

Outcome of children operated for congenital anorectal malformations: A prospective single center study

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Abstract

Introduction: Anorectal malformations (ARM) have an incidence of 1 in 5000 live births and affect male and female almost equally. Operative correction of pediatric ARM is of potential clinical interest; however, long-term outcome of patients in respect to probable complications requires precise follow up and surveillance. The aim of our study was to assess the outcomes of children undergoing surgical correction of ARMs.

Materials and Methods: In a prospective follow-up study, we wanted to assess occurrence of incontinence, constipation, soiling, abdominal distension, diarrhea, stenosis, dilated sphincter and failure to thrive (FTT) in ARM patients. In addition, management of these conditions has been discussed. Reoperations have also been reviewed. The primary outcome of the study was determination of occurrence of incontinence at follow-up visits. Secondary outcomes were occurrence of constipation, anal stenosis, soiling, abdominal distension, dilated sphincter, diarrhea and FTT at follow-up visits. In addition, the decision of research team on patients at follow-up visits was considered as secondary outcomes.

Results: Two hundred and ninety patients were studied. Of the study patients, 174 (60.4%) were boys and 114 (39.6%) were girls. Mean age of boys was 4.8 ± 2.0 years and mean age of girls was 5.0 ± 2.0 years. The mean follow-up period of our patients was 39.5 ± 29.1 months. During the study follow-up period, 63 patients (21.7%) had complications. The most common complication was constipation. It was present in 21 patients (33.3%). Soiling, incontinence, dilated sphincter, FTT, stenosis, abdominal

Keywords

- Anorectal malformation
- Congenital anomaly
- Incontinence
- Constipation

distension and diarrhea were present in 21 (33.3%), 11 (17.5%), 9 (14.3%), 6 (9.5%), 6 (9.5%), 5 (7.9%), 3 (4.8%) and 2 (3.3%) patients respectively.

Conclusion: We found that the most common complications following ARM surgery are constipation, soiling, incontinence, dilated sphincter, FTT, stenosis, distension and diarrhea. The overall complication rate was 21.7%. 7.2 % and 3.1% of study population experienced constipation and incontinence respectively. In 3.1% of the study population reoperation was required. We revealed that outcome of surgical correction of ARMs is considerably good and complication rates are acceptable. Continence rate was also acceptable.

Introduction

Anorectal malformations (ARM) have an incidence of 1 in 5000 live births and affect male and female almost equally¹. Imperforated anus embodies a wide range of anorectal malformations which require surgical correction based on anatomical locations². Operative correction of pediatric ARM is of potential clinical interest and would improve function and quality of life of children suffering from it³⁻⁵. However, long-term outcome of patients in respect to probable complications requires precise follow up and surveillance.

Children who underwent surgical correction of ARM may suffer from incontinence, constipation, soiling and inappropriate feeding in short-term and long-term periods after their surgery^{3,6,7}. In addition, anomalies of the urinary tract in patients with ARM could create potential problems comparable to fecal incontinence in the long-term follow-up⁸. Constipation and incontinence have been indicated as the main late complications of ARM surgery^{3,9,10}.

Overall, it is believed that results of surgical correction of ARMs are good and the rate of acceptable continence is high. Poor results are

related to associated anomalies rather than surgical techniques and background characteristics¹¹. However, the severity of anomaly could affect surgical outcomes and future life style¹². Management of complications which arise during the short-term and long-term follow-up is of potential concern and may necessitate precise investigation and consultations. Educational consults, pharmacologic interventions and reoperations have been proposed to manage incontinence in children operated for ARM¹³.

The aim of our study was to assess the outcomes of children undergoing surgical correction of ARMs. In a prospective follow-up study, we wanted to assess occurrences of incontinence, constipation, soiling, abdominal distension, diarrhea, stenosis, dilated sphincter and failure to thrive (FTT) in ARM patients. In addition, management of these conditions has been discussed. Reoperations have also been reviewed. Long-term follow up of patients in this study is of potential clinical interest and surgical importance.

Materials and Methods

In a prospective single-center study, we assessed children older than one year who were candidates

of anorectal malformation surgeries. All studied patients were admitted to and operated at Tabriz Children's Hospital, Tabriz University of Medical Sciences, Tabriz, Eastern Azerbaijan, Iran from March 21, 2011 to February 28, 2015. Tabriz Children's hospital is the main pediatric hospital in East Azerbaijan province and it is the referral pediatric surgery center of northwest of Iran. The study sample consisted of 290 children.

The patients were followed after their discharge. The conductors of the present study called patients' parents every six months for three years. They were asked for any new or persistent symptoms and were called to the pediatric surgery outpatient clinic so as to conduct physical examination and probable further investigations. Further follow-ups were also conducted 42, 48 and 54 months after discharge.

A comprehensive history was taken from the patients and their parents at follow-up visits. Physical examination of anus and anal sphincter was conducted at pediatric surgery clinic of Tabriz Children's hospital. The patients and their parents were asked about any symptoms of incontinency, obstructive complaints, soiling, constipation, abdominal distension and diarrhea. The patients were also investigated for signs of stenosis in anal sphincter, dilation of the sphincter and presence of failure to thrive (FTT). The patients and their parents were assured if the results of follow-up visits had not clarified any significant complication. In patients with remaining problems or complications, consultation programs, drug administration, examination under anesthesia or surgery were planned. **Figure 1** illustrates the follow-up diagram of the study patients.

Inclusion criteria of the study were age older than one year, diagnosed anorectal malformation requiring surgery, first operation of patient due to his or her congenital anorectal malformation and parents' informed consent to enter the study. Exclusion criteria were age younger than 12 months, history of previous anorectal surgeries, congenital spinal malformations affecting anal tone and continence and patients in whom follow-up was impossible for any reason.

Background variables consisted of age, sex, history of preterm labor and follow-up period. The primary outcome of our study was to determine the occurrence of incontinence at follow-up visits. Secondary outcomes were occurrence of constipation, anal stenosis, soiling, abdominal distension, dilated sphincter, diarrhea and FTT at follow-up visits. In addition, the medical diagnosis and management of the research team for patients at follow-up visits was considered as secondary outcomes. Patients were divided into five groups according to their management requirements at follow-up visits: Reassurance, Consultation, Consultation & Medication, Examination under Anesthesia and Reoperation. Consultation included education to patients and their parents regarding defecation habits, nutritional patterns and sleep programs.

We analyzed our data using SPSS version 19.0. Demographic data are shown by mean \pm standard deviation (SD) and frequency (%). Associations of quantitative and qualitative variables were compared by T-test and chi-square test respectively. A p value 0.05 was considered statistically significant. Assessment of association of complications with primary operation was conducted by Kaplan-Meier

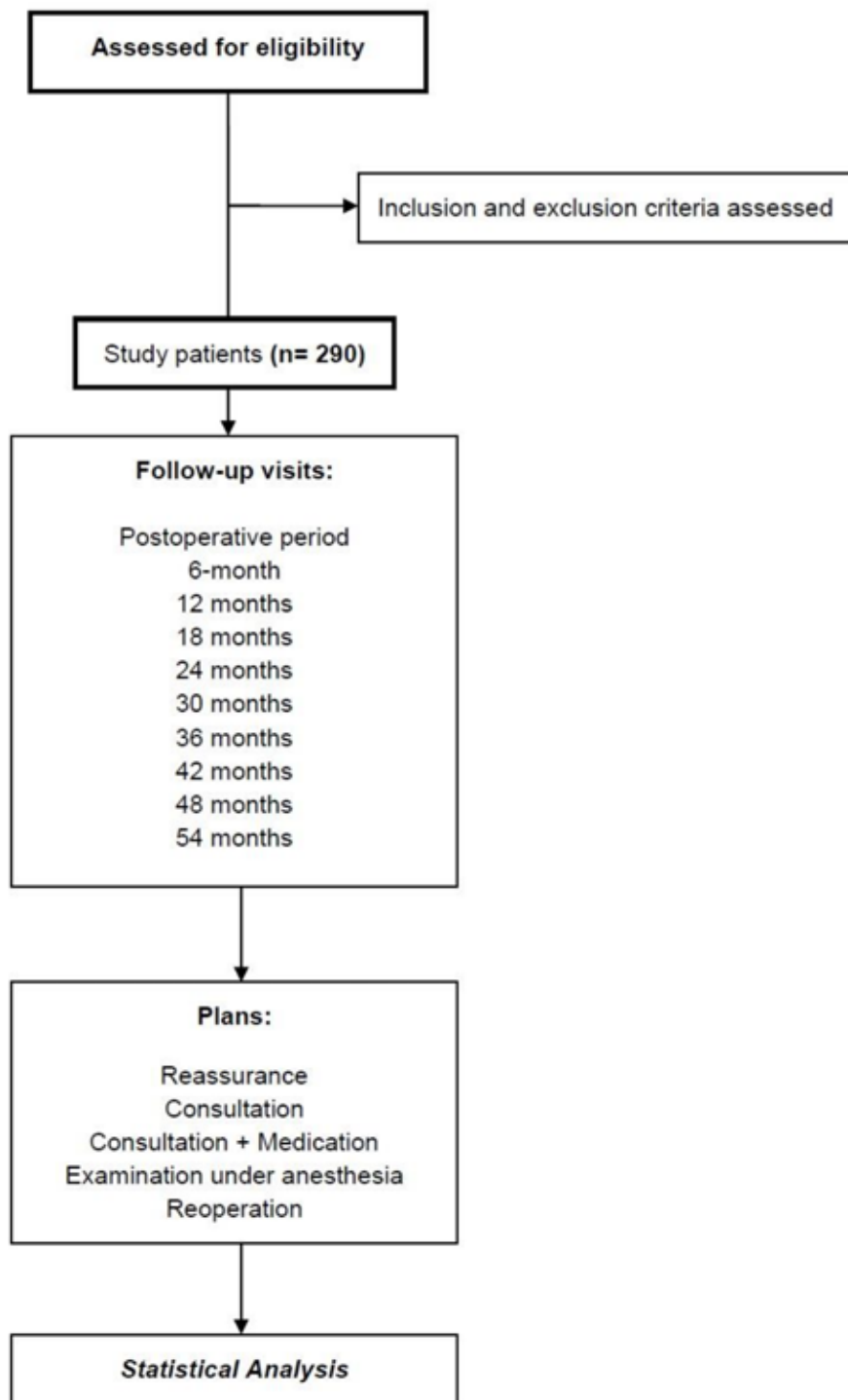


Figure 1: Follow diagram of study patients at Tabriz Children’s hospital, Tabriz, Iran.

survival analysis and pattern of observation of each complication was investigated.

Our study protocol was approved by research deputy of Faculty of Medicine, Tabriz University of Medical Sciences, Tabriz, Iran and also it was re-approved by the ethics committee of vice chancellor of research office of Tabriz University of Medical Sciences. Informed consent was obtained from parents of our patients. It was explained that patients were allowed to leave the study in any stage of the research and their participation does not deprive them from routine medical and surgical care. All data were used, published and analyzed without patients' names.

Results

Two hundred and ninety patients were studied. Of the study patients, 174 (60.4%) were boys and 114

(39.6%) were girls. Mean age of boys was 4.8 ± 2.0 years and mean age of girls was 5.0 ± 2.0 years. The mean follow-up period of patients was 39.5 ± 29.1 months. During the study follow-up period, 63 patients (21.7%) had complications. Of the entire patients with complications, 39 cases (61.9%) were boys and 24 cases (38.1%) were girls. Of these patients with complications, 10 patients (16.4%) had a history of preterm labor.

During the study follow-up period, incontinence, constipation, anal stenosis, soiling, abdominal distension, dilated sphincter, diarrhea and FTT were assessed. The most common complication was constipation. It was present in 21 patients (33.3%). **Table 1** demonstrates the frequency and percentage of complications in all study patients and also in boys and girls. P values are presented to compare the frequencies in boys and girls.

Table 1- Frequency of complications in study patients during follow-up period. The analyses were done by chi-square test.

Complication	Total	Boys	Girls	p value
Constipation	21 (33.3%)	12 (57.1%)	9 (42.9%)	0.547
Soiling	11 (17.5%)	9 (81.8%)	2 (18.2%)	0.162
Incontinence	9 (14.3%)	7 (77.8%)	2 (22.2%)	0.299
Dilated sphincter	6 (9.5%)	1 (16.7%)	5 (83.3%)	0.395
FTT	6 (9.5%)	1 (16.7%)	5 (83.3%)	0.594
Stenosis	5 (7.9%)	4 (80.0%)	1 (20.0%)	0.641
Distension	3 (4.8%)	3 (100.0%)	0 (0.0%)	0.284
Diarrhea	2 (3.3%)	2 (100.0%)	0 (0.0%)	0.522

FTT, Failure to Thrive

Kaplan-Meier survival analysis was conducted to investigate event free survival in respect to studied complications at follow-up intervals at 6, 12, 18, 24, 30, 36, 42 and 48 months. **Table 2** demonstrates the event free percentages. In addition, **Figure 2** illustrates complication free ratios for overall

complications and in each complication. Cox regression analysis was conducted to assess the effects of confounding variables on complication free events. Gender, order of birth and history of preterm labor were considered. The association was not significant ($p = 0.082$).

Table 2- Kaplan-Meier Analysis demonstrating occurrence of complications at follow-up intervals. Percentages show patients without presence of complication.

Complication	Early PO	6 months	12 months	18 months	24 months	30 months	36 months	42 months	48 months	54 months
Incontinency	100%	100%	99%	99%	98%	97%	96%	94%	94%	94%
Constipation	99%	97%	97%	94%	93%	93%	90%	89%	86%	69%
Stenosis	100%	100%	100%	98%	98%	98%	98%	98%	98%	88%
Soiling	98%	98%	98%	98%	97%	96%	95%	93%	90%	90%
Distension	100%	100%	100%	100%	100%	100%	98%	98%	98%	98%
Dilated Sphincter	100%	100%	100%	100%	100%	99%	97%	96%	96%	96%
Diarrhea	100%	99%	99%	99%	99%	99%	99%	99%	99%	99%
FTT	100%	99%	99%	99%	99%	99%	99%	97%	94%	84%

PO, Postoperative – FTT, Failure to Thrive

A total of 63 patients out of 290 patients had complications (21.7%). We did consultation with patients and their parents in 47 patients (74.6%). In three patients (4.8%) consultation was conducted and medications were used. In four patients (6.3%) examination under anesthesia was required. In nine patients (14.3%) reoperation was required. **Table 3** demonstrates types of necessary surgical operations at follow-up visits. Postoperative consultations were also conducted for them.

Discussion

We found that the most common complications following ARM surgery are constipation, soiling,

incontinence, dilated sphincter, FTT, stenosis, distension and diarrhea. The overall complication rate was 21.7%. In 7.2 % and 3.1% of the study population we found constipation and incontinency respectively. In 3.1% of our cases reoperation was required. We revealed that outcome of surgical correction of ARMs is considerably good and complication rates are acceptable. Continence rate was also acceptable.

We did our survey in a single center at Tabriz Children's hospital, Tabriz, Iran. We studied 290 patients in a prospective setting. A number of other studies have studied long-term outcome of patients undergoing surgical repair of ARM. Huang et al.³

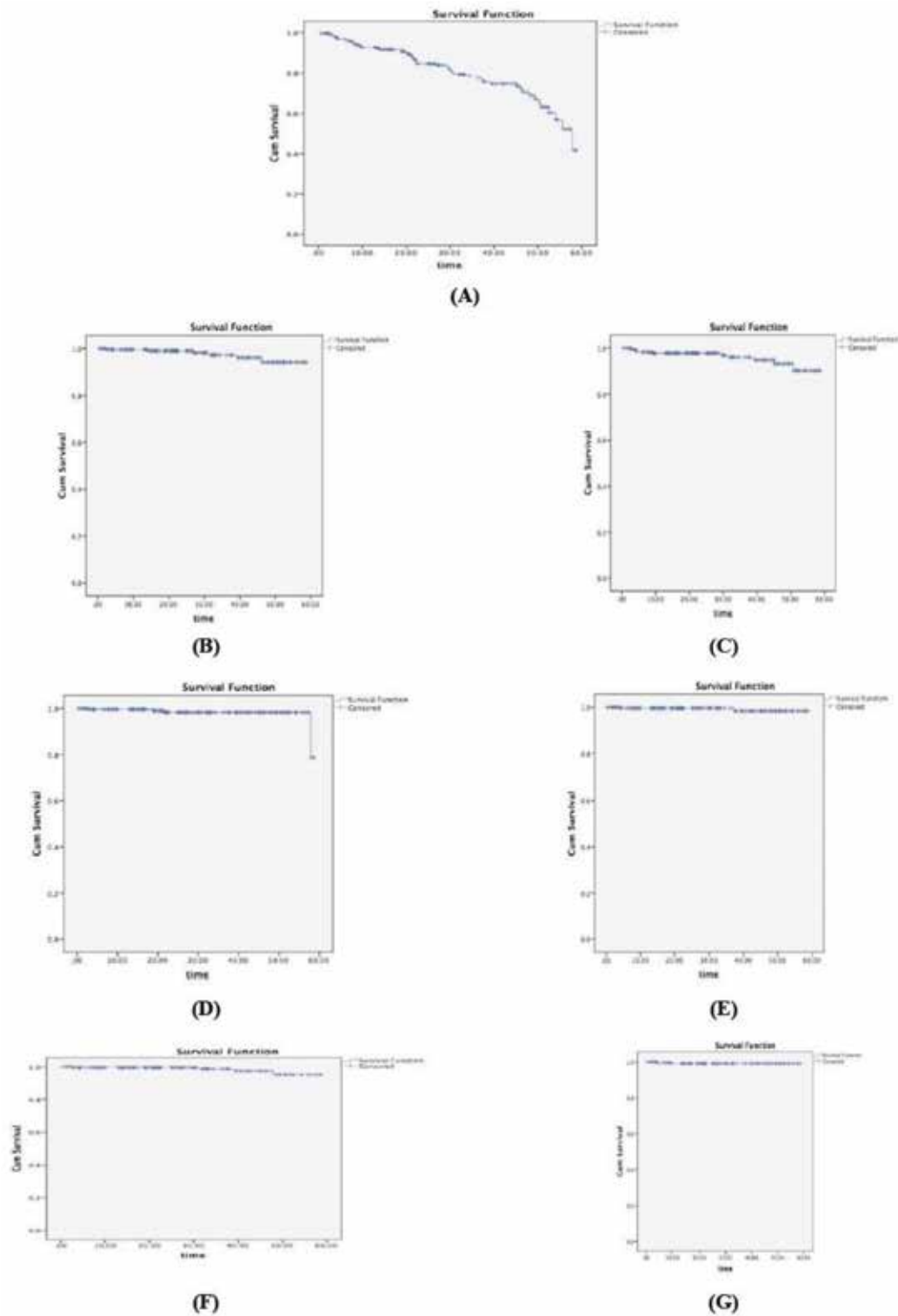


Figure 2: Kaplan-Meier event-free survival analysis for overall complications and separately for each complication; (A), Overall complications; (B), Incontinency; (C) Stenosis; (D), Soiling; (E), Distension; (F), Dilated Sphincter; (G), Diarrhea

Table 3-Types of reoperations in study patients. Nine patients (6.3% of patients with complications and 3.1% of the entire study patients) required reoperation.

No. of Patient	Type of operation
No. 1	Sphincteroplasty
No. 2	Sphincteroplasty + Colostomy
No. 3	Sphincteroplasty + Anoplasty
No. 4	Anoplasty
No. 5	Mucosectomy of prolapsed mucosa
No. 6	Anal mucusectomy
No. 7	Excision of prolapsed mucosa + Proctopexy + Anoplasty
No. 8	Repair of prolapse
No. 9	Mucosectomy of prolapsed mucosa + Repair of prolapse + Proctopexy

No., Number

studied 188 children who underwent operation for repair of imperforated anus. They followed their patients for 4.3 years. The most common complication was found to be constipation. We also found that the most complication following ARM surgery is constipation.

High continence rate have been reported in previous studies on children undergoing ARM surgery^{8, 14-16}. We found also a high continence rate. Quality of life after surgical correction of ARM is reported to be acceptable and satisfactory^{2, 16}. However, insufficient treatment of constipation and inappropriate management of incontinence could potentially compromise life style of children with ARM¹⁷. Thus, it is crucial to find conservative treatments of constipation and fecal incontinence for standardize ARM follow-up managements. In addition, an important part to improve quality of care is to develop strategies to strengthen parents' contribution to support patient care¹⁸.

According to Rintala & Lindahl⁵ 68% of patients undergoing anal repair due to ARM experience

constipation. However, they revealed that constipation resolves during puberty and in the meantime, continence improves significantly. We found that constipation is a major complication in ARM surgery. Nonetheless, the association between decline of constipation severity and improvement of continence was not found in our study. Besides, surgery can always be used for improving incontinence in selected patients⁹.

In a study by Nam et al.¹⁰ in 311 children who underwent surgery for ARM, the overall rate for constipation and soiling was 30.7% and 6.5% respectively and in 82.2% of children the outcome of continence was very good. Julià et al.¹¹ also studied ARM surgery results in 107 children. In their study, the most frequent complication was prolapsed rectal mucosa and they reported good continence in 62 cases. We also found a high continence rate. This is in contrast to our study in which the most common complication was constipation. Although the rate of continence following ARM surgery is high in literature review, a long-term clinical follow-up of

affected children is warranted ¹⁹.

Fecal incontinence potentially impairs the quality of life in children and adolescents just like adults and even more ²⁰. Thus, precise attention to the signs of recurrence of symptoms after ARM surgery is crucial. We carried out a close follow-up of patients and visits were planned to be conducted every six months. In cases of any suspicion or uncertainty; examination under anesthesia was planned to further assessment of patient's condition. According to Kaselas et al. ²¹, functional outcomes of children with anorectal malformations in the long-term, is significantly lower than normal controls. Filho et al. also reported a low quality of life particularly in children with ARM who experienced fecal incontinence ²². Thus in order to maintain a socially accepted day life for them; regular follow-ups are required ²³.

Conclusion

In conclusion, many complications could occur after surgical treatment of ARM in children specially constipation and incontinence. Although continence rate after ARM surgery is high and

acceptable, follow-up visits are essential to screen the patients with incontinence. Reoperation may be necessary in a number of them. To our knowledge, they are few studies of long-term outcomes of children undergoing surgical correction of ARM in Iran, therefore the results are of potential interest.

Conflicts of interest

There are no conflicts of interest.

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