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Research Article

Outcomes of Anorectal Malformations in Northern of Ghana 6 years of Studies

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Abstract

The study conducted a retrospective and prospective analysis of 22 patients diagnosed with anorectal malformation between January 2009 and June 2016, continuing until June 2020, in the surgical departments of the Regional Hospitals of Sunyani, Wa, and Tamale Teaching Hospital in northern Ghana. The findings revealed that 59.1% of cases exhibited low birth weights, with a predominance of male cases. Associated malformations were present in 35.2% of cases, affecting the gastrointestinal and skeletal systems. High anorectal malformation was observed in 37% of male cases. The most frequently performed surgical technique was posterior sagittal anorectoplasty and 50% of the children underwent surgery before their first birthday. Postoperative complications were recorded in 31.2% of cases, with a mortality rate of 19.1%. In conclusion, the overall outcomes were deemed satisfactory.

Introduction

Anorectal malformations, congenital anomalies of the gastrointestinal tract, necessitate meticulous surgical evaluation [1]. Historical records dating back to approximately thirteen centuries reveal early awareness of this condition. Paulus Aegineta, in ancient Greece, documented the initial surgical correction of these malformations, which was followed by dilations employing a spark plug-a method that remained effective until modern times [2,3].

Current global statistics indicate that one in every 5000 live births is afflicted with anorectal malformations, with a higher prevalence among males, constituting 58% of cases. Among these children, 72% present with intermediate or high-grade malformations, which pose greater complexity in terms of resolution [2].

Over the past two decades, pediatric surgery has achieved remarkable progress. In the latter half of the 20th century and into the early years of the 21st century, technological advancements, such as the introduction of the electrostimulation method and the utilization of laser technology alongside measurement tools for assessing tone and motility, have significantly improved the management of these malformations in both neonates and infants. By the 1980s, Alberto Rock and Devries pioneered the posterior sagittal anorectoplasty as the primary treatment for high anomalies, revolutionizing the approach to these conditions [3]. Anorectal malformations represent a clinically significant entity, the management of which has greatly benefited from technological advancements. Early and precise diagnosis, facilitated by these developments, has led to improved treatment outcomes.

The progress in neonatal surgery and the promising results achieved in the treatment of anorectal malformations are emblematic of the quality of surgical care provided by specialized pediatric surgeons. Consequently, we have undertaken a study to scrutinize the behavior and clinical management of anorectal malformations. This investigation aims to elucidate the key clinical aspects and therapeutic outcomes associated with anorectal malformations within the scope of our study period.

Method

This study was conducted in the departments of General Surgery at the Regional Hospital Sunyani, Regional Hospital Wa, and Tamale Teaching Hospital in the northern region of Ghana. The study period spanned from November 2008 to November 2010, and then from June 2016 to June 2020, with the aim of comprehensively understanding the behavior and outcomes of anorectal malformations. In preparation for this research, prior coordination was established with the respective departments and record-keeping units of the aforementioned hospitals to facilitate the collection of primary data.

Methodological approach

This study is characterized as descriptive, cross-sectional, and incorporates both retrospective and prospective elements to accomplish its defined objectives. The study population comprised 22 patients, representing 100% of those under the care of pediatric surgery specialists across the different hospitals.

To gather general demographic data, information regarding the sex and birth weight of the subjects was extracted from their respective clinical records. Birth weight was categorized according to the guidelines outlined in the Ghana Pediatrics Manual. Additionally, infants weighing less than 1500 grams at birth were classified as very low birth weight.

Prenatal history related to anorectal malformations was assessed through a thorough examination of clinical records. Prenatal ultrasounds and alpha-fetoprotein studies were not considered in this

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evaluation, as they were not consistently conducted as part of prenatal care.

The presence of associated malformations was determined based on information available in the clinical records. This assessment encompassed malformations of the alimentary canal and other organ systems, excluding rectourinary and rectovaginal fistulas, which were classified under genitourinary malformations. In cases where a cardiovascular anomaly was suspected, a chest X-ray and, if available, an echocardiogram was prescribed to provide a more comprehensive diagnosis.

In order to diagnose genitourinary system malformations, a comprehensive evaluation was conducted, which included renal ultrasound, intravenous pyelogram, and distal colostogram. The diagnosis of anorectal malformations was determined in accordance with the anatomical classification outlined by Wingspread [2,4,5]. This diagnosis was established through a combination of physical examination, radiographic studies including invertogram and prone cross-table lateral X-ray, as well as macroscopic peri-operative findings.

Technical and procedures

Data collection techniques:

- Scientific information for this study was gathered through extensive bibliographical research involving scientific articles and specialized books in the field.
- Primary data was obtained from clinical records and operative reports of patients who underwent surgery for anorectal malformations during the specified study period. A dedicated form, provided as an annex to this study, was utilized for this purpose.

Data analysis and processing:

- Data analysis was conducted utilizing the statistical method of simple tabulation. The results are presented as percentage values and depicted in graphical representations.

Discussion and synthesis technique:

- The objectives of the study were justified, and a thorough discussion was undertaken, leading to the formulation of conclusions and the provision of relevant recommendations. This process was crucial in achieving the goals of the study.

Result and Discussion

In the period under analysis, a total of 22 cases of anorectal malformations were identified, constituting 100% of the morbidity in the regions studied. This observation is attributed to the presence of a specialized pediatric surgeon with expertise in neonatal surgery in these regions. Throughout this same period, there were a total of 114,655 live births, resulting in an incidence rate of 1 in every 5,211 live births for anorectal malformations. This incidence rate aligns with findings reported in global literature [2,6,7].

The significance of antenatal pathological history in the context of anorectal malformations is detailed in Table 1. It is noteworthy that only 36.3% of neonates with anorectal malformations had notable prenatal antecedents. However, it is crucial to acknowledge that while this is a meaningful index, it does not conclusively establish a direct causal relationship with the specific malformation under study.

Upon reviewing the literature, we did not find notable antenatal antecedents, except for a potential genetic transmission, indicated by the presence of affected members spanning up to the third generation [2,3,8].

In our cases, we were unable to establish a clear pattern of familial recurrence of the condition. The most frequently recurring antecedent was urinary tract infections, a common occurrence in pregnant women. As such, we do not attribute significant importance to it in the genesis of anorectal malformations.

The distribution of patients with anorectal malformations based on birth weight is presented in Table 2. Notably, 59.1% of the patients were born underweight, signifying a considerable difference from recently eutrophic newborns. Additionally, two patients were premature, suggesting that in some instances, anorectal malformations may contribute to low birth weight, akin to malformations of the digestive apparatus.

In the extensive literature we reviewed, we did not find any studies that specifically correlated birth weight with anorectal malformations. Consequently, we are unable to make direct comparisons with existing research. However, it is worth noting this observation, as it may hold importance for future studies.

Table 2 provides an overview of the patient distribution by gender, with males comprising the majority at 54.5%. This represents a slight but statistically insignificant difference compared to the female population. These findings align with similar studies in the field [2,3,6,9,10].

The distribution of associated malformations is presented in Table 3, highlighting the prevalence of gastrointestinal and skeletal anomalies in our study, each observed in six patients with associated malformations. It is crucial to note that only seven patients (31.8%) exhibited associated malformations. Among digestive system anomalies, the most common association was between esophageal atresia and anorectal malformations, including one patient diagnosed with the VATER association.

In relation to associated malformations, the literature shows variability in their incidence and the systems affected. Some studies suggest that 50% to 60% of patient's exhibit associated malformations, with 30% of these pertaining to the genitourinary system [1].

It is generally believed that the genitourinary and gastrointestinal systems are the most commonly affected [1,2,11-13]. However, our findings deviate from this trend, as we observed associated malformations in only 31.8% of cases, and genitourinary anomalies were diagnosed in only 3 patients, accounting for 17.8% of the

Table 1: Antenatal care.

Antenatal	Frequency	%
With antenatal history	8	36
Without antenatal history	14	64
Total	22	100

AH.*antenatal history

Table 2: Distribution of patients of anorectal malformation per the new born weight.

New born weight/g	Frequency	%
-1.449	2	9.10%
1500-2449	11	50
2500	9	40.9
Total	22	100

Table 2A: Distribution of Patients with anorectal malformations per sex.

Sex	Frequency	%
Male	12	55
Female	10	46
Total	22	100

associated malformations.

The classification and distribution of anorectal malformations based on specific diagnoses are presented in Table 4. Notably, there was a higher incidence of high anomalies, particularly in males, accounting for 37% of cases. Additionally, in female patients, intermediate and low anomalies were significant, representing 18% of cases.

In males, the most common type of fistula was rectourethral, observed in 5 out of 12 cases within this gender group. Conversely, in the group of malformations without urethral involvement, there were 3 patients. Among the 10 female patients, recto-vestibular fistulas were the most frequently observed, accounting for 4 cases, while only one case lacked a fistula.

Our findings align with similar results reported in a series conducted in Liverpool from 1953-1975 [14]. Specifically, in regard to rectourethral fistulas, they constituted the most common anorectal anomaly, corroborating findings from other studies [2,3,10,15-17] and confirming our own results.

Regarding the malformations identified in females, the most common was the vestibular-vaginal fistula, which is consistent with findings in other foreign literature [10,18], but differs from reports by Rickham, Lister, and Irving [14] who noted these malformations as rare in females. In two cases, anal stenosis was diagnosed in patients with the anus in an ectopic position. However, after confirming the presence of an external sphincter at this level, it was decided not to pursue conversion surgery, as the issue primarily involved aesthetic concerns of the perineum, which could be addressed through dilations, effectively resolving the functional dysfunction.

Similar to our observations, Husberg and colleagues [19] and Tuggle and colleagues [20] also encountered cases with an ectopic anus and stenosis.

The surgical procedures employed in the therapeutic management of anorectal malformations, as well as the utilization of preoperative colostomy, are detailed in Table 5.

It is worth noting that, in cases involving a higher number of high and intermediate malformations in adults, technical procedures were performed with the aim of lowering the blind digestive termination (rectum) to the surface of the perineum. This group included procedures such as posterior sagittal anorectoplasty and combined abdominoperineal procedures of Stephens, which were employed in 14 out of the 18 patients receiving definitive treatment. Unfortunately, one patient passed away without undergoing any procedures, while two others succumbed to complications after receiving a derivative colostomy. Additionally, one patient who had undergone colostomy experienced complications necessitating region transfer prior to the definitive operation.

Overall, of all the surgical interventions conducted, 52.4% involved posterior sagittal anorectoplasty, with only one case (4.8%) undergoing the combined abdominoperineal procedure of Stephens.

As evident, the surgical techniques employed align with the more modernized procedures advocated by the majority of authors [2,3,4,10,18,21,22]. Among the patients, six did not undergo a prior colostomy. In two of these cases, a low or intermediate malformation with a perineal fistula was present, necessitating a prior anorectoplasty. In the remaining four cases, anorectoplasty was performed in the neonatal period, yielding satisfactory results as there were no incidences of sepsis in the surgical area. This advantage of neonatal surgery has been corroborated by other authors in recent years, who have reported similar outcomes [21,23,24].

Only colostomy* two of this patients died before the anorectoplasty and the other once transferred to the other region for definite treatment.

The age at which anorectoplasty was performed is detailed in Table 5A. It is noteworthy that 50% of the patients underwent surgery within the first year of life, and among them, 4 cases (25% of the total) received definitive surgical treatment in the neonatal period. Additionally, 6 patients underwent surgical intervention between 13 months and 2 years (37.5%), while only 2 patients were operated on at 3 years or older.

Importantly, none of the patients who underwent anorectoplasty within the first year of life experienced complications, and their post-operative recovery was satisfactory, even in cases where prior colostomy was not performed in 4 of them.

As mentioned earlier in the analysis presented in Table 5, in recent years, some authors have advocated for anorectoplasty in the first few months of life due to its positive outcomes [21,23,24]. This

Table 3: Associated malformation according to specific diagnosis.

Malformations	Specific Diagnosis	Frequency	Total	%
Gastrointestinals	Oesophageal atresia with con TED fistula	3		
	Intestinal malrotation			
	Cleft palate	1	6	35.2
	Palate Fisura	1		
Squeleticas	Club foot	1		
	Cogenital hip luxation	2		
	Esternocleidomastoid agenesia	1		
	Hemivertebras Dorsales	1	6	35.2
	Ausencia de 3 costillas	1		
Genitourinary	Renal agenesia	1		
	Horseshoekidney	1	3	17.8
	Bilateral Criptorquidia	1		
Neurological	regresión Caudal sindrome	1	1	5.9
Cardiovasculars	Fallottetralogy	1	1	5.9
Total		17	17	100

Table 4: Anorectal malformation according to wingspread classification.

Male	Frequency	Total	%
High Rectal Agenesia		8	37
With fistula	5		
Recto-urethral	3		
Without Fistula			
Intermedia		2	9
With Fistula	1		
Rectourethral	1		
Without fistula			
low Anal Agenesia		2	9
With rectoperineal Fistula	2		
FEMALE			
High Rectal Agenesia		2	9
With Fistula	1		
Rectovaginal	1		
Rectovestibular			
Intermedia Rectal Agenesia		4	18
With Fistula	3		
Rectovestibular	1		
Without Fistula			
low Anal Agenesia		4	18
With Fistula	2		
Anoperineal	2		
Anal Estenosis			
Total	22	22	100

Table 5: Surgical technique used according to the frequency.

Surgical technique	With colostomía	without colostomía	Total	%
Posterior sagital Anorectoplasty	7	4	11	52
Onlycolostomy*	3	-	3	14
Dilatation	-	-	2	9.5
Anterior Anorectoplasty	-	2	2	9.5
Fistula Transposition	1	-	1	4.8
Cut- Back	1	-	1	4.8
Stephen Technique	1	-	1	4.8
Total	13	6	21	100

Table 5A: Age at the moment of the anorectoplasty

Age	Frequency	%
0-29days	4	25
30 days-12 months	4	25
13 months-2years	6	38
3years and more	2	13

observation aligns with our findings, particularly in the neonatal period, where this surgical technique can be carried out without the necessity of colostomy.

Regarding post-operative complications specific to the surgical technique, they are outlined in Table 6. While the absolute numbers of patients in each group with a specific surgical technique may not be suitable for direct comparison, it is worth noting that the technique of Posterior Sagittal Anorectoplasty and prior anorectoplasty demonstrated a higher incidence of complications (18.1% and 0%, respectively). It is important to mention that fecal incontinence is included among the post-operative complications. Overall, we recorded complications in 25% of patients following definitive surgical treatment.

Regarding complications, the literature review did not yield specific overall complication rates. However, in relation to posterior sagittal anorectoplasty, Gonzalez Pérez and colleagues [3] reported a 50% complication rate in a series of 10 cases, while Nakayama and colleagues [25] found a 26% post-operative complication rate in a series of 23 patients treated with posterior sagittal anorectoplasty, which is less favorable compared to our outcomes.

The specific types of complications, corresponding to the particular diagnoses, are presented in Table 6 A. Instances of posterior urethral opening, fecal incontinence, wound sepsis, and intestinal obstruction comprised the five cases in our series.

As observed, there were no significant differences in the frequency of complications, although fecal incontinence was the most common, occurring in 40% of cases. In the majority of the literature we reviewed, genitourinary complications [2,3,12,14,26,27] and post-operative fecal incontinence were frequently reported [2,3,6,7,14,28-30]. These included urethral stenosis, bladder dysfunction, and recurrent urinary tract infections, which were not observed in the postoperative follow-up of our patients, although the observation period ranged from 2 to 9 years. Overall, we interpret our results as satisfactory given the low incidence of complications and the specific types encountered.

The final status of patients diagnosed with anorectal malformations at the conclusion of this study is outlined in Table 7. Out of the 16 patients who underwent definitive anorectoplasty, one unfortunately succumbed, constituting a 7% mortality rate within this group.

Overall, there were four unfortunate fatalities, constituting a 19.1% mortality rate for anorectal malformations. Regrettably, three of these patients passed away prior to receiving definitive treatment; one of them, who did not undergo colostomy, succumbed within the first few hours of life. Another passed away at 24 hours of age, and the third at 71 days. All four deceased patients presented with multiple malformations, with three of them exhibiting esophageal atresia, one displaying VATER association, and two being born underweight.

As outlined in the literature, it is widely acknowledged that associated malformations are the primary contributors to mortality in patients with anorectal malformations [1,2,7,17,31], a finding which aligns with our own observations. Specifically, the patient with VATER association succumbed at the age of three due to intestinal obstruction with perforation and subsequent peritonitis, following the successful correction of all malformations.

Conclusion

As the most frequently associated malformation in the study population. High anorectal malformations were the predominant type, affecting 10 out of the 22 cases examined, with a notably higher occurrence among male patients, constituting 37% of the total diagnosed cases.

Table 6: Complications patients in the postoperative according to the surgical techniques used

Surgical technique	Complication	%	No Complication	%	Total	%
Posterior Sagital Anorectoplasty	2	18	9	82	11	100
Anterior Anorectoplasty	-	-.	2	100	2	100
Stephen Technique	1	100	-	-	1	100
Fistula transposition	1	100	-	-	1	100
Cut-back	-	-	1	100	1	100
Total	4	25	12	75	16	100

Table 6A: Transoperative and postoperative complications according to the diagnosis.

Diagnosis	Frequency	%
Posterior urethral Fistula recurence	1	20
Fecal Incontinence	2	40
Surgical site Infection	1	20
Postoperatory Intestinal Obstruction	1	20
Total	5	100

Table 7: Relation between to definitive surgical treatment with the discharge.

	alive	%	death	%	Total	%
With Anorectoplasty	15	93	1	7	16	100
Without Anorectoplasty	2*	40	3	60	5	100
Total	17	81	4	19	21	100

*Exclusión the patient was transferred the other region without definitive surgical treatment.

Among male patients, rectourethral fistula was the most frequently encountered type, while among female patients, vestibular vaginal fistula prevailed. The surgical procedure most commonly performed was the posterior sagittal anorectoplasty, which accounted for 52.4% of the cases. Impressively, a quarter of patients with anorectal malformations underwent anorectoplasty in the neonatal period without prior colostomy.

Complications were observed in a quarter of patients after definitive procedures, with cases of posterior sagittal anorectoplasty showing a lower rate of 18.1%. Finally, the mortality rate for anorectal malformations was determined to be 19.1%, with a higher incidence noted among patients with more significant associated malformations, indicating a direct correlation between the complexity of cases and increased mortality.

Recommendation

1. Conduct a comprehensive investigation of associated malformations, particularly focusing on esophageal atresia, within patients diagnosed with anorectal malformations. This will provide valuable insights into potential coexisting conditions involving the gastrointestinal, skeletal, and urogenital systems.
2. Enhance the emphasis on studying and training in surgical techniques aimed at addressing high malformations associated with rectourethral fistula and vestibular vaginal fistula. Notably, prioritize the utilization of the posterior sagittal anorectoplasty technique, which has demonstrated effectiveness in such cases.
3. Advocate for early anorectoplasty procedures, ideally within the first month of life, provided the patient's clinical condition permits the surgical intervention without significant risks. This approach, supported by its positive outcomes during this early period, should be considered fundamental in the treatment protocol for anorectal malformations.

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