

## ANORECTAL INCONTINENCE FOLLOWING REPAIR OF CONGENITAL ANORECTAL MALFORMATIONS

By

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**Background/Purpose:** Anorectal incontinence (ARI) is a serious problem in patients operated for congenital anorectal malformations (ARM). The aim of this study was to evaluate the various diagnostic tools and therapeutic options for patients with ARI after primary repair of ARM.

**Material & Methods:** Twenty-two patients (17 males & 5 females) were treated for various degrees of ARI after repair of congenital ARM. The initial surgical procedures used for repair of the ARM were: abdominoperineal pull through (n= 12), posterior sagittal anorectoplasty (PSARP) (n=8) and perineal approach (n=2). Templeton & Ditesheim fecal continence score was used to quantify the degree of ARI in both pre and post treatment periods. Detailed clinical assessment, radiological investigation and motility studies were used for selection of the treatment modality.

**Results:** The ages of the 22 patients ranged from 4 to 19 years. The clinical and imaging studies revealed a sphincteric defect (N=8), anteriorly displaced anus (n=2), laterally displaced anus (n=2), rectal mucosal prolapse (n=3), contracted perineal scar (n=1), anal stenosis (n=1) and no apparent clinical abnormalities (n=5). Conservative treatment (enema program, biofeedback therapy, and self-perineal exercises) was applied initially for all patients. Satisfactory results were noted in 10 patients (45.5 %). Twelve patients (54.5%) required secondary surgical procedures for treatment of ARI after failure of the conservative treatment. These secondary surgical procedures included: relocation of anorectum through posterior sagittal approach (n=4), PSARP & sphincteroplasty (n=3), excision of prolapsed mucosa (n=3), and anooplasty (n=2). Two patients achieved complete continence following surgery, 4 had a significant improvement, 2 showed mild to moderate improvement and 4 had no improvement. The follow-up periods ranged from 6 to 30 months.

**Conclusions:** 1. Conservative treatment should be tried initially in the majority of patients of ARI following repair of congenital ARM; 2. When properly selected, surgical treatment proved to be a viable option in the management of ARI following repair of congenital ARM particularly in patients with mislocated anorectum outside the muscle complex; 3. A less favorable postoperative functional result is expected in cases with significant damage and scarring of the levator and muscle complex

*Key Words:* Imperforate anus, Anorectal Incontinence, Posterior Sagittal Anorectoplasty

### INTRODUCTION

Fecal incontinence, in variable degrees, continued to be an unpleasant and frequent postoperative sequel after the surgical repair of many patients with anorectal malformation (ARM) (1,2). At least 25 to 30% of patients with high imperforate anus will suffer from fecal incontinence (3). Another 30% will suffer from other functional defecation disorders such as constipation, occasional soiling and fecal

incontinence during periods of diarrhea (4). Fecal incontinence may improve with age and by toilet training in some patients. However, significant number of patients continues to be incontinent to varying degrees throughout childhood, adolescence and adulthood (5,6).

Imaging studies plays a key role in defining the site of defect. Endoanal ultrasonography, and computed tomography (CT) have been the studies of choice (7).

Recently, magnetic resonance imaging (MRI) has proven to be of considerable value in evaluating those patients (8,9).

Numerous secondary operative procedures and medical treatments have been described to establish fecal continence (1-5), all have achieved varying degrees of success. This study was undertaken to define management strategies and outcome of children with ARI following repair of ARM in our institution

## PATIENTS AND METHODS

Twenty-two patients (17 males & 5 females) with ARI after repair of imperforate anus were treated at the Department of Pediatric Surgery, Tanta University Hospital, and affiliated hospitals from May 1997 to March 2003. Each patient was evaluated as regard to age, sex, details of primary operative procedure, and presenting symptoms and signs. Spinal and sacral radiographs, contrast enema, endorectal ultrasound, EMG, anorectal manometry and CT and/or MRI were performed. Templeton and Ditesheim fecal continence score (6) was used to quantify the degree of ARI in both pre and post treatment periods (Table 1).

The anatomical defects were elucidated after detailed clinical examination, endoanal ultrasound and CT or MRI. Eight patients had variable degrees of sphincteric defects along the anal circumference, two had anteriorly misplaced anus, two had laterally displaced anus, three had mucosal rectal prolapse, one had contracted perineal scar, and one had anal stenosis. No significant anatomical abnormalities were found in 5 patients (Table 2). The radiological investigation and motility studies were used for selection of the appropriate treatment modality.

Conservative treatment (enema program, biofeedback therapy, and self perineal exercises) was applied initially for all patients. Ten patients were managed successfully while twelve patients required surgical intervention after failure of conservative therapy.

The secondary surgical procedures were planned according the clinical findings and imaging studies. Four patients with either anteriorly displaced anus (n=2) or laterally displaced anus (n=2) were treated by PSARP, which involved relocation of the rectum and anus to proper position within the muscle complex. Three of the 8 patients, who had documented sphincteric defect, were treated by sphincteroplasty through PSARP, while the remaining five patients responded to conservative treatment. All the three patients with mucosal rectal prolapse undergone excision of prolapsed mucosa. Anoplasty was performed for each of the 2 patients with contracted perineal scar and anal stenosis (Table 3).

**Table (1):. Fecal Continence Score (Templeton and Ditesheim)**

Parameter	Score
<b>1. Toilet training for stool</b>	
(A) Successful	1.0
(B) Occasionally successful (awareness of impending stool)	0.5
(C) No awareness of impending stool	0
<b>2. Accidents</b>	
(A) None, or rare	1.0
(B) 3 per week or less	0.5
(C) More than 3 per week	0
<b>3. Extra underpants (or liners) needed</b>	
(A) Never	1.0
(B) Only when having diarrhea	0.5
(C) Always	0.0
<b>4. Social problems</b>	
(A) None	1.0
(B) Infrequent order; does not miss school, but no overnights, dates, camping	0.5
(C) Frequent order affects school and play	0
<b>5. Activity restrictions</b>	
(A) None	0.5
(B) Avoids swimming, sports	0
<b>6. Rashes</b>	
(A) No current problems	0.5
(B) Some current problems	0
<b>Total Score (range)</b>	0-5

*Scoring system is as follows:*

- *Good, 4 to 5 points;*
- *Fair, 2 to 3.5 points;*
- *Poor, 0 to 1.5 points.*

*Operative technique:*

The bowel was prepared by repeated colonic washout the day before surgery. Only one patient underwent a diverting colostomy prior to the secondary surgical procedure. During surgery, the patient was placed in a jackknife position. A skin incision was made in the natal cleft, starting in the coccyx and continuing down to the posterior margin of the anus, then running around the anus. Multiple 5-0 silk sutures were placed at the mucocutaneous junction of the anus as described by Pena. (1,3) The incision was deepened through the levator muscle

and muscle complex until the posterior wall of the rectum was identified. Then, the dissection was carried out around the anus, staying as close as possible to the bowel wall without injuring it. All muscle structures were dissected away from the bowel wall and preserved using needle cautery. The rectum was raised out from the wound in case of anterior or lateral displacement. Direct inspection of the anatomic relationship between the previously pulled-through rectum and the pelvic muscles was then performed. The anatomy was explored with an electronic muscle stimulator.

The distal portion of the rectum was mislocated out of the ring of the muscle complex and close to the posterior urethra in 2 cases and lateral to the distal part of the muscle complex in another 2 patients. The rectum was repositioned within the limits of the external sphincter muscle and the anterior and posterior borders of the

muscle complex were approximated. In another 3 patients, the levator muscle and muscle complex were damaged partially or became scarred. The damaged muscle fibers were approximated with 6-0 polydioxanone absorbable monofilament (PDS) sutures. A mesenteric fat around the rectal wall was noted in two patients, who had undergone abdominosacroperineal approach initially for the repair of imperforate anus. This mesenteric fat was resected and the levator muscle was snugly repaired around the rectum. Limited posterior tapering of the rectum was required in 2 cases.

The follow-up periods ranged from 6 to 30 months (mean 17.6 months). The outcome of various diagnostic and therapeutic modalities was analyzed.

**Table (2): Clinical & Imaging findings of the 22 patients**

<i>Clinical findings</i>	<i>Number</i>
Defect of the sphincteric complex	8
Anteriorly misplaced anus	2
Laterally misplaced anus & sphincteric defect	2
Mucosal prolapse	2
Mucosal prolapse & patulous anus	1
Contracted old perineal scar	1
Anal stenosis	1
No apparent clinical or imaging sphincteric defect	5

**Table (3): Secondary surgical procedures**

<i>Secondary surgical procedure</i>	<i>Anatomical defects</i>	<i>Number</i>
PSARP & sphincteroplasty	Defect of the sphincteric complex	3
PSARP& relocation of the rectum and anus	Anteriorly misplaced anus	2
PSARP& relocation of the rectum and anus	Laterally misplaced anus & sphincteric defect	2
Excision of prolapsed mucosa	Mucosal prolapse	3
Anoplasty	Contracted old perineal scar	1
Anoplasty	Anal stenosis	1

**PSARP: posterior sagittal anorectoplasty**

**RESULTS**

The ages of the 22 patients ranged from 4 to 19 years (mean 6.5 + 4.1). Twelve patients undergone abdominoperineal pull through as an initial primary procedure for treatment of high ARM. Two patients had anterior perineal anoplasty for imperforate anus and rectovestibular fistula, and 8 had posterior sagittal anorectoplasty (PSARP) for repair of high or intermediate ARM. The pretreatment incontinence score index ranged from 0-3 according to Templeton and Ditesheim scoring system.

**Imaging studies**

Associated sacral anomalies were found in 6 patients and sacralization of the last lumbar vertebra was found in one patient. Contrast enema study revealed huge dilatation and loaded colon in 6 patients, who were presented with anorectal incontinence with constipation. A normal sized colon was noted in the remaining patients.

Variable degrees of sphincter defects were noted by endoanal ultrasound in 8 patients. (Fig. 1) These defects included: fragmentation of anal sphincter at multiple sites of its circumference (n=4), defect of anal sphincter at one

site (n= 3), and thinning of the sphincter (n=1). Endoanal ultrasound revealed no apparent structural defect in 5 patients. In 9 patients, this study was not feasible because of either stenosed or small sized anus, or due to lack of compliance of the patients.

Computed tomography was performed in 9 patients, while MRI has been done to another 8 patients. Although the initial interpretation was not accurate in early studies, but MRI proven to be of considerable value in evaluating those patients. (Figs. 2&3)

#### *EMG & Motility Studies:*

Anorectal manometry studies were performed in 13 patients. Reduction of both resting and squeeze pressure was noted in all of them. (Fig. 4) Resting pressure ranged between 16-50 mm Hg with a mean value of 26.4 mm Hg. It was markedly reduced in 10 patients. Squeeze pressure ranged from 28 to 124 mmHg (mean 54.3 mm Hg). It was markedly low in 9 patients, and moderately low in 4 patients. Anorectal sensation was impaired in all patients except one. The maximum tolerable volume ranged from 280 to 400 ml (mean 236 ml). The rectoanal inhibitory reflex was intact in all patients except one.

Sphincter mapping by either electromyography (EMG) and/or electrogalvanic stimulator were performed in 14 patients. Reduced amplitude of action potential was recorded in the external anal sphincter during both rest and voluntary contraction of the pelvic floor muscle in 8 patients indicating variable degrees of either neurogenic or myogenic lesions (Fig 5). A satisfactory contraction with no noticeable defect in the muscle contraction was detected in the remaining studied 6 patients.

#### *Conservative treatment*

Conservative treatment including dieting, enema, constipating or laxative agents was used according to the presentation. Self-perineal exercise, and biofeedback therapy were practiced whenever feasible.

Regular bowel washout has been used as a method of management of fecal incontinence. Patients with continence during daytime were instructed to introduce 250-1000 ml of warm water (38c) within 5 to 10 minutes after they have their first meal. Older children and adolescents were advised to wait until urge to defecate was felt. Patients with soiling during overnight sleep were advised to irrigate during evening. Frequency of irrigation varied from two times per day to two times per week.

Biofeedback therapy was tried in 14 patients. Two to eight sessions were used. Significant clinical improvement

was noted in 5 patients, moderate improvement in 6, and no significant improvement was note in remaining patients.

In addition to the expensive equipments needed for doing biofeedback, we used a novel cheap method for practicing biofeedback at home or when the machine of biofeedback was not available. The CVP manometry scale attached to a balloon filled with colored saline was used. The balloon was inserted into the rectum and inflated with the colored saline. The patient was instructed to squeeze while watching the elevation of the colored saline column in the CVP manometry scale. Many patients were able to do biofeedback therapy alone at home by this simple method (Fig 6).

#### *Surgical treatment*

Twelve patients (54.5%) required surgical treatment after failure of conservative management. (Table 3)..These secondary surgical procedures were planned according to the clinical findings and imaging studies.

Four patients with either anteriorly or laterally displaced anus required relocation of the rectum and anus to a proper position within the muscle complex through the posterior sagittal approach. (Figs. 7-9). Three of those four patients showed a significant improvement of their continence score from 1.5- 2.5 at preoperative period to 3.5- 4 postoperatively. The fourth patient who had lateral misplaced anus associated with a significant damage and scaring of the sphincteric complex did not show any significant improvement of his continence score (Table 4).

Three patients with documented sphincteric defect at one or more sites of the circumference were treated by sphincteroplasty through the posterior sagittal approach. The continence score has not improved in any of those three patients.

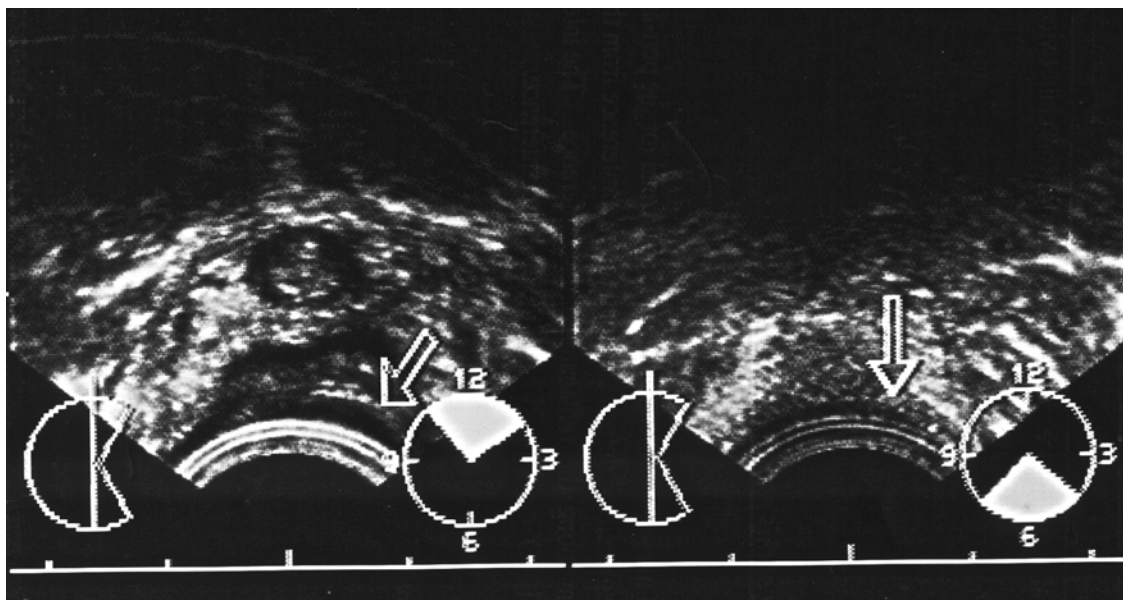
Three patients with mucosal rectal prolapse had undergone excision of the prolapsed mucosa. Two of them regained almost normal fecal continence, while the third patient, who had associated sphincteric weakness and patulous anus has mild improvement of his continence status (Fig. 10)

Anoplasty was performed for each of the 2 patients with contracted perineal scar and anal stenosis. These 2 patients had low type anomaly, and were treated initially by one stage perineal anoplasty at neonatal period. Both of them showed an excellent functional result with regaining of near normal continence after the secondary surgical procedure.

**Table( 4): Continence scoring index (Templeton and Ditesheim) at both pre and post-surgical treatment**

No	Secondary surgical procedure	Indication for surgery	Pre treatment C. S.	Post treatment C. S.
1	PSARP	Anteriorly misplaced anus	2.5	4
2	Redo PSARP	Anteriorly misplaced anus	1.5	3.5
3	PSARP	Laterally misplaced anus	1.5	4
4	PSARP& sphincteroplasty	Laterally misplaced anus & sphincteric defect	1	1.5
5	PSARP& sphincteroplasty	Sphincteric defect	1	1.5
6	PSARP& sphincteroplasty	Sphincteric defect	1.5	1.5
7	PSARP& sphincteroplasty	Sphincteric defect	1.5	1.5
8	Excision of prolapsed rectal mucosa	Prolapsed rectal mucosa	3.5	5
9	Excision of prolapsed rectal mucosa	Prolapsed rectal mucosa & patulous anus	1	2.5
10	Excision of prolapsed rectal mucosa	Prolapsed rectal mucosa	2	4
11	Anoplasty	Anal stenosis	3.5	5
12	Anoplasty	Contracted old perineal scar	2.5	4

*Continence Score (C.S.) is as follows: Good, 4 to 5; Fair, 2 to 3.5 points; Poor, 0 to 1.5 points*



**Fig. (1): Endo anal ultrasound of a 6-year-old child with anorectal incontinence after abdominoperineal pull-through for congenital ARM showing defect of the sphincteric complex in more than half of the circumference.**



Fig.(2): Sagittal T2 MRI section showing a patulous anal canal with poorly defined, hypoplastic muscle complex

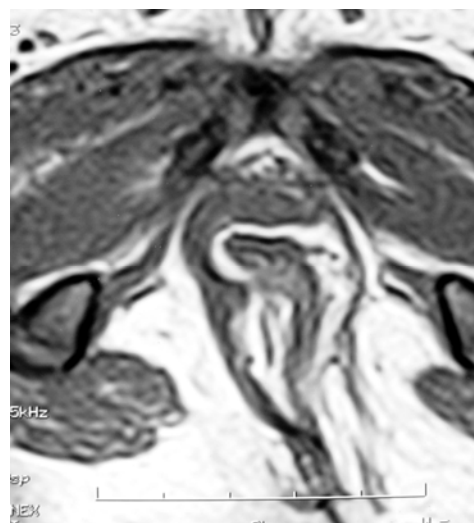


Fig.(3): Coronal T2 MRI showing a significant defect of the atrophic levator ani and muscle on the left side

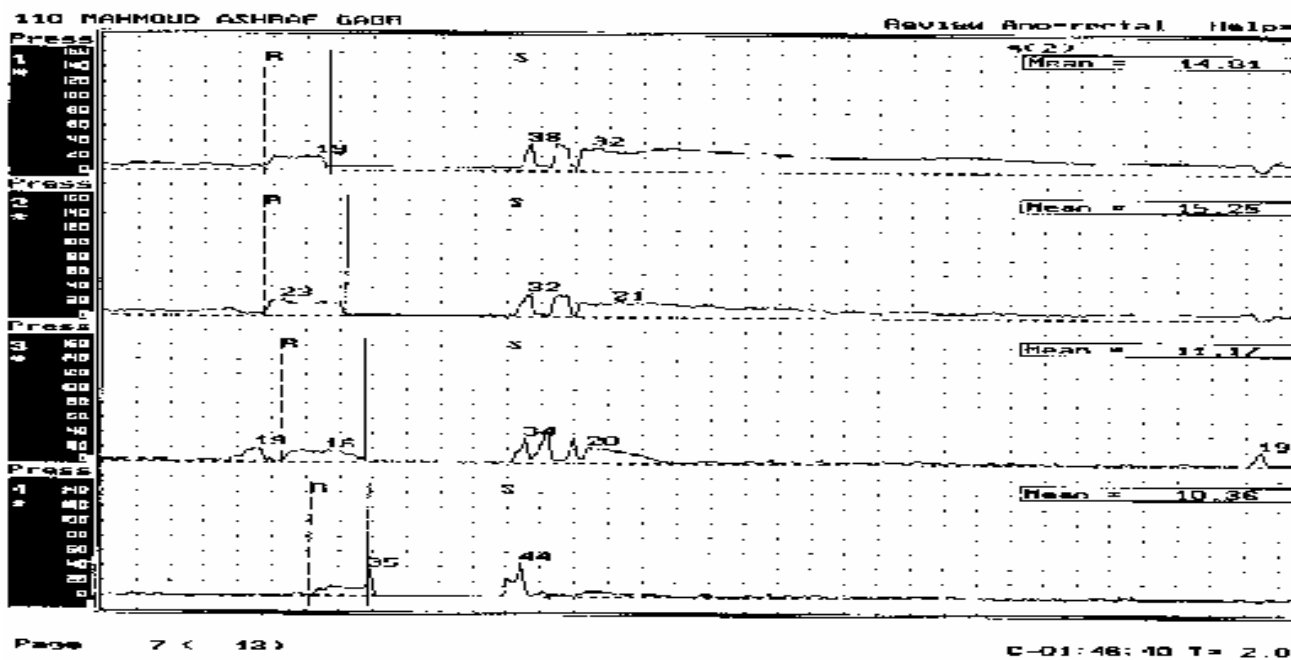


Fig. (4): Anorectal motility of a male child with anorectal incontinence after pull-through for congenital ARM showing low resting pressure

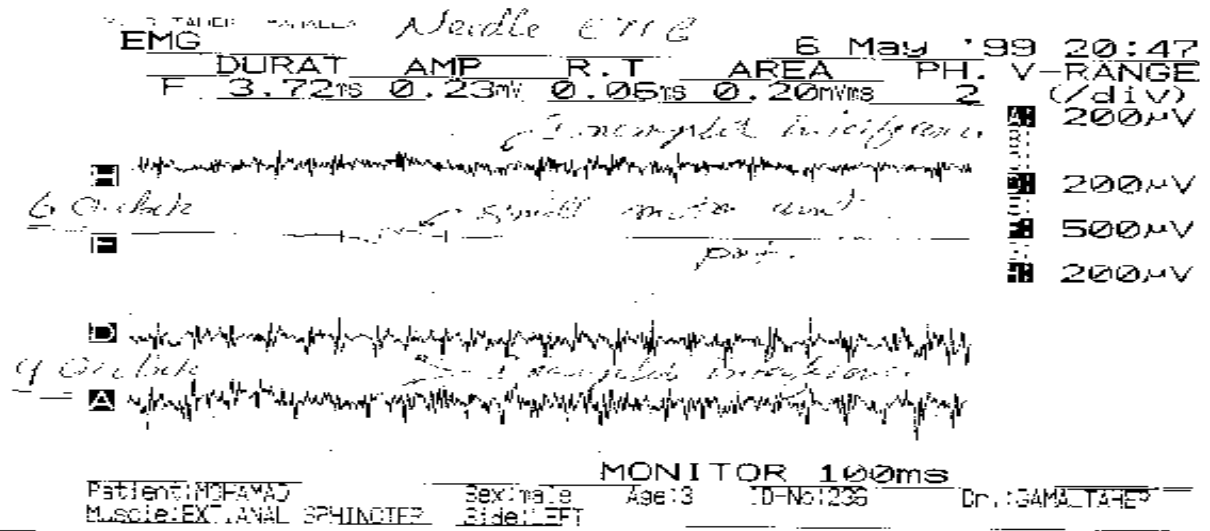


Fig. (5). EMG of a 7 year old child with anorectal incontinence after abdominoperineal pull-through showing very weak contraction of the external anal sphincter

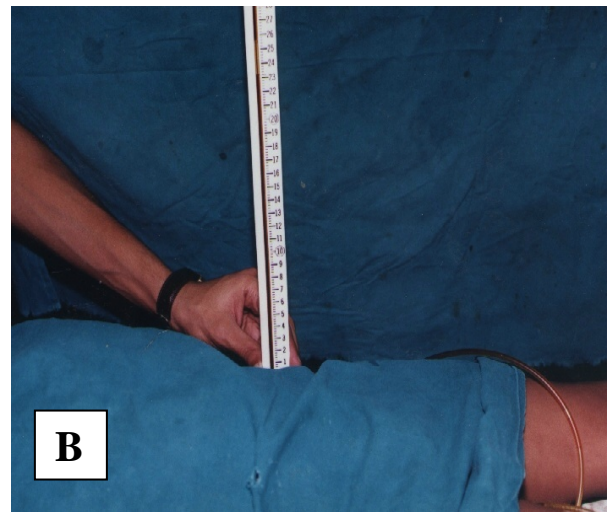
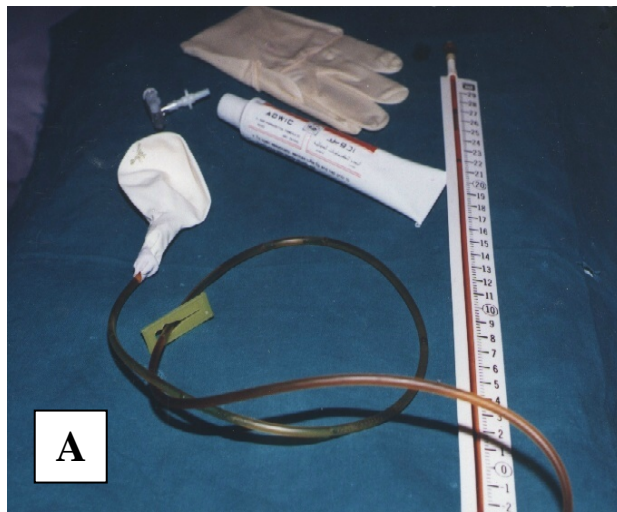


Fig. (6 A&B): Biofeedback: A novel cheap method for biofeedback. The CVP manometry scale attached to a balloon filled with colored saline. B. The balloon is inserted into the rectum and inflated with the colored saline. The patient is instructed to squeeze while watching the elevation of the colored saline column in the CVP manometry scale.



Fig. (7 A). Laterally misplaced and stenotic anus

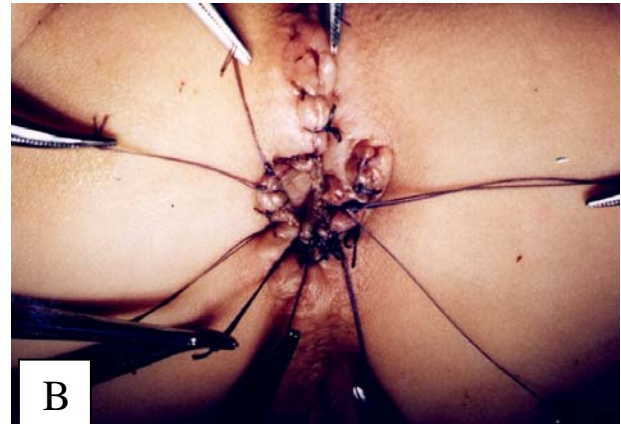


Fig. (7B) : The anorectum was relocated within the muscle complex through posterior sagittal approach



Fig. (8A): Anteriorly misplaced anus. The patient presented with constipation associated with encopresis . B, Contrast enema showed stenosis of the distal part of the neo annual canal associated with megarectum. The patient regained normal anorectal continence after redo PSARP

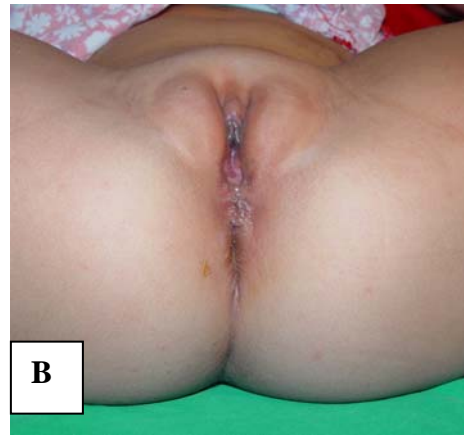
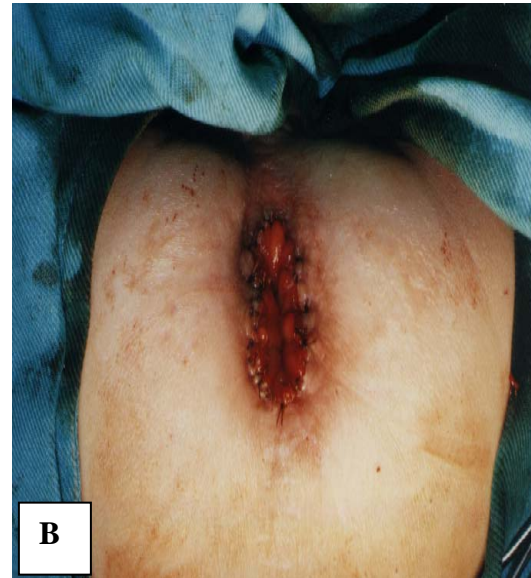
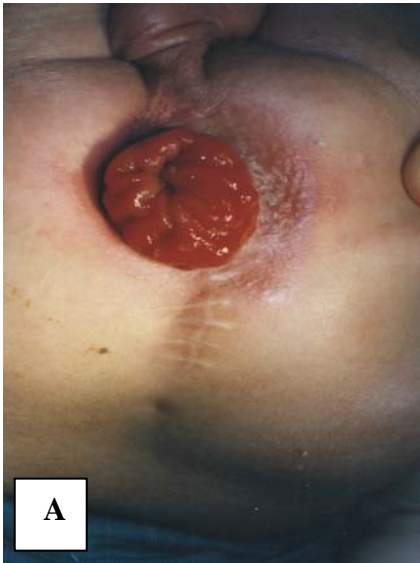


Fig. (9): 5 year old child presented with incontinence after failed primary repair of imperforate anus and rectovestibular fistula. A. The anus is almost retracted to the original site of rectovestibular fistula. B. The anorectum was relocated within muscle complex through posterior sagittal approach . The perineum looks normal , and the patient regained normal anorectal continence





**Fig. (10 A): Rectal prolapse after PSARP for high imperforate anus. B. Excision of the prolapsed mucosa**

## DISCUSSION

Fecal incontinence occurs after repair of congenital ARM is a tragedy to the patients and a challenge to the surgeon. The etiology of incontinence in these patients may be related to the congenital defects of the sphincteric muscles and its innervations, or due to technical errors during the primary repair or both <sup>(1-5)</sup>.

In the current study various diagnostic and imaging modalities proved to be of help in choosing the most suitable management plan for each individual case. In contrast, Tsugawa et al <sup>(2)</sup> reported that the preoperative radiologic evaluations did not always show the anatomy as seen in the operation and concluded that a lack of correlation between local findings of the anorectum, defecograms, CT or MRI, and operative findings does exist. The appearance of the anus and determination of the existence of muscles through digital palpation is significantly correlated to the operative findings. <sup>(2)</sup>

Electrogalvanic stimulation showed weak sphincter activity in all studied patients. Low manometry readings and impaired rectal sensation was detected also in those patients. These abnormal motility findings are quite similar to those reported by others <sup>(10)</sup>. Nagashima et al <sup>(10)</sup> reported a series of 32 patients with previous surgery for ARM, 12 of them had ARI with inadequate anal pressure, in addition to loss of the optimal rectal sensation or rectal reservoir function. According to their findings, the authors concluded that fecal continence is maintained not only by sphincter function but also by recto sigmoid functions.

Conservative treatment with enemas, laxatives, and medications are often given by most clinicians in an

indiscriminate manner and without a demonstrated benefit. <sup>(11)</sup> It is vital to determine the type of fecal incontinence from which the patients suffer and to target their treatment accordingly. Pena et al <sup>(11)</sup> advocated a well-defined systematic diagnostic approach and bowel management program for patients suffering from fecal incontinence after primary repair of ARM. A successful rate ranging from 79%-93% has been achieved by them when the bowel management and medications were administered in an organized manner <sup>(11)</sup>. Conservative treatment (enema program, biofeedback therapy, and self perineal exercises) was successful in 10 patients (45.4%) while twelve patients required surgical intervention after failure of conservative therapy.

Biofeedback therapy reinforces voluntarily sphincter function and improves conditioning rectal sensation. Iawai et al <sup>(12)</sup> reported that biofeedback therapy can be effective in children who have adequate anal resting pressure and is not effective in those patients who have congenitally absent or weak sphincter resting pressure. In the current series almost half of our patients showed significant improvement on biofeedback combined with other conservative measures. The implementation of our simple novel method for biofeedback will ensure the availability of this modality of conservative therapy to all these poor patients in the future.

Rectal prolapse is a frequent complication after pull through operation for imperforate anus. When mucosal prolapse is not associated with a sphincteric defect, a remarkable improvement is expected after surgical trimming of the prolapsed mucosa. All the three patients who undergone excision of the mucosa showed either significant improvement or gained complete continence.

Seven patients with ARI were treated by posterior sagittal approach, which was advocated by Pena <sup>(13)</sup> for primary repair of high and intermediate ARM as well as a secondary operation after failure of primary surgery for ARM <sup>(1&3)</sup>. Several other surgeons used PSARP as secondary surgical procedure for repair of various complications that developed after primary repair of the ARM <sup>(13-17)</sup>. The posterior sagittal approach provides a direct approach and expose the anatomic relation between the misplaced anorectum and pelvic muscles and allows surgeons to perform proper repositioning of the neoanorectum within the muscle complex. <sup>(1,13)</sup>

Four patients in this series had either anterior or lateral mislocated rectum. Pena <sup>(1)</sup> noted that surgeons tended to pass the rectum as close as possible to the urethra because they believed that the puborectal portion of the levator muscle has the most important role in continence. The functional results following repositioning of the neoanorectum within the muscle complex is very encouraging both in our limited series as well as in previously reported similar cases <sup>(2)</sup>. In contrast, Brain & Kiely <sup>(16)</sup> reported disappointing results of PSARP as secondary operation for management of fecal incontinence and concluded that PSARP, when used as a secondary procedure, is good for correcting anatomical defects but not for improving fecal continence.

Limited posterior tapering of the rectum was required in 2 cases, who had incontinence associated with constipation and megarectum. Tapering of the rectum is emphasized in the previous report, by Pena <sup>(1)</sup> because the size of the rectum must be such that it can fit into the levator muscle and muscle complex. In other operated cases, the lower portion of the levator muscle and muscle complex was repaired behind the rectum without tapering. Tsugawa et al <sup>(2)</sup> reported that when the rectum is pulled and stretched, it could fit into the muscular structure without tapering <sup>(2)</sup>

Only one of our patients had diverting colostomy before surgical treatment of the incontinence. Pena <sup>(3)</sup> emphasized the importance of a protective colostomy in his early report, although he performed secondary operations without a colostomy in a recent report. <sup>(18)</sup>. A meticulous colonic irrigation before surgery was performed. We believe that colostomy seems to be unnecessary when good bowel preparation is achieved particularly when tapering of the rectum is not planned <sup>(2)</sup>.

Finally, our study has proven that the secondary operations for ARI through a posterior sagittal approach may restore fecal continence in properly selected patients with definite mislocated anorectum outside the muscle complex. A good functional result is usually achieved following a limited redo anoplasty for patients with

complicated primary perineal repair of their low anomalies. A less favorable result is expected in cases with significant damage and scarring of the levator and muscle complex. The procedure can be safely performed without a diverting colostomy. In cases with mucosal rectal prolapse with otherwise good sphincter tone, simple excision of the prolapsed mucosa should be attempted before trying other more complicated procedures.

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