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Evaluation of anorectal malformations in a tertiary care Centre: The role of a radiologist

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Abstract
Anorectal malformation (ARM) are of important common congenital anomaly with regard to final outcomes since the anorectal lesions with associated lesions that cause intestinal obstruction are correctable with operative intervention requires prompt radiological investigations.
Aims: To study the presentation, types of anomalies, associated malformations, role of radiological investigations and procedures performed in relation to the type of anomaly and early outcomes analysis of ARM patients in the neonatal period.
Settings and Design: A prospective study was performed from January 2017 to September 2020.
Subjects and Methods: The study included all patients with ARM admitted in the Neonatal Intensive Care Unit.
Results: There were 60 neonates having ARM with 32 males and 28 females. High-type ARM was seen in 38 (63.33%) cases, while low type in 22 (36.66%) neonates. Associated malformations were documented in 4 cases of (6.66%) neonates. All neonates were diagnosed on the 1st day of life. There were 01 (1.66%) deaths, 59(98.33%) survivors.).
Conclusions: Radiology plays an important role in diagnosing ARM, associated fistulas, classification and associated anomalies.

Keywords: ARM, PSARP

Introduction
Anorectal malformations (ARMs) comprise wide spectrum of diseases, affecting both sexes [1]. ARM is diagnosed because of the absence of an anus or the presence of an ectopic anus at birth [1]. It is essential to diagnose accurately all the associated malformations in ARM and with plain radiograph lateral invertogram, contrast studies (MCUG), erect radiograph and cross table lateral radiograph, associated lesions as soon as possible, as these have important implication with mortality and morbidity, since the anorectal lesions (majority of cases) that cause intestinal obstruction are correctable with operative intervention [2]. Surgical management of a neonate with ARM is directed according to the level of anomaly as well as the presence or absence of a fistula between the ano-rectum and urinary tract or vagina. We aimed to study the presentation, types of anomalies, associated malformations, and radiological investigations in the diagnosis, procedures performed in relation to the type of anomaly and associated malformations, and early outcomes analysis of ARM patients in the neonatal period at our tertiary care institute.

Subjects and Methods
The study was carried out in Department of Radiodiagnosis, Sapthagiri Institute of Medical Sciences and Research, Bengaluru 560090.

Study design
Type of the study**:** A prospective Hospital Based Study.

Sample size
60 patients Patients referred by the clinician from our hospital and from outside, to radiology department of SIMSRC with suspected ARM, with complaints of non-passage of stool since 24 hours, Abdominal distension, Excessive crying, Passage of stool through any other opening than anus admitted in the Neonatal Intensive Care Unit (NICU) of the department of pediatric surgery over a period of 3 year from January 2017 to September 2020 were prospectively studied.
A detailed history and careful physical examination was completed. An invertogram, contrast studies (MCUG), erect radiograph and cross table lateral radiograph was performed in whom the examination did not reveal the type of ARM. In female patients with cloacal malformation/vestibular fistula in whom the diagnosis was established by perineal examination, a babygram was performed to diagnose the presence of congenital pouch colon (CPC).
ARM was classified into high and low types according to the Krickenbeck classification [3]. Ultrasonography of the abdomen,

spinal USG was performed to evaluate for cloacal malformation.

Inclusion criteria.

1. Not passing stools since birth
2. Absence of normal anal opening
3. Passing stools from abnormal routs i.e. vagina, urethra or ectopic sites,
4. Abdominal distension,
5. Vomiting, excessive crying,
6. Off and on constipation,
7. Thin pipe stools

Exclusion criteria

Patient who did not give consent for inclusion in the study & other causes of abdominal distension and vomiting.

Results

Table 1: Sex distribution

Gender	Number	%
Male	32	53.33
Female	28	46.67

Table 2: Age distribution.

0-5days	5days-1month	1month-3months	3months-1year	>1yr
21	5	5	13	16

Table 3: Type of anorectal malformation

Type of lesion	No of cases	%
Anorectal agenesis with rectovaginal fistula	9	15
Anal agenesis with recto vestibular fistula	4	6
Anteposed anus with anal stenosis	3	5
Anal agenesis with recto perineal fistula	11	18
Anal agenesis without Fistula (pouch colon=2)	30+2	53
Cloaca	0	0
Total	60	

Table 4: Sex Incidence and level of lesion

Level of lesion	Present study n=60		Indian study[6] n=30		Chau Series[4] n=108	
	n	%	n	%	n	%
High	30	50	14	46.66	39	36.1
Intermediate	8	13.3	4	13.33	32	29.62
Low	22	36.6	9	30	35	32.04
Cloaca	0	0	3	10	2	1.85
Total	60		30		108	

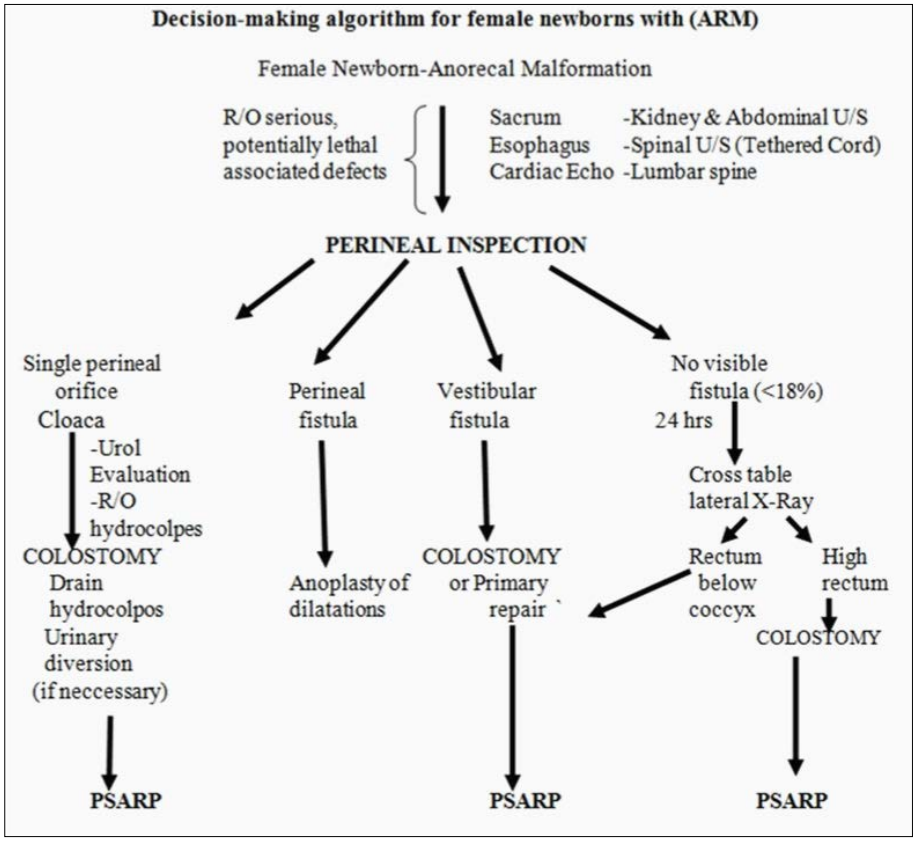

Fig 1

Fig 2

Discussion

Anorectal malformations (ARMs) are relatively rare congenital anomalies which affect both sexes with an incidence of around 1 in 5000 live births [7]. ARMs are often associated with other congenital anomalies with an approximate incidence of 50 % and are presented in a great number of syndromes It has a wide spectrum of muscle sphincter complex development from near-normal muscles to complete absence of the sphincter muscle. Routinely, depending on the level of the obstruction in above and below of muscle sphincter, this anomaly is categorized into two groups; high and low type [8].

Low variety: Blind end of rectum terminates below the level of puborectalis sling of levator ani muscle,

Radiological – gas shadow is seen below the line drawn from end of coccyx to pubic symphysis.

High variety: Blind end of rectum terminates above the level of puborectalis sling of levator ani muscle.

Radiological – gas shadow is seen above the line drawn from end of coccyx to pubic symphysis

An invertogram, contrast studies (MCUG), erect radiograph and cross table lateral radiograph was performed in whom the examination did not reveal the type of ARM.

Plain film - Lateral invertogram is used to investigate the extent of defect in the anal or rectal atresia. The anus is marked with radio opaque marker, the baby is inverted. A lateral radio graph is taken in the air, the rectum will rise to the highest point, giving an indication of the extent of the atresia.

Abdominal radiograph: Can be variable depending on the site of atresia (e.g. high or low), level of impaction with meconium and physiological effects such as straining. May show multiple dilated bowel loops with absence of rectal gas

Prone cross table lateral radiograph: Provides equal or sometimes better information, compared to the invertogram, for demonstration of the level of rectal atresia in neonates.

Its advantages are - easy positioning, better cooperation of the patient, elimination of the effect of gravity, and better delineation of the rectal gas shadow. The infant would be placed in a prone position with the hip flexed and elevated up to 45 degrees. The radiographic center was placed around the greater trochanter. A radiologic marker was routinely placed at the perineal area where there should be anal dimpling.

Fluoroscopy - contrast study

- To detect recto-vesical, recto-vaginal or rectoperineal fistula.
- The fistula is considered low (below levator ani plane) if it is below the pubo-coccygeal line and considered high fistula if above the pubo-coccygeal line.

Ultrasound

- The anus may be seen as an echogenic spot at the level of the perineum and in an atresia this echogenic spot may be absent.
- May show bowel dilatation
- An infra coccygeal or transperineal approach may allow differentiation between a high or low sub-type [4].

Sonography is usually used to determine the level of the disorder (low and high) indirectly based on the distance between location of anoplasty and pouch of rectum. Although this approach is not very determinative and there are a lot of diagnostic overlaps in this field. Our experience was limited [8, 9]. MRI is more sensitive but less specific in fistula detection when compared with distal cologra [10].

In the present prospective study of 60 patients, 18 (60%) patients presented in neonatal period and 12 (40%) in post neonatal period as compared to the British Columbia Hospital study comprising 120 patients, in which 105 (87.50%) patients presented in neonatal period and 15 (12.50%) in post neonatal period[11]. Factors responsible for higher rate of delayed presentation in present study were lack of awareness, illiteracy, poverty and negligence of parents. The common presenting symptoms in the present study were Non passage of meconium per anus since birth (76%), abdominal distension (100%), constipation (70%), vomiting (20%) and passing of stools per vagina (3.70%) were presenting symptoms [6, 12]

Anorectal agenesis without fistula was the most common type of anorectal malformation n=32(43.33% and associated anomalies were shown in Table 3.

One case is associated with VACTERL, two case with Down's syndrome & a case with choanal atresia [13, 14]. In the present study, most of associated congenital anomalies were seen in high variety of ARM. Similar findings have been reported by other authors [13].

In the present study, in 40 patients PSARP (Posterior sagittal anorectoplasty) was done, PSARP was combined with abdominoperineal pull through in 14 cases. Cut back anoplasty in 6 and anal dilatation done in 6 cases. In Chau series of 108 patients of imperforate anus, PSARP was done in 65.74% patients, PSARP with pull through in 1.85%, cut back anoplasty in 27.77% and anal dilatation in 4.62% patients [1]. One case died

because of post op sepsis.

Limitation of Study

The present study suffered from the following limitations:

- Small sample size
- Short follow-up period.

Conclusion

It was concluded that plain radiograph lateral invertogram proves to be the most important modality in diagnosing imperforate anus. It is followed by prone cross table lateral radiograph and abdominal erect radiograph. Micturating cystourethrogram proves to be important in diagnosing associated anomalies such as rectovesical fistula.

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Ethical clearance: Obtained.

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