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Anoplasty for low anorectal malformation

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ABSTRACT

Background: The presence of a slightly anterior, stenotic anus is a low anorectal malformation. It is a type of rectoperineal fistula. The cardinal symptoms are constipation and straining at stool. Hendren described a technically simple anoplasty for this condition in 1978. Controversy about terminology has led to a lack of clarity about this condition.

Methods: Clinical data were reviewed about the authors' patients, who had Hendren anoplasty, from 2009 to 2015. Followup data were obtained from office visits and telephone interviews with families and primary care doctors.

Results: Seven patients (4 boys) presented with intractable constipation. All had a stenotic anal orifice, located anterior to the center of the anal wink, within the pigmented perianal skin. In 4 of 7 cases, the lesion was not recognized at birth. Anoplasty was performed at a median age of 8 months (range 6–28). Late followup information was obtained on six of the seven patients at a median of 32 months (range 28–61). Four reported no or minimal laxative requirement and two reported daily laxative use but good symptom control.

Conclusions: Low anorectal malformation/rectoperineal fistula may be overlooked in the newborn. When symptomatic, it may be corrected by a simple anoplasty with excellent results.

The clinical entity of constipation caused by anterior location of the anus was described by Bill in 1958 [1]. Hendren described a technically simple anoplasty for anterior anus in 1978 [2]. There has been controversy about the definition and descriptive terminology of the condition [3,4,5]. Slight anterior displacement of the anus has been variously described as *anterior ectopic anus (AEA)*, *covered anus*, or *anteriorly displaced anus*. The most recent English language texts include this abnormality in the term *rectoperineal fistula* [6].

The term *rectoperineal fistula* is now used to describe a situation where the anus opens anteriorly to the normal location, passing anterior to, or partially through, the sphincter mechanism [6] (Figs. 1a,2a,4a). The anus in these lesions is usually stenotic. The most minor, proximal form may be corrected by a simple anoplasty, whereas a more displaced fistula is usually treated by a procedure to relocate the rectum within the sphincters [6] (Fig. 3).

A source of confusion in this area is the entity of *anterior ectopic anus* [7,8]. Consensus has emerged that this term should only be used to describe a correctly formed anus and sphincter mechanism, located more anteriorly than normal [4,5,6,7]. Its prevalence is disputed. It is not the lesion described in this report.

1. Materials and methods

The project was approved by the Albany Medical Center Institutional Review Board (Certificate Number 4394). Records were obtained of the authors' cases of Hendren anoplasty from 2009 to 2015. Clinical photographs were reviewed. All patients were under the care of a single clinician (CW). Followup data were obtained from office visits, telephone calls, or from primary care practitioners. A standardized questionnaire was administered to family members who were interviewed.

2. Surgical technique

The technique has been described previously [2]. The patient is placed in lithotomy position. Antibiotics are administered. The center of the anal sphincter is defined visually and by muscle stimulation (Fig. 4a). A circular skin flap is marked out, extending back from and including the posterior margin of the anus and encircling the area of the anal muscle contraction (Fig. 4b). Holding sutures are placed at each side of the flap and suspended over hemostat clamps placed on the drapes at each side, exerting moderate traction (Fig. 4b).

The skin is incised and the edge of the flap is grasped with hemostats. The skin flap is elevated. The sphincter fibers are pushed bluntly off the back of the flap with scissors, but they are not cut. This dissection is facilitated by placing a finger in the rectum and is carried just proximal to the dentate line, so that the upper part of the flap is full thickness of rectum (Fig. 4b, 4c).

The flap of skin is excised at the anocutaneous junction, by first cutting it vertically in the midline, placing the first stitch in the six o'clock position, and then removing the excess skin at each side (Fig. 4c). The

Abbreviations: VACTERL, vertebral anomalies, anorectal malformation, cardiac anomalies, tracheoesophageal fistula, renal anomalies, limb anomalies; PSARP, posterior sagittal anorecto plasty; AEA, anterior ectopic anus; AGI, anogenital index; API, anal position index.

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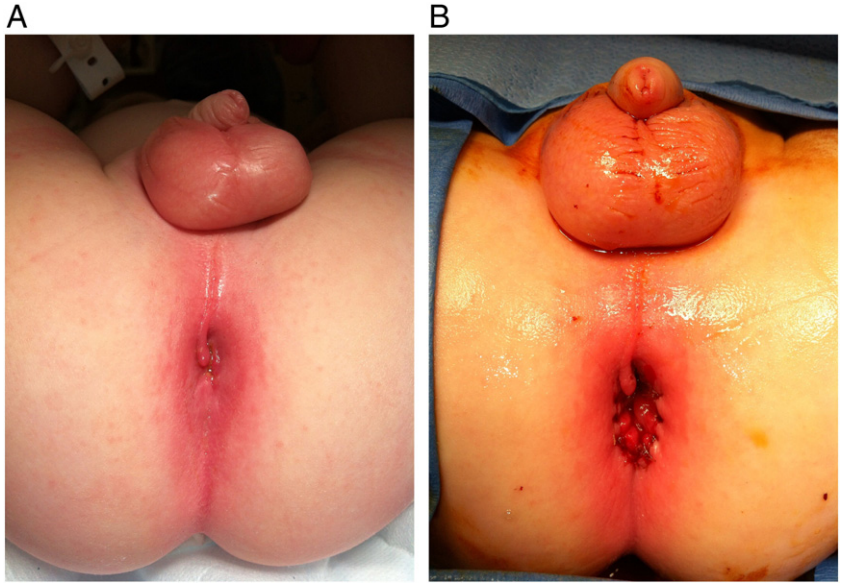

Fig. 1. a. Newborn male with rectoperineal fistula: stenotic anus located anteriorly within the pigmented skin of the anal dimple. Posterior shelf present. b. Same infant as 1a, after anoplasty procedure.

rectal wall is then sutured to the posterior skin edge with chromic catgut. The visible anal mucosa retracts inward afterwards. The procedure effectively opens out the stenotic orifice, straightens the path of the anus, and eliminates the posterior shelf by displacing the sphincter fibers posteriorly, without disrupting them.

Normal diet is resumed immediately. The patient is discharged home on laxatives. No dilatations are required. During follow-up, laxatives are weaned according to clinical response.

3. Results

Over a six-year period, in our community pediatric surgery practice, the authors have encountered seven cases of a distinct clinical problem characterized by the following features (Figs. 1a, 2a, 4a):

- Anal stenosis
- Constipation
- Severe straining at stool
- Location of the stenotic anal orifice just anterior to the center of the anal contraction, within the anterior margin of the pigmented skin of the anal dimple,
- Shelf located posterior to the anal orifice

There were four boys and three girls, operated at the ages of 6–28 months (median 8 months). All patients had a trial of laxative management prior to operation. Median age of onset of symptoms was 5 months (range 0–7).

Other anomalies were present in two of the seven cases, who exhibited features of the VACTERL association. One VACTERL patient had esophageal atresia and tracheoesophageal fistula, left diaphragmatic hernia (Bochdalek type), and vertebral anomalies. The other, born at 27 weeks, had duodenal stenosis and annular pancreas, atrial and ventricular septal defects.

In three cases the lesion was recognized at birth and they were initially managed nonoperatively. Two of these three were the babies

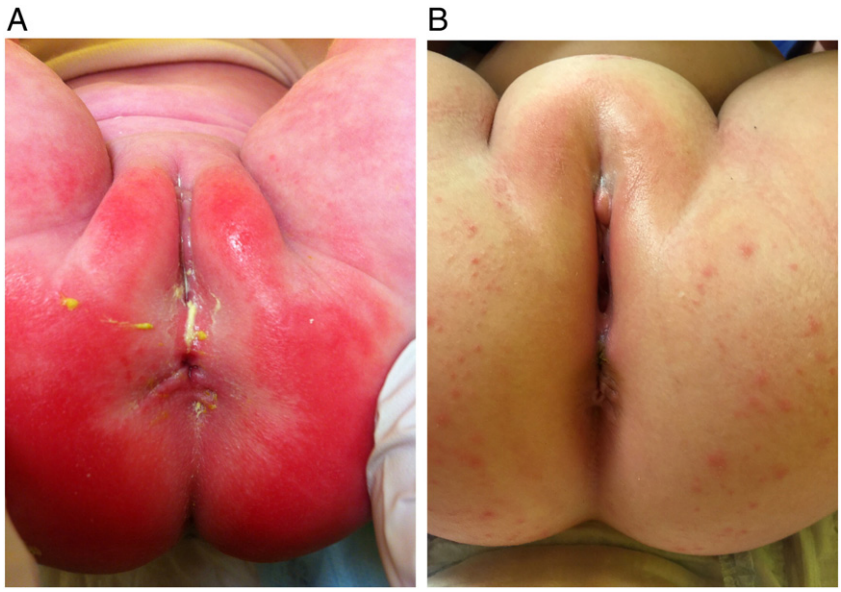

Fig. 2. a. Newborn female with rectoperineal fistula, anoplasty at 8 months of age. b. Same patient as 2a, appearance at 3 years of age.