

## Original article

# Surgical management of anorectal malformations based on Krickenbeck classification: 13 - year experience from a single institute

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**Background:** Anorectal malformations (ARMs) represent a wide spectrum of disease with various operative managements. Posterior sagittal anorectoplasty (PSARP) has been developed as the standard technique. However, there are still controversies in the management of ARMs.

**Objectives:** The objective of this study was to assess the operative managements including associated anomalies and functional outcome of anorectal malformations at King Chulalongkorn Memorial Hospital over a 13-years.

**Methods:** A retrospective study was conducted at King Chulalongkorn Memorial Hospital between January 2004 and December 2016. Patients' information was classified based on Krickenbeck classification. Statistical analysis was performed using IBM SPSS statics 22.0 version for MAC.

**Results:** A total of 96 patients underwent repair of ARMs. The most common ARMs in male patients were rectourethral fistulas (32.0%) and in female patients were vestibular fistulas (45.0%). Seventy-five (78.0%) patients had at least one associated anomaly. Thirty-four patients (36.0%) had VACTERL association. There were 74 (77.0%) patients were managed with colostomy as staged procedures. All patients with rectourethral bulbar fistulas underwent PSARP (11/11) while patients with rectourethral prostatic fistulas or rectovesical fistulas underwent abdominoperineal pull-through (12/17), PSARP with laparotomy (4/17) and PSARP (1/17). ARMs with perineal fistulas underwent cut back operation (7/13), PSARP (3/13) and ASARP (3/13) depending on surgeons' preferences. ARMs with vestibular fistulas underwent PSARP (15/21) or ASARP (6/21). Mostly cloacal malformations underwent PSARVUP with laparotomy (6/13). ARMs with no fistula underwent PSARP (13/13). H-type ARMs underwent fistulectomy (4/5) and ASARP with protective colostomy (1/5). Fifty-three patients (61.0%) had no postoperative complications. Continence was achieved in 80.0% of the patients. The median time of follow up is 4.5 years.

**Conclusion:** In our series, the incidences of ARMs according to Krickenbeck classification are quite similar compared to other studies. Operative managements of ARMs are highly variable depending on types of ARMs. Functional outcome is also comparable to other studies.

**Keywords:** Anorectal malformations, Krickenbeck classification, posterior sagittal anorectoplasty, posterior sagittal anorectovaginourethroplasty, abdominoperineal pull-through.

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Anorectal malformations (ARMs) are congenital anomalies where the anus and rectum do not develop properly. ARMs occurs in one out of every 4,000 to

5,000 newborns and is slightly more common in males.<sup>(1)</sup> Defects vary from minor lesions to complex anomalies. Previously, anorectal malformations were classified as high, intermediate or low anomalies according to the Wingspread classification in 1984. This classification was widely accepted over the years and was based on detailed embryological, anatomic studies performed and radiographic investigations. However, some details of the Wingspread classification still remained questionable.

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In 1995, Pena A.<sup>(2)</sup> proposed a classification based on the type of the fistula presented. With this approach, a unique opportunity arose to correlate the external appearance of the perineum with the operative findings and, subsequently, with the clinical results. With the presence of different classification systems, it was difficult to compare the outcome in patients with ARMs between different centers. An International Conference for the Development of Standards for the Treatment of Anorectal Malformation<sup>(3)</sup> was organized at Krickenbeck Castle, Germany, on 2005. The participants developed a new international classification for ARMs and a new grouping for follow-up assessment and standard surgical procedures. This new international classification enables the different operative procedures to be more comparable to each other than with the other classifications. At present, the Krickenbeck classification has become the gold standard for the classification of ARMs. All pediatric surgeons had multiple lessons learned in order to improve the quality of life of children born with all different types of ARMs. The posterior sagittal approach for the treatment of ARMs was performed first in 1980, and its description was published in 1982 according to Pena A, *et al.*<sup>(4)</sup> and De Vries P, *et al.*<sup>(5)</sup> Posterior sagittal anorectoplasty (PSARP) has developed as a standard technique for numerous surgeons worldwide. However, there are still controversial topics in the management of ARMs, including various diagnoses and operative managements.

In Thailand, most of the studies categorized ARMs based on Wingspread Classification that could not compare the operative techniques and postoperative results between the different centers of pediatric surgery internationally.<sup>(6)</sup> The aim of this study was to review the operative management including associated anomalies and functional outcome over 13 years of experience at King Chulalongkorn Memorial Hospital based on Krickenbeck classification.

## Materials and methods

A retrospective study was conducted by reviewing of the medical records of patients with ARMs aged 0 -15 years whom were surgically treated at King Chulalongkorn Memorial Hospital between January 2004 and December 2016. The study has been approved by the Ethics Committees of the institute

(COA No.214/2016). Patients' data were collected including demographics, types of ARMs according to the Krickenbeck classification, associated anomalies, operative management, complications and postoperative results. Patients who had redo-operation or incomplete data were excluded. Statistical analysis was performed by using IBM SPSS statistics 22.0 version for MAC.

## Results

### Demographic data

From January 2004 to December 2016, 96 patients with anorectal malformations were recruited. They were 49 females and 47 males. Types of anorectal malformations were classified according to Krickenbeck classification. In females, the majorities of cases were vestibular fistulas (22 patients, 45.0%). The second most common type was cloacas (13 patients, 27.0%). In males, the most common type of ARMs was recto-urethral fistulas (15 patients, 32.0%). There were 11 patients with recto-urethral bulbar fistulas (23.0%) and 4 patients with recto-urethral prostatic fistulas (9.0%). The second most common was rectovesical fistulas (13 patients, 28.0%)(Table 1). Seventy-five patients (78.0%) had at least one associated malformation or syndrome. The most common associated anomalies were renal anomalies (40 patients, 50.0%). Proximal esophageal atresias with tracheoesophageal fistulas were found in 6 patients (6.0%) and duodenal atresias/stenoses were found in 5 patients (5.0%)(Table 2). VACTERL associations were found in 35 patients (36.0%). Twelve patients (12.0%) were syndromic: Trisomy 21 (10 patients) and Currarino syndrome (2 patients). All patients with Down's syndrome had no fistulas.

### Operative managements

The choice of surgical management for ARMs varies depending on the type of ARMs, patient's underlying status and surgeons' preferences. At the initial neonatal evaluation, the patients with perineal fistulas, a few patients with vestibular fistulas and without fistula were identified, and later, definitive operations were usually performed on them. Otherwise colostomy would be the first operative procedure. The flow of operative management is shown in Figure 1.

Two patients died due to severe underlying disease before definitive surgeries were done. Summary of definite surgery is shown in Table 3.

**Types and locations of colostomy**

The median time for creating a colostomy was 2 days old (1 - 5, 130 days). Seventy-four patients (74/96, 77.0%) had colostomies. Loop colostomies and double end colostomies were performed in 44 patients

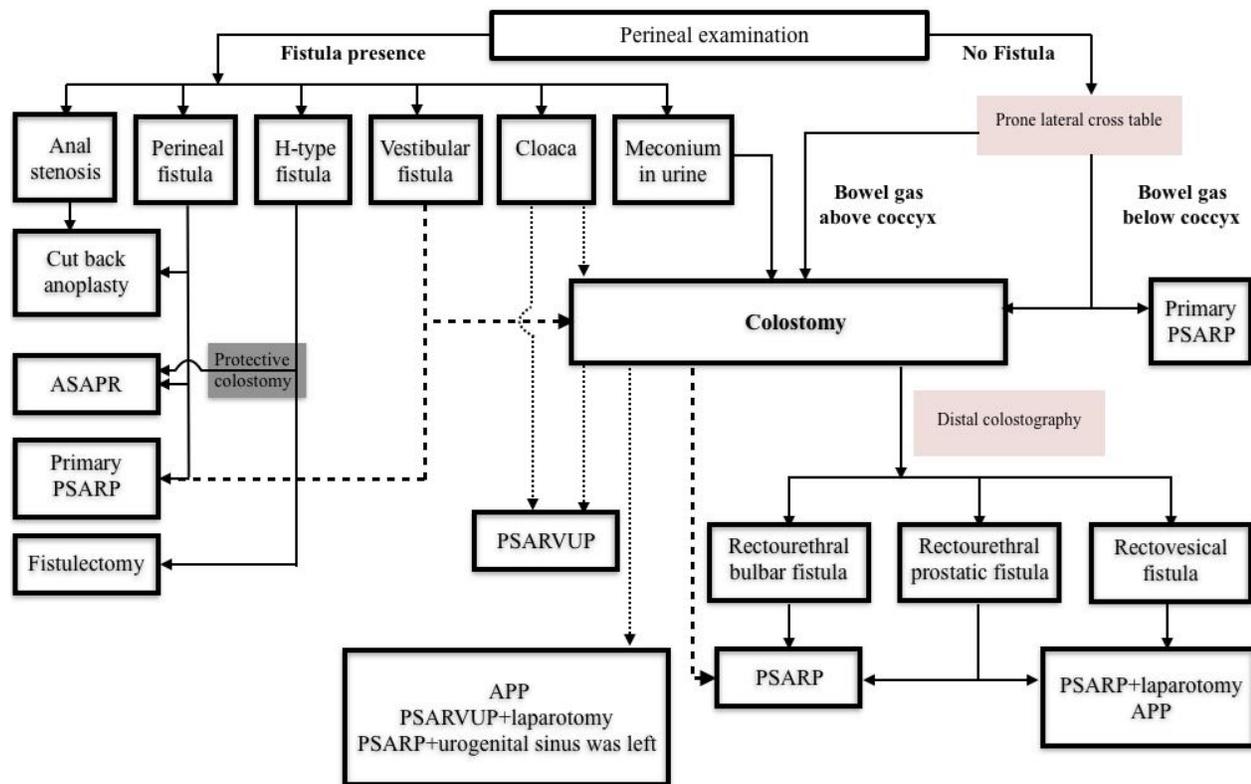
(44/74, 60.0%) and 27 patients (27/74, 36.0%), respectively. Descending-sigmoid colostomies were performed in 61 patients (61/74, 82.0%), while transverse colostomies were performed in 13 patients (13/74, 18.0%).

**Table 1.** Incidence of type of anorectal malformations according to Krickbeck classification.

| Male                         | Total (n) | %    | Female                 | Total (n) | %    |
|------------------------------|-----------|------|------------------------|-----------|------|
| Perineal fistula             | 10        | 21.0 | Perineal fistula       | 4         | 8.0  |
| <b>Rectourethral fistula</b> | 15        | 32.0 | Vestibular fistula     | 22        | 45.0 |
| Prostatic                    | 4         | 9.0  | <b>Cloaca</b>          | 13        | 27.0 |
| Bulbar                       | 11        | 23.0 | < 3 cm. common channel | 4         | 9.0  |
| Rectovesical fistula         | 13        | 28.0 | > 3 cm. common channel | 9         | 18.0 |
| No fistula                   | 8         | 17.0 | No fistula             | 5         | 10.0 |
| Anal stenosis                | 1         | 2.0  | Rare: H type fistula   | 5         | 10.0 |
| <b>Total (n = 96)</b>        | <b>47</b> |      |                        | <b>49</b> |      |

**Table 2.** Frequency of associated anomalies in patients with anorectal malformations.

| Type of associated anomalies          | Anomaly/Total (n) | %    |
|---------------------------------------|-------------------|------|
| <b>Vertebral and spinal anomalies</b> | 23/80             | 28.0 |
| Sacrum or coccyx anomalies            | 13/80             | 16.0 |
| Tethered cord                         | 9/80              | 11.0 |
| Vertebral anomalies                   | 8/80              | 10.0 |
| Syringomyelia                         | 4/80              | 5.0  |
| <b>Cardiac anomalies</b>              | 30/96             | 31.0 |
| PDA                                   | 16/96             | 17.0 |
| VSD                                   | 10/96             | 10.0 |
| ASD                                   | 7/96              | 7.0  |
| Tetralogy of Fallot                   | 3/96              | 3.0  |
| <b>Esophageal atresia</b>             | 6/96              | 6.0  |
| <b>Renal anomalies</b>                | 40/79             | 51.0 |
| Renal agenesis                        | 9/79              | 11.0 |
| Hydronephrosis                        | 16/79             | 20.0 |
| VUR                                   | 17/79             | 22.0 |
| Dysplastic kidney                     | 7/79              | 9.0  |
| <b>Genital anomalies</b>              | 10/47             | 21.0 |
| Undescended testis                    | 3/47              | 6.0  |
| Hypospadias                           | 7/47              | 15.0 |
| <b>Limb anomalies</b>                 | 12/96             | 12.0 |
| Absence radius                        | 1/96              | 1.0  |
| Polydactyly                           | 4/96              | 4.0  |
| Club foot                             | 2/96              | 2.0  |
| Others                                | 5/96              | 5.0  |
| <b>Others</b>                         |                   |      |
| Duodenal atresia/stenosis             | 5/96              | 5.0  |
| Hypothyroid                           | 5/96              | 5.0  |
| Cleft lip/palate                      | 5/96              | 5.0  |



FASARP: anterior sagittal anorectoplasty; PSARP: posterior sagittal anorectoplasty; APP: abdominoperineal pull-through; PSARVUP: posterior sagittal anorectovaginoureteroplasty

**Figure 1.** Flow of operative managements for the anorectal malformations.

**Table 3.** Definitive surgery for the anorectal malformations.

| Procedure                              | Anomaly/<br>total (n) | Vestibular<br>fistula<br>(n = 22 <sup>a</sup> ) | Perineal<br>fistula<br>(n = 14 <sup>a</sup> ) | Anal<br>stenosis<br>(n = 1) | H type<br>fistula<br>(n = 5) | No<br>fistula<br>(n = 13) | Bulbar<br>fistula<br>(n = 11) | Prostatic<br>fistula<br>(n = 4) | Rectovesical<br>fistula<br>(n = 13) | Cloaca<br>(n = 13) |
|--|-----------------------|---|---|-----------------------------|------------------------------|---------------------------|-------------------------------|---------------------------------|-------------------------------------|--------------------|
| Cut back anoplasty                     | 8/94                  |   | 7/13  | 1/1                         |                              |                           |                               |                                 |                                     |                    |
| Fistulectomy                           | 4/94                  |   |   |                             | 4/5                          |                           |                               |                                 |                                     |                    |
| ASARP                                  | 10/94                 | 6/21  | 3/13  |                             | 1/5                          |                           |                               |                                 |                                     |                    |
| PSARP                                  | 43/94                 | 15/21   | 3/13  |                             |                              | 13/13                     | 11/11                         | 1/4                             |                                     |                    |
| PSARP +<br>laparotomy                  | 4/94                  |   |   |                             |                              |                           |                               | 1/4                             | 3/13                                |                    |
| APP                                    | 15/94                 |   |   |                             |                              |                           |                               | 2/4                             | 10/13                               | 3/13               |
| PSARVUP                                | 3/94                  |   |   |                             |                              |                           |                               |                                 |                                     | 3/13               |
| PSARVUP +<br>laparotomy                | 6/94                  |   |   |                             |                              |                           |                               |                                 |                                     | 6/13               |
| PSARP+<br>urogenital<br>sinus was left | 1/94                  |   |   |                             |                              |                           |                               |                                 |                                     | 1/13               |

<sup>a</sup>: one patient of each group was dead before definite surgery due to irrelevant causes. ASARP: anterior sagittal anorectoplasty; PSARP: posterior sagittal anorectoplasty; APP: abdominoperineal pull-through; PSARVUP: posterior sagittal anorectovaginoureteroplasty

### **Perineal fistulas and anal stenosis**

Cut-back anoplasty was commonly performed in 8 patients (7 patients with perineal fistula and 1 patient with anal stenosis). Anterior sagittal anorectoplasty (ASARP) and limited posterior sagittal anorectoplasty (PSARP) were performed in 6 patients (3/6, 3/6). Most of the procedures (10/13) were performed as a primary surgery in neonatal period. However, some

patients underwent colostomies (3/13) due to severe associated anomalies and delayed diagnosis (1.5 year) (Table 3).

### **Vestibular fistulas**

Most patients (14/21) underwent 3-stage operation (colostomy, definitive surgery and closure of colostomy) due to surgeons' preferences. Protective

colostomies at the same time of definitive surgery were performed in two cases of delayed diagnosis (23 days, 14 years). Some surgeons performed PSARP (5 cases) as a primary surgery in the neonatal period followed by 10 - 14 days of total parenteral nutrition. PSARP were commonly performed in 15 (15/21) (Table 3). ASARP was performed in the other patients. (6/21).

#### ***Rectourethral fistulas (bulbar fistulas and prostatic fistulas)***

All patients underwent 3-stage operation. Colostomies were done in neonatal period and then distal colostography were usually achieved later within 3 months of age. PSARP was performed in all patients with rectourethral bulbar fistulas (11/11). Abdominoperineal pull-through (APP) was performed in 2 patients with rectourethral prostatic fistulas (2/4) (Table 3). The others underwent in PSARP (1/4) and PSARP with laparotomy (1/4).

#### ***Rectovesical fistulas***

All patients underwent 3-stage operation. APP was performed in 10 patients (10/13) and PSARP with laparotomy were performed in 3 patients (3/13) (Table 3).

#### ***No fistulas***

Most patients (12/13) underwent 3-stage operation. Prone cross-table lateral radiographies were investigated 24 hours after birth in all patients (13/13) (Table 3). PSARP was performed as definitive surgery and as a primary surgery in one patient during neonatal period.

#### ***H type fistulas***

All patients (5/5) had delayed diagnosis (3, 7, 7, 8 and 9 months). Fistulectomy was performed in 4 patients (4/5) while ASARP with protective colostomy was performed in one patient (1/5) (Table 3).

#### ***Cloacas***

##### ***Common channel length of shorter than 3 cm***

Posterior sagittal anorectovaginourethroplasty (PSARVUP) with total urogenital mobilization (TUM) was performed in 3 patients (3/4) and PSARP with delayed repair of urogenital sinus in 1 patient (1/4) (Table 3).

##### ***Common channel length of longer than 3 cm***

PSARVUP with laparotomy was performed in 6 patients (6/9) and APP was performed in 3 patients

(3/9). Vaginal replacements using the rectum as neovagina was performed in 2 patients (2/9).

#### ***Operative management of patients with ARMs and associated anomalies***

##### ***ARMs and duodenal atresias***

Five patients had ARMs and duodenal atresias. Duodenoduodenostomy with colostomy was performed in 4 patients (2 no fistula, 1 rectourethral bulbar fistula and 1 cloaca). Duodenoduodenostomy with cut-back anoplasty was performed in 1 patient with perineal fistula.

##### ***ARMs and esophageal atresia with tracheoesophageal fistulas (EA with TE fistulas)***

Five patients had ARMs and EA with TE fistulas. Esophagoesophagostomy with colostomy were performed in 3 patients with vestibular fistulas and 2 patients with rectovesical fistulas. One patient had ARMs with perineal fistula, duodenal atresia and EA with TE fistula. She underwent cutback anoplasty with duodenoduodenostomy as the first surgery and esophagoesophagostomy seven days later.

#### ***Complications***

##### ***Complications of colostomy***

Complication of colostomies occurred in 21.6% (16 /74), including prolapse (5/74), improper length (1/74), incomplete diversion which caused urinary tract infection or fecal impaction (8/74), gut obstruction (1/74) and retraction (1/74).

##### ***Postoperative complications***

Thirty-five patients (35/94, 37.0%) had postoperative complications (Table 4). The most common complication was wound infection (15/94, 15.9%). Twenty-two patients (22/94, 23.4 %) had redo-operations but only 10 patients (10/94, 10.6%) underwent major operations. The most common redo-operation was mucosectomy for mucosal prolapse (12/94, 12.7%).

##### ***Postoperative results***

Fifty-two patients were older than 3 years of age and were assessed for functional outcome according to Krickenbeck classification (Table 5). The median time of follow-up is 4.5 years. Total continence had been achieved in 42.0% of all. Constipation occurred in 31.0% of the patients, mostly in perineal fistulas, vestibular fistulas and no fistulas, respectively. Soiling occurred in 27.0%, mostly in cloacas, rectourethral prostatic fistulas and rectovesical fistulas.

**Table 4.** Post-operative complications.

| Types                           |         | Wound infection | Mucosal/rectal prolapsed | Anal stricture |
|---------------------------------|---------|-----------------|--------------------------|----------------|
| Vestibular fistula              | (n= 21) | 5               | 0                        | 2              |
| Perineal fistula                | (n= 13) | 0               | 0                        | 1              |
| Anal stenosis                   | (n= 1)  | 0               | 0                        | 0              |
| H type fistula                  | (n= 5)  | 0               | 0                        | 0              |
| No fistula                      | (n= 13) | 1               | 1                        | 1              |
| Rectourethral bulbar fistula    | (n= 11) | 2               | 1                        | 1              |
| Rectourethral prostatic fistula | (n= 4)  | 1               | 2                        | 0              |
| Rectovesical fistula            | (n= 13) | 1               | 6                        | 1              |
| Cloaca                          | (n= 13) | 5               | 2                        | 2              |
| Total                           | (n= 94) | 15              | 12                       | 8              |

**Table 5.** Functional outcome of the 52 children older than 3 years of age according to the Krickenbeck assessment system.

| Type of ARMs                    | Total continence (%) | Constipation (%) | Soiling (%) | Total (n) |
|---------------------------------|----------------------|------------------|-------------|-----------|
| Perineal fistula                | 50.0                 | 50.0             | 0.0         | 4         |
| Vestibular                      | 55.0                 | 45.0             | 0.0         | 11        |
| H-type fistula                  | 100.0                | 0.0              | 0.0         | 1         |
| No fistula                      | 29.0                 | 57.0             | 14.0        | 7         |
| Rectourethral bulbar fistula    | 63.0                 | 12.0             | 25.0        | 3         |
| Rectourethral prostatic fistula | 33.0                 | 0.0              | 66.0        | 8         |
| Rectovesical fistula            | 44.0                 | 28.0             | 33.0        | 9         |
| Cloaca                          | 11.0                 | 22.0             | 66.0        | 9         |
| Total                           | 42.0                 | 31.0             | 27.0        | 52        |

## Discussion

The worldwide incidence, prevalence and presentation of patients with ARMs are heterogeneous. Pena A. reported rectourethral fistula as the most common lesion in males and vestibular fistula as the most common lesion in females.<sup>(7)</sup> The first report of the ARM-NET (European consortium on anorectal malformations) reported perineal fistula as the most common lesion both in males (43.0%) and females (41.0%) followed by rectourethral fistula in males (29.0%) and vestibular fistula in females (28.0%).<sup>(8)</sup> In our series, rectourethral fistula is the most common lesion in males (32.0%) and vestibular fistula is the most common lesion in females (45.0%). Followed by rectovesical fistula (28.0%) and cloaca (27.0%), respectively. The high incidence of cloacas in our series owing to our institution as a tertiary care center. The incidence of ARMs without fistula in our series was higher from previous reports.<sup>(9)</sup> Seventy-five patients (78.0%) had at least one associated anomaly, higher than which has been described

in literatures, they vary between 20.0 to 70.0%.<sup>(10)</sup> The most common associated anomalies were renal anomalies (50.0%). Associated anomalies in our series show no difference from recent literatures.<sup>(10-16)</sup>

The operative management depends on the types of ARMs. The decision making between a primary repair or staged operation depends on the experience of the surgeon, the condition of the patient and the surrounding circumstances. We usually perform primary repair in most patients with perineal fistulas (10/13), H type fistulas (4/5), in addition, in a patient with vestibular fistula (1/13) and another patient with no fistula (1/13). Colostomy is traditionally performed as part of staged operation. Accordingly, 77.0% of cases had colostomies. We performed both loop colostomies and double end colostomies. It has been suggested that divided sigmoid colostomy may result in better radiological studies and a lower incidence of prolapse.<sup>(17)</sup> In our series, the incidence of prolapse showed no difference between the two groups but

the incomplete diversion from loop colostomies caused urinary tract infection and fecal impaction more than double end colostomy. Therefore, the authors currently perform divided sigmoid-descending colostomy. Minimal PSARP is the current standard technique for perineal fistulas. However, the alternative technique, anoplasty may not affect the functional or cosmetic outcome.<sup>(18)</sup> In our series, surgeons also perform cut back anoplasty and ASARP. In vestibular fistulas (17/21) and no fistulas (12/13), we performed staged operation in most of the patients. The surgeon preferred to performed protective colostomy because of the risk of wound infection and wound dehiscence.<sup>(17-22)</sup> In recent studies, PSARP performed without a protective colostomy have low morbidity and good continence.<sup>(23)</sup> Our surgeons started to perform PSARP without a protective colostomy (4/21) with post operative total parenteral nutrition for 10 - 14 days in 2014. Also, in no fistulas, the surgeon started to performed PSARP (1/13) as the primary surgery in neonatal period, if prone lateral crossable x-ray film showed rectal gas below the coccyx. Various surgical techniques have been used for H type fistulas.<sup>(24-27)</sup> In the majority of H type fistulas (4/5), we performed fistulectomy. This procedure is less invasive, better visualization, and easier to perform, with reliable results.<sup>(26)</sup>

The most common postoperative complication in our series was wound infection, mostly in cloacas. Second most common complication was mucosal prolapse, largely in rectovesical fistulas. The length of rectal dissection in the intra-abdominal space may play an important role in developing postoperative rectal prolapse. Longer dissections in rectovesical fistulas may lead to higher rate of rectal prolapse.<sup>(27)</sup> There was no wound infection in perineal fistulas, anal stenosis and H-type fistulas owing to the fact that wound problems after neonatal treatment of low defects are very uncommon and urological complications following surgery as they are unacceptable.<sup>(28)</sup>

Postoperative incontinence and constipation remain major problems that impede social and psychological development. We assess the functional outcome according to international classification (Krickenbeck) to evaluate voluntary bowel movement, soiling and constipation. In our series, voluntary bowel movement was presented in 42.0% of patients, comparable to other studies.<sup>(29, 30)</sup> Soiling was presented in 27.0% and mainly found in cloacas, rectourethral prostatic fistulas and rectovesical fistulas.

The causes of worst prognosis are the more marked hypoplasia of the voluntary sphincter muscles and the presence of severe sacral anomalies including tethered cord.<sup>(32)</sup> Constipation was presented in 31.0% and found 50.0% with perineal fistulas, 45.0% with vestibular fistulas, and 57.0% with no fistulas, comparable to the 1995 study of Pena and the others.<sup>(2, 33)</sup> The cause of constipation is unclear: colonic motility disorder in patients with low defects and vestibular fistulas.<sup>(30)</sup> Also, rectosigmoid hypomotility and generalized colonic motility disturbance have been suggested.<sup>(34)</sup> Our series didnot establish the relationship between types of surgical procedures and functional outcome due to small sample size in each groups.

### Conclusion

In our series, an incidence of types of ARMs according to Krickenbeck classification was quite similar compared to the other studies. Surgical managements were highly variable depending on the types of ARMs and the surgeons' preferences. Functional outcome was consistent with other studies. Prospective study and long-term follow up are needed to provide evidence on the outcome of different surgical procedures.

### Conflict of interest

The authors, hereby, declare no conflict of interest.

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