

Original Research Article

Pattern of anorectal malformation in a tertiary care hospital of Bihar

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ABSTRACT

Background: Anorectal malformation (ARM) is common cause of neonatal intestinal obstruction and is one of the most common neonatal surgical emergency encountered by paediatric surgeons. There is no published study about the prevalence of ARM cases within the referral zone of the hospital. This study was done to demonstrate the burden of Anorectal malformation and their surgical intervention as seen in Indira Gandhi Institute of medical science, Patna, Bihar.

Methods: Data were retrospectively collected over a period of four years between January 2015 and December 2018. The data collected were demographic status of the patient, diagnosis, associated anomaly, duration of hospital stay and their surgical intervention.

Results: There were 395 cases of anorectal malformation out of 93203 OPD patients (0.42%). Male to female ratio of 3.01:1. ARM also represented 25.3% of neonatal emergency surgical procedures. The majority of the patient had presented in the neonatal period (51.6%) with mean weight of 2.6 kg. The most common type of ARM found in male patient was rectourethral fistula n=215 (54.4%) in which 164 (76.2%) patients were with rectobulbar fistula and 51 (23.8%) patients had rectoprostatic fistula. Sixty nine (17.4%) female patients presented with perineal fistula.

Conclusions: We found a clear male preponderance of anorectal malformation in our study. Patients affected were mostly from poor socioeconomic strata of the society. Present study confirms huge work load of anorectal malformation in this region. This will be helpful in future planning and effective management including financial constraints of the treatment for our poor patients.

Keywords: Anorectal malformation, Bihar, Hospital, Pattern, Tertiary care

INTRODUCTION

Anorectal malformation (ARM) is common cause of neonatal intestinal obstruction, which is usually diagnosed clinically by the absence, or ectopic location of the anus. Incidence of the anorectal malformations (ARM) varies between 1/1500 to 1/5000 live births.¹ Anorectal malformation is one of the most common neonatal surgical emergency encountered by paediatric surgeons.² To the extent of our knowledge there is no published data about the prevalence of ARM cases within the referral zone of the hospital. This article reflects the pattern of ARM cases at our tertiary care centre and their surgical intervention.

METHODS

This was a retrospective study in which all cases of anorectal malformation admitted to Indira Gandhi Institute of Medical science, Patna between January 2015 and December 2018 were included. The study was approved by Ethics committee of the institute. The hospital is only super specialty tertiary care hospital with teaching facility in government setup where paediatric surgery emergencies are being performed. It has a paediatric surgical unit with a 50 bedded ward that includes surgical neonatal intensive care unit. The records of all patients inflicted with ARM were reviewed along with their surgical intervention and associated co-morbid

conditions. The ARMs were categorized according to the Krickenbeck classification. In all babies routine blood count, babygram, prone cross table lateral x-ray focusing on greater trochanter, echocardiography, and abdominal sonography were performed. Chromosomal study was done in cases of syndromal association. We did primary surgeries for both male and female babies except patient presenting with sepsis, gut perforation, complex anomalies including cloaca and injudicious perineal exploration done outside hospital in which we did diverting colostomy. Records of 395 patients were available for study. The age at presentation was between one day and 9 years. Medical and radiological reports of these patients were reviewed. Data were collected regarding patient gender, age of presentation, type of anorectal lesions, associated anomalies and the operative procedure. Isolated anomalies were categorized by different organ system e.g. CVS, Genito-urinary, GIT and syndromal association. Data was entered and evaluated using Microsoft excel Version: 2019 16.0.6742.2048.

RESULTS

Three hundred ninety five patients of anorectal malformation were admitted out of 93,203 OPD patients. ARM represented 0.4% of patients coming to our hospital and 25.3% of neonatal surgical emergencies. Age of the patients varies from one day to nine years. Out of 395 children of anorectal malformations, there were 75.7% males (n=299) and 24.3% females (n=96) (Figure 1).

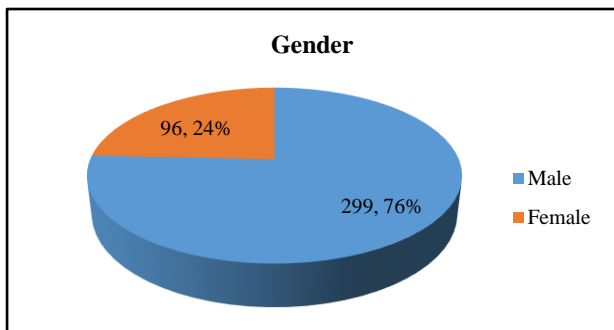


Figure 1: Gender distribution of anorectal malformation.

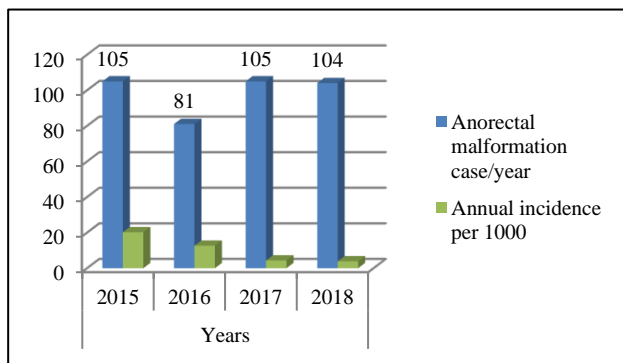


Figure 2: Annual incidence of anorectal malformation per 1000.

Table 1: Types of anorectal malformation gender crosstabulation.

Types of anorectal malformation		Gender		Total
		Male	Female	Male
Perineal fistula	count	62	12	74
	% within types of anorectal malformation	83.8%	16.2%	100.0%
	% within gender	20.7%	12.5%	18.7%
Recto-urethral fistula	count	215	0	215
	% within types of anorectal malformation	100%	0%	100.0%
	% within gender	71.9%	0%	54.4%
Recto-vasical fistula	count	9	0	9
	% within types of anorectal malformation	100%	0%	100.0%
	% within gender	3.0%	0%	2.3%
Recto-vestibular fistula	count	0	69	69
	% within types of anorectal malformation	0%	100%	100%
	% within gender	0%	71.9%	17.5%
Colaca	count	0	11	11
	% within types of anorectal malformation	0%	100%	100%
	% within gender	0%	11.5%	2.8%
No fistula	count	2	1	3
	% within types of anorectal malformation	66.7%	33.3%	100%
	% within gender	0.7%	1.0%	0.8%
Pouch colon	count	9	0	9
	% within types of anorectal malformation	100%	0%	100%
	% within gender	3.0%	0%	2.3%
Rectal artesia	count	2	0	2
	% within types of anorectal malformation	100%	0%	100%
	% within gender	0.7%	0%	0.5%
H-type fistula	count	0	1	1
	% within types of anorectal malformation	0%	100%	100%
	% within gender	0%	1.0%	0.3%
Recto-vaginal fistula	count	0	2	2
	% within types of anorectal malformation	0%	100%	100%
	% within gender	0%	2.1%	.5%
Total	count	299	96	395
	% within types of anorectal malformation	75.7%	24.3%	100%
	% within gender	100%	100%	100%

Male to female ratio was 3.11:1. The majority of the patient had presented in the neonatal period (52%) with mean weight of 2.6 kg. Most of the babies were from poor socioeconomic strata of the society. Annual incidence rate of ARM cases per year was shown in the (Figure 2).

Table 2: Types of operation performed.

Types of operation		Gender		Total
		Male	Female	Male
Anoplasty	Count	57	7	64
	% within type of operation	89.1%	10.9%	100%
	% within gender	19.1%	7.3%	16.2%
ASARP	Count	0	13	13
	% within type of operation	0%	100%	100%
	% within gender	0%	13.5%	3.3%
PSARP	Count	114	61	175
	% within type of operation	65.1%	34.9%	100%
	% within gender	38.1%	63.5%	44.3%
Abdomino PSARP	Count	2	0	2
	% within type of operation	100%	0%	100%
	% within gender	0.7%	0%	0.5%
APP	Count	79	2	81
	% within type of operation	97.5%	2.5%	100%
	% within gender	26.4%	2.1%	20.5%
Colostomy	Count	22	8	30
	% within type of operation	73.3%	26.7%	100%
	% within gender	7.4%	8.3%	7.6%
PSARVP	Count	0	3	3
	% within type of operation	0%	100%	100%
	% within gender	0%	3.1%	0.8%
Closure	Count	25	2	27
	% within type of operation	92.6%	7.4%	100%
	% within gender	8.4%	2.1%	6.8%
Total	Count	299	96	395
	% within type of operation	75.7%	24.3%	100%
	% within gender	100%	100%	100%

Average duration of hospitalization was 9 days. The most common type of ARM found in male patient was rectourethral fistula n=215 (54.4%) in which 164 (76.2%) patients were with rectobulbar fistula and 51 (23.8%) patients had rectoprostatic fistula. Sixty nine (17.4%) female patients presented with rectovestibular fistula. High anorectal malformation was found in 18 (4.5%) male and 13 (3.2%) female patients. High ARM male patients were mostly associated with rectovesical/baldder neck fistula and female patients were with common cloaca. Low malformation in form of cutaneous fistula and covered anus was found in 62 (20.7%) cases of male babies and in 12 (12.5 %) of female patients (Table 1). Most common type of surgery performed in male patient was PSARP (Posterior sagittal anorectoplasty) 65.2% followed by APP (abdomino-perineal pull through) 36.7% and anoplasty 19.1%. Among the female patient commonly performed surgeries were PSARP 63.5%, ASARP in 13.4% cases followed by anoplasty in 13.5% cases (Table 2). Among the associated anomalies urogenital and cardiovascular anomalies were found to be predominant (Table 3).

Table 3: Associated anomalies with anorectal malformation.

Associated anomaly	No. of anomaly	percentage of total patient
Cardiovascular anomaly	29	7.4%
ASD	14	
VSD	7	
PDA	3	
Teratology of fallot	1	
Dextrocardia	2	
Perinealhemangioma	2	
Genitourinary	39	9.9%
Hydronephrosis	6	
Absent kidney	5	
Undescended testes	7	
Hypospadias	6	
Penoscrotal transposition	14	3.8%
Aphallia	1	
GIT	15	
Tracheoesophageal fistula	7	
Duodenal atresia	3	
Ileal atresia	2	
Meckles diverticulum	2	6.6%
Malrotation	1	
Syndrome	26	
Down	8	
Trisomy 18	1	
VACTERL	12	6.6%
Currarino	5	

Among the urogenital anomalies, penoscrotal transposition 3.5% (14) was the most common anomaly found in our study followed by undescended testes 2.34% (7) and hypospadias 1.5% (6). Among the patients of hydronephrosis 4 cases presented with vesicoureteric reflux disease and 2 cases were associated with PUJ obstruction.

Gastrointestinal anomalies constituted 3.8% of the total anomalies. Commonest association was tracheo-oesophageal fistula 2.02% (8) cases. Among the syndromes, VACTERL (3%) was the commonest association found, in which one case was associated with trisomy 18 and unilateral proximal focal femoral deficiency. Down syndrome was found in 8 (2%) cases mostly in the male babies.

DISCUSSION

ARM have been recognized in animals since the time of Aristotle way back in the third century BC.³ The first attempt to classify ARMs date back to eighteenth century when Amussat described several groups of ARM.⁴ Present study is based on classification system based on the Krickenberg conference.⁵ It was developed because of dissatisfaction with previous classification systems; the relatively arbitrary definitions of 'high', 'intermediate' and 'low' ARMs were difficult to translate into clinical practice.⁶ The Krickenberg classification reflects the anatomical findings of ARMs, as they were identified through the experience with the posterior sagittal anorecto-plasty and the radiological studies of the relationship between the renal tract, the genital area and the hindgut structures (e.g. fistula).⁷ In our study annual incidence of anorectal malformation per 1000 shows declining trend over the years studied though the total number of cases of ARM was fairly constant over the years of study. We found a clear male preponderance resulting in a male to female ratio of 3.11:1 which was greater than observation made by other authors with male to female ratio ranging from 1.46: 1 to 2.4: 1.^{8,9}

Separating the cohort along gender lines the rectourethral fistula continues to be the most frequent ARM in boys followed by perineal fistula but in girls the most dominant ARM is a vestibular fistula (Figure 2). These results are similar with the findings of Nah et al.⁸ In our study, syndromal association was common in low birth weight babies similar to the study by Stoll et al.¹⁰ Overall, syndrome and associations accounted for 6.6% in this study this was in line with other series.¹¹ Down syndrome was the most common chromosomal anomaly present in 2% of the cohort with male predominance. Cuschieri et al and Chen observed chromosomal anomaly (trisomy 21) in 2 to 9% of the cases in their study.^{11,12} The two most frequently affected organ systems were the renal system (9.9%) followed by the CVS (7.4%). In renal system the most common association found was penoscrotal transposition comprising of 3.5% of total anomalies in which one case had complete transposition. In CVS,

commonest association was atrial septal defect followed by ventricular septal defect and patent ductus arteriosus. Tracheo-oesophageal fistula was the most common gastro intestinal tract anomaly (1.7%) detected which was in accordance with other study.¹³ A VACTERL association was only considered when three of the possible seven criteria were acknowledged together.^{10,12,14} VACTERL was the most frequent syndromal association and it was seen in 12 infants (5.5%), other investigators noted VACTERL association ranging from 5-15% in their respective studies.^{8,13} In our study a baby of monozygotic twin delivery was found to have aphallia and atretic urinary bladder. No familial association was noted in our study.

The commonly adopted technique in male/female patient for perineal fistula was anoplasty/ASARP (anterior sagittal anorectoplasty). For rectourethral fistula (rectoprostatic and rectobulbar) in male and rectovestibular fistula and rectovaginal fistula in female primary PSARP were performed (Table 1). The PSARP technique, as popularized by Peña et al is easy to teach and adopt and follows anatomical landmarks.¹⁵ In two cases (male) while doing primary PSARP surgery we could not locate rectal pouch so we did abdomino-psarp after changing the position of patient. We did primary surgery in most of the cases with the exception of the patients presented with gut perforation, complex anomalies such as common cloaca and in patients with injudicious perineal exploration done outside of the hospital in total 30 (7.5%) cases, where we did perform diverting colostomy. The overall mortality in our study was 3.5% (14 cases) mostly associated with sepsis and low birth weight. Others reported mortality rate of 4.5% Gangopadhyia et al and 12.6% Godse et al in their respective series.^{16,17} However, the present study was a retrospective and results were hospital based which is not truly representative of population is a limitation of this study.

CONCLUSION

We found a clear male preponderance of anorectal malformation in our study. Patients affected were mostly from poor socioeconomic strata of the society. Isolated case of ARM was prevalent than syndromal association. The genitourinary tract anomalies were found to be commonest one in anorectal malformations. Correction of this malformation in single stage offers satisfactory outcome at affordable cost. Present study confirms huge work load of anorectal malformation in neonatal surgical emergencies in this region. This will be helpful in future planning and effective management including financial constraints of the treatment for our poor patients.

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Conflict of interest: None declared

Ethical approval: The study was approved by the Institutional Ethics Committee

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