

Case Report

Anorectal Malformations: Epidemiological, Clinical, Therapeutic and Evolutionary Aspects in the General Surgery Department of the Sominé DOLO Hospital of Mopti

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Abstract: We conducted a retrospective and prospective study. In our study, we collected 35 cases of MAR, including 21 boys and 14 girls, over a period of 4 years, from January 2017 to December 2020. We recorded an average of 8.7 cases of ADR per year. They are more frequent in boys, with a sex ratio of 1.5. The majority of our patients came from the Mopti region and were of Dogon ethnicity. Most of the malformations were discovered by the parents, given that 54.3% of the mothers had given birth in a health centre and the malformed children were most often seen in a state of intestinal obstruction or sub-occlusion requiring an emergency colostomy. The diagnosis was made during the systematic examination of the newborn in the delivery room in 8.57% of our patients. Low forms were more frequent (48.6%). Invertograms were performed in 15 patients (42.9%). Abnormalities associated with MAR were dominated by spinal malformations in 4 cases (11.4%). In terms of treatment, the operative techniques used, in order of frequency, were: ASARP 8 cases (30.8%); Pena PSARP 15 cases (57.7%), YV Proctoplasty 2 cases (7.7%) and PENA PSARP plus MOLLARD AP 1 case (3.8%). Average follow-up was 12 months. Four (4) patients (15.4%) were lost to follow-up. Post-operative follow-up was favourable in 17 patients (65.4%); 4 cases of death and 1 case of anal stenosis.

Keywords: Anorectal malformations, Somine Dolo, Hospital, Mopti.

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INTRODUCTION

Anorectal malformations represent a very wide range of malformations, from simple defects in the resorption of the anal membrane to anorectal agenesis with fistulas in the urinary or genital tracts and sacral anomalies [1].

There are several anatomical forms, classified as high, intermediate and low, depending on the position of the intestinal pouch in relation to the levator ani muscles.

The incidence of anorectal malformations (ANMs) varies from 1:2500 to 1:5000 births and may

vary according to geographical, ethnic and genetic variations [2].

The aetiology of MAR remains unknown but appears to be multigenic [3]. However, favourable factors such as vascular defects and medication taken by the mother during pregnancy (drugs, toxins, etc.) are thought to be involved.

Diagnosis of MARs is generally straightforward and is usually made in the delivery room, although the diagnosis of associated malformations is not as straightforward [4].

The diagnosis must be made in the delivery room during the first examination of the newborn,

although certain additional examinations are still necessary for classification. In the African context, the diagnosis is made late, often in the presence of a picture of occlusion.

Treatment is only considered once a precise topographical diagnosis has been made, with low forms requiring only a simple perineal operation, whereas high and intermediate forms require relatively lengthy and complex operations under colostomy cover [5].

While the immediate vital prognosis depends above all on the severity of the associated malformations, the subsequent functional prognosis depends much more on the type of MAR and the quality of the surgical repair.

Studies have been carried out on anorectal malformations in particular:

In the United States, Pena achieved over 90% in 2000 [8].

In Japan, Endo M collected 86% in 2000 [9].

In Spain, Nazer identified 43% of fistula forms in all MAR in 2000 [10].

In Burkina Faso, Wandaogo found a 42.2% frequency of fistula in a series of 45 cases of MAR in 2005 [7].

In Mali, Konandji found 23.33% of MAR with fistula in his series of 30 cases in 2004 [6].

The lack of data at regional level prompted the initiation of this study at the Somine Dolo hospital in Mopti, with the following objectives:

GENERAL OBJECTIVES

Study of anorectal malformations in the general surgery department of the Sominé Dolo Hospital in Mopti.

SPECIFIC OBJECTIVES

- To determine the hospital frequency of anorectal malformations at the Mopti DSH.
- To describe the epidemiological and clinical aspects of anorectal malformations at the HSD in Mopti.
- To describe the management of anorectal malformations at the HSD in Mopti.

METHODS

Inclusion criteria

All patients aged between 0 and 15 years who came for MAR at the HSDM were included in our study.

Non-inclusion criteria: No non inclusion criteria.

RESULTS

We recorded 35 cases of ADR during our study period.

1. Epidemiological aspects of MAR

The overall frequency of children hospitalised during our study was 291.25 cases/year.

1.1 Annual MAR frequency

The average frequency of ADR was 8.7 cases/year (extremes: 5 and 16 cases).

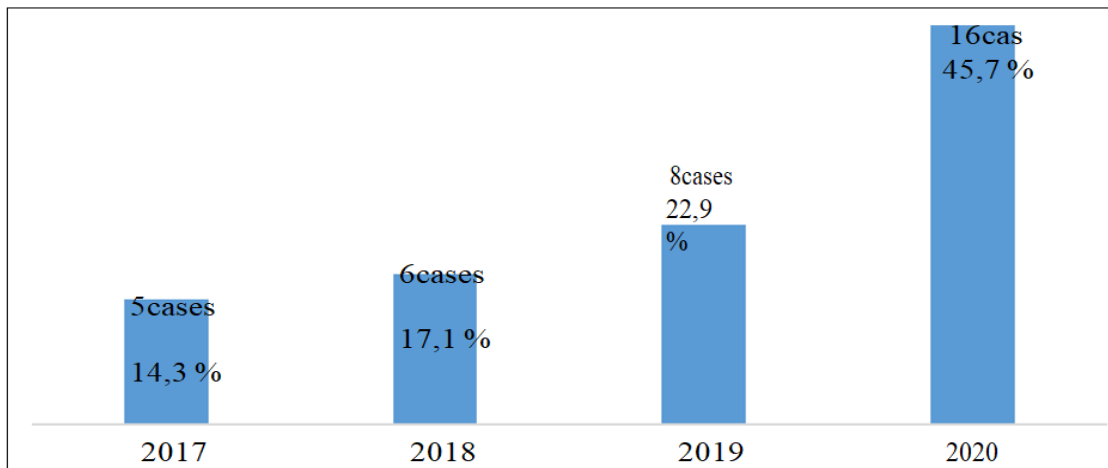
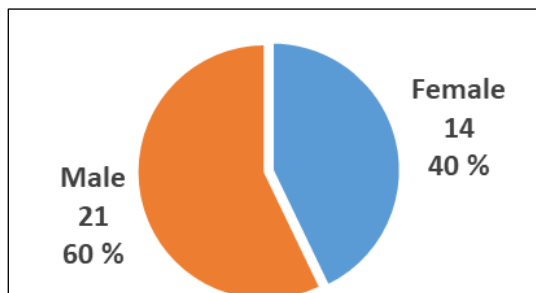


Table I: Breakdown of patients by age group

Age	Workforce	%
Newborns (0 - 28 days)	23	65,7
Infant (1 month-2 years)	11	31,4
Older children (8 - 15 years)	1	2,9
Total	35	100

The age of our patients ranged from 1 day to 13 years, with a median of 4 months.

Gender



The sex ratio is 1.5

Table II: Breakdown of patients by type of hospital admission

Method of admission to hospital	Workforce	%
Brought by parents	23	65,7
Referral by a health agent	12	34,3
Total	35	100

65.7% of our patients were brought in by their parents

2.1.5. Admissions department

Table III: Breakdown of patients by admission department

Admissions department	Workforce	%
Emergencies	21	60
External consultation	14	40
Total	35	100

60% of our patients were admitted as emergencies

Table IV: Breakdown of patients by reason for consultation

Reasons for consultation	Workforce	%
Occlusion or sub-occlusion	22	62,8
Emission of faeces through the vulva	10	28,6
Absence of anus with perineal fistula	3	8,6
Total	35	100

Occlusion or sub-occlusion was frequent in 62.8% of cases.

Table V: Breakdown of patients by ASA stage

ASA	Workforce	%
ASA I	13	37,1
ASA II	14	40
ASA III	8	22,9
ASA U	35	100

40% of our patients were ASA II.

Table VI: Distribution of patients according to the result of the perineum examination

Examination of the perineum	Workforce	%
Covered anus with flat perineum	15	43,8
Rectal fistula in the vulvar forchette	7	20
Anus fully covered with rounded perineum	6	17,1
Incomplete covered anus with anocutaneous fistula	2	5,7
Incomplete covered anus with ano-vestibular fistula	2	5,7
Anus covered with 1 vaginal opening	2	5,7
Recto-vaginal fistula	1	2,9
Total	35	100

An invertogram was performed in 15 patients (42.9%).

Table VII: Distribution of patients according to invertogram result

Invertogram	Workforce	%
MAR high	8	53,4
MAR low	5	33,3
MAR Intermediates	2	13,3
Total	15	100

High MARs were common in 53.4% of cases.

Table VIII: Distribution of patients according to whether colography and fistulography were performed

ASP	Numbers (n=35)	%
Colography		
Realized	15	42,9
Not carried out	20	57,1
Fistulography		
Realized	11	31,4
Not carried out	24	68,6

Colonography was performed in 15 patients (42.9%) and fistulography was performed in 11 patients (31.4%) to diagnose the type of MAR.

Table IX: Distribution of patients according to the results of the abdominal-pelvic ultrasound scan

Abdo-pelvic ultrasound	Workforce	%
Multicystic kidney	2	7,1
Right renal agenesis	1	3,6
Right renal ectasia	1	3,6
Right ectopic kidney	1	3,6
Total	5	17,9

A multi-cystic kidney was found in 7.1% of cases.

Type of MAR by gender

Table X: Breakdown of patients by type of MAR by sex

Types of MAR	Sex		Total (%)
	Male (%)	Female (%)	
MAR High	11/15 (71,4)	4/15 (28,6)	15/35 (42,9)
MAR intermediates	2/3 (75)	1/3 (33,3)	3/35 (8,5)
MAR low	8/17 (47,1)	9/17 (52,9)	17/35 (48,6)

High and intermediate MARs were more frequent in boys and low forms in girls.

Treatment and Development of MAR

A colostomy was performed in 19 patients (54.3%) before the operation, including:

- 17 cases (89.5%) of high and intermediate MAR

- 2 cases (10.5%) of low MAR
- 3 patients died after colostomy;
- 5 patients were lost to follow-up;
- 1 patient was referred to another health facility at the parents' request.

Table XI: Distribution of patients of Surgical technic

Surgical technic	MARB		MARHI		Total	
	Numbers	%	Numbers	%	Numbers	%
ASARP	8	30,8	-	-	8	30,8
PSARP	6	23,1	9	34,6	15	57,7
Proctoplastic YV	2	7,7	-	-	2	7,7
PSARP+AP de Mollard	-	-	1	3,8	1	3,8
Total	16	61,6	10	38,4	26	100

Colo-anal lowering by perineal sagittal anorectoplasty was performed in 57.7% of patients.

Table XII: Breakdown of patients according to the Clavien-D classification

Clavien classification	Numbers	%
Grade I	18	69,2
Grade III a	3	11,5
Grade III b	1	3,9
Grade V	4	15,4
Total	26	100

69.2% of our patients were Clavien Grade I.

Table XIII: Distribution of patients according to postoperative evolution after 6 months

Evolution at 6 months	Number	%
Favourable	15	57,7
Deaths	4	15,5
Lost from sight	3	11,5
Anal stenosis	3	11,5
Recurrence	1	3,9
Total	26	100

Progression was favourable in 57.7% of cases after 6 months' follow-up.

COMMENTS AND DISCUSSION

This was a retrospective and prospective study of ADR in the general surgery department of the HSD in Mopti. Over a period of 4 years, we identified 35 patients who met our inclusion criteria.

In our study, the average annual incidence of ADR was 8.7 cases. This figure is close to that found by Bandré [18] in Burkina Faso but lower than those reported by Habou [3] in Niger, Mouafo [19] in Côte d'Ivoire and Pranshu [20] in India.

The incidence of ADR in our regions probably does not reflect reality. It may be underestimated for a variety of reasons: ignorance, difficulties in accessing healthcare facilities (insecure areas), lack of resources and qualified staff.

In our series, 60% of patients were male. This male predominance is found in many other series [4, 7, 19]. Although no obvious explanation has been put forward, some authors [8, 9, 22] believe that this male influence is partly based on genetic factors which have yet to be demonstrated.

MAR was diagnosed late in 34.3% of our patients. Wandaogo [7] found a late diagnosis in 20% of cases in Burkina Faso in 2005 and KIM [23] in 18.8% of cases in the USA in 2000.

The delay in diagnosing ADR is due to the clinical form of ADR, which, when associated with a sufficiently large fistula, can delay the onset of transit disorders, in particular occlusive syndrome and constipation. Secondly, the absence of a thorough examination of the perineum in the delivery room, which should end with a rectal examination, because the presence of a normal-looking anus does not immediately mean that it is permeable. The position of the anus,

normally between the two ischials, should be checked; an anteriorly positioned anus should not be overlooked.

In our study, 45.7% of our patients were born at home. This rate is significantly higher than those of LANOU [25] in Burkina Faso and MOUAFO [19] in Côte d'Ivoire, who had rates of 6.7% and 17.9% respectively.

The high frequency of home births is thought to be linked to a number of factors, including the absence or remoteness of health centres, certain traditional beliefs and a lack of resources.

However, the influence of the place of birth on the earliness of diagnosis has not been demonstrated [25], as shown by our 91.4% rate of diagnosis outside the delivery room. This suggests that efforts should be made to raise awareness among traditional birth attendants, matrons and health workers of the importance of systematically inspecting the perineum of all newborns.

An occlusive or sub-occlusive syndrome is the reason for consultation in 62.9% of our patients.

UBA [60] in Nigeria and MOUAFO [19] in Abidjan have clearly established the occlusive picture as the first complaint prompting consultation by parents. Intestinal obstruction is a frequent complication of ADR, which explains the delay in diagnosing this pathology.

The positive diagnosis of MAR is generally obvious. However, it is the diagnosis of clinical forms that determine the surgical procedure that requires further investigations. This explains why invertograms were performed in 42.9% of our patients, colography in 42.9% and fistulography in 31.4%.

MOUAFO [19] in the Ivory Coast performed an invertogram in 60% of cases combined with

opacification of the colon in the case of a fistula. LANOU [25] in Burkina Faso performed an invertogram in 28.9% of cases.

The invertogram (Wagensteen and Rice film) is essential for determining the level of the intestinal cul-de-sac, and can be used to differentiate the clinical forms of MAR for surgical management.

Low MAR (MARB) is more frequent in our study, followed by High MAR (MARH). Our results are similar to those found in other series.

We found a predominance of MARB in girls, especially of the vulvar anus type. The frequency of ano-vulvar fistula in MARB in girls has been reported by other authors [3, 14].

High and intermediate MAR are frequent in boys in our study, as reported in other series [3, 17, 25].

Associated malformations were found in 8 patients (22.9%) in our study. LANOU [25] reported an associated malformation rate of 25.4% in Burkina Faso. MERIÈME [5] reported 31.03% association with other malformations in Morocco. Politi [16] generally described a rate of 30 to 50% of association of other malformations with MAR.

Our frequency may be underestimated because not all of our patients were systematically screened for associated malformations due to a lack of financial resources for the parents.

A colostomy was performed in 54.3% of our patients before anoplasty. MOUAFO [19] in Côte d'Ivoire performed an emergency colostomy in 43% of cases. In Burkina Faso, LANOU [25] performed a standby colostomy in 68.9% of these patients.

This emergency or waiting colostomy, depending on the series, is justified even in low forms, on the one hand in the presence of a picture of intestinal occlusion, as found in 62.9% of our patients on admission to the centre, and on the other hand by the difficulties of imaging in many cases to locate the rectal cul de sac. Also, some of our patients had already had a colostomy before being referred to our centre.

The surgical treatment of low-lying MAR is well codified and the subject of a consensus [3, 17]. We have performed anterior sagittal anorectoplasty, Pena posterior sagittal perineal anorectoplasty and YV proctoplasty in these low forms.

The treatment of high and intermediate MAR is controversial due to the complexity of their correction. We have performed anorectoplasty using the posterior sagittal perineal approach of Pena or combined with the abdominoperineal approach of Mollard in one case,

because of the advantages of these techniques, such as precise identification of the muscular elements, removal of the recto-urinary or genital fistula under direct visual control, and rectal docking in the deep plane to avoid prolapse.

MOUAFO [19] in the Ivory Coast and LANOU [25] in Burkina Faso used the Stephens and Douglas technique. This technique does not allow identification of the more superficial structures of the external sphincter and carries a high risk of prolapse because the rectum is not attached to the various muscular structures. However, it appears to be less damaging than the previous two techniques.

Generally speaking, most authors agree on the choice of techniques for low-lying MAR. This choice depends on the surgeon's convictions and preferences in high and intermediate forms.

The outcome at 12 months after colorectal reduction was favourable in 77.3% of our patients. Mortality was 15.5% and anal stenosis 4.5%.

Our results are comparable to those of: HABOU [3] in Niger who reported a good result in 53.04% of cases, anal stenosis in 14.78% and a mortality rate of 13.3% after 14 months. LANOU [25] in Burkina Faso reported a favourable outcome in 62.5% of cases and a mortality rate of 34.6% after 3.6 years.

The percentage of favourable results varies from 50 to 70% according to authors [5, 18, 20]. In our case, a favourable result is defined as the patient being able to lead an acceptable social life (without major soiling).

CONCLUSION

Anorectal malformations are uncommon pathologies in our context. They are more common in boys. At birth, the majority of cases were discovered by the parents at home. Children presenting with MAR were generally seen with intestinal obstruction, leading to emergency colostomy.

Clinical and radiological examinations were used to identify the different types and varieties of MAR. High and intermediate MAR were dominated by boys and low MAR by girls. Associated malformations were dominated by spinal deformities.

In our series, the surgical technique most frequently used was anorectoplasty via the posterior sagittal perineal approach of Pena.

The average follow-up was 12 months, and 77.3% of patients with anal stenosis had a favourable post-operative outcome. The mortality rate was 15.5%.

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