

ORIGINAL ARTICLES

Anorectal malformations treatment results: Multicentric research on 32 cases in Abidjan

Aké Yapi Landry*¹, Sophie Kouassi¹, Kokoe Midekor¹, Rebecca Bonny¹, Ello Moh¹, Eric Yebouet², Sanny Bankole², Cosme Aguehoude¹

¹Paediatric Surgery Department, UTHC of Cocody, Abidjan R.C.I., Côte d'Ivoire

²Paediatric Surgery Department, UTHC of Treichville, Abidjan R.C.I., Côte d'Ivoire

Received: December 28, 2016

Accepted: February 2, 2017

Online Published: February 14, 2017

DOI: 10.5430/css.v3n2p1

URL: <http://dx.doi.org/10.5430/css.v3n2p1>

ABSTRACT

Objective: Evaluate results of treatment of anorectal malformations (ARM).

Methods: Multi-centric research on results of treatment of ARM in two university teaching hospital centres of Abidjan, from the period of January 2000 to December 2010. Sixty-three children operated for an anorectal malformation have been recorded. Thirty two of these children have been re-examined at an average age of 4 years old. The clinical evaluation was on anorectal function, physical condition of anorectal complex, urinary continence and quality of life according to the Ditehseim's score.

Results: Sixteen patients (50%) out of the 32, had normal faecal continence; 4 had severe constipation; 10 cases (31.25%) were of complete soiling and 8 cases (25%) were of faecal incontinence. The clinical examination of anorectal complex was normal for 22 patients (68.75%). For the other 10 we could notice 5 cases of anal stricture and 2 cases of mucous ectropium. The urinary incontinence was associated with 2 cases. The low form has been observed as a factor of good prognosis for those operated of ARM ($p = .04$). During the evaluation of quality of life, 4 out of 12 patients aged of more than 5 years old, had a bad score in it.

Conclusions: The improvement of the medium and long term results of treatment of ARM in our context requires the setting up of a multidisciplinary follow-up (paediatric surgeons, gastro-enterology paediatrics and paediatric psychologists) and raising the consciousness of parents to ensure an adapted nursing.

Key Words: Anorectal malformations-treatment-results

1. INTRODUCTION

The medical care for anorectal malformations (ARM) remains a challenge for the paediatric surgeon with numerous and varied operating techniques, from the proctoplasty made by Amussat in 1835 to the Posterior Sagittal Ano Recto Plasty (PSARP) of Peña and De Vries, and today the laparoscopic way proposed since the years 2000 by Georgeson et al.^[1] This medical care must be instantly perfected to improve the anorectal functional prognosis on the long term which, in some forms, mainly high, is poor.^[1] This state

of facts leads to the problem of post-operative monitoring of ARM, on the medium and long term. The evaluation of these post therapeutic results of ARM has been the subject of numerous publications with diverse and varied evaluation criteria. The variety of evaluation criteria shows lack of consensus on the subject up to date.^[2] Some results of long term monitoring have been reported by authors of great series (Rintala, Peña, Davies). These results remains marked by the prevalence of faecal incontinence and its therapeutic difficulties. In African literature, the subject is broached but not

*Correspondence: Aké Yapi Landry; Email: akeyapilandry@yahoo.fr; Address: 03BP 2172 Abidjan 03, Abidjan R.C.I., Côte d'Ivoire.

sufficiently described, particularly when it is about the future of these young patients and their quality of life. Through this bi-centric research, we have assessed these ARM treatment results in our paediatric surgery departments.

2. PATIENTS AND METHOD

This is a multi-centric research on the ARM treatment results in the University Teaching Hospital Centres of Cocody and Treichville (in Abidjan) for a period of 11 years (January 2000 to December 2010). Sixty-three children (22 in Cocody and 41 in Treichville) operated of ARM have been registered. The ARM type was ranked according to the Wingspread ranking (see Table 1). The average age during the intervention was 15.96 months (a continuum from 6 months to 72 months). The cure of ARM has taken some times, for the low forms, or preceded by a colostomy in neonatal period, for the high and intermediate forms (see Table 2).

Table 1. Type of ARM (n = 63)

Type of ARM	Total number	Percentage (%)
Low form	25	39.68
Intermediate form	26	41.28
High form	10	15.87
Cloacal form	02	3.17
Total	63	100

Note. ARM: anorectal malformations

Table 2. Distribution of the patients according to the operative technique carried out (n = 63)

Operative Techniques	Total number	Percentage (%)
Low form		
• Proctoplasty in Y or VY	13	20.63
• Peñã Anoplasty	12	19.04
Intermediate, high and cloacal Forms		
• Peñã Anorectoplasty	28	44.44
• Peñã modified Mollard	01	1.58
• Stephens	08	12.69
• Santuli	01	1.58
Total	63	100

Out of the 63 patients operated for ARM, 4 died, we lost sight of 27, and 32 responded to our appeal for evaluation with an average elapsed time of 2 and a half years (a continuum of 3 months to 9 years). It involved 16 boys and 16 girls with an average age of 4 years old, at the time of our evaluation.

The clinical evaluation was about:

- Anorectal function: normal continence, research of complications (incontinence, soiling, constipation).
- Physical condition of the anorectal complex.
- Urinary continence.

- Quality of life according to the Ditesheim's score (quantitative assessment of quality of life).^[3]

The evaluation of the continence was done with the Kelly's score (see Figure 1).

Clinical findings	score
Soilure	
often clean	2
accidental stain	1
frequent stain	0
Sphincter tonus	
strong	2
weak	1
lacking	0
Faecal control	
in all circumstances	2
some accidents	1
no control	0
Kelly's Score	<input type="text"/>

Figure 1. Kelly's score

Normal faecal continence (3 to 6 on Kelly's score); Faecal incontinence (0 to 2 on Kelly's score).

Ditesheim (quantitative assessment of quality of life):

- (1) Schooling: full time = 1; part time = 0.5; never = 0
- (2) social relationships: no limitation= 1; some restrictions (example: does not spend the night at some friend's place, does not go camping) = 0.5; very limited (example: no party, nodating, no contact) = 0
- (3) physical capacities: toilet free (can stay at 1 h distance of the toilets, ex: travelling by car) = 0.5; take part in all sport, no limits for swimming according to the age = 0.5; no limitation for work (according to the age) = 0.5

Total = 0 to 3.5.

At the end of this clinical evaluation we've noticed the following results, according to Davies et al. works criteria:^[4]

- Poor results: patients having a final stoma and incontinent patients
- Average results: constipated patients having to resort to evacuating enema
- Good results: continent patients, having a spontaneous defecation, even if there was sometimes a tendency to constipation

The research of the factors that could influence anorectal functional prognosis of our operated patients, has been done by some correlations, through the Khi 2 test, with some patient's data (type of ARM, type of associated malformation, gender, plastic surgery technique used, the operative effects, type of post-operative complication) and the occurrence of good or poor results.

3. RESULTS

3.1 Clinical Evaluation of Anorectal function

3.1.1 Normal continence

Sixteen children (50%) out of the 32 assessed had a normal faecal continence.

3.1.2 Functional complications

Constipation: Four patients out of the 32 showed signs of chronic constipation (12.5%).

Soiling (stools leakages): Stools leakages were observed in 10 patients out of the 32 (31.25%).

Faecal incontinence: Eight of the evaluated children (32) showed signs of faecal incontinence (25%).

3.2 Physical condition of the anorectal complex

3.2.1 Normal examination

The examination of the anus and of anorectal area was normal for 22 children out of the 32.

3.2.2 Complications

Local complications have been observed in our evaluated patients (see Table 3).

At urinary level, incontinence has been observed in two of the patients evaluated.

3.3 ARM operated and quality of life

Quality of life score (n = 12) (see Table 4).

3.4 Anorectal functional complications treatment

See Table 5.

3.5 Analysis of treatment results

See Table 6.

3.6 Prognosis Factors

3.6.1 Type of ARM

The low form in ARM is a factor of good prognosis ($p = .04$) (see Table 7).

3.6.2 Associated malformations

The presence or not of associated malformation has not influenced the treatment results of our operated for ARM ($p = .37$).

Gender: The gender was not a prognosis factor in our study ($p = .44$).

Plastic Surgery technique used (see Table 8): The type of plastic surgery technique has not influenced the treatment results of those operated for ARM in our research.

Operatives effects: The nature of the operative effects has not influenced the treatment results of our operated for ARM ($p = .26$).

Table 3. Post anorectoplasty local complications (n = 32)

Local complications	Types of ARM				Total
	Low	Intermediate	High	Cloacal	
Ectropium	00	01	01	00	02
Dehiscence	00	00	01	00	01
Prolapsus	00	01	00	00	01
Ulceration	00	01	00	00	01
Total	01	05	04	00	10

Note. ARM: anorectal malformations

Table 4. Type of ARM and quality of life (n = 12)

Ditesheim's score	Types of ARM				Total
	Low	Intermediate	High	Cloacal	
0-1	00	01	01	00	02
1-2	00	02	00	00	02
2-3, 5	04	02	00	02	08
Total	04	05	01	02	12

Note. ARM: anorectal malformations

Table 5. Type of ARM and therapeutic means used (n = 32)

Therapeutics means	Types of ARM				Total
	Low	Intermediate	High	Cloacal	
Enema	05	06	02	01	14
Dilatation	03	03	02	01	09
Defecation training	03	01	02	–	06
Food diet	04	02	02	01	09
Medecines (laxatives)	–	02	02	–	04
Surgery	–	01	–	–	01
Traditherapy	02	–	–	–	02
Biofeedback	–	–	–	–	–
None	03	01	06	–	10

Note. ARM: anorectal malformations

Table 6. Type of ARM and of treatment results according to Davies et al. criteria (n = 32)

Treatment results	Type of ARM				Total
	Low	Intermediate	High	Cloacal	
Good	11	03	01	01	16 (50%)
Average	06	04	01	00	08 (25%)
Poor	02	02	03	01	08 (25%)
Total	15	10	05	02	32 (100%)

Note. ARM: anorectal malformations

Table 7. Results according to type of ARM (n = 32)

Type of ARM	Result		p
	Good	Poor	
Low Form	13	02	.04
• Intermediate Form	06	04	.43
High Form	02	03	.16
• Cloacal Form	01	01	.53
Total	22	10	

Note. ARM: anorectal malformations

Table 8. Results according to the plastic surgery techniques used (n = 32)

Plastic Surgery techniques used	Results		p
	Good	Poor	
Peña Proctoplasty	09	05	.26
Peña Anorectoplasty	04	04	.18
Peña anorectoplasty + urogenital mobilization + proctoplasty	01	01	.53
Y or VY Proctoplasty	04	00	.20
Santuli Technique	01	00	.68
Total	22	10	

4. DISCUSSION

The decision to research functional future and physical condition of anorectal complex, on the medium and long term, for patients operated of ARM is to be encouraged in our context, because few pieces of writing talk about it. We have to go

beyond simple evaluation of faecal continence to appreciate the quality of life of these children after an anorectoplasty. Functional problems and local complications of anorectal complex are real and in good proportion for those operated of ARM. The anorectal function has been evaluated by the study of faecal continence. Out of the 32 patients evaluated 13 had normal faecal continence that is, having wilful control of stools and lack of soiling. This probability which involve hardly half of our re-evaluated patients is the ideal desired in the effects of anorectal malformation surgery. The percentage of continent children in our study varies according to the type of anorectal malformation: out of the 13 continent patients 10 had a low form, two had an intermediate form and one had a high form. This influence of the continence by the type of anorectal malformation is found in literature: Diseth reports 41% of the low forms and none of the high forms;^[6] Rintala reports 52% of the low forms and 35% of the high and intermediate forms.^[7,8] The anorectal function problems observed in our series have been chronic constipation (12.5%), stools leakages or soiling (31.25%) and faecal incontinence (25%). This is about real problems being the focus of medium and long term monitoring of the patients operated for ARM. It is necessary to give a good explanation to the parents of the patients on the probability of the occurrence of such complications, particularly in the high and intermediate forms. These anorectal functional problems are also observed in diverse proportions in the literature.^[7,9]

With respect to the faecal incontinence, which refers to the inability to wilfully hold back the stools in the rectum, it remains the focus of our worries for this evaluation of the treatment results of ARM, considering the plethora of publications on this subject.^[5,7,8,10-13] We've numbered 25% in our study; results that may be superimposed to those of Peña (25%) and of Ludman (28%).^[14] The evaluation of this faecal incontinence is more difficult because of the disparity in the criteria studied in the publications.^[2] We have used Kelly's evaluation score for its simplicity and its objectivity.^[15] It has enabled us to appreciate the functional results of our patients.

These results call for caution, in regard to the announcement of a prognosis, children should be given all the available care, so as to improve this, in short and long term.

The local aspect of the anorectal complex was normal with 22 patients out of the 32 evaluated. The complications observed with the other 10 were dominated by the anal stenosis (5 cases) and the mucous ectropium (2 cases). These complications are frequent and reported by some authors like Ngom,^[16] Mollard^[17] and Nixon.^[18] According to these authors, the mucous ectropium is a frequent cause for re-operating, particularly with the high forms. We have observed at the end of the evaluation that the results were good or acceptable with two third of the patients, and bad with the remaining one third. These results are encouraging since they are closer to that of a meta-analysis reported by Davies et al, while analyzing the digestive post-effects of ARM.^[4] They observed 45% good, 39% average, and 15% poor results. We are of the same opinion with the authors who mentioned in the literature the heterogeneous nature of results publicized. Some such as Liem^[19] reported on them as good and excellent results (58%), average (38%), and (4%) poor results. These results are influenced by factors, among which some are known: the type of anorectal malformation (high forms are of less good prognosis than the low forms, the presence of fistula, cloacal malformations), the associated malformations particularly the sacral agenesis with its corollary radicular deficit. Other factors have been reported by some authors: Liem underlines the influence of the depth of external sphincter complex on the anorectal function; Fiogbé in his study, compares the medical treatment given in

Abidjan, Côte d'Ivoire to the one given in Lausanne, Switzerland, he underlines the influence of the geographical origin of patients on the results.^[13] The analysis of the treatment results of the ARM also takes into account the urinary abnormalities. Thus on the functional aspect in our study, the urinary incontinence has been the more observed abnormality (5 patients over 32 evaluated). We agree with Cortes et al. who claim that the incidence of urogenital malformations associated to ARM is more important in case of high malformation than in the case of low malformation.^[20] Concerning the management of anorectal functional disorders, it's done with our patients by enema, the anal dilations, defecation training, food diet and medicines (laxative, anti-diarrheic etc...). We have no experience of the biofeedback techniques, of stimulation by sacral neuromodulation described by some authors. The purpose of the surgical treatment and of the active monitoring of these children is to enable them to acquire faecal continence compatible with an acceptable social life. Hence the importance of evaluating the life quality of these children operated for ARM in our study. We have chosen the Ditesheim^[3] score, which through the ability of schooling, social relationships and the physical capacity, has enable us to evaluate the quality of life of our patients which are of schooling age. The quality of life was better with those operated for low forms than those operated for high forms. The impact of anorectal functional disorders on patients' quality of life has been observed with one-third of children in schooling age, in our study. It is a threat for the psychosocial future of the children. Ditesheim shows the influence of functional disorders namely the faecal incontinence on the patients' quality of life and it becomes decisive from the age of 10. A psychosocial assistance of the children and even of their parents becomes necessary.

5. CONCLUSIONS

The improvement of the results in medium and long term of the ARM treatment in our context is influenced by the setting up of a multi-disciplinary follow-up (paediatric surgeon, gastro-enterology paediatric and paediatric psychologist) and raising awareness of parents to provide adapted.

CONFLICTS OF INTEREST DISCLOSURE

The authors declare they have no conflict of interest.

REFERENCES

- [1] Georgeson KE, Inge TH, Albanese CT. Laparoscopically assisted anorectal pull-through for high imperforate anus: a new technique. *J Pediatr Surg.* 2000; 35: 927-30. PMID: 10873037. <https://doi.org/10.1053/jpsu.2000.6925>
- [2] Hassett S, Snell S, Amy HT, et al. 10 years outcome of children born with anorectal malformation, treated by postero sagittal anorectoplasty, assessed according to the Krickbeck classi-

- fication. *J Pediatr Surg.* 2009; 44: 399-403. PMID: 19231543. <https://doi.org/10.1016/j.jpedsurg.2008.10.092>
- [3] Ditesheim A, Templeton J. Short-term versus long term quality of life in children following repair of high imperforate anus. *J Pediatr Surg.* 1987; 22: 581-7. [https://doi.org/10.1016/S0022-3468\(87\)80103-3](https://doi.org/10.1016/S0022-3468(87)80103-3)
- [4] Davies M, Creighton S, Wilcox D. Long-term outcomes of anorectal malformations. *Pediatr Surg Int.* 2004; 20: 567-72. PMID: 15309468. <https://doi.org/10.1007/s00383-004-1231-6>
- [5] Ranke A. Study of anal continence in a child operated of anorectal malformation: characteristics of questionnaires. Project Work for the Interuniversities Diploma of Coloproctology. 2002; 21.
- [6] Diseth TH, Emblem R. Somatic function, mental health, and psychosocial adjustment of adolescents with anorectal anomalies. *J Pediatr Surg.* 1996; 31(5): 638-43. [https://doi.org/10.1016/S0022-3468\(96\)90664-8](https://doi.org/10.1016/S0022-3468(96)90664-8)
- [7] Rintala R, Lindahl H. Is normal bowel function possible after repair of intermediate and high anorectal malformations? *J Pediatr Surg.* 1995; 30(3): 491-4. [https://doi.org/10.1016/0022-3468\(95\)90064-0](https://doi.org/10.1016/0022-3468(95)90064-0)
- [8] Rintala RJ, Lindahl HG, Rasanen M. Do children with repaired low anorectal malformation have normal bowel Function? *J Pediatr Surg.* 1997; 32(6): 823-6. [https://doi.org/10.1016/S0022-3468\(97\)90628-X](https://doi.org/10.1016/S0022-3468(97)90628-X)
- [9] Labouré S, Besson R, Lamblin MD, et al. Incontinence and constipation after low anorectal malformations in a boy. *Eur J Pediatr Surg.* 2000; 10: 23-29. PMID: 10770243. <https://doi.org/10.1055/s-2008-1072318>
- [10] Rintala R, Lindahl H, Marttinen E, et al. Constipation is a major functional complication after intestinal sphincter-saving posterior sagittal anorectoplasty for high and intermediate anorectal malformations. *J Pediatr Surg.* 1993; 28(8): 1054-8. [https://doi.org/10.1016/0022-3468\(93\)90518-P](https://doi.org/10.1016/0022-3468(93)90518-P)
- [11] Holschneider AM, Pfrommer W, Gerresheim B. Result in treatment of anorectal malformations with special regard to histology of the rectal pouch. *Eur J Pediatr Surg.* 1994; 4(5): 303-9. PMID: 7857888. <https://doi.org/10.1055/s-2008-1066122>
- [12] Holschneider AM, Ure BM, Pfrommer W, et al. Innervation patterns of the rectal pouch and fistula in anorectal malformations: a preliminary report. *J Pediatr Surg.* 1996; 31(3): 357-62. [https://doi.org/10.1016/S0022-3468\(96\)90738-1](https://doi.org/10.1016/S0022-3468(96)90738-1)
- [13] Fiogbé M. therapeutic medical care for anorectal malformations: multicentric study of 356 cases in Abidjan and Lausanne. CES project work of paediatric surgery. 2006; 86.
- [14] Ludman L, Spitz L. Coping strategies of children with faecal incontinence. *J Pediatr Surg.* 1996; 31(4): 563-7. [https://doi.org/10.1016/S0022-3468\(96\)90497-2](https://doi.org/10.1016/S0022-3468(96)90497-2)
- [15] Ong NT, Beasley SW. Long-term continence in patients with high and intermediate anorectal anomalies treated by sacroperineal (Stephens) rectoplasty. *J Pediatr Surg.* 1991; 26(1): 44-8. [https://doi.org/10.1016/0022-3468\(91\)90424-R](https://doi.org/10.1016/0022-3468(91)90424-R)
- [16] Ngom G, Fall I, Sanou A, et al. Medical care of anorectal malformations in Dakar: Relating to 84 cases. E-project work for the Académie Nationale de Chirurgie. 2002; 1(4): 47-9.
- [17] Mollard P. the high anal imperforations, treatment and results. *Pediatric surgery.* 1984; 25: 305-10.
- [18] Nixon H, Puri P. The results of treatment of anorectal anomalies: a thirteen to twenty follow up. *J Pediatric Surgery.* 1977; 12(1): 27-37. [https://doi.org/10.1016/0022-3468\(77\)90292-5](https://doi.org/10.1016/0022-3468(77)90292-5)
- [19] Liem N, Hau B. Long-term follow-up results of the treatment of high and intermediate anorectal malformations using a modified technique of posterior sagittal anorectoplasty. *Eur J Pediatric Surgery.* 2000; 11: 242-45. PMID: 11558014. <https://doi.org/10.1055/s-2001-17155>
- [20] Cortes D, Thorup J, Nielsen O, et al. Cryptorchidism in boys with imperforate anus. *J Pediatric Surgery.* 1995; 30(4): 631-5. [https://doi.org/10.1016/0022-3468\(95\)90148-5](https://doi.org/10.1016/0022-3468(95)90148-5)