PRIMARY POSTERIOR SAGITTAL ANORECTOPLASTY WITHOUT COLOSTOMY IN NEONATES WITH HIGH IMPERFORATE ANUS

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Abstract- The standard approach to management of high imperforate anus is colostomy in the newborn period followed by posterior saggital anorectoplasty (PSARP) at 6 to 12 months of age. The purpose of this study was to determine whether a one-stage repair by primary PSARP in the newborn period could be performed without clear determent to the patient's functional result. Totally 30 newborns with high imperforate anus who underwent primary PSARP without colostomy were studied retrospectively. All cases were studied by sonography, echocardiography, lower vertebra X-ray and finally routine blood and urine laboratory tests. Incontinence was defined as fecal soiling at least twice a day, and patients requiring more than office dilatation were described to have stricture formation. All patients recovered well and were followed for periods ranging from 1.5 to 10 years. There were 3 cases of postoperative wound infection, but no anastomotic dehiscence, stricture formation and fistula recurrence were seen. In cases of constipation and fecal incontinence there were associated anomalies such as sacral dysplasia, kidney dysplasia, ventricular septal defect and vesicoureteral reflux. There were no complications in other cases. The most important factor is patient selection, and one-stage PSARP spares the patient the morbidity of additional surgeries in the standard multistage approach for high imperforate anus. These preliminary results suggest that one-stage PSARP is a safe and viable approach to the management of high imperforate anus without clear determent to future bowel function.

Acta Medica Iranica, 45(2): 121-125; 2007

Key words: Posterior saggital anorectoplasty, high imperforate anus, colostomy

INTRODUCTION

In 1982 Pena and De vries introduced the posterior saggital anorectoplasty (PSARP) operation, in which the rectourogenital connection is divided under direct vision, and the neonatal rectum anastomosed to the perineum (1). Since then, PSARP has become the most common surgical procedure performed for patients with imperforate anus (2).

Received: 19 Nov. 2005, Revised: 28 Dec. 2005, Accepted: 3 Jan. 2006

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With the exception of those with perineal fistulas, colostomy at birth is strongly recommended in all patients with "high" imperforate anus with PSARP to be performed selectively sometime during the first year of life (3, 4). In addition to PSARP, a variety of surgical approaches for imperforate anus have been described, including the combined abdominoperineal operations of Rhodes, Pipes, and Randall (5), and the sacroperineal approaches by Aluwihare (6). Many of these operations are performed at birth as a primary procedure without need for colostomy. Most of these techniques are blind, and potentially injurious (7). In 1990, Moore reported that saggital anorectoplasty can be safely and successfully carried out in the newborn with high imperforate anus without the

need for colostomy (8). Recently, Albanese *et al.* and Liu and Hill have reported on 12 newborns successfully underwent primary PSARP without colostomy (9, 10).

This report describes our experience with primary PSARP for high imperforate anus and, unlike similar studies (8, 9), provides long-term follow-up data on bowel continence.

MATERIALS AND METHODS

From January 1993 to September 2003 totally 30 newborns with high imperforate anus underwent primary PSARP. We obtained informed consent from parents of all children.

In all cases, the presence of a high imperforate anus was diagnosed initially by clinical examination at birth, then repeated at 24 hours to rule out a perineal fistula. Serial urinalysis was done and a piece of gauze was placed on the tip of the penis to check for particles of meconium in all cases. Standard anterior-posterior and lateral films were routinely performed to rule out vertebral abnormalities. In all cases, abdominal ultrasound (to rule out associated obstructive uropathy) and echocardiography (to rule out lethal cardiac defects) were performed shortly after birth.

All patients underwent PSARP as described by De vries and Pena. Postoperatively, the "neo-anus" was gently dilated with an 8 size Hegar dilator on 12th postoperative day. Parents were instructed in gentle Hegar dilatation to be done once a day beginning on 14 Hegar caliber and gradually increasing by the surgeon based on biweekly examinations.

Follow-up, achieved in all patients, ranged from 2 to 10 years (mean, 6 years). Surgical outcomes of all patients were obtained by a combination of retrospective in and outpatient chart review, and personal telephone communication when appropriate. Post-operative complications of wound infection, anastomotic dehiscence, stricture formation, and recurrent fistulae were duly recorded.

Patients requiring more than office dilatation were considered to have stricture formation. All patients were observed for bowel function notably the presence or absence of constipation, fecal incontinence, or both. Patients > 3 years of age were clinically assessed for continence and considered incontinent if they had fecal soling at least twice a day. Patients were considered constipated if they required a medical regimen to have daily bowel movements.

RESULTS

There were 17 males (15 with recto-urethro-bulbar fistula and 2 with recto-urethro-prosthetic fistula) and 13 females (3 with low recto-vaginal fistula and 10 with rect-vestibular fistula). Birth weight averaged 3.2 kg (range 2.5-4.0 kg). All males were operated upon at the age of 1 to 2 days and females at mean age of 8 days (range 1 to 20 days). Preoperative abdominal ultrasounds were normal except for a single case with significant bilateral renal dysplasia, and one case of vesico-urethral-reflux. Associated anomalies are summarized in Table 1.

Table 1. Associated anomalies based on sex and type of fistula

	Females		Males	
Type of anomaly	**Fistula type**	**Number**	**Fistula type**	**Number**
Ventricular septal defect	rectovestibular	1	recto-urethro-bulbar	2
Vesicoureteral reflux	recto-vaginal	3	recto-urethro-bulbar	2
	recto-vestibular	1		
Sacral dysplasia	-	-	recto-urethro-bulbar	2
			recto-urethro-prostatic	1
Bilateral renal dysplasia	-	-	recto-urethro-prostatic	1
Tethered cord	recto- vaginal	1	-	-
Hypospadias	-	-	recto-urethro-bulbar	1
Undescended testis	-	-	recto-urethro-bulbar	1
Inguinal hernia	-	-	recto-urethro-bulbar	1
Vaginal septum	recto-vestibular	1	-	-

In no case was it necessary to taper the rectum for pull-through. All patients recovered uneventfully. There were 3 cases of postoperative wound infections, but anastomotic dehiscence, stricture formation, and recurrent fistula did not develop in any patients. No patient has required a subsequent colostomy. Twenty five of 30 patients were continent of stool with no soiling episodes (13 male, 12 female). Postoperative bowel functions are summarized in Table 2.

DISCUSSION

Since early 1980s, PSARP as described by De vries and Pena has become the most commonly performed surgical procedure in many medical centers for patients with imperforate anus (7). Although PSARP is intended as a primary procedure in the newborn with a low imperforate anus, specifically perineal fistula, it had been strongly recommended that early colostomy followed by PSARP during the first year of life should be performed for "high" malformations, including vestibular fistula (7).

The rationale behind the 2-stage approach included the following: the anatomy of this area in newborn babies could in theory be less well-defined, specifically where the sphincteric mechanism resides, making dissection less precise and more prone to straying from the midline where exact placement of the neo-anus through the center of the sphincteric mechanism is essential to achieve optimal bowel function. A distal colostogram, which cannot be performed in a child without a colostomy, would accurately show the actual site of the rectourinary fistula, the true height of the rectal pouch, and the presence, if any, of significant urinary tract anomalies, such as vesicourethral reflux. There is a risk of dehiscence and infection in a primary repair. Colostomy may be essential for the survival of some newborns, especially low-birth weight new-borne, to permit continued growth while awaiting later definitive repair.

Moore first described saggital anorectoplasty without colostomy in newborns with rectourinary tract fistulas with excellent surgical results (8). In his series, saggital anorectoplasty was performed through an anterior approach with the baby in the supine, modified-lithotomy position. Albanese *et al.* recently described 5 newborns who underwent successful primary PSARP (9). Liu and Hill series of 7 male newborns with rectourinary tract fistula who underwent primary PSARP also showed good results (10). Our series of 30 newborns with high fistula of imperforate anus who underwent primary PSARP also showed that anorectoplasty can be performed safely and effectively in newborns with "high" fistula.

In all cases the superficial parasagittal muscle fibers and the "muscle complex" as described by Pena (7) were easily identified during the dissection with the help of a nerve stimulator, and the midline was strictly adhered to. In newborns with rectourethral fistula, the fistula was easily identified and repaired through the posterior saggital approach. However, unlike Moore and Liu, we were unable to repair a rectovesical fistulae solely through the saggital approach, and added laparotomy for identification and repair of the fistula.

Table 2. Postoperative bowel function in 30 patients

Males (n=17)			Females (n=13)		
Bowel function	**Fistula type**	**Associated anomaly**	**Bowel function**	**Fistula type**	**Associated anomaly**
Continent (n=13)	-	–	**Continent** (n=12)	–	–
Soiling (n=5)	RUB (n=3) RUP (n=2)	sacral dysplasia (n=3) renal dysplasia (n=1) VUR (n=1)	Soiling (n=3)	RV (n=3)	tethered cord (n=1) VUR (n=2)
Constipation (n=5)	RUB (n=5)	sacral dysplasia (n=2) VUR (n=2) VSD (n=2)	Constipation (n=1)	RV (n=1)	VUR (n=2)
Incontinent (n=4)	RUB (n=2) RUP (n=2)	sacral dysplasia (n=3) renal dysplasia (n=1)	**Incontinent** (n=1)	RV (n=1)	tethered cored (n=1)

Abbreviations: RUB, recto-urethro-bulbar; RUP, recto-urethro-prostatic; RV, recto-vaginal; VUR, vesicoureteral reflux.

We believe laparotomy is necessary for rectovesical fistulae, because it represent a true supralevator malformation, which is inaccessible through the perineum. There were 3 cases of postoperative wound infection, but no anal strictures, recurrent fistula, or anastomotic dehiscence were seen in our series. In no case was it necessary to taper the rectum for pull-through. Furthermore, no patient required a subsequent colostomy.

Unlike the reports by Moore and Albanese, Liu cases and our series provided long-term follow-up to assess the effect of primary PSARP on bowel continence: 25 of 30 patients had excellent fecal continence and voluntary control of bowel movements without soiling; 5 patients were incontinent (2 recto–urethro-prosthetic fistula, 2 recto-urethro–bulbar fistula and one recto–vaginal high type fistulae), which all 5 patients had severe associated anomalies. This was not surprising, as patients with supralevator malformations have been shown to have high rates of fecal incontinence (11).

We had 6 cases of constipated patients, and 8 cases of soiled patients among our series. All cases of incontinent, soiled and constipated patients mostly had severe associated anomalies, such as sacral dysplasia (3), bilateral renal dysplasia (1), tethered cord (1), and vesicoureteral reflux (6).

Judgment on the usefulness of any surgical procedure for imperforate anus rests ultimately on the effect on future bowel function, especially the level of continence. Traditional PSARP performed in the first year of life after neonatal colostomy was still associated with significant rates of incontinence (10-14). Only 42% of all patients born with imperforate anus and subjected to a PSARP will be totally continent (11). Pena described a 65% rate of soiling in patients with rectourethral fistula with increasing rates among patients with higher malformations such as rectovesical fistula (84%); 20% of patients with rectourethral fistulas did not have voluntary bowel movements, the rates increasing with higher malformation (11). Importantly, as Pena suggested, traditional PSARP performed during the first year of life after neonatal colostomy is an effective and satisfying way to repair high defects and, together with an effective bowel management program, achieves acceptable bowel function and quality of life for these patients.

Although our series of 30 patients is larger than previous reports, it is still too small to reach definite conclusions on the effect of primary PSARP on future bowel function. It is encouraging that some of our patients on long-term follow-up are completely continent of stool. This is not entirely suppressing as it has been reported that PSARP performed in children under 6 months of age resulted in improved rates of continence (9). Furthermore, primary PSARP avoids the frequently cited complications that can occur with performance of a colostomy, such as wound infection, dehiscence, and silage (15).

We conclude that primary PSARP is a safe and viable approach to management of high imperforate anus without clear detriment to future bowel function deserving further investigation.

Conflict of interests

The authors declare that they have no competing interests.

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