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Anorectal Malformations Operated at University Hospital Brazzaville

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Abstract

Aim: To appreciate the care of children operated for an anorectal malformation. **Materials and methods**: It was a retrospective study carried out from January 2014 to December 2018 (5 years) in the department of pediatric surgery of university hospital of Brazzaville. **Results**: We retained 35 files including 20 boys and 15 girls. The mean age of discovery of the malformation was 1.7 days (range 0 and 4 days). On physical examination, we found intestinal occlusion in 22 patients (62.9%): the absence of an anal opening (62.9%), abdominal bloating with tympanism were the most represented signs. (57.1%). There were 13 cases (37.1%) of anorectal malformations high, 12 cases (34.3%) of low and 10 cases (28.6%) of intermediate. There were 20 cases (57.1%) of anorectal malformations without fistula and 15 cases (42.9%) with fistula. The mean age at the time of the anal plasty was 12.7 months (range 2 days and 14 years). We performed the anorectoplasty according to the Peña and De Vries technique in 26 cases (74.3%), perineal anoplasty in 5 cases (14.3%) and a transposition of the fistula in 4 cases (11.4%). Twenty-seven patients (77.1%) were reviewed with an average follow-up of 2.7 years (1 year and 5 years extremes). The anus had a normal aspect in 20 cases (74.1%). We evaluated anal continence according to the Krickenbeck criteria in 10 patients aged over 3 years, and six had good results.

Keywords

Anorectal Malformations, Anorectoperineoplasty, Child

1. Introduction

Anorectal malformations (ARM) are congenital anomalies that partially or com-

pletely interrupt the continuity of the terminal portion of the digestive tract or modify its topography [1]. Anorectal malformations (ARM) are the result of an abnormal development of the terminal part of the digestive tract interesting anus and/or rectum that occur early between the sixth and tenth week of embryonic development. They carry a malformation spectrum of severity depending on the level of disruption of the anorectal canal and of the associated. They are among the most frequent congenital surgical anomalies of the digestive tract [2]. In western countries, their incidence is estimated at one case per 5000 live births [3]. The sex ratio is 2/1 in favor of girls [4]. In the Democratic Republic of Congo, Ngondo *et al.* [5] reported a hospital frequency of 16.2%. There are high, low, and intermediate ARM according to the position of the rectal cul-de-sac in relation to the levator ani muscles [6]. Their treatment is based on a precise topographic diagnosis and the functional prognosis depends on the type of ARM and the quality of its surgical repair; while the vital prognosis depends on the severity of the associated malformations [3] [6]. However, the functional results of ARM surgery have improved considerably with the Peňa and De Vries technique or anorectoplasty by posterior sagittal perineal route, with section of the perineal muscles followed by their reconstruction [6]. Recently, a consensus was agreed upon by the International Conference for the Development of Standards for the Treatment of Anorectal Malformations at Krickenbeck Castle, Germany, in May 2005 [2]. We carried out this study in order to assess the management of ARM in our environment.

2. Patients and Methods

It was a retrospective study carried out over a 5-year period from January 2014 to December 2018 at the pediatric surgery of the university hospital of Brazzaville. We included children from zero to 16 years old operated for an anorectal malformation. We excluded non-operated patients. We have collected data from medical records, admissions registers and operational reports from the department. The study population consisted of 35 patients including 20 boys and 15 girls, a sex ratio was equal to 1.3; the mean age was 10.6 months (range 1 day and 14 years). The variables studied were:

- Diagnosis: the consultation period, the circumstances of discovery, the clinical signs and the results of the radiological examinations, the associated malformations;
- Therapeutic: surgical procedures, length of hospital stay, postoperative treatment, long-term anatomical and functional results according to the Krickenbeck criteria [7].

The quantitative variables were expressed as percentages and the qualitative variables as an average.

3. Results

3.1. Diagnostic Aspects

Circumstances of discovery

- The average age of discovery of the malformation was 1.7 days (range 0 and 4 days);
- The absence of an anal opening represented the most frequent reason for consultation with 62.9% of the patients (Figures 1-3).

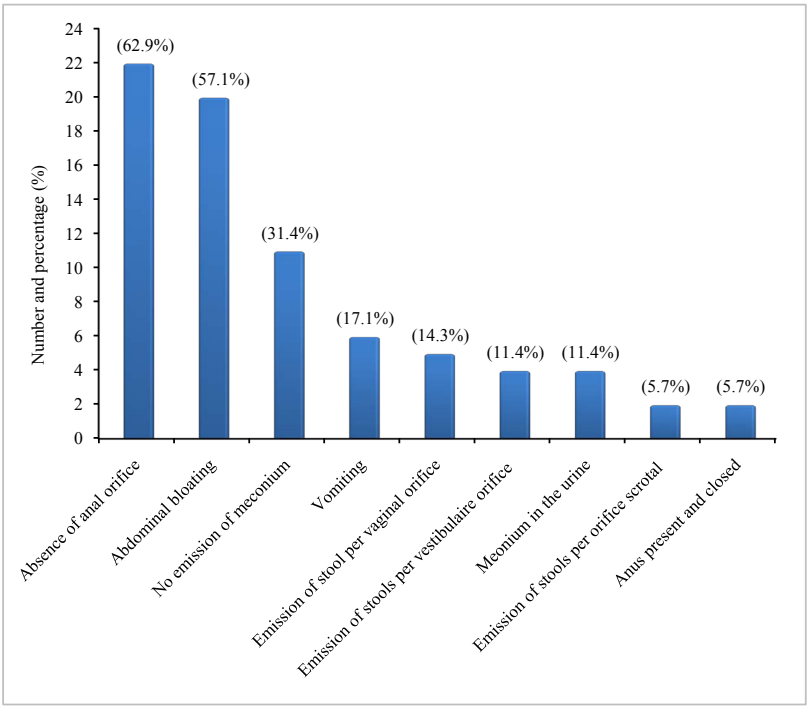

Figure 1. Reasons for consultations.

(a) (b)

Figure 2. Abdominal bloating (a) and absence of anal orifice in a newborn (b).

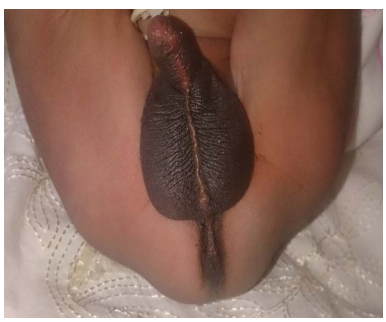

Figure 3. Perineal swelling related to low MRA in newborn.

Physical signs

On physical examination, we found an intestinal obstruction in 22 patients (62.9%): the absence of an anal opening (62.9%), abdominal bloating with tympanism were the most represented signs (57.1%), as shown in Table 1.

Radiologic examinations

- Twenty-five (57.1%) patients performed an invertogram. The distal colostogram by colostomy was performed in three patients, and had shown two upper rectovaginal fistulas (Figure 4) and one rectovesical fistula (Figure 5).
- The x-ray of the spine had to detect two cases of spinal malformations including agenesis of the coccyx. There was no presacral tumor.
- The abdomino-pelvic ultrasound did not show any associated malformations of the intra-abdominal organs.
- Cardiac ultrasound revealed interventricular communication in two patients.

Table 1. Distribution of patients according to physical signs.

Physical signs	**Effective**	**%**
Abdominal bloating	**20 (N = 35)**	**57.1**
meteorism	**20 (N = 35)**	**57.1**
Anus	**N = 35**	**100**
Absent	22	62.9
Ectopic	11	31.4
Present and obstructed	2	5.7
Types of fistula	**15**	**42.9**
Recto-vaginal	5	33.3
Recto-urinary	4	26.7
Recto-vestibular	4	26.7
Recto-scrotal	2	13.3
Associated malformations	**7**	**20**
Rachidian	2	5.7
Cardiac	2	5.7
Ears	1	2.9
finger	1	2.9
Ocular (cat eye)	1	2.9

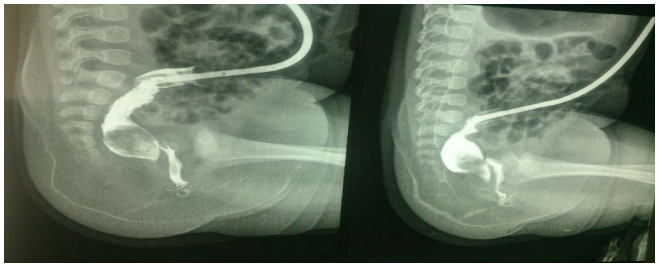

Figure 4. Distal cologram showing rectovaginal fistula in girl of 5-month-old.

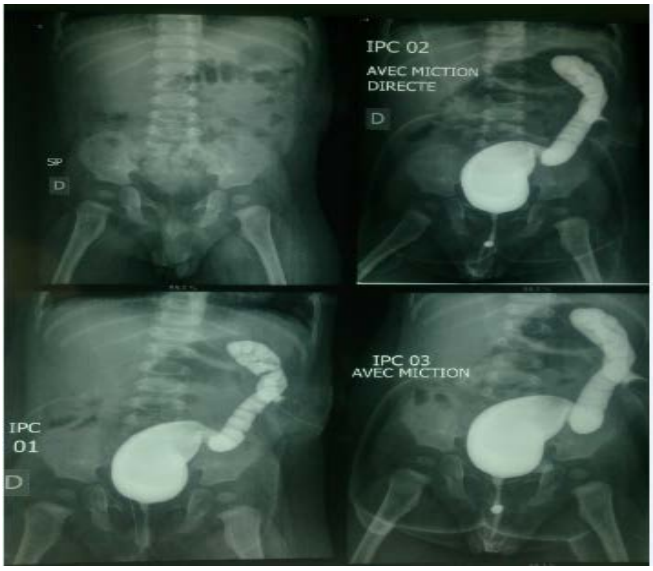

Figure 5. Distal colostogram showing rectovesical fistula in a 6-month-old boy.

Diagnosis retained

At the end of the clinical and radiological assessment, we distributed the patients into high MAR with 13 cases (37.1%), low ARM with 12 cases (34.3%) and intermediate ARM with 10 cases (28.6%). There were 20 cases (57.1%) of ARM with no fistulas and 15 cases (42.9%) of ARM with fistula. ARM were associated with another malformation in 7 cases (20%) (Table 1). Figure 6 illustrates cat eye in an infant of 3-month-old with an anorectal malformation.

3.2. Therapeutic Aspects

Colostomy

It was performed in 20 patients (57.1%). All colostomies were of sigmoid seat including 14 double terminalized (or with cutaneous bridge) and six on rods.

Anoplasty

The mean age at the time of the anarectoplasty was 12.7 months (range 2 days and 14 years). We performed the Peňa and De Vries technique in 26 cases (74.3%), perineal anoplasty in 5 cases (14.3%) and a transposition of the fistula in 4 cases (11.4%).

We performed some anal dilatations in 25 patients (71.4%) at one to two sessions per week.

All the patients had had a colon preparation before surgery: antibiotic therapy, deworming and colonic enemas either by fistula or by stoma.

Evolution during hospitalization

The average length of hospital stay was 6 days with extremes of 3 and 12 days. The postoperative follow-up was simple in the majority of cases (62.9%), as shown in Table 2.

Long-term evolution

Among the 27 patients reviewed (77.1%) with an average follow-up of 2.7

years (extremes of 1 year and 5 years). The anus aspect was normal in 20 cases (Table 3). Anal continence was evaluated according to the Krickenbeck criteria in 10 patients aged over 3 years (Table 4). On anal continence, our results were good in 6 out of 10 patients evaluated.

Table 2. Distribution of patients according to the evolution during hospitalization.

Evolution during hospitalization	Effective	%
Simples	**22**	**62.9**
Complications	**11**	**31.4**
Suture dehiscence	*8*	
Suppuration	*3*	
Death	**2**	**5.7**
Total	**35**	**100**

Table 3. Distribution of patients according to long-term development.

Anus aspect	Effective	%
Normal	20	74.1
Anal gaping	4	14.8
Anal stenosis	2	7.4
Mucous ectropion	1	3.7
Total	**27**	**100**

Table 4. Postoperative results according Krickenbeck criteria.

N° Voluntary bowle movement	Soiling	Constipation
1	Yes	No
2	Grade 1	
3	Yes	No
4	No	
5	Yes	No
6	No	
7	Yes	Grade 1
8	No	
9	No	Grade 2
10	No	
11	Yes	No
12	No	
13	Yes	No
14	No	
15	No	Grade 2
16	No	
17	Yes	No
18	No	
19	Yes	Grade 1
20	No	

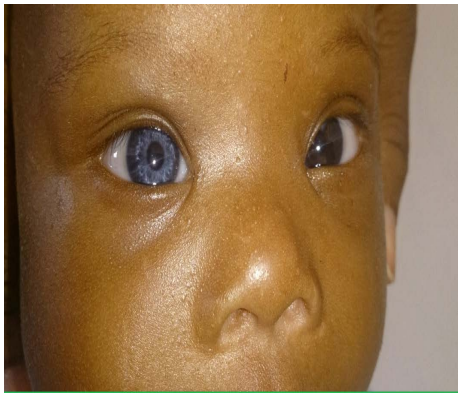

Figure 6. Cat eye in a 3-month-old infant followed for an anorectal malformation.

4. Discussion

This is a retrospective study; like all studies of this kind, we have been confronted with limits in the collection of certain data and the appreciation of certain parameters. However, the results obtained during this study may allow a review of the literature on ARM.

Diagnostic aspects

All cases of ARM were discovered in the neonatal period in our series, the average age being 1.7 days (0 and 4 days), while Wandaogo *et al.* [8] as well as Kim *et al.* [9] reported a discovery rate in the neonatal period of 80% and 79.2% respectively.

The most frequent reason for consultation was the absence of an anal opening in 62.9% of cases. In the series by Bandre *et al.* [10] it also constituted the first reason for consultation but at a higher rate (93.3%).

Two distinct clinical pictures were observed:

- more frequent forms without fistula represent 57.1% of cases. This rate is close to that reported by Chabal *et al.* [11] in Senegal (56.7%), Bandre *et al.* [10] in Burkina Faso (57.8%) and Nazer *et al.* [12] in Spain (61%).
- forms with fistula: in our series, they represent 42.9% of cases. While for Niedzielski *et al.* [13], Endo *et al.* [14], and Peňa *et al.* [15], ARM with fistulas are more frequent with 53.3%, 90% and 95% respectively.

Rectovaginal fistulas are the most frequent with 33.3% compared to 26.7% of rectovestibular fistulas in our series. Rosen *et al.* [16] in the United States highlighted a much lower frequency of rectovaginal fistulas (1% against 29% for rectovestibulars) in a series of 617 girls.

Intestinal obstruction was found in 22 patients (62.9%). This rate is close to that reported by Luhiriri *et al.* [17] with 66.7%. It consists of an absence of meconium emission, progressive bloating of the abdomen within a few hours, and vomiting which is late and reflects the severity of the occlusion [18]. It was objectified in all patients with ARM without fistula and in two patients (33.3%) with ARM with narrow or non-functional fistula.

Associated malformations

In our series, ARM are associated with other malformations in 17.1%. This rate is far lower than that found in the literature. Indeed, Nazer [12], Endo [14] and Hager [19] respectively reported 64%, 45.2% and 59% of ARM associated with other malformations. In the literature according to which the spinal and cardiac malformations come in second and third position after the urogenital malformations [3] [18] [20]. Cho *et al.* [21] found 49% respectively; 43% and 27% in their series; Nazer *et al.* [12] noted 42.5%; 26% and 18.5%. Heart defects are clinically expressive and are more easily detected from this point of view. In our patients, this is inter-ventricular communication, also found by Bandre *et al.* [10].

- Imaging

The invertogram performed on twenty patients (57.1%). This essential examination for determining the level of the rectal cul-de-sac has been abandoned by certain authors [22] [23] who consider that its interpretation is tainted by several sources of error:

- the coccyx may be absent in the event of an anomaly sacral (consequently, the position of the pubo-coccygeal line will be distorted),
- in the event of meconium impacted in the rectal cul-de-sac, the air will not reach the end of this cul-de-sac and by consequently the reference mark of the latter's position will be false;
- the child's crying and cries will be responsible for movements of the pelvic diaphragm which modify the anatomical landmarks.

According to a recent study in 2005, the sensitivity of this examination in assessing the distance from the rectal cul-de-sac is 27%. This is significantly lower than that of perineal ultrasound (86%) and opacification by a colostomy (100%). In our series, the colostogram is performed in three patients (8.6%). This rate of completion of the colostogram is lower than that reported by Ayyadi [24] with 55.2% of the cases. It was also systematic for all those who underwent a colostomy in the Peňa series [15].

Therapeutic aspects

We performed the colostomy in 57.1% of the patients. It interested patients who had a high or intermediate form of MAR as recommended by some authors [25] [26]. This rate is comparable to that of Patwardhan *et al.* [27] in England with 61.25% and de Bandre *et al.* [10] in Burkina Fasso with 68.9%; while Chabal *et al.* [11] performed a plastic surgery straight away, because of the difficulties posed by colostomy. The double terminal sigmoid colostomy was the most performed. It is the most recommended colostomy currently in order to avoid the occurrence of a mega rectum; on the one hand, it allows the evacuation of stool through the stoma mouth upstream; and on the other hand, the opacification of the rectal cul-de-sac and the enemas by the stoma mouth downstream [28].

The average age at the time of final treatment is 12.7 months (range 2 days and 14 years).

The most used technique (88.6%) is the posterior sagittal anorectoplasty ac-

cording to the Peña and De Vries technique [6]. Alumeti *et al.* [29] reported 66.7%, in Dakar with 68.5% in older children.

In our series, complications during hospitalizations occurred in 37.1% of patients marked by loose sutures and suppurations.

We observed two cases (5.7%) of death by septic shock. This rate is lower than those reported by other authors [10] [11] [17].

Long-term evolution

We reviewed 27 patients (77.1%) with an average follow-up of 2.7 years (1 and 5 year extremes). The anus aspect is normal in most cases (74.1%). On anal continence, our results were good in 6 out of 10 patients evaluated. For Tong *et al.* [30], Alumeti *et al.* [29], and Bandre *et al.* [10], results were good in respectively 23%, 87.6% and 62.5% cases.

5. Conclusion

Anorectal malformations are diagnosed and treated early in the neonatal period or in infant in our country. Pena and Vries recto-anoplasty improved their functional prognosis.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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