The Effect of Post War Limited Resources in Management of Anorectal Malformations in Mosul City

Obay Abdulaziz Edan^{*}, Bassam Khalid Al-Hajjar^{*}, Awss Amjad Yehya^{**}

ABSTRACT:

BACKGROUND:

Anorectal malformations (ARMs) include a wide spectrum of diseases affecting male and female neonates and involve the rectum, anal canal, and genitourinary tract.

OBJECTIVE:

To know the actual incidence of anorectal malformations in Mosul city, upgrade clinical knowledge about ARMs between health workers and midwives who look after newborn babies, and assess the related mortality in the limited resources zone.

METHOD:

A closed cohort study was conducted on patients with ARMs admitted to the pediatric surgery center between January 2018 and January 2019. Patients included in this study are those with ARMs in the neonatal period, those admitted for pull-through or closure colostomy. We excluded from this study any Patients born before 2018 or those not operated.

RESULTS:

Sixty-two patients were included in this study with a Male: female ratio of 1.3:1. The estimated incidence was 4 per 5000 live birth. Twenty-eight (45.2%) patients presented after 48 hours of birth (Delayed diagnosis), with a median age at diagnosis of two days. Twenty-five (40.3%) from a total of 60 operated patients had associated congenital anomalies. Forty-three (69.3%) patients had colostomy (mainly transverse colostomy), with some cases have completed their three-stage procedure. Mortality was recorded in 5 (8%) cases (four males and one female, with major congenital anomalies). **CONCLUSION**:

ARMs represent one of t

ARMs represent one of the stressful GIT anomalies to pediatric surgeons. The overall outcome is greatly influenced by age at diagnosis, surgical experience, and associated anomalies. The major subtypes that need three stages surgeries require great efforts and more hospital resources. We recommend the need for widespread teaching programs for midwives and health workers to diagnose and refer ARMs cases to the tertiary centers as soon as possible to minimize complications and hopping lessen the mortality rate.

KEYWORDS: post war, limited, resources, anorectal malformations.

INTRODUCTION:

Anorectal malformations (ARMs) include a wide spectrum of defects affecting male and female neonates and involve the rectum, anal region, and genitourinary tract ^[1]. These malformations occur in approximately one per 4000-5000 births with a slight male predominance with a risk of 1% for families to have a second baby with ARM^[2,3], It also can happen as an isolated anomaly or in association with other malformations^[4].

An imaging study (including ultrasonography, MRI) is necessary to evaluate the associated urologic anomalies, which should be done after 72 hours of birth because the earlier study may miss abnormalities due to physiological oliguria in the early hours after birth^[5]. Colostography is also necessary to delineate the anatomy preoperatively ^[6]. The limitation in these tests, antenatal care, genetic lab, and lack of proper nerve stimulators all adversely affect the overall outcome in the management of ARMs cases. In 2017, Mosul city (the second biggest one in Iraq) was subjected to a massive damage during the processes of liberation and left with a very limited medical resources that adversely affected the overall health outcome.

^{*} Department of Surgery, College of Medicine,

University of Mosul, Mosul, Iraq.

^{*}Department of Pediatric Surgery, Al-Khansaa Teaching Hospital, Mosul, Iraq.

POST WAR LIMITED RESOURCES IN MOSUL CITY

A large number of patients, in addition to a limited resource (including lack of proper medical knowledge), were stressful and may lead to delay in diagnosis causing more morbidity or even mortality even in the presence of a tertiary facility.^[7]

Pediatric surgeons face additional stress when dealing with ARMs cases associated with major congenital heart disease without proper coronary care unit or real-time cardiac intervention.

<u>Vater no vater does not matter</u>, a stimulant statement by professor Alberto Pena that encourages pediatric surgeons to work in limited resources centers and provide an excellent anatomic repair together with the management of all sequel, mainly fecal and urinary incontinence^[1].

OBJECTIVE:

To know the actual incidence of ARMs in Mosul city, upgrade clinical knowledge about ARMs between health workers and midwives who look after newborn babies, and assess the related mortality in limited resources zone.

MATERIALS AND METHODS:

Study design and Setting

A closed cohorts study (over one year) for patients with ARM admitted to the pediatric surgery center from January 2018 till January 2019.

This study was conducted in the pediatric surgery department in Al-Khansaa Teaching Hospital, the only governmental tertiary health institution in Mosul city, serving nearly 3.5 million people with annual live birth (in 2018) of about 832000.

The surgical ward contains 24 beds, including six infant incubators and two ICU beds. Three consultant pediatric surgeons and 13 specialists (with different experiences) were shared to manage ARMs cases.

Study populations

The study included all patients presented to Al-Khansaa Teaching Hospital with ARMs in the neonatal period, those for a pull-through procedure, colostomy closure, and any other procedure related to ARMs. Patients born before January 2018 or after January 2019 or those not operated were excluded from this study.

Patients who enrolled in this study were diagnosed clinically and radiologically, classified according to male or female with the corresponding type of lesion based on information from the patient file and responsible surgeon. Lateral invertogram, cross-table lateral decubitus film (in limited cases), abdominal ultrasound, and echocardiography were done to check for associated anomalies and congenital heart disease. Nasogastric intubation was performed for every patient with suspicion of esophageal atresia. MRI was not available in Al-Khanssa Teaching Hospital or even in other governmental hospitals as an adverse effect of war on Mosul city; so, screening of vertebral column for the presence of tethered cord was not performed in these patients.

Preoperatively, all patients were admitted for iv fluid, antibiotics, and preoperative laboratory evaluation. Each patient operated either as emergency diverting colostomy, mainly transverse seldom pelvic colostomy, or one stage anoplasty for cutaneous fistula, and some have vaginostomy. The same for elective surgery, like posterior sagittal anorectoplasty (PSARP), posterior sagittal ano-recto-vagino-urethroplasty (PSARPVU), or one-stage anal transposition for Anovestibular fistula.

Postoperatively, the patients were kept on nil by mouth, intravenous fluid, antibiotics (iv cefotaxime with metronidazole), and analgesia, and start oral once bowel sound is positive with follow up of wound till discharge, to be followed in the clinic of the responsible surgeon. Dilatation started on day 14 postoperatively, with the closure of colostomy when the neo-anus reached the desired size, usually 2-3 months post PSARP.

Data collection

Data of each patient was collected from the file and responsible surgeon, coded on an excel sheet including name, age, age at diagnosis, gender, weight, type of ARMs, associated anomalies, type of feeding, the treatment offered, morbidity, and mortality.

Ethical consideration

The approval to carry out this study was sought from the institutional ethical committees of Mosul Medical College – University of Mosul. All information, including names and pictures, were kept under strict confidentiality. The study did not interfere with the decision of the responsible surgeons.

RESULTS:

Incidence

Mosul is the second biggest city in IRAQ, with annual live birth for 2018 (832000). Total cases of ARMs received were 62, making the disease

incidence in Mosul city nearly 4 for each 5000 live birth.

Socio-demographic characteristic

Between January 2018 and January 2019, 62 patients were admitted with ARMs, 35(56.4%) males and 27 (43.6%) females, with a male: female ratio of 1.3:1.

The age at diagnosis ranged from one to four days with a median age of two days of life.

Type of lesion according to clinical classification Out of 35 males, 10(16.1%) patients presented with recto-perineal fistula, the remaining 25 (40.3%) patients presented with high type that needed a colostomy, while out of 27 females, 23 (37.4%) presented with three perineal orifices (Anovestibular fistula (AVF)), and only two cases (3.2%) with two orifices (Rectovaginal fistula), the other two (3.2%) had single cloacal orifice (table 1).

Type of lesion at presentation (clinical)	Number	%
Male	35	
Recto perineal fistula	10	16.1
(high type) need colostomy	25	40.3
Female	27	
Three orifices (urethra, vagina& anal fistula)	23	37.4
Two orifices (urethra & rectovaginal confluence)	2	3.2
Cloaca single orifice (urethrorectovaginal confluence)	2	3.2
Total	62	

Table 1: Types of the lesion at presentation.

Associated anomalies

Out of 62 patients, 25 (40.3%) cases had other associated anomalies as follows; cardiac in 9 (14.5%) patients, five males and four females, followed by urological in 7 (11.2%) patients,

all were male, gastrointestinal in 6 (9.6%) patients, four male and two females, chromosomal in three (4.8%) patients, two males and one female, and lastly skeletal anomalies in one (1.6%) male patient.

Delay in diagnosis

Delay in diagnosis is defined for any case diagnosed after 48 hours of life.

In this study, 28 (45.2%) patients had delayed diagnosis (beyond 48 hours of life), while the remaining 34 (54.8%) patients were diagnosed earlier.

Surgical treatment

Two (3.2%) patients from a total of 62 patients with ARMs were not operated upon, one due to parental refusal (rare sacral fistula) and the other had multiple anomalies unfit for surgery.

Out of 35 (56.4%) male patients, 23 (37.4%) patients had a transverse colostomy, two (3.2%) patients had a pelvic stoma, and ten patients with

cutaneous fistula had their anoplasty as emergency operations.

From a total of 27 (43.5%) female patients, 23 (37.4%) patients with anovestibular fistula (three orifices) received treatment as follow; three (4.8%) waiting for surgery, 6 (9.6%) had one stage perineal transposition while the remaining 14 (22.5%) patients completed their three stages operations. Two females (3.2%) with two orifices had a colostomy, and two patients with cloaca (3.2%), the first had a colostomy and the other completed PSARVUP.

Mortality and morbidity

The morbidity that may be happened whether secondary to disease or surgical intervention, were all managed inside the surgical department like wound sepsis, wound dehiscence, colostomy prolapse, parastomal hernia, bleeding, stenosis and intestinal obstruction. These complications were not included in this study.

Mortality was recorded in 5 (8%) out of 60 operated patients, one female with associated TEF and four males, three with pouch colon, and the last one had down syndrome with congenital heart disease (table 2).

THE IRAQI POSTGRADUATE MEDICAL JOURNAL

no	age	Gen.	Type of lesion with anomalies	Surgery
1	1d	fe	TEF +ano-cutanous fistula	One stage
2	2d	m	Pouch colon +tetralogy of Fallot	colostomy
3	4d	m	Pouch colon	Resects pouch
4	4d	m	Pouch colon	colostomy
5	4d	m	Down+ CHD+ need colostomy	colostomy

Table 2: Mortality with ARMs.

DISCUSSION:

Anorectal malformations occur in one of every 4000 to 5000 live newborns. It is affecting males slightly more than females. The most frequent type of defect in males is anal atresia with a recto urethral fistula, while in females, it is anal atresia with a rectovestibular fistula ^[1]. The incidence in our closed cohort study is high (4 in every 5000 live newborns), which is still similar to (mooree et al.) ^[8] and other parts of the world with low living standards. We could not find the actual reason, but the larger population may carry more incidences of such anomalies or may be due to the consageous marriage, but we still need more information and investigation to figure out the exact cause.

We found that the affected male patients were slightly more than females, which is similar to (alabbasi et al.) ^{[9],} but not in other studies like (Ramadan et al.) ^[10] who found females more involved. (Bilal Mirza et al.)^[11] recorded a marked difference in male: female ratio of 3.4:1, here we need more investigation to see why this happened. Regarding types of defect, the significant finding was the great variability in the occurrence of the main ARMS subtypes among different cultures and races. In male patients, we recorded rectoperineal fistula in 16.1%, rectourethral fistula (high type) in 40.3%, which is inversely related to a study done in the United States on Hispanic populations by (Rosas-Blum et al.)^[12] in 2020. who stated that 46% of cases had rectoperineal fistula, and only 16% had a rectourethral fistula. Another study in Kenya by (P Kuradusenge et al.) ^[13] in 2014 reported that 42% of the male subtype was imperforate anus without fistula. In females, (85%) of cases had AVF in our data, which was higher than what was mentioned by (Theron A et al.)^[14] (70%).

Associated congenital anomalies in the neonate with ARMs are assumed as a significant factor for survival (Ahmad, leva, et al.) $^{[15,16]}$. In our study, the associated anomalies are (40.3) % which is in the middle of a range between (30-70%) as found

in (Ahmad j., Leva E., Ratan S. K., et al.) ^[15,16,17]. Thus, it highlights the need for more investigation and screening for those patients. We never diagnosed a case of tethered cord simply because, after the war, we were left with no functional MRI machine in the city.

Delay in diagnosis again contributes to the survival rate adversely. (Birabwa-Male et al.), (Makanga, M. et al.), and (S. Govender et al.) ^[18,19,20], all showed a significant percentage of delayed diagnosis as in our study, which was explained by the difficulty in accessing health care facilities in the war zone with a lot of curfews and closed roads. The limited medical knowledge among midwives and health workers regarding ARMs care with low economic state families also play a role in such delay, making these patients present to the hospital only due to intestinal obstruction or long-term constipation in a female with AVF.

Colostomy to divert stool could be fashioned as an emergency or elective way. A study was done in IRAQ in 2016 (similar situation as in our center) reported a 70% complication rate (Amer abd allah ejrish)^[21] probably because we still practice the transverse loop stomas for most of our patients and this practice needs to be changed to pelvic complete defunctioning one to avoid acidosis and dilated distal colon. For these reasons, some of our surgeons practicing one stage pull through exclusively for females with AVF depending on a study done in Baghdad city (Murtadha et al. 2017)^[22], with promising results for both patients (less family burden) and surgeons with fewer resources used in our center, this needs to be evaluated and reported scientifically.

The mortality rate in this study accounts for 8% (five patients out of 60 operated on), which is higher than Haider et al. (4%) and S. Govender et al. (5.7%) $^{[7,20]}$. We believe it is an acceptable rate with the adverse circumstances associated with the management of ARMs in our center. The high mortality rate among male patients with pouch

POST WAR LIMITED RESOURCES IN MOSUL CITY

colon in this study necessitates further study and more medical knowledge for this rare entity (Ahmad z et al. 2015)^[23]. The delay in diagnosis and associated diagnosed or undiagnosed anomalies are another additive factor for mentioned mortality.

CONCLUSION:

ARMs represent one of the stressful GIT anomalies to pediatric surgeons. The overall outcome is greatly influenced by age at diagnosis, surgical experience, and associated anomalies, and despite having scarce resources, we could manage those cases with minimal complications. The major type that needs three stages surgeries requires great efforts and more hospital resources. We recommend the need for widespread teaching programs for midwives and health workers to diagnose and refer ARMs cases to the tertiary centers as soon as possible to minimize complications and hopping lessen the mortality rate.

Acknowledgement:

Great thanks to all pediatric surgeons working in our pediatric surgery Centre for their participation in data collection and support throughout our project.

REFERENCES:

- 1. Marc A Levitt, Alberto Pena .Anorectal malformation. Orphanet Journal of rare disease.2007;2:33.
- **2.** Mundt E, Bates MD. Genetics of Hirschsprung disease and anorectal malformations. Semin Pediatr Surg. 2010;19:107-17.
- **3.** Wood RJ, Levitt MA. Anorectal malformations. *Clin Colon Rectal Surg* 2018; 31:61–70.
- **4.** Byun SY, Lim RK, Park KH, et al. Anorectal malformations associated with esophageal atresia in neonates. Pediatr Gastroenterol Hepatol Nutr 2013;16:28–33.
- 5. Hosokawa T, Yamada Y, Tanami Y, Hattori S, Sato Y, Tanaka Y, et al. Sonography for an Imperforate Anus: Approach, Timing of the Examination, and Evaluation of the Type of Imperforate Anus and Associated Anomalies. J Ultrasound Med. 2017; 36:1747-58.
- **6.** Kraus SJ, Levitt MA, Peña A. Augmentedpressure distal colostogram: the most important diagnostic tool for planning definitive surgical repair of anorectal malformations in boys. Pediatr Radiol. 2018;48 :258-69.

- 7. N Haider, R. Fisher. Mortality and morbidity associated with late diagnosis of anorectal malformation in children. Surgeon , 2007; 5: 327-30.
- **8.** Moore, S. et al . The spectrum of anorectal malformation in Africa. Pediatric surgery international.2008; 24:677-83.
- **9.** Bassam Khalil AL-Abbasi, Abdul Rahman Abdul Aziz AL-Shahwani. Management of anorectal malformation A study of 100 cases.2004; IPMJ 3.
- **10.** Ramadhan j. et al . Clinical profile and outcome of surgical management of anorectal malformation at a tertiary care hospital in Tanzania. 2018;20.
- **11.** Bilal Mirza, Lubna Ijaz, Muhammad Saleem, Muhammad Sharif, Afzal Sheikh. Anorectal malformations in neonates. Afr J Paediatr Surg. May-Aug 2011;8:151-54.
- 12. Rosas-Blum, Eduardo D.; Reddy, Ajay; Shaban, Mohamed A.; Aziz, Sandra; Do, Ailinh; Spurbeck, William; Francis, Denease. Characteristics of Anorectal Malformations in Children at the United States-Mexico Border: A 3-Year Study. Journal of Pediatric Gastroenterology and Nutrition: July 2020;71- e12-e15.
- **13.** P Kuradusenge, R T Kuremu, G Jumbi, P W Saula. Pattern of anorectal malformations and early outcomes of management at MOI teaching and referral hospital Eldoret-Kenya. East Afr Med J. 2014 Dec;91(12):430-4.
- 14. Theron A, Numanoglu A. Birth prevalence of anorectal malformations for the western cape province, South Africa, 2005 to 2012. Eur J Pediatr Surg. 2017; 27:449–54.
- **15.** Ahmad j. et al. Association of urogenital anomalies with anorectal malformation; a review of 200 cases. Pakistan Journal of Surgery.2005; 21:89-92.
- **16.** Leva E. et al. single-stage surgical correction of anorectal malformation associated with rectourinary fistula in male neonate. Journal of Neonatal Surgery.2013; 2:3.
- **17.** Ratan S. K. et al. Associated congenital anomalies in patients with anorectal malformation a need for developing a uniform practical approach. Journal of Pediatric Surgery.2004;39:1706-11.

THE IRAQI POSTGRADUATE MEDICAL JOURNAL

POST WAR LIMITED RESOURCES IN MOSUL CITY

- Birabwa-Male, D. Anorectal malformation in Mulago Hospital, Kampala-Uganda. East and Central African Journal of Surgery.2004; 9:1.
- **19.** Makanga, M . et al. Anorectal malformation at university teaching hospital of Butare in Rwanda: A review of 46 operative cases. East and Central African Journal of Surgery .2007;12:110-115.
- 20. S. Govender, R Wiersma. Delayed diagnosis of anorectal malformations (ARM): causes and consequences in a resource-constrained environment. Pediatric Surgery International.2016; 32:369-75.
- **21.** Amer abd allah ejrish .Medical Journal of Babylon .2016;13 : 294-306.
- 22. Murtadha Abdul Kadhim, Kawther Fakhri, Hassan k Gatea. Iraqi Academic Scientific Journal. 2017;: 438-43.
- **23.** Ahmad Z. Zain , Salah S. Mahmood , Raghad J. Aboalhab. Congenital pouch colon: A rare presentation of Anorectal malformation. Journal of the Faculty of medicine .2015;57: 193-97.