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Bowel perforation in newborn with anorectal malformation and no fistula at presentation ☆

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ABSTRACT

Purpose: Anorectal malformation (ARM) in newborns with no fistula at presentation resembles intestinal obstruction. The aim of this study is to study the factors associated with bowel perforation in this group of patients.

Methods: From 2000 to 2012, 106 newborns with ARM were managed in our hospital. Thirty neonates without fistula at presentation were included in this study. Demographic data and the incidence of bowel perforation were studied.

Results: Twenty-nine male and 1 female were included in the study. Five patients were born premature and six patients had low birth weight. Six patients had Down's syndrome and 12 patients had associated anomalies. Cross-table lateral x-ray in prone position was performed from 20 to 24 hours after birth. All operations were performed within 48 hours after birth. One neonate underwent primary anoplasty. Twenty-nine neonates underwent colostomy. Two males developed bowel perforation before surgery (at 33 and 36 hours after birth). Perforation was associated with low birth weight ($p = 0.034$) and was not associated with prematurity ($p = 0.31$), Down's syndrome ($p = 0.634$) or the presence of other associated anomalies ($p = 0.687$).

Conclusions: In newborns with ARM, bowel perforation can occur within 36 hours after birth. Forty-eight hours of waiting is too long as it risks perforation. In this study, a neonate with low birth weight was trended toward bowel perforation.

In newborns with anorectal malformation (ARM), the diagnosis is usually made after birth. The initial management includes work-up for the associated anomalies and to decide the type of initial operation [1]. Although there are different classification systems in ARM [2,3], the simplest classification that guides the initial management in males is to classify the imperforate anus as high or low type. In high type, a colostomy is performed in the neonatal period while in low type, primary anoplasty is performed [4].

Clinical examination was performed to classify the types of malformation in 90% [5]. Perineal fistula or bucket handle deformity in males is suggestive of a low lesion (Fig. 1A), while meconium-stained urine or a flat buttock is suggestive of a high lesion (Fig. 1B). However, if there is no clinical clue, a cross-table lateral x-ray in prone position is required to differentiate the types of malformation (Fig. 2). It is suggested the x-ray be taken 24 hours after birth in order to build up adequate pressure in the distal colon although others suggested that x-ray can be taken at 12 hours after birth [1].

In this group of patients, since there is no passage of meconium at presentation, this condition mimics intestinal obstruction and poses a risk of intestinal perforation. This study aims to examine the perinatal management and characteristics of this particular group of neonates without fistula clinically at initial presentation and to determine any factor associated with bowel perforation.

1. Materials and methods

A retrospective review was performed in all newborn males with ARM from January 2000 to November 2012. One hundred six newborns with ARM were managed in our hospital in the study period. Thirty-two neonates did not have any fistula clinically before operation. Of the 32 patients, 2 patients who underwent colostomy without cross-table lateral x-ray were excluded from this study: 1 had associated esophageal atresia, which was repaired in the first day of life, and the other one was born at 32 weeks of gestation and was small for gestational age. He underwent colostomy within 24 hours after birth.

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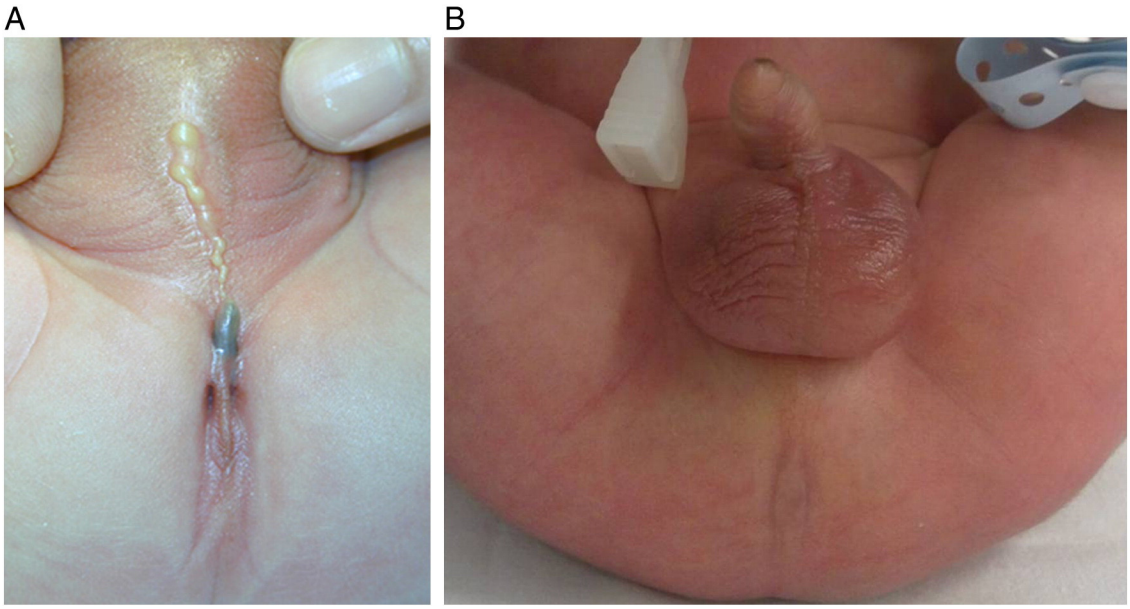

Fig. 1. (A) Low-type anorectal malformation with bucket handle deformity and mecnoium track at the perinium. (B) High-type anorectal malformation with a flat buttock and meconium-stained urine at the meatus.

The remaining 30 patients who underwent cross-table lateral x-ray were subjected to further analysis. Clinical characteristics including birth weight, maturity of the neonates and the presence of any associated anomalies were recorded. Prematurity is defined as a gestation age <37 weeks. Low birth weight is defined as birth weight <2.5 kg. The diagnosis of Down's syndrome was confirmed by chromosome assay. All had orogastric tube insertion for gastric decompression and continuous SaO_2 monitoring before surgery.

The timing of the cross-table lateral x-ray was recorded. Primary anoplasty was performed if the distance of the most distal bowel gas and the perineal skin marker is less than 1 cm in the cross-table lateral x-ray in prone position (Fig. 2A). If the distance is greater than 1 cm (Fig. 2B), colostomy was performed. The type and timing of initial operation and the finding of loopogram and cystourethrogram were reviewed. The incidence of bowel perforation and any associated factor were studied.

Statistical analysis was accomplished using the SPSS program for Windows 15.0 (SPSS, Chicago, IL, USA). Fisher's exact test was used to compare categorical data. A *t*-test was used to compare continuous data. $p < 0.05$ was considered statistically significant. The study was approved by the local clinical research ethical committee (reference number: CRE-2013.148).

2. Results

Thirty patients (male/female, 29:1) were included in this study. Five patients were born premature. Five patients had a low birth weight. Six patients had Down's syndrome and 12 patients had associated anomalies (Table 1).

Cross-table lateral x-ray in prone position was performed between 20 and 24 hours after birth in all patients. One patient had an x-ray feature suggestive of low-type ARM and primary anoplasty was performed. The rest of the 29 children underwent colostomy. All initial operations were performed <48 hours after birth. Three children were noticed to have perineal fistula after colostomy. The fistula was not apparent on on-table assessment at the time of colostomy and was detected soon after the operation.

One child died after colostomy. Distal loopogram and cystourethrogram were performed in the remaining 28 patients who had a colostomy (Fig. 3). Three patients had perineal fistula. Fifteen neonates did not have any fistula including the six neonates with Down's syndrome. Among these 15 patients, loopogram showed that 2 children had the distal rectum ends at the prostatic urethra level. The rest of the 13 children including all 5 patients with Down's syndrome had the distal rectum ends at the bulbar urethra level.

Ten neonates demonstrated the presence of rectourethral fistula. Eight had rectourethral prostatic/rectobladder neck fistula and two had rectourethralbulbo fistula (Fig. 4). The only female patient in this study had Down's syndrome who had no fistula demonstrated in the loopogram.

Two children developed bowel perforation while awaiting surgery. The timing of relief of the obstruction in the perforated group (mean 34.5 ± 2.1 hours, range 33–36 hours) was significantly longer than the nonperforated group (mean 28.9 ± 7.5 hours, range 24–48 hours) ($p < 0.05$). Desaturation was noticed while awaiting surgery at 33 and 36 hours after birth respectively. One patient underwent anoplasty and the perforation was noticed after the anoplasty. Retrospective review of the chest x-ray after desaturation showed free gas under the diaphragm. He subsequent underwent colostomy and repair of the perforation. Another child who was scheduled for colostomy developed desaturation while awaiting operation. The x-ray showed free gas under the diaphragm. Colostomy together with repair of the perforation was performed. He died of sepsis after colostomy. Perforation was located at the rectosigmoid junction with dilated sigmoid proximal to the site of perforation in both cases.

Perforation was associated with low birth weight ($p = 0.034$) and was not associated with prematurity ($p = 0.31$), Down's syndrome ($p = 0.634$) or the presence of associated anomalies ($p = 0.681$) (Table 2).

3. Discussion

Patients with anorectal malformation with no fistula at presentation are considered a special group of patients. There is no passage of meconium before operation. The anal appearance does not always indicate the exact nature of the anomalous anatomy. A cross-table x-ray in prone position is required in the decision of the initial operation. However, the operation is usually performed more than 24 hours after birth because it was recommended that