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SURGICAL OUTCOME IN INFANTS BORN WITH ANORECTAL MALFORMATION

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ABSTRACT

Background: Anorectal malformations in infants is not an uncommon finding raising concerns. The presence of the associated anomalies, delayed referral, and diagnosis can further negatively affect the outcomes of the anorectal malformations.

Aim: The present study aimed to assess the early outcomes of anorectal malformations seen in the neonate.

Methods: The study prospectively assessed all the neonates admitted to the NICU (neonatal intensive care unit) with anorectal malformations. The subjects were assessed postoperatively for 1 month to evaluate the short-term surgical outcomes in infants with anorectal malformations.

Results: Among 157 neonates, 75.15% (n=118) were males and 24.84% (n=39) were females. High anorectal malformations and low anorectal malformations were seen in 84.07% (n=132) and 15.92% (n=25) study subjects respectively. Associated congenital anomalies along with anorectal malformations were seen in 38.21% (n=60) of study subjects. The most common associated anomalies were gastrointestinal in 40.76% (n=64) subjects followed by esophageal and genitourinary in 31.21% (n=49) and 19.74% (n=31) subjects respectively. The most common surgical procedure was transverse colostomy done in 63.69% (n=100) subjects and the most common complication seen was thrombocytopenia in 36.30% (n=57) subjects followed by sepsis in 31.21% (n=49) subjects. Mortality was seen in 28.02% (n=44) subjects with 31.06% (n=41) in HARM and 12% (n=3) in LARM. A significant association was seen in mortality with birth weight and associated malformations with p=0.0001 and 0.003 respectively.

Conclusion: The study concludes that in subjects with anorectal malformations, a higher mortality rate is seen in neonates with cardiac anomalies, esophageal anomalies, sepsis on admission, GIT perforation, double/triple atresia, and low birth weight. The majority of deaths are associated with anomalies warranting a detailed systematic evaluation of the subtypes.

Keywords: Anorectal malformations, colostomy, neonatal ICU, atresia, mortality

INTRODUCTION

Anorectal malformations or ARMs represent a diverse group of congenital anomalies comprising the genital system along with the urinary tract and/or the lower gastrointestinal tract. Anorectal

malformations affect a large group of infants globally including India with the incidence of anorectal malformations being reported as 1:5000 live births.[1] The anorectal malformations have a varied and diverse presentation ranging from a simple low perineal fistula to high anomalies presenting and demanding complex surgical management. Anorectal malformations are usually associated with anomalies affecting the other organ system which is known as the VACTERL association, and this association is seen in nearly 5% to 30% of the affected subjects.[2]

The presence of the associated anomalies, delayed referral, and diagnosis can further negatively affect the outcomes of the anorectal malformations.[3] An adverse impact on the prognosis of the anorectal malformations is seen with gut perforation, septicemia, delay in a consultation with the pediatric surgeons, associated septicemia, low birth weight, and/or the associated complex anomalies.[4]

However, few modifiable risk factors can help in reducing neonatal mortality in subjects with anorectal malformations including sepsis, poor immunity, hypoglycemia, and hypothermia.[5] The final good functional outcomes can result from efficient surgical repair, management of the associated anomalies, timely diagnosis, and accurate treatment.[6]

Hence, the present clinical study aimed to assess the early outcomes concerning the mortality and morbidity statistics in neonates present with anorectal malformations in an Indian healthcare center.

MATERIALS AND METHODS

The present prospective clinical study was aimed to assess the early outcomes concerning the mortality and morbidity statistics in neonates present with anorectal malformations in an Indian healthcare center. The study was done at after the clearance was given by the concerned Institutional Ethical committee. Informed consent was taken from the wards of all the participants before the study participation.

The study included all the neonates admitted to the NICU (neonatal intensive care unit) with anorectal malformations. The exclusion criteria for the study were subjects that underwent any surgical outcome outside the institute, subjects with Cloacal regression syndrome, syringomyelia, and subjects with cloacal exstrophy.

After final inclusion, all the subjects underwent a comprehensive clinical assessment and the anorectal malformations were classified based on the Krickenbeck classification.[7] Anomalies were classified as miscellaneous, syndromic, skeletal, neurological, genitourinary, gastrointestinal, and/or cardiac anomalies.

This was followed by the radiographic assessment using the Babygram. X-ray-prone cross-table view with pelvis raised was done after 18-24 hours of life in subjects with no visible fistula. Any other abdominal pathology associated with the anorectal malformations was assessed with the Ultrasound (Figure 1). In subjects having cardiac anomalies, echocardiography was done. Esophageal atresia was classified following the Gross anatomic classification[8] and VACTERL association[9] was also recognized.

The majority of the subjects underwent transverse colostomy following the conventional procedure. Postoperatively, all subjects were given broad-spectrum antibiotic coverage,

nasogastric suction, and intravenous fluids to correct the electrolyte and fluid deficits. Optimal respiratory support, hypoglycemia correction, and hypothermia control were adopted in all the subjects. colostomy following the conventional procedure. Broad-spectrum antibiotic coverage, nasogastric suction, and intravenous fluids were continued postoperatively.

In all the neonates with LARM, primary angioplasty without a protective diverting colostomy was performed, whereas, in HARM, either high sigmoid loop colostomy (SLC) or LTC (left transverse colostomy) was done. Based on the type of colostomy, the neonates underwent the fistula ligation with either excision of the pouch with end ileostomy/ end colostomy or type 1 and type 2 pouch ostomy.

The subjects were kept nil orally till the child had passed meconium (Figure 3) or the stoma started to function. The mother was explained about the colostomy care. Postoperatively, distal loop washes were done. In neonates having esophageal atresia, oral feeds were only given following the contrast study on the 7^{th} day postoperatively to rule out any anastomotic leak. The subjects were followed for 1 month postoperatively to assess the early outcomes of the study.

The data gathered were analyzed statistically using the SPSS software version 21.0 and the one-way ANOVA. The significance levels were kept at $p<0.05$ and the data were expressed as numbers and percentages.

RESULTS

The study assessed 157 neonates, 75.15% (n=118) were males and 24.84% (n=39) were females with anorectal malformations. Low anorectal malformations were seen in 15.92% (n=25) study subjects with Covered anus/Anteriorly placed anus (Figure 2) in 0.63% (n=1) female subjects and Bucket handle/covered anus/anal stenosis/anteriorly placed anus in 15.28% (n=24) male subjects respectively. For HARM, it was seen in 84.07% (n=132) study subjects with rectal atresia, HARM with/without rectovaginal fistula, vestibular fistula, persistent cloaca, and HARM with/without rectourinary fistula seen in 3.18% (n=5), 7.64% (n=12), 828% (n=13), 7% (n=11), and 57.96% (n=91) study subjects respectively as shown in Table 1.

On assessing the predictors of mortality in the study neonates, primary and secondary perforations were seen in 3 subjects each and with the survival of 33.3% (n=1) subject each from primary and secondary perforations which were non-significant with p=0.521. LARM had malformations including LARM with inguinal hernia, LARM with skeletal abnormality, LARM with PS hypospadias, LARM with EA (esophageal anomalies), and isolated LARM seen in 1, 2, 1, 2, and 19 subjects with mortality of 1 subject each with inguinal hernia, skeletal abnormalities, and isolated LARM respectively which was non-significant with p=0.008. LARM was seen in 15.92% (n=25) subjects with 12% (n=3) deaths. The vestibular fistula was seen in 8.28% (n=13) subjects with death in 30.76% (n=4) study subjects with 1, 2, and 1 death from VF with EA and DA, VF with EA, and isolated VF respectively which was significant with p=0.001. Cloaca has seen in 6.36% (n=10) study subjects with mortality in 30% (n=3) subjects from the isolated cloaca, Cloaca with vaginal atresia/ hydrometrocolops, and cloaca with CPC 1 respectively (p=0.846). Rectal atresia was seen in 3.18% (n=5) subjects with isolated and with EA in 4 and 1 subject respectively and death was seen in 60% (n=3) subjects with 2 and 1 death from isolated

rectal atresia and Rectal atresia with pure EA respectively (p=0.623). Atresia has been seen in 12.73% (n=20) subjects with mortality in 85% (n=17) subjects with 14, 2, and 1 death from Double atresia (EA+ARM), Triple atresia (EA+DA+ARM), and Double atresia (DA+ARM) respectively (p=0.11). Mortality was seen in 27.28% (n=43) subjects with 16, 23, 3, and 1 subjects with birth weights of >2500, 1500-2499, 1000-1499, and <1000 respectively which was statistically significant with p=-0.0001. Associated malformations were seen in 38.21% (n=60) subjects with mortality from miscellaneous, neurologic, down syndrome, skeletal malformations, urogenital malformations, cardiac malformations, and GI malformations in 1, 1, 2, 5, 4, 8, and 20 subjects respectively. This was statistically significant with p=0.002 (Table 2).

Concerning the malformations associated with the ARM in the study subjects, in miscellaneous malformations in 1.91% (n=3) subjects, cleft lip, umbilical polyp, and hernia was seen in 33.3% (n=1) subjects each. Neurologic malformations and Down syndrome were seen in 0.63% (n=1) subjects each. Skeletal malformations were seen in 4.45% (n=7) subjects with cleft palate, multiple limb anomalies, and vertebral defects in 14.28% (n=1), 57.14% (n=4), and 28.57% (n=2) subjects respectively. Urogenital malformations were seen in 7.64% (n=12) with hydronephrosis in 16.66% (n=2) subjects and developmental sex disorders, bicornuate uterus, vaginal atresia, renal agenesis, ureteral duplication, posterior urethral valve, scrotal transportation, undescended testis, proximal hypospadias, and distal hypospadias each in 1 subject each. Cardiac malformations were seen in 7% (n=11) study subjects with teratology of Fallot, right aortic arch, dextrocardia, and septal defects in 9.09% (n=1), 9.09% (n=1), 9.09% (n=1), and 72.7% (n=8) subjects respectively. GI malformations were seen in 15.92% (n=25) subjects with caecum and appendix duplication, Meckel's diverticulum, and Malrotation in 4% (n=1) subjects each. Duodenal atresia, EA type A, and EA type C in 12% (n=3), 4% (n=1), and 72% (n=18) subjects respectively (Table 3).

In the gastrointestinal perforations in study subjects with anorectal malformations, In HARM, 4 subjects died and 2 subjects were discharged from the Institute. In one subject discharged, perforation was seen at the distal jejunum which was secondary to LTC (left transverse colostomy) and was repaired with pouchostomy and excision of the gangrenous pouch and colostomy with the birth weight of 1600 grams. In other discharged subjects, perforation was seen at CPC 1 with a perforated pouch which was primary perforation repaired with fistula ligation, CPC excision, and ileostomy, and had a birth weight of 2000 grams. Another neonate that died had distal jejunum perforation in 2400 grams weight seen secondary to LTC and repaired with Jejunal perforation repair with divided colostomy and Omenatl pouch and drain placement. In two subjects with 2500 grams and 1800 grams weight respectively and primary sigmoid perforation, perforation repair, and LTC was done and the subject died. Another neonate, with 2600 grams weight and secondary caecal perforation which was repaired died (Table 4).

DISCUSSION

The present study aimed to assess the early outcomes of anorectal malformations seen in the neonate. The study assessed 157 neonates, 75.15% (n=118) were males and 24.84% (n=39) were females with anorectal malformations. Low anorectal malformations were seen in 15.92%

(n=25) study subjects with Covered anus/Anteriorly placed anus in 0.63% (n=1) female subjects and Bucket handle/covered anus/anal stenosis/anteriorly placed anus in 15.28% (n=24) male subjects respectively. For HARM, it was seen in 84.07% (n=132) study subjects with rectal atresia, HARM with/without rectovaginal fistula, vestibular fistula, persistent cloaca, and HARM with/without rectourinary fistula seen in 3.18% (n=5), 7.64% (n=12), 828% (n=13), 7% (n=11), and 57.96% (n=91) study subjects respectively. These data were similar to the studies of Nah SA et al[10] in 2012 and Thapa B et al[11] in 2013 where authors assessed neonates with demographic data comparable to the present study.

It was seen that concerning the predictors of mortality in the study neonates, primary and secondary perforations were seen in 3 subjects each and with the survival of 33.3% (n=1) subject each from primary and secondary perforations which were non-significant with p=0.521. LARM had malformations including LARM with inguinal hernia, LARM with skeletal abnormality, LARM with PS hypospadias, LARM with EA (esophageal anomalies), and isolated LARM seen in 1, 2, 1, 2, and 19 subjects with mortality of 1 subject each with inguinal hernia, skeletal abnormalities, and isolated LARM respectively which was non-significant with p=0.008. LARM was seen in 15.92% (n=25) subjects with 12% (n=3) deaths. The vestibular fistula was seen in 8.28% (n=13) subjects with death in 30.76% (n=4) study subjects with 1, 2, and 1 death from VF with EA and DA, VF with EA, and isolated VF respectively which was significant with p=0.001. Cloaca has seen in 6.36% (n=10) study subjects with mortality in 30% (n=3) subjects from the isolated cloaca, Cloaca with vaginal atresia/ hydrometrocolops, and cloaca with CPC 1 respectively (p=0.846). Rectal atresia was seen in 3.18% (n=5) subjects with isolated and with EA in 4 and 1 subject respectively and death was seen in 60% (n=3) subjects with 2 and 1 death from isolated rectal atresia and Rectal atresia with pure EA respectively (p=0.623). Atresia has been seen in 12.73% (n=20) subjects with mortality in 85% (n=17) subjects with 14, 2, and 1 death from Double atresia (EA+ARM), Triple atresia (EA+DA+ARM), and Double atresia (DA+ARM) respectively (p=0.11). Mortality was seen in 27.28% (n=43) subjects with 16, 23, 3, and 1 subjects with birth weights of >2500, 1500-2499, 1000-1499, and <1000 respectively which was statistically significant with p=-0.0001. Associated malformations were seen in 38.21% (n=60) subjects with mortality from miscellaneous, neurologic, down syndrome, skeletal malformations, urogenital malformations, cardiac malformations, and GI malformations in 1, 1, 2, 5, 4, 8, and 20 subjects respectively. This was statistically significant with p=0.002. These results were consistent with the studies of Mathur P et al[12] in 2009 and Gubbi S et al[13] in 2018 where authors similar predictors for neonatal mortality in anorectal malformations were seen in their respective studies.

The study results showed that for the malformations associated with the ARM in the study subjects, in miscellaneous malformations in 1.91% (n=3) subjects, cleft lip, umbilical polyp, and the hernia was seen in 33.3% (n=1) subjects each. Neurologic malformations and Down syndrome were seen in 0.63% (n=1) subjects each. Skeletal malformations were seen in 4.45% (n=7) subjects with cleft palate, multiple limb anomalies, and vertebral defects in 14.28% (n=1), 57.14% (n=4), and 28.57% (n=2) subjects respectively. Urogenital malformations were seen in

7.64% (n=12) with hydronephrosis in 16.66% (n=2) subjects and developmental sex disorders, bicornuate uterus, vaginal atresia, renal agenesis, ureteral duplication, posterior urethral valve, scrotal transportation, undescended testis, proximal hypospadias, and distal hypospadias each in 1 subject each. Cardiac malformations were seen in 7% (n=11) study subjects with teratology of Fallot, right aortic arch, dextrocardia, and septal defects in 9.09% (n=1), 9.09% (n=1), 9.09% (n=1), and 72.7% (n=8) subjects respectively. GI malformations were seen in 15.92% (n=25) subjects with caecum and appendix duplication, Meckel's diverticulum, and Malrotation in 4% (n=1) subjects each. Duodenal atresia, EA type A, and EA type C in 12% (n=3), 4% (n=1), and 72% (n=18) subjects respectively. These results were in agreement with the findings of Gupta R et al[14] in 2015 and Kamal JS et al[15] in 2013 where authors reported similarly associated malformations in neonates with anorectal malformations as in the present study.

It was also seen that in the gastrointestinal perforations in study subjects with anorectal malformations, In HARM, 4 subjects died and 2 subjects were discharged from the Institute. In one subject discharged, perforation was seen at the distal jejunum which was secondary to LTC (left transverse colostomy) and was repaired with pouchostomy and excision of the gangrenous pouch and colostomy with the birth weight of 1600 grams. In other discharged subjects, perforation was seen at CPC 1 with a perforated pouch which was primary perforation repaired with fistula ligation, CPC excision, and ileostomy, and had a birth weight of 2000 grams. Another neonate that died had distal jejunum perforation in 2400 grams weight seen secondary to LTC and repaired with Jejunal perforation repair with divided colostomy and Omenatl pouch and drain placement. In two subjects with 2500 grams and 1800 grams weight respectively and primary sigmoid perforation, perforation repair, and LTC was done and the subject died. Another neonate, with 2600 grams weight and secondary caecal perforation which was repaired died. These findings were in line with the studies of Menon P et al[16] in 2017 and Gupta R et al[17] in 2019 where authors comparable GI perforations, their repair, and outcomes were seen in the studies by authors.

CONCLUSION

Considering its limitations, the present study concludes that in subjects with anorectal malformations, a higher mortality rate is seen in neonates with cardiac anomalies, esophageal anomalies, sepsis on admission, GIT perforation, double/triple atresia, and low birth weight. The majority of deaths are associated with anomalies warranting a detailed systematic evaluation of the subtypes.

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TABLES

Anomaly type	**Number (Males + females)**	**Percentage (%)**
LARM	25	15.92
Covered anus/Anteriorly placed anus (female)	1	0.63
Bucket handle/ covered anus/ anal stenosis/anteriorly placed anus	24	15.28
HARM	132	84.07%
Rectal atresia	5 (3+2)	3.18
HARM with/without rectovaginal fistula	12	7.64
Vestibular fistula	13	8.28
Persistent cloaca	11	7

HARM with/without rectourinary fistula	91	57.96
Total	157	100

Table 1: Anorectal malformation types in the present study

Associated malformations	N %	Survival n (%)	Mortality n (%)	p-value
Primary/secondary perforation				
Primary	3	1 (33.3)	2 (66.6)	0.521
Secondary	3	1 (33.3)	2 (66.6)	
Total	6	2 (33.3)	4 (66.6)	
LARM				
LARM with inguinal hernia	1	0	1	0.008
LARM with skeletal abnormality	2	1	1	
LARM with PS hypospadias	1	1	0	
LARM with EA	2	2	0	
LARM isolated	19	18	1	
Total	25(15.92)	22 (88)	3 (12)	
Vestibular fistula (VF)				
VF with dextrocardia	1	1	0	0.001
VF with EA and DA	1	0	1	
VF with EA	3	1	2	
VF isolated	8	7	1	
Total	13(8.28)	9 (69.23)	4 (30.76)	
Cloaca				
Cloaca with umbilical polyp	1	1	0	0.846
Cloaca with CPC 3	1	1	0	
Cloaca with CPC 2	1	1	0	
Cloaca with CPC 1	2	1	1	
Cloaca with vaginal atresia/ hydrometrocolpos	1	1	1	
Isolated cloaca	4	2	1	
Total	10(6.36)	7 (70)	3 (30)	
Rectal atresia				
Isolated Rectal atresia	4	2	2	0.623
Rectal atresia with pure EA	1	0	1	
Total	5 (3.18)	2 (40)	3 (60)	
Atresia type				0.911
Triple atresia (EA+DA+ARM)	2	0	2	
Double atresia (EA+ARM)	17	3	14	
Double atresia (DA+ARM)	1	0	1	
Total	20(12.73)	3 (15)	17 (85)	
Body weight (gms)				0.0001
>2500 (Norma)	81	65	16	
1500-2499 (LBW)	72	48	23	
1000-1499 (VLBW)	3	1	3	
<1000 (ELBW)	1	0	1	
Total	157 (100)	114 (72.61)	43 (27.38)	
Associated malformations				
Miscellaneous	3	2	1	0.002
Neurologic	1	1	1	
Down syndrome	1	0	2	
Skeletal malformations	7	2	5	
Urogenital malformations	12	6	4	
Cardiac malformations	11	3	8	
GI malformations	25	5	20	

Total	60 (38.21)	19 (31.66)	41 (68.33)	

Table 2: Predictors of Neonatal mortality in study subjects

Malformations associated	N	%
Miscellaneous	3	1.91
Cleft Lip	1	33.3
Umbilical polyp	1	33.3
Hernia	1	33.3
Neurologic	1	0.63
Down syndrome	1	0.63
Skeletal malformations	7	4.45
Cleft palate	1	14.28
Multiple limb anomalies	4	57.14
Vertebral defects	2	28.57
Urogenital malformations	12	7.64
Sex development disorders	1	8.33
Bicomuate uterus	1	8.33
Vaginal atresia	1	8.33
Renal agenesis	1	8.33
Ureteral duplication	1	8.33
Posterior urethral valve	1	8.33
Hydronephrosis	2	16.66
Scrotal transportation	1	8.33
Undescended testis	1	8.33
Proximal hypospadias	1	8.33
Distal hypospadias	1	8.33
Cardiac malformations	11	7
Teratology of Fallot	1	9.09
Right aortic arch	1	9.09
Dextrocardia	1	9.09
Septal defects	8	72.7
GI malformations	25	15.92
Caecum and appendix duplication	1	4
Meckel's diverticulum	1	4
Malrotation	1	4
Duodenal atresia	3	12
EA type A	1	4
EA type C	18	72
Total	60	38.21

Table 3: Malformations associated with anorectal malformations in the present study

ARM	Perforation site	Perforation type (primary/secondary)	Repair type	Outcome	Weight (grams)
HARM	Distal jejunum	Secondary to LTC	Pouchostomy Excision of gangrenous pouch and colostomy	Discharge	1600
	Distal jejunum	Secondary to LTC	Jejunal perforation repair with a divided colostomy. Omental pouch and drain placement	Death	2400
	Sigmoid	Primary	Perforation repair and LTC	Death	2500
	Sigmoid	Primary	Perforation repair and LTC	Death	1800
	CPC 1 with	Primary	Fistula ligation, CPC excision, and	Discharge	2000

	perforated pouch		ileostomy		
	Caecal	Secondary	Perforation repair	Death	2600
Total	N=6	3= primary 3= secondary		Death=4 Discharge =2	

Table 4: Gastrointestinal perforations their repair and outcomes in the study of neonates

ATLAS

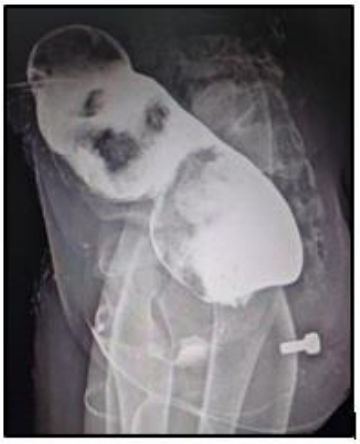

Figure 1: Abdominal pathology associated With anorectal malformations

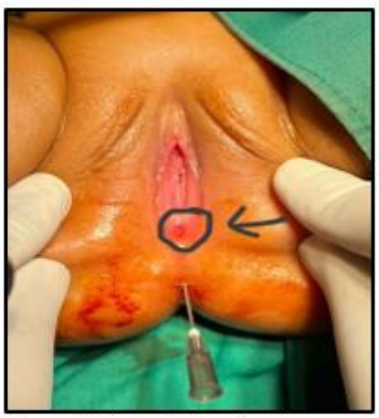

Figure 2: Anteriorly placed anus with Vestibular fistula

Figure 3: Meconium pearl