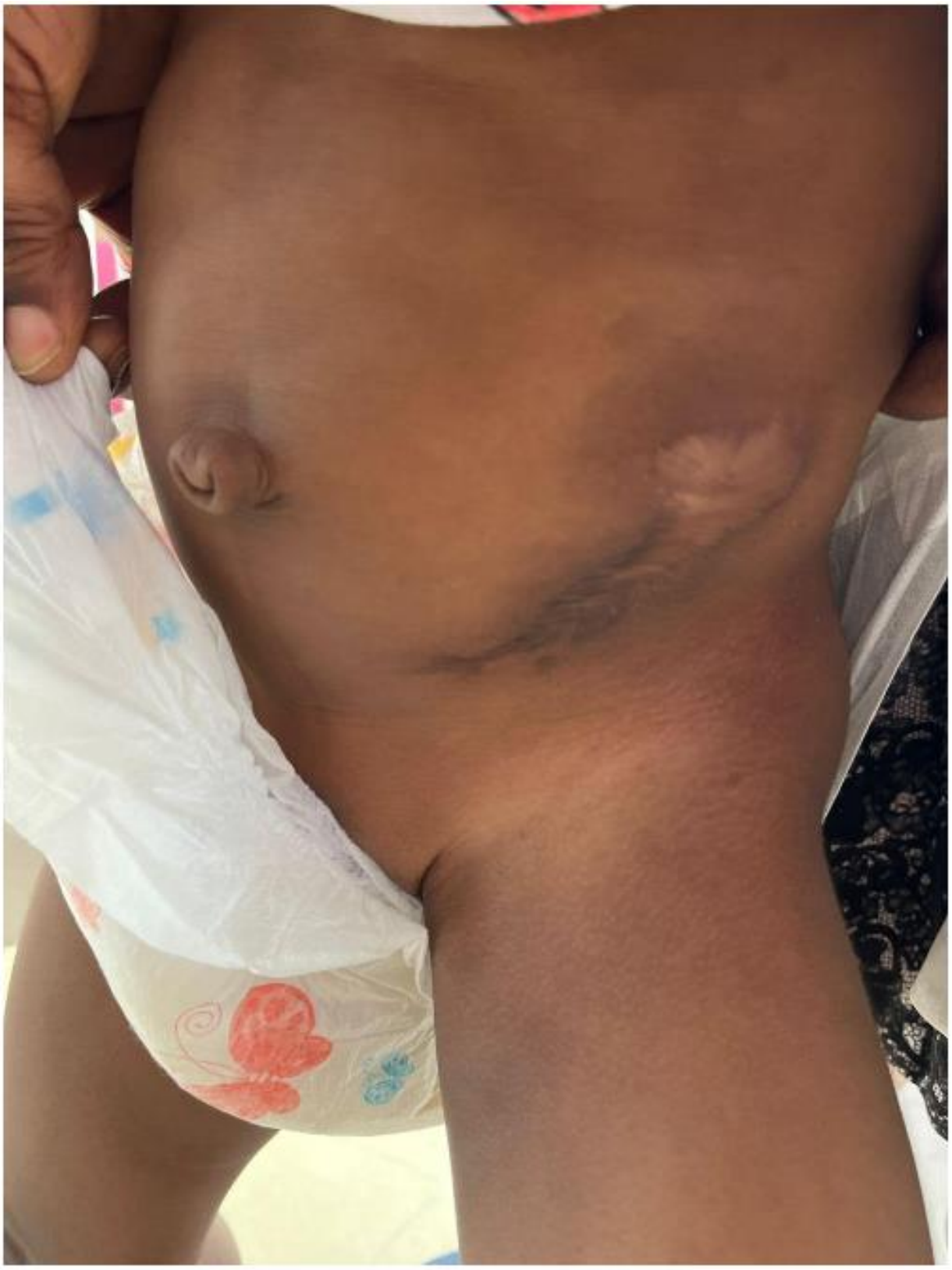

Figure 4.11 Child with Colostomy Reversal Performed without Complications

4.1.15 Complications post closure of colostomy

27.59% of the surgeries had surgical complications after closure and reversal of colostomy. 37.5% had surgical site infection, and 25% had an anastomotic breakdown of the sigmoid colon when the colostomy was reversed. Others were electrolyte imbalance 1(12.5%), rectal mucosal prolapse 1 (12.5%) and faecal incontinence 1(12.5%). The second mortality was from this section. The mortality rate during reversal of colostomy: is 3.44%. Figure 4.12 shows the distribution of complications post reversal of colostomy.

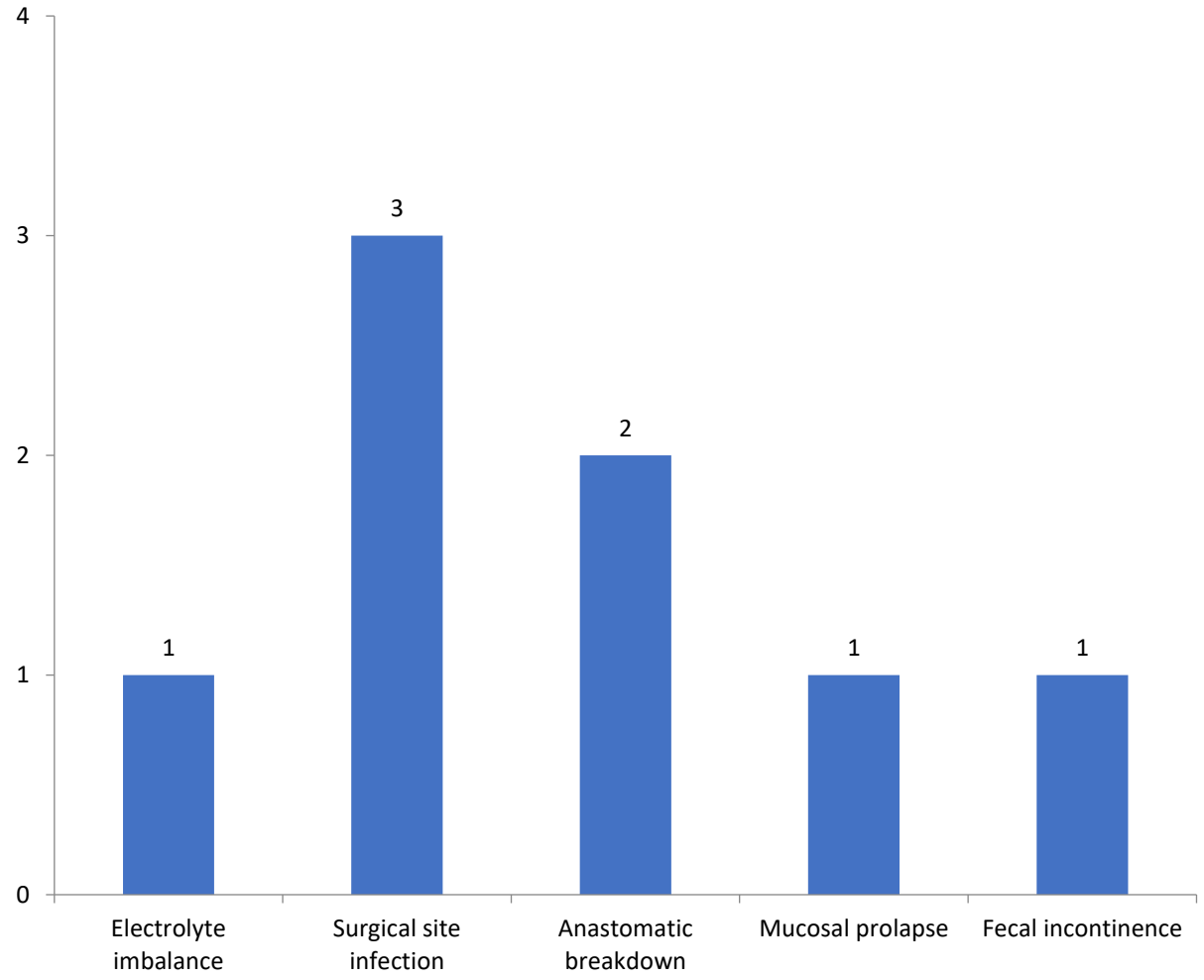

Figure 4.12 Distribution of Complications Post Reversal of Colostomy

Three redo surgeries were done. For faecal incontinence, a redo psarp was done. (The patient had anorectal malformation with a recto bulbar fistula). The two other patients had an anastomotic breakdown, so a relaparotomy was done and stomas re-sited to closed three months later. One succumbed to sepsis on post-operative day 10.

4.1.16 Univariate association between independent and dependent variables

The overall complication rate during the study period was 39. 3%. Relationship was determined between sex, birthweight, time of presentation and the presence of fistula and the development of complications as shown in Table 4.8

Table 4.8 Relationship between Sex, Weight, Time of Presentation and Presence of Fistula or not in Development of Complications

Complications in Anorectal Malformation repair					
Variable	Complication – YES No. (%)	Complication – NO No. (%)	Total No. (%)	X^2	p-value
Weight					
Normal Weight	21(91.30)	33(86.84)	54(88.52)	0.2808	0.596
Underweight	2(8.70)	5(13.16)	7(11.48)		
Total	23(100.00)	38(100.00)	61(100.00)		
Time of Presentation					
Early (< 24hrs)	4 (57.1)	3 (42. 9)	7 (100.0)	1.0497	0.306
Late (> 24hrs)	20 (37.0)	34 (63)	54 (100.0)		
Total	24 (39.3)	37 (60.7)	61 (100.0)		
Type of ARM					
With fistula	2(8.70)	11(28.95)	13(21.31)	3.5041	0.061
Without fistula	21(91.30)	27(71.05)	48(78.69)		
Total	23(100.00)	38(100.00)	61(100.00)		

There was no significant association between birth weight (P value of 0.596), the presence of a fistula (P value of 0.061), and the time of presentation with a (P value of 0.306)

4.1.17 Rescheduling of surgeries

Some surgeries were rescheduled for the PSARP procedure and the closure of colostomy.

Out of the 45 patients who had the PSARP procedure 27 (60% of patients) of them had their surgery rescheduled.

Out of the 29 patients booked for closure of colostomy 16 (55.2%) of them also had their surgery rescheduled.

The reasons for the rescheduling for PSARP included financial challenges which accounted for 11% of the rescheduled cases. The COVID 19 crisis alone accounted for 29.6% of the cases to be rescheduled while institutional challenges alone; including theatre and equipment issues, accounted for 18.5% of the rescheduled cases. Two cases (representing 7%) of the rescheduled cases were as a result of other reasons including the patient's nutritional state (severely malnourished) at the time surgery was planned. COVID 19 crises as well as institutional challenges accounted for 29.6%. COVID 19 crises, financial and institutional challenges accounted for 3.7%.

The reasons for rescheduling surgeries for the closure of colostomy were; malnourished state of the patient is 12.5%, financial constraints 18.75%, COVID 19 crises 50%, institutional challenges is 6.25% and COVID 19 crises, financial constraints, and institutional challenges making 12.5%. This is summarised in the Figure 4.13.

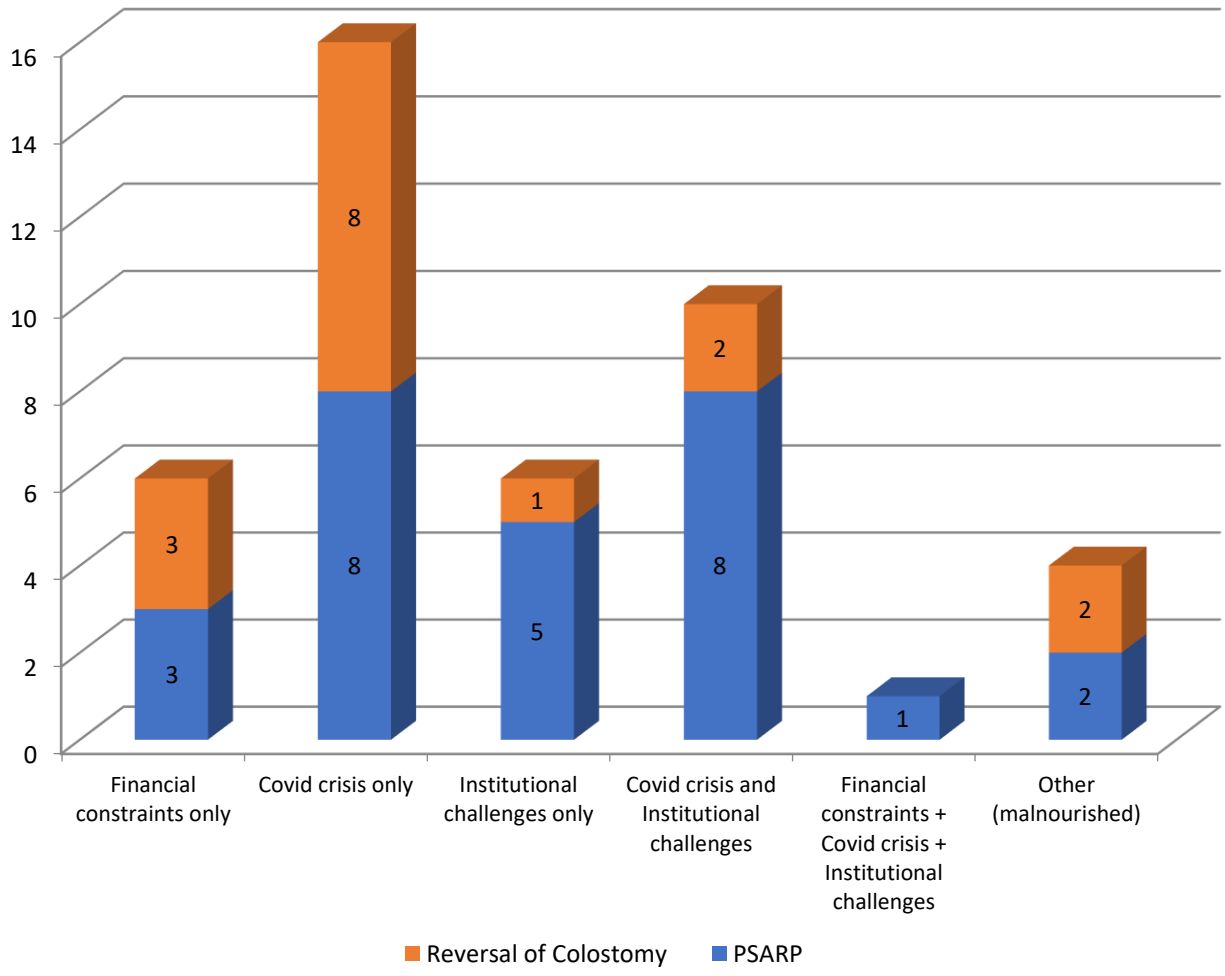

Figure 4.13 Reasons why Surgeries were rescheduled

4.1.18 Cost analysis of the management of anorectal malformation.

Cost analysis of in-hospital management of the various stages of anorectal malformation repair is presented. Direct cost associated with drugs, investigations and surgical procedures was obtained from hospital billing sheets. All payments made by the child's guardians aside from those covered by the National Health Insurance Scheme are termed Out-of-pocket payments (OOP). The analysis does not include the loss of productivity cost. All values quoted are in Ghana Cedis and reflect prices in the year 2020. All the participants in the study had NHIS.

The average cost of initial colostomy, PSARP and colostomy closure was GHC2955.69, GHC5031.83 and GHC3287.75, respectively. Out of pocket payments accounted for 56.12% of during the creation of a colostomy, 66.9% during PSARP and 58.56% during the colostomy reversal. Table 4.8 summarises these findings.

Table 4.9 Direct in-hospital Cost of Managing Anorectal Malformation

Direct cost						
	NHIS (%)	OOP (%)	Total	NHIS average	OOP average	Average per procedure
Colostomy (n = 61)	79112.38 (43.88%)	101184.73 (56.12%)	180297.1	1296.92	1658.77	2955.69
PSARP (n = 45)	74944.83 (33.10%)	151487.45 (66.90%)	226432.3	1665.44	3366.39	5031.83
Reversal of colostomy n= (29)	39382.49 (41.31 %)	55962.24 (58.69%)	95344.73	1358.02	1929.73	3287.75

The average cost of colostomy + PSARP for the 45 children was GHC9038.43, while the average cost of the complete three-stage procedure (colostomy + PSARP + reversal of colostomy) was GHC11604.93.The patient that had a two stage procedure paid 8600.20 for the overall cost of management. The PSARP plus colostomy cost 6300 cedis and the closure of colostomy cost 2300.20 cedis.

Mothers were asked to rate the cost of in-hospital admission. Figure 4.14 shows their responses.

18% rated it high, 44% rated cost as a medium, and 38% rated it average.

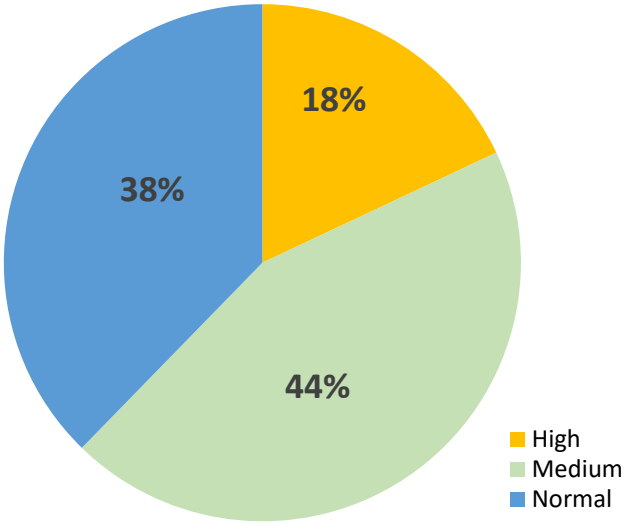

Figure 4. 14 Rating of Cost of in-hospital Management of Anorectal Malformation

4.2 Qualitative study results

This section focuses on the findings of the data obtained from interviews with mothers. It aimed to explore the mother's perspective on the challenges with identification, presentation, management and taking care of children with anorectal malformations. Three main themes emerged from the data gathered. The themes and sub-themes have been presented with the support of anonymised quotes from the participants.

4.2.1 Demographic characteristics of participants

Fifteen (15) mothers taking care of their children with anorectal malformation were sampled for interviews. Saturation was reached at the tenth interview. Thus ten interviews were included in the analysis. Most participants were in the age brackets of 25-34 years.

All participants were females. Most participants have more than one child, with 9 participants having more than 2 children. Seven (7) participants were single, while three (3) were married. Nine (9) were employed.

4.2.2 Identification and presentation of anorectal malformation

The narrations on the identification of anorectal malformation are unique to each mother. There were challenges with the referral after the identification of anorectal malformation. Some themes identified were lack of knowledge on anorectal malformation, accidental identification by relative or nurse or identification because of complications.

4.2.3 Lack of knowledge

Some of the mothers did not know about anorectal malformation.

A mother had this to say:

> “I never knew that a child can be born without an anus. So, I assumed everything was fine with my boy until I noticed that my child had not passed stools after three days, so I checked. I saw that the child did not have an (anus)” – M4.

Another mother narrated this;

> "I was bathing my child, and I noticed that the anal opening is not where it should be. Rather I saw a tiny hole at the side. I thought that was the anus until my child's stomach started becoming big” – M2.

The attending physician or nurse noticed some mothers' anorectal malformation.

One participant had this to say:

> "My baby was born through an operation (Caesarean section). I did not see my baby for 24 hours. I was informed that my child did not have an anal opening. It came as a shock to me." – M8.

The mother in the following narration got to know after the child developed intestinal obstruction.

> "My child's abdomen had become very tense and big on the third day after birth. He started vomiting, so I sent him to a nearby clinic. The nurse informed me the child does not have an anal opening, which is why the abdomen is big." - M5.

4.2.4 Referral challenges

Mothers emphasised the bottlenecks of getting their children to KATH for specialised care. All the participants in the study were enrolled in the NHIS.

A mother who had had a caesarean section complained about the referral policy at the facility she delivered.

> "They told me it will not be advisable to refer me until I am fit enough to be with the child. So, I waited three days to get well before they referred my child. They had noticed child did not have an anus a day after the surgery". - M10

A few complained about financial challenges in getting their children to KATH.

> " … I am from the Northern part of Ghana. They insisted I come in an ambulance. However, I did not have money to cover the cost. I had to beg relatives for money to come to Kumasi" - M8.

4.2.5 Challenges with management

This theme highlighted the challenges with the various stages of management. Mothers demonstrated unique challenges at varying stages of the management. Sub-themes identified are challenges with pre-operative workup, stoma care, rescheduling of surgeries and financial challenges.

4.2.6 Challenges with pre-operative workup

Several mothers acknowledged that surgery was not delayed. However, they had to make any out-of-pocket payments for some investigations.

> "I thought insurance covers this lab, only to be told, it does not. I had to pay with my own money. I had heard some mothers not were not able to pay for some investigations (augmented pressure distal colostogram), so the surgery was not done for them" – M3.

The second narration provided more information on issues with getting investigations done.

> "… I was asked to do some scans and labs. When I went to where they do the scan, I was told the scan machine at this hospital was not functioning. My husband and I pay for ambulance to send us out of the facility for the scan because my child was not well. This added to our cost" – M1

4.2.7 Rescheduling of surgeries

Some mothers said their surgeries were rescheduled either because of the child's state or logistic issues at the theatre.

> "My surgery was postponed because the doctor said my child's weight is down and he has not been eating well. The surgery will worsen my child's condition in his state. I was referred to the child welfare clinic where I was taught to prepare and give my son some meals to help him grow and gain weight"- M6.

Another mother had this to say:

> "My surgery was postponed because the doctors said there was a problem at the theatre. The machines they will use for the baby during surgery were faulty, so only emergency cases were being done at that time." – M5.

4.2.8 Stoma care

All the participants indicated they were taught how to care of the stoma. They expressed minimal challenge with care for the stoma. There was general satisfaction with how to care for the stoma was taught.

> "I was taught how to care for the colostomy. It was strange, though, putting diapers on my child's abdomen. The method the nurses taught me is what I still use today in managing my baby" - M10.

One of the mothers was relieved she was taught how to care for her child after the colostomy appropriately.

"… You know, seeing your child with faeces from the stomach is not emotionally easy. However, you still have to care for this child. The nurses cannot always do it. I am glad I was taught how to place the nappy diapers properly so my child does not soil himself. Learning was not difficult. I easily picked it up once I psyched myself for the task." – M1

4.2.9 Financial challenges

All the participants indicated they made substantial top-up payments for surgeries. They expressed disappointment in the co-payment scheme.

This is a narration by one mother:

"I was told by the nurse where I was referred that I should come to the bigger hospital because it is a child. Everything will be free once I have insurance. However, I am paying extra money from my pocket. They say some investigations and some drugs are not covered by insurance, so you must pay. I have spent as much as the insurance scheme made me pay at the end of the day (National Health Insurance)." – M2

On how the parents sourced funds to cover their bills, a few said they paid from their own pockets while others had to solicit funds from relatives and church.

"Life is 'hard'. I have to go to my pastor and ask for help. The church raised some funds for me to pay my bills. I do not have anyone to help so that was the only thing I could do" – M10.

Another mother said she had to borrow money from relatives to cover her bills.

"I initially thought it would be small money. But when they gave me my bill, I had to call my relatives for help immediately. Fortunately, my brother was around, so he came to my aid". M7

Donations were a primary source of monetary support for a few mothers.

4.2.10 Psychosocial dimension of having and treating a child with anorectal malformation

This theme explored the pertinent issues with giving birth to a child with anorectal malformation and how guardians manage the period of surgical intervention. One key observation was stigmatisation.

Stigmatisation was a chief concern for several of the mothers. Their concerns centred on two key areas: the stigma of giving birth to a child born with anorectal malformation. The other end of the stigma was noted in having a child with a colostomy.

Some mothers complained that they feared or have been stigmatised for having a child with anorectal malformation.

"In my community, children born without anus are seen as a curse. They will tell you to neglect that child. They will say negative things about your family, that you will bring a bad omen to your extended family. When some of my extended family members noticed that my son does

not have an anus, they did not visit again, and I heard them calling me all manner of names." - M2.

Another mother narrated how she felt noticing the child had anorectal malformation.

> "I feared sending my child home. I did not want them to know my child had an issue. If I did, they would call me all kinds of negative names; I was glad when they referred me early". - M6

However, a few mothers did not experience ostracism. Instead, they received support during that period.

> "When my mother noticed that my daughter did not have an anus, she was the first person to encourage me not to give up. I did not experience any rejection from my parents or husband. They rather pushed me to bring the child to the hospital." M5

The stigmatisation and social withdrawal were between the first colostomy and reversal of the colostomy. All the mothers experienced some form of the stigma associated with caring for a child with a colostomy.

> "I felt out of place when I send my child to the weighing clinic. I cannot stand how the nurse looks at my baby and the comments passed by onlookers whenever I expose my child fully for him to be weighed" luckily on admission I get to catch upon the vaccinations I missed.- M8.

Having a child with colostomy had negative social implications. It affected where, when and what guardians of children with anorectal malformation could do or be. The most crucial effect highlighted by mothers was social withdrawal. Some guardians were unwilling to participate in any social activity until the colostomy was reversed.

Below are narrations by two mothers.

> "I do not want to go anywhere with my daughter because of her abdomen's opening (colostomy). I feel uncomfortable letting another person see what my child has." -M5
>
> "I thought things would be better after surgery. However, it is rather worse. The child now has some openings in the stomach, and I cannot go anywhere with him. I have not been to church in a while. I will avoid it until the opening is closed." - M3.

Those who can attend social functions do not intermingle with their peers.

> "At times, I go to church. But I also sit at the back because my baby would be soiling himself through the opening [stoma]. I do not want the other people to know what has been done to my child" – M5.

Another woman narrated this story:

> "…. because I do not have anyone to attend to my needs in the house, I go to the market myself. I am not comfortable leaving the child with anyone. When I want to go to the market,

I go at dawn, buy my items quickly, and then return home. I have been doing this after the surgery.” – M7.

4.2.11 Psychological support

The mothers complained of a lack of proper psychological support. They usually get encouragement from those who have been through managing their children with anorectal malformation. Psychological support also came from their religious leaders.

> "Madam Adwoa has been very helpful to me. She came on a visit with her child, who had an anorectal malformation that has been corrected. That alone gave me hope that something could be done about mine. Honestly, after I saw the openings created by the doctors, I gave up. After sharing her experience with me, I am ready to face the challenges ahead." M10

Another woman narrated this:

"After my child was admitted and sent for surgery, another woman who had been on the ward for two months was my major source of motivation. If she could go through it, I go also do the same. There was no psychologist to speak to us. It is not easy seeing your only child go through this.”- M-9.

CHAPTER 5

DISCUSSION

5.1 Socio-demographic characteristics of patients and their parents

Data from this study shows that mothers were between the ages of 25-35 years. This reflects the fertility age range characteristics of the Ghanaian population. Most caregivers had education up to Junior High School**.** A significant positive relationship between higher education status with early presentation of anorectal malformations has been noticed by Lawal et al.[77]

The average weight for this study was 3.12 +/- 0.51Kg with (8)13% of the children being underweight. Variable weight distribution has been documented for the presentation of anorectal malformation. However, no statistical association has been found with the development of anorectal malformation. Birthweight has been linked to functional voluntary bowel outcomes after definitive repair. Makrufardi et al.[126] observed good functional outcomes associated with average birth weight than those with low birth weight.

Early intake of folic acid (before 6 weeks of gestation) has been associated with a reduction in congenital anomalies, including anorectal malformations.[33] In this study, most mothers (80.33%) did not take folic acid in the early stages of the development of the foetus but started antenatal clinic early (before 8 weeks). Statistically, analysis of its effects was beyond the scope of the study.

Sex distribution of anorectal malformation varies in the literature. This current study had a male preponderance (male: female ratio = 1:0.96). This aligns with some studies [8,92,127] while in dissonance with others[7,118].

5.2 Identification and Presentation of Anorectal Malformation

Early identification of anorectal malformation is crucial in managing anorectal malformations. Improper identification of the anomaly may lead to delayed presentation, which has a higher mortality rate.[3,128]

In this study, the majority (88.5%) of patients presented late (after 24hrs). The reasons given in this study by mothers were a general lack of awareness of the condition, and some referral bottlenecks. Only 29.5 %(18) of the defect were detected by health care professionals, with 70.5 %(43) of the cases being detected by the mother or close relatives. This is consistent with previous data in Nigeria. Adejuyigbe et al [6], in their review of children admitted to a Nigerian tertiary centre, noted that 86% of the children having anorectal malformations presented after 24 hours. Delays in presentations may be possible because of lack of awareness by caregivers, false assurance of normal anus when a child passes meconium, inadequate examination of the neonates after birth and the rarity of specific malformations. Delayed presentation of anorectal malformation is also common in Sub-Saharan

Africa.[6,128] . Similarly, a review of 263 children by Govender et al.[5] noted that 57.9% of the patients presented after 24hours. The problem of delayed presentation is peculiar to developing countries. Twenty-one percent of all children with anorectal malformations admitted to a Children's hospital in Ireland presented after 48hours after noticing malformations. Isolated cases of late presentation in adulthood are observed. Ogundoyin et al.[84] reported a case where a female adult was successfully managed in a Nigerian centre. One patient excluded from the study was 16- year old who presented with a perineal fistula. She had been battling with constipation her whole life and underwent a three stage procedure successfully.

Lack of awareness has been noted to be one of the significant contributory factors to delayed presentation. The majority (91.8%) of mothers were unaware of anorectal malformations in this study. This study agreed with the findings of Eltayeb et al.[129] and Mavrou et al.[130] Awareness of anorectal malformations contrasts with awareness of neonatal jaundice and sickle cell disease among mothers. (Lang et al., 2009) The most often cited reason for the low awareness is the relative rarity of anorectal malformations[77] Mavrou et al[130] have noted that higher income and better education are associated with awareness of anorectal malformations. These authors suggest that higher education and socio-economic factors are essential to enlightenment and access to media and other sources of information. In this study, only 11.48 % had higher education. It is therefore not surprising the low level of awareness among mothers in this study.

Statistical analysis done did not show any significant relationship between the sex, birth weight, time of presentation and when the malformation was detected.

The apparent lack of awareness is not only limited to mothers and caregivers. It reflects in the low percentage of detection (29.5%) by health professionals in this study. In this study, all the mothers delivered at a health facility with supervised skilled delivery by a midwife or a medical doctor either through a caesarean section or spontaneous vaginal delivery. However, most of the anorectal malformations were first observed by non-health professionals. This suggests that health care professionals do not actively look for congenital disabilities at birth. Turowski et al [78]also noted a similar pattern of non-health professionals being the first to notice anorectal malformations. The authors observed that 82.8% of children with anorectal malformations were born in the hospital.[78] 52.7% of these malformations were first detected by relatives after being discharged home. Some studies have also suggested home deliveries as a significant contributor to delayed presentation. [132]. This phenomenon was not observed in this study as all the deliveries occurred under skilled management in the health centre.

Despite observed late presentation in this study, the majority (55.74%) of the children with anorectal malformations presented in a stable state at the emergency department—this contrasts with other

studies where most of the children with delayed presentation presented with severe complications. There is still a high percentage of patients (32.79%) presenting with Intestinal obstruction.

Other contributory factors to the late presentation are rural location, delayed referral and financial constraints**.** Additional contributory factors were ignorance and the inability to afford hospital bills. Makanga et al [51] explained that hospitals were far from the rural settings where patients lived. This study agrees with the noted financial constraints in getting patients to tertiary centres for proper management.

Notably, none of the children in this study presented with any prior surgery at the peripheral health centre. Lawal[3] had cited worse outcomes with prior surgeries by general practitioners for correction of anorectal malformation as a significant factor in mortality.

The spectrum of presentation is expectedly gender biased. In this study, anorectal malformations with recto prostatic urethral fistula were predominant in males. This is consistent with observations in Ethiopia[133], Uganda[83], Nigeria[6] and Malawi[134]. The recto-vestibular fistula was the dominant type in this study, representing 63.3% of malformations in girls. This is consistent with the literature in sub-Saharan Africa.[6,83,133,134]

5.2 Screening Congenital Anomalies and other Diseases

Congenital anomalies are usually observed in anorectal malformations. It assumes clinical significance as prognosis depends on the severity and number of associated anomalies. [84,135]In the present study, associated anomalies were recorded in 11.5% of the patients, a figure lower than reported findings in the literature.[19,135] This disparity can be attributed to incomplete screening for the associated anomalies.

Notably, only 45.9% in this study were entirely screened. Screening varies by type of anorectal malformation.[58] Analysis of results from this study identified the primary reason for the apparent lack of complete workup to be physician's discretion in waiving the investigation. This is because most of the patients were fit to undergo the first surgery without these investigations. There appears to be the belief by practitioners that complete screening of patients with less severe forms of malformation is unnecessary because of the relatively low incidence in these groups.[58,136]

Furthermore, screening appeared to vary by organ system in this study. The spine was the most frequently screened part of the body (68.9%). This could be explained by the relatively cheap cost and availability of x-ray services in the hospital. Of particular note is the low screening for genitourinary anomalies in this study (57.4%), missing close to half of the patients although anorectal malformations have been observed to have nearly 50-80% association with gastrointestinal and urinary systems. [52] The rate of screening by echocardiogram was 62.3% of the cases. This is slightly higher than in other studies in Sub-Saharan Africa[3] but lower than reported in advanced

countries.[137]To limit the financial burden in the management of the condition, physicians in LMIC's choose to skip echocardiogram when there is no abnormal feature in the history or physical examination.

Identifying a cardiac pathology could be lifesaving. associated cardiac anomalies in anorectal malformations are associated with poor outcomes.[138] Screening for limb deformities was the least frequently done in this study. Limb deformities (Equino varus) identified in this study were noted on clinical examination. The patients were subsequently referred to the paediatric orthopaedic surgical team for management of the deformity.

Although screening permits the detection of structural deformities for later correction by orthopaedic surgeons, paediatric surgeons often waive it. Minneci et al [58] have suggested that because congenital limb deformities likely do not affect the decision to correct the anorectal malformation, they are not treated as urgent.

Furthermore, in explaining the reasons for lower congenital anomalies in Africa, some authors[6,12] have suggested that children with complex malformation have high mortality in the neonatal period.

5.3 Surgical Management

The initial surgical management of anorectal malformation is very crucial. Several approaches for correcting different types of anorectal malformation have been suggested in the literature.[103] There is an ongoing debate among surgeons on the best approach for corrective procedures for anorectal malformations.

The staged method of repair was used in this study. The three-stage procedure was the preferred method for repair of anorectal malformations in this study accounting for 93.63% of all cases. This is even though the majority presented in a stable state at the emergency. Colostomy anorectal malformation are mostly urgent procedures.[99,105] The argument favouring the three-staged repair of an anorectal malformation is that it affords better pre-operative anatomical knowledge; a child would have aged by then.[103] It also enables the performance of the distal colostogram in delineating fistulas.[90] The two-staged procedure has been described for a group of patients with anorectal malformation, where colostomy and definitive surgery are done in one operating session. The colostomy is taken down at another operating session. One (1) patient with a perineal fistula who presented on the 3rd day of life underwent the two-staged procedure in this study. Colostomy was returned 14 months later with no sequelae. No complications were noted with the colostomy or neo anus. The delay in closure of colostomy was as a result of financial challenges the parents had.

The literature discusses two main approaches: primary repair versus staged repair. The choice of repair depends on the type of anorectal malformation, availability of logistics and qualified personnel for anaesthesia and the surgical procedure.[52] The argument for primary repair is that it avoids the

complications of colostomy. Primary repair is growing interest as it is feasible for some selected low and intermediate anomalies[103]. Primary repair avoids the complications of colostomy[139].

It however presents unique challenges, like urogenital tract injuries, surgical site infections and wound dehiscence. Even though there were low types of anorectal malformation example the anorectal malformation with recto perineal fistula and anorectal malformation with rectovestibular fistula that could have met the criteria for a single-stage procedure, none was carried out because they came with sepsis and significant intestinal obstruction plus the lack of dedicated paediatric anaesthesiologists made it a prudent choice to do a stage procedure to ensure survival of the patient. Creating a stoma is often the first step in the three-stage procedure. It is done for all patients in whom faecal diversion is required. Divided colostomy was the colostomy of choice for all patients in this study. This was preferred over loop colostomy as it had better functional outcomes and less rate of prolapse and urinary tract infections.[7,99]

The main arguments against colostomy are its complications and socially unacceptability in specific populations[85,93]. The complication rates are high and associated with higher morbidity and mortality.[7] Complication rate observed by Mfinanga et al,[7] was 31.8%. A relatively lower complication rate (26.6%) after colostomy was recorded in this study. Most of the complications were superficial surgical site infections managed in the ward mainly by the stoma nurses. The colostomy type could explain the low complication rates. Divided colostomies rather than loop colostomies were done for these patients. Loop colostomies are associated with higher rates of complications[106,112,113]. Mfinanga et al[7] attributed a higher complications rate in their study to the majority being done by junior doctors. Colostomy revision was performed in 5% of the patients in this study. This is lower than the observed 8% of revision surgeries by Oda et al[106]. and 52.9% by Mfinanga et al.[7]. The time of reversal of colostomy after creation of the neo anus was 8 months in this study. Which is consistent with Mfinanga et al[7]. The unit policy is to reverse all colostomies in 3 months if patient is fit for surgery. However, the unique challenges in 2020, with the covid 19 pandemic, and some few institutional challenged lead to the delay that was observed.

Reoperation was done for two patients who had leaking anastomosis after closure of their colostomies. One succumbed to sepsis and the other survived. Both had a new colostomy re-sited. The one who survived had the colostomy reversed after 3 months. One patient had a revision of his PSARP done. He had anorectal malformation with a Rectobulbar fistula with a well-developed muscle sphincter complex. Patient was assessed and found to be incontinent of stool after the closure of colostomy. The neo anus on examination was patulous and a narrowing to the correct hegar dilator size made him continent. He was discharged 3 weeks after the revision surgery.

5.4 Psychosocial Aspects of the Management of Patients with an Anorectal Malformation

Aside from the financial burden of managing a child with an anorectal malformation, the experience is emotionally stressful for parents. Most studies concentrate on the psychosocial dimension in the growing child for whom repair has been done.[140,141,142]

There is a gap in the literature on the psychosocial effects on of parents during the initial management period. This study highlighted the stigma associated with having a child with anorectal malformation. Some caregivers were shunned by their families.

Prevailing cultural and societal beliefs could explain bad omens and associated curses. Stigma and the social withdrawal of colostomy were dominant themes in the qualitative analysis. Social withdrawal and stigmatisation have been documented in older patients having colostomies.[143]

However, the literature about caregivers' social withdrawal in the initial management phase of anorectal malformation is scanty. Social stigmatisation and social withdrawal of caregivers of children with colostomies impact their lifestyle. The withdrawal lasts until the colostomy is taken down.

Goudarzi et al[144] observed problems in raising children with anorectal malformations. These problems start early, especially with the care of the colostomy. Caring for a child with a colostomy can be stressful. Helping caregivers give colostomy care is essential in relieving some of the stress.[144] During the qualitative interviews, mothers stated they are less stressed about colostomy care because they were well educated and they applauded the support from the stoma care nurses. The impact of stress on caregivers in managing anorectal malformation has not been thoroughly investigated in sub-Saharan Africa. An objective assessment is thus warranted.

No formal psychological support was given to caregivers in this current study. The lack thereof can be explained by the few psychologists available and the lack of patient –support groups for children with this congenital anomaly.. Support to parents was through informal means, where those with prior experience encouraged caregivers. The importance of education and family support cannot be overemphasised.

5.5 Length of Stay

The average length of stay in the hospital from the time of admission to the time of creation of colostomy was found to be 12 days. The average length of stay from admission to discharge for the creation of the neo anus was found to be about 30 days. And the average length of stay in the hospital from the time of admission to the time of discharge was found to be about 18 days for patients who came for the reversal of their colostomy. The overall length of stay in the hospital for a patient undergoing a three stage procedure for an anorectal malformation is around 60 days. That is saying parents have to spend two months in the hospital for correctible surgeries to be done on their children.

Even though loss of productivity was not captured in this study, carers spending that amount of time in the hospital been unproductive at their workplace and not being at home causes significant strain on the families and may worsen the financial state of some carers. Lack of stoma care nurses at the peripheral facilities, trained personnel for stoma care, lack of disposable dilators, unavailability of peripheral centres where anal dilations can be done safely, and loss to follow up are some of the reasons that contributed to the prolonged length of stay of patients in this study.

5.6 The Financial Aspect of Treatment

Health is expensive and surgery more so. The Teaching Hospital allows a co-payment system with the NHIS which has gone long way to make surgery affordable to Ghanaians. In this study, most of monies paid were out of pocket payments for laboratory work, radiological investigations and top up payments for the NHIS. The parents paid almost an equivalent amount and sometimes slightly higher amount than the National health insurance scheme. (61.1% of the total amount of monies was out of pocket-payments for three-staged surgery for ARM). The average cost of treatment in Ghana cedis is GHC2995.69, GHC5031.83 and GHC3287.75 for having a colostomy, a PSARP and closure of colostomy respectively.

The study revealed that caregivers were disappointed in the co-paying system and were surprised by how much they had to pay to get their children operated on 18%(11) parents rated the amount they have to pay as high, 44%(27) of the parents said the amount was medium (high but expected) and the remaining 38%(23) said the amount was average. The cost of managing congenital malformations is expensive as indicated by Kovacic et al[124] in Wisconsin USA. In their review, an estimated $45.5 million was spent on managing patients with an anorectal malformation. Such comparisons are difficult to make to the Ghanaian system because of the difference in billing systems used. That said carers were able to source funds from relatives and various organisations to get treatment.

CHAPTER 6

CONCLUSION

Anorectal malformation is correctible. However, it challenges the paediatric surgeon and the mothers and guardians alike. We must continually assess how we are managing this group of patients and seek out interventions to make life easier for everyone involved.

6.1 Limitation

This was a cross-sectional study for only 8 months and did not consider the long-term complications associated with managing this malformation.

6.2 Recommendations

The following recommendations are made to health care providers, Komfo Anokye Teaching Hospital, and the National Health Insurance Authority.

Health practitioners

1. Thorough and detailed examination of the new born must be emphasised through periodic in-service trainings in all hospitals, clinics and health facilities especially among midwives, obstetric doctors and nurses and paediatricians. This will aid in identifying anorectal malformations early before complications develop.
2. There should be prompt referrals at the peripheral health facilities and efficient transport systems implemented to avoid complications

Komfo Anokye Teaching Hospital

1. There should be broader engagement in the facilitating the training of highly specialized personnel like paediatric anaethesiologist, intensivist and paediatric critical care nurses so that more two-stage or even one stage repair of ARM can be done for selected patients
2. The paediatric unit should be provided with dedicated psychosocial teams to help caregivers deal with the psychological aspects of the management of ARM.
3. More public education must be made about anorectal malformation and colostomies. This will reduce the stigma and social withdrawal associated with anorectal malformation and its treatment.

National Health Insurance Authority

1. The National Health Insurance Authority should review the tariffs to keep up with current cost of performing surgery for anorectal malformation. This would reduce the out-of-pockets payments made by caregivers.

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APPENDICES

Appendix I: Statement of Consent

Information Sheet and Informed Consent Form

Consent to participate in a prospective study titled: “Types, presentation and challenges with initial management of anorectal malformations at Komfo Anokye Teaching Hospital, Kumasi:”

This section should be read by or to caregivers or legal representatives of participants before enrolment into the study.

The purpose of the study is to know the types, presentations and initial management challenges of ARM in KATH.

Our study would enrol at least 57 participants. This consent form should be read to/by you carefully, and you should take your time to understand and then agree to participate. After the study has been fully explained to you, and if you agree to allow your ward to participate, you will be asked to sign this consent form to indicate your willingness to permit your child's participation in this study.

What do I have to do if I decide to participate?

If you agree to allow your child to participate, we will ask for your child's name, age, sex, and current illness and then conduct a physical examination of the child before surgery. As long as the child stays in the hospital after the surgery, examinations would be done whenever appropriate.

What about my right to decline/withdraw from the study?

You are free to refuse to participate in the study or withdraw your child from the study at any time without giving a reason. This will not affect the care of your child, nor would it result in any penalty or loss of benefits to which your child is entitled.

Whom do I contact if I have questions?

If you have any questions, you may ask them now or later. If you wish to ask questions later, you may contact Dr Fareeda Agyei on 0206836404.

Will my information be kept confidential?

All study records will be kept confidential. Access to computerised and paper files will be secured and only accessible to the investigator. Auditors, ethical review boards and regulators may be granted access to your child's information if necessary.

Parents/Guardians Consent Form

Study Title: Types, presentation and challenges with initial management of anorectal malformations at Komfo Anokye Teaching Hospital, Kumasi

Investigator: Dr Fareeda Agyei

Child's name: ……

This study has been explained to me by:

Dr/Mr/Mrs/Ms. ……...

1. I confirm that I understand the information on the information sheet. This has been explained to me, and I have had the opportunity to ask questions.

2. I understand that my child's participation is voluntary, and I am free to withdraw from the study at any time, without giving reasons and without my child's medical care or legal rights being affected.

3. I understand that the study's investigator would have access to my child's medical records. Some portions of the records may be made available to regulatory authorities if deemed necessary and appropriate. I permit these individuals to have access to those records.

4. I agree for my child to participate in the above study.

Name of Parent/Guardian/Legal Representative: ……

Signature/(R) /Thumbprint: ……

Telephone number: …….

Date: ……

Name of the person administering the consent…

Signature: ……..... Date.....

Appendix II: Data Sheet

QUANTITATIVE STUDY

TYPES, PRESENTATION AND CHALLENGES WITH THE INITIAL MANAGEMENT OF ANORECTAL MALFORMATION IN A TERTIARY TEACHING HOSPITAL KATH, KUMASI, GHANA

SECTION A

DEMOGRAPHY OF PATIENT

This section describes the demographic characteristics of parents.

(Please ask mother to bring along her antenatal card)

1. Name
2. Age
3. Current weight (kg):
4. Birth weight…….
5. sex: a. male b. female
6. I. gestational age @ birth
7. ii. Patient was born at :

a. term b. pre-term c. post-term

8. mode of delivery a. SVD b. assisted VD c. caesarean section
9. day of discharge from health facility a. same day b… detained/admitted

SECTION B

DEMOGRAPHY OF PARENTS

This section de-scribes the demographic characteristics of both parents (if present)

1. Age of mother.

2. Age of father
3. Occupation of mother…….
4. Occupation of father…….
5. The educational level of the mother.
    a. Uneducated
    b. Primary school
    c. Junior high school
    d. secondary school
    e. tertiary education
6. The educational level of the father…….
    a. Uneducated
    b. Primary school
    c. Junior high school
    d. secondary school
    e. tertiary education
7. Did the mother take folic acid routinely before pregnancy? A) yes B). No
8. When was the booking of pregnancy made?
    a. Before 8 weeks
    b. After 8 weeks
9. If after 8 weeks, when was it done

    Please state weeks……..
10. Did the mother routine take all the antenatal medications?

    a. Yes

    b. No
11. If no, what was the reason

……..…

……..

12. Did the mother have any pregnancy-related conditions?

    a. Yes

    b. No

13. If yes, please state the condition…. ……..

14. Has either parent heard of an ARM before the presentation

    a. Yes

    b. No

15. If yes, what were you told (please summarise)

    …….

    ……..

    …….

SECTION C

PRESENTATION AT THE HOSPITAL

This section describes the timing and mode of presentation of the patient to the hospital

1. Day of presentation. …….. (in days or months)
2. Mode of presentation ……
    a. Stable
    b. Intestinal obstruction
    c. Sepsis
    d. Vomiting
    e. Other ……

3. When was the malformation detected? (Please state in days) …….

4. Who noticed the malformation?

    a. Health personnel

    b. Mother

    c. Other (PLEASE STATE) …….

5. Was there a delay (if a patient presents after 24 hours)?

    a. Yes

    b. No

6. What was the cause of the delay?

    a. Late detection

    b. Referral issues

    c. Financial constraints

    d. Other (please state) …….

7. What was the type of ARM that the patient presented with

    i) Male presentation

        A. recto-bladder fistula

        B. recto-bladder neck fistula

        C. recto bulbar fistula

        D. arm with recto-prostatic urethral fistula

        E. arm with recto-perineal fistula

        F. arm without fistula

        G. other please specify …

    ii). Female presentation

        A. recto-perineal fistula

        B. recto-vestibular fistula

        C. arm with recto-vaginal fistula

D. arm without fistula

E .persistent cloaca

F. other please state……

SECTION D

This section describes the pre-operative assessment and investigations.

Screening for Associated anomalies…

1. Was a vacterl anomaly workup done? Please tick the ones that were done for the patient. (answer can be more than one)

A. X-ray of spine vertebrae
B. Cross-table lateral X-ray
C. KUB ultrasound scans
D. Echocardiogram
E. N-G tube was passed/patient could feed
F. Other (specify) ……..…

2. Can you explain why the vacterl anomaly screen was not done?

A. Financial
B. Broken down equipment
C. Lack of technical personnel
D. Other….(PLEASE STATE)

3. Did the patient have another congenital abnormality (apart from vacterl anomaly)

a. Yes (if yes, please specify ……)

b. No

4. I) did the patient have any syndrome?

a. Yes (if yes, name the syndrome ……)

b. No

SECTION E SURGERY

Section E provides details on the surgical procedure done. It is subdivided according to the type of repair done by the surgeon.

1. When was the first surgery done?

a) Day of admission

b) 24 to 48 hours

c) 48 -72 hours

d) After 72 hours

2. If surgery was not done on the same day, what was the reason?

a) Labs work/radiological workup not ready

b) Lack of funds to purchase medications

c) Refusal to consent to surgery

d) Lack of theatre space

e) Other

3. Type of correction?

a. Primary repair

b. Staged repair (diverting colostomy)

4. Type of surgery?

a. Emergency

b. Elective

SECTION E I. PRIMARY REPAIR

I. Did the patient receive parenteral nutrition after primary repair?

a. Yes

b. No

II. The patient was anaesthetised by……

a. Doctor anaesthesiologist

b. Nurse Anaesthesist

III. Type of anaesthesia administered……

a. General

b. Spinal

c. Local

IV. Was there any anaesthetic complication?

a. Yes

b. No

V. If yes, pleases select the type of anaesthetic complication?

a. Drug reaction

b. Dislodged tube

c. Cardiac arrest

d. Airway oedema

e. Other (please specify……)

VI. Where was the patient taken to after surgery?

A. Ward B3……

B. Mother and baby unit

C. Neonatal or Paediatric intensive care unit

D. Other ……..

VII. Was there any surgical complication after surgery?

a. Yes

b. No

VIII. If yes, please specify the type of complication?

a. Breakdown

b. Retraction

c. Surgical site infection

d. Sepsis

e. Other

IX. Was there a need for a redo surgery after primary repair?

a. Yes

b. No

X. If yes, please state the reason for the redo surgery; …….

XI. What was the duration of treatment from the time of admission to discharge? ……

XII. What is the interval between admission and primary repair? ……

XIII. How long did you stay in the hospital after primary repair? (State in days) ……

XIV. How much did the NHIS pay? ……

XV. How much did you pay as top up to the NHIS? …….

XVI. Were you able to pay?

a. Yes

b. No

XVII. If not, were you……

a. Detained

b. Discharged

c. Referred to the social welfare clinic

d. Other

XVIII. Total cost of primary repair? ……

SECTION E: II STAGED REPAIR

1. Type of surgery?

a. Emergency

b. Elective

2. If staged repair, what was the stage?

a. 2 stage (colostomy + psarp) and reversal of colostomy

b. 3 stage (colostomy + psarp+ reversal of colostomy)

SECTION E III;

A) Two staged repair

1. a. (colostomy+ psarp) was done by……..

a. Junior resident

b. Senior resident

c. Fellow

2. Closure of colostomy was done by ……..

a. Junior resident

b. Senior resident

c. Fellow

3. (Colostomy + psarp) was anaesthetised by……

a. Doctor anaesthesiologist

b. Nurse Anaesthesist

4. Reversal of colostomy was anaesthetised by…….

a. Doctor anaesthesiologist

b. Nurse Anaesthesist

5. Type of anaesthesia

a. General

b. Spinal

c. Local

6. Was there any anaesthetic complication?

a. Yes

b. No

7. If yes, pleases select the type of anaesthetic complication?

a. Drug reaction

b. Dislodged tube

c. Cardiac arrest

d. Airway oedema

e. Other (please specify……)

8. Where was the patient taken to after surgery?

A. Ward

B. Mother and baby unit

C. Neonatal or paediatric intensive care unit

D. Other……

9. Was there any surgical complication after (colostomy + psarp)?

a. Yes

b. No

10. If yes, please specify……

I) Colostomy complications

a. Superficial surgical site infection

b. Deep surgical site infection

c. Skin excoriation

d. Prolapse

e. Retraction

f. Other ……..

II) Psarp complications

a. Breakdown

b. Superficial surgical site infection

c. Retraction of neo-anus

d. Other……

11. Was there a need for a redo surgery after (colostomy + psarp)?

a. Yes b. No

12. If yes, what was the indication for the redo surgery?

……

……

……

13. Was there any complication after closure of colostomy?

a. Yes

b. No

14. If yes

a. Superficial surgical site infection

b. Breakdown (anastomosis)

c. Deep surgical site infection

d. Other

15. Was there a need for a redo surgery after closure of colostomy?

a. Yes

b. No

16. If yes, what was the indication for the redo surgery?

……

……

……

17. What is the treatment duration from admission to (creation of colostomy and neo-anus?)

……..

18. What is the interval between the creation of (colostomy and neo-anus) and closure of colostomy? …….

19. How long did you stay in the hospital after the closure of the colostomy? ……

20. How much did the NHIS pay for the creation of (colostomy and psarp)? ……

21. How much did the NHIS pay for the closure of colostomy? ……

22. How much did you pay as top up to the NHIS for the creation of (colostomy and psarp)?

……

23. How much did you pay as a top-up to the NHIS for colostomy closure? ……

24. Were you able to pay?

a. Yes

b. No

25. If not, were you

a. Detained

b. Discharged

c. Referred to solicit funds

d. Other (please specify ……..…)

26. If you were detained, where did you solicit your funds from?

a. Social welfare

b. NGO

c. Other (please specify ……..…)

27. Total cost of stage 2 repair? ……

SECTION E II: C; THREE-STAGE REPAIRS

1. Colostomy was done by……..

a. Junior resident

b. Senior resident

c. Fellow

2. Patient was anaesthetised by…….

a. Doctor anaesthesiologist

b. Nurse Anaesthesist

3. Type of anaesthesia

a. General

b. Spinal

c. Local

4. Was there any anaesthetic complication?

a. Yes

b. No

5. If yes, pleases select the type of anaesthetic complication?

a. Drug reaction

b. Dislodged tube

c. Cardiac arrest

d. Airway oedema

e. Other (please specify……..)

6. Where was the patient taken to after surgery?

A. Ward B3

B. Mother and baby unit

C. Neonatal /paediatric intensive care unit

D. Other ……

7. Was there any surgical complication after surgery?

a. Yes

b. No

8. If yes, please select the type of complication:

a. Superficial surgical site infection

b. Breakdown (anastomosis)

c. Deep surgical site infection

d. Other

9. Was there a need for a redo surgery after the creation of colostomy?

a. Yes

b. No

10. If yes, what was the indication for the redo surgery?

……

……

……

11. What is the treatment duration from admission to discharge from the hospital after the creation of the colostomy? …….

12. How much did Nhis pay from admission to discharge? ……

13. How much did you pay as top up to the NHIS payment? ……

14. Were you able to pay?

a. Yes

b. No

15. If no, were you……

a. Detained

b. Discharged

c. Referred to the social welfare clinic

d. Other (Please specify…….)

16. PSARP was done by……..

a. Junior resident

b. Senior resident

c. Fellow

17. Anaesthesia for the Psarp procedure was done by …….

a. Doctor anaesthesiologist

b. Nurse Anaesthesist

18. Type of anaesthesia

a. General

b. Spinal

c. Local

19. Was there any anaesthetic complication?

a. Yes

b. No

20. If yes, please select the type of anaesthetic complication?

a. Drug reaction

b. Dislodged tube

c. Cardiac arrest

d. Airway oedema

e. Other (please specify……)

21. Where was the patient taken to after surgery?

A. Ward B3

B. Mother and baby unit

C. Neonatal or paediatric intensive care unit

D. Other ……

22. Was there any surgical complication after the creation of neo-anus?

a. Yes

b. No

23. If yes, please select the type of complication;

a. Breakdown

b. Superficial surgical site infection

c. Retraction of neo-anus

d. Other ……

24. Was there a need for a redo surgery after the creation of neo-anus?

a. Yes

b. No

25. If yes, what was the indication for the redo surgery? ……

26. What is the interval between the creation of colostomy and neo-anus? ….……

27. How long did you stay in the hospital after the creation of neo-anus? ……

28. How much did NHIS pay for the creation of neo-anus? ……

29. How much did you pay as top up for the creation of neo-anus? ……

30. Were you able to pay?

a. Yes

b. No

31. If no, were you……

a. Detained

b. Discharged

c. Referred to the social welfare clinic

d. Other (please specify ……..)

32. Reversal of colostomy was done by……..

a. Junior resident

b. Senior resident

c. Fellow

33. Anaesthesia was given by …….

a. Doctor anaesthesiologist

b. Nurse Anaesthetist

34. Type of anaesthesia

a. General

b. Spinal

c. Local

35. Was there any anaesthetic complication?

a. Yes

b. No

36. If yes, pleases select the type of anaesthetic complication?

a. Drug reaction

b. Dislodged tube

c. Cardiac arrest

d. Airway oedema

e. Other (please specify ……..…)

37. Where was the patient taken to after reversal of colostomy?

a. Ward B3

b. M.B.U

c. N.I.C.U

d. Other …….

38. Was there any complication after closure of colostomy?

a. Yes

b. No

39. If yes, please specify……

a. Superficial surgical site infection

b. Breakdown (anastomosis)

c. Deep surgical site infection

d. Other

40. Was there a need for a redo surgery after reversal of colostomy?

a. Yes

b. No

41. If yes, what was the indication for the redo surgery? ……..……

42. What is the duration of treatment from the creation of neo-anus to reversal of colostomy?

…….

43. How long did you stay in the hospital for the reversal of colostomy? ……..

44. How much did NHIS pay for the reversal of colostomy? …...……

45. How much did you pay as top up to the NHIS for the reversal of colostomy? ……

46. Were you able to pay?

a. Yes

b .No

47. If no, were you……

a. Detained

b. Discharged

c. Referred to the social welfare clinic

d. Other (please specify ……)

48. Total cost of 3 stage repair? …….

SECTION F. DELAYS IN MANAGEMENT

This section describes the problems encountered during the surgical procedure

1. Did any of your surgical treatment delays

    a) Yes

    b) No

2. If yes, what accounted for it

    a) Financial constraints

    b) Covid crises

    c) Institutional challenges ….please tick where appropriate

        i) Broken-down anaesthetic equipment

        ii) Theatre fumigation

        iii) Non-available surgeon /theatre staff

        iv) Other

3. Which part of your surgical management was delayed?

        a) Creation of colostomy

        b) PSARP

        c) Reversal of colostomy

        d) PSARP + colostomy

        e) Primary repair

4. How long did you have your colostomy…….. (Please state in months if the patient has a colostomy)

5. How long have you had your colostomy……. (Please state in months if colostomy is yet to be reversed)

6. State the challenges you had, or you have with the colostomy

……

……

.....

7. Was any of your surgeries rescheduled?

a) Yes b) No

8. If yes, what was the reason?

a) Institutional challenges

B) COVID crises

c) Patient was not fit

d) Other (please specify ……)

9. How many times was your surgery rescheduled?

A) Once

B) Twice

C) 3 or more

10. If you were to rate the cost of managing your child/ward with an anorectal malformation, what would be your rate.

A) Normal (easy to pay)

B) Medium (costly but manageable)

C) High (very costly … challenging to manage)

QUALITATIVE STUDY

SEMI-STRUCTURED INTERVIEW GUIDE

Study Title: Types, presentation and challenges with initial management of anorectal malformations at Komfo Anokye Teaching Hospital, Kumasi

Investigator: Dr Fareeda Agyei

Child's name: ……

This study has been explained to me by:

Dr/Mr/Mrs/Ms. ……...

1. I confirm that I understand the information on the information sheet. This has been explained to me, and I have had the opportunity to ask questions.

2. I understand that my child's participation is voluntary, and I am free to withdraw from the study at any time, without giving reasons and without my child's medical care or legal rights being affected.

3. I understand that the study's investigator would have access to my child's medical records. Some portions of the records may be made available to regulatory authorities if deemed necessary and appropriate. I permit these individuals to have access to those records.

4. I agree for my child to participate in the above study.

Name of Parent/Guardian/Legal Representative: ……

Signature/(R) /Thumbprint: ……

Telephone number: …….

Date: ……

Name of the person administering the consent: …….

Signature: ……... Date .....

Section A. Background Information

Interview ID

Date of Interview

Gender

Age

Level of Education

Marital Status

Interview Guide:

Introductory component of in-depth interview

Greet and start with a brief introduction of yourself and the study, then relay it to the participant

Probe for the following:

1. How the condition was first noticed?

2. What was done for the patient?

3. How the referral of the patient was done

4. What has been done for the patient so far?

5. Challenges with the financial aspect of the management and how they are managing currently?

6. Did the participant have challenges with managing the colostomies? How did you overcome it?

7. Did the participant face any stigmatisation and isolation?

8. What other issues were worrisome or bothersome to you?

9. Was your surgery rescheduled and why?

10. Advice, lessons learnt and encouragement for other mothers

Appendix III: Ethical Clearance

KWAME NKRUMAH UNIVERSITY OF SCIENCE AND TECHNOLOGY
COLLEGE OF HEALTH SCIENCES
SCHOOL OF MEDICAL SCIENCES / KOMFO ANOKYE TEACHING HOSPITAL
COMMITTEE ON HUMAN RESEARCH, PUBLICATION AND ETHICS

Our Ref: CHRPE/AP/370/20 — 15th October, 2020.

Dr. Fareeda Agyei
Department of Paediatric Surgery
Komfo Anokye Teaching Hospital
Post Office Box 1934
KUMASI.

Dear Madam,

LETTER OF APPROVAL

Protocol Title: ***"Types, Presentation and Challenges with the Management of Anorectal Malformation in a Tertiary Teaching Hospital (KATH)."***

Proposed Site: ***Komfo Anokye Teaching Hospital.***

Sponsor: ***Principal Investigator.***

Your submission to the Committee on Human Research, Publications and Ethics on the above-named protocol refers.

The Committee reviewed the following documents:

- A notification letter of 15th July, 2020 from the Komfo Anokye Teaching Hospital (study site) indicating approval for the conduct of the study at the Hospital.
- A Completed CHRPE Application Form.
- Participant Information Leaflet and Consent Form.
- Research Protocol.
- Questionnaire.

The Committee has considered the ethical merit of your submission and approved the protocol. The approval is for a fixed period of one year, beginning **15th October, 2020** to **14th October, 2021** renewable thereafter. The Committee may however, suspend or withdraw ethical approval at any time if your study is found to contravene the approved protocol.

Data gathered for the study should be used for the approved purposes only. Permission should be sought from the Committee if any amendment to the protocol or use, other than submitted, is made of your research data.

The Committee should be notified of the actual start date of the project and would expect a report on your study, annually or at the close of the project, whichever one comes first. It should also be informed of any publication arising from the study.

Thank you, Madam, for your application.

Yours faithfully,

Rev. Prof. John Kuupiel Pobee.
Honorary Secretary
FOR: CHAIRMAN

Room 7 Block J, School of Medical Sciences, KNUST, University Post Office, Kumasi, Ghana
Phone: +233 3220 63248 Mobile: +233 20 5453785 Email: chrpe.knust.kath@gmail.com / chrpe@knust.edu.gh

Appendix IV: Gazette – Evidence of Name Change

GHANA GAZETTE

REPUBLIC OF GHANA

Published by Authority

No. 85 FRIDAY, 4TH SEPTEMBER 2015

CONTENTS

CHANGE OF NAMES—*contd.*

8712. **Miss Nyefele Rose,** *a.k.a.* **Miss Rose Ama Nyefele,** a Student and of P.O. Box 98, Nsawam, wishes to be known and called **Miss Rose Ama Nyefele** with effect from 12th August, 2013. All documents bearing her former names are still valid.

8713. **Mr. Abubakari Nasiru,** *a.k.a.* **Mr. Moussa Aboubakari,** a Welder of Faccine SRL, Italy and of P.O. Box 156, Kwashieman, Accra, wishes to be known and called **Mr. Moussa Aboubakari** with effect from 2nd September, 2015. All documents bearing his former names are still valid.

8714. **Miss Addo Jemima Oparebea,** *a.k.a.* **Miss Ewurakua Addo,** a Secretary of Gloryship Services and of H/No. 1/N72, Site 16, Community 1, Tema, wishes to be known and called **Miss Ewurakua Addo** with effect from 3rd August, 2015. All documents bearing her former names are still valid.

8715. **Mr. Openti Odei-Lartey,** an Unemployed and of H/No. Plot 91, Block BX, Ejisu-Besease, wishes to be known and called **Mr. Kwadwo Openti Odei-Lartey** with effect from 3rd September, 2015. All documents bearing his former name are still valid.

8716. **Mr. Gustav Mikki Anku,** an Electrician of One-G Enterprise and of P.O. Box WJ 53, Weija, Accra, wishes to be known and called **Mr. Selorm Komla Anku** with effect from 3rd September, 2015. All documents bearing his former name are still valid.

8717. **Miss Juniter Afia Antwi,** a Seamstress and of H/No. D. 352, Ashaiman, Accra, wishes to be known and called **Mrs. Juniter Afia Sam** with effect from 18th April, 2001. All documents bearing her former name are still valid.

8718. **Miss Priscilla Thompson,** a Hairdresser and of H/No. PT 90, Parescoa, Bakakyir, Secondi, wishes to be known and called **Mrs. Priscilla Kulasie** with effect from 19th August, 1998. All documents bearing her former name are still valid.

8719. **Miss Salma Alhassan Ali,** *a.k.a.* **Miss Salma Alhassan,** a Trader, c/o Alima Ahamed, P.O. Box 914, Tema, wishes to be known and called **Miss Salma Alhassan Ali** with effect from 3rd December, 2014. All documents bearing her former names are still valid.

8720. **Miss Elizabeth Mensah,** an Interior Decor and of P.O. Box 2724 and of H/No. 1, Kaneshie, Accra, wishes to be known and called **Mrs. Elizabeth Mensah Sheidu** with effect from 15th December, 1990. All documents bearing her former name are still valid.

8721. **Mr. Morsindi Nnamelim Chukwuemeka,** *a.k.a.* **Mr. Morsindi N. Chukwuemeka,** *a.k.a.* **Mr. Jude Yao Emmy Dzakpata,** a Businessman and of H/No. 44, Volta Avenue, East Kwabenya, Legon, Accra, wishes to be known and called **Mr. Morsindi Nnamelim Chukwuemeka** with effect from 3rd September, 2015. All documents bearing his former names are still valid.

8722. **Miss Gifty Asare,** a Public Health Officer of St. John's Ambulance, c/o Liberty Centre A/G Church, P.O. Box AN 12242, Accra, wishes to be known and called **Mrs. Gifty Asante Wiafe** with effect from 29th December, 2013. All documents bearing her former name are still valid.

8723. **Mr. Seth Benney Tsiase,** a Graphic Designer of Grand Grapix and of P.O. Box 2448 and of H/No. AX 29, Community 7, Tema, wishes to be known and called **Mr. Seth Benney Onyame Tsiase** with effect from 3rd September, 2015. All documents bearing his former name are still valid.

8724. **Mr. Phreeman Osei-Bonsu Bismark,** *a.k.a.* **Mr. Phreeman Bismark Osei-Bonsu,** *a.k.a.* **Mr. Bismark Osei-Bonsu,** *a.k.a.* **Mr. Bismark Osei Bonsu,** a Fraud Examiner/Investigator of Quemic Ghana Limited/Bluesec and of P.O. Box KN 904, Kaneshie, Accra, wishes to be known and called **Mr. Phreeman Osei-Bonsu Bismark** with effect from 3rd September, 2015. All documents bearing his former names are still valid.

8725. **Dr. Fareeda Agyei,** a Medical Doctor with Staff No. 701336 of Komfo Anokye Teaching Hospital and of H/No. 25, Odencho, Kwadaso, Kumasi, wishes to be known and called **Dr. (Mrs.) Fareeda Galley** with effect from 10th April, 2015. All documents bearing her former name are still valid.

8726. **Miss Eunice Accam,** a Businesswoman and of H/No. C 27, Sakumono, Tema, wishes to be known and called **Mrs. Eunice Accam Seade** with effect from 25th April, 2015. All documents bearing her former name are still valid.

8727. **Miss Adriana Arhu,** a Student Nurse of Western Hills School of Nursing, Ofankor, Accra and of H/No. V33, Community 8, Tema, wishes to be known and called **Mrs. Adriana Hoenyedzi** with effect from 5th March, 2015. All documents bearing her former name are still valid.

8728. **Miss Regina Quansah,** a Cashier of Electricity Company of Ghana, Accra and of H/No. Block B, 13D, Akuse, Eastern Region, wishes to be known and called **Mrs. Regina Dzogbetsah** with effect from 8th August, 2015. All documents bearing her former name are still valid.