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Gynecologic anatomic abnormalities following anorectal malformations repair



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ABSTRACT

Background/aim: Patients may present with gynecologic concerns after previous posterior sagittal anorectoplasty (PSARP) for repair of an anorectal malformation (ARM). Common findings include an inadequate or shortened perineal body, as well as introital stenosis, retained vaginal septum, and remnant rectovestibular fistula. An inadequate or shortened perineal body may impact fecal continence, sexual function and recommendations regarding obstetrical mode of delivery. We describe our experience with female patients referred to our center for evaluation of their previously repaired ARM, with a specific focus on perineal body anatomy and concomitant gynecologic abnormalities. We outline our collaborative evaluation process and findings as well as subsequent repair and outcomes.

Material/methods: A single site retrospective chart review from May 2014 to May 2016 was performed. Female patients with a history of prior ARM repair who required subsequent reoperative surgical repair with perineoplasty were included. The decision for reoperation was made collaboratively after a multidisciplinary evaluation by colorectal surgery, urology, and gynecology which included examination under anesthesia (EUA) with cystoscopy, vaginoscopy, rectal examination, and electrical stimulation of anal sphincters. The type of original malformation, indication for reoperative perineoplasty, findings leading to additional procedures performed at time of perineoplasty, postoperative complications, and the length of follow up were recorded.

Results: During the study period 28 patients were referred for evaluation after primary ARM repair elsewhere and 15 patients (60%) met inclusion criteria. Thirteen patients (86.6%) originally had a rectovestibular fistula with prior PSARP and 2 patients (13.4%) originally had a cloacal malformation with prior posterior sagittal anorectovaginourethroplasty. The mean age at the time of the subsequent perineoplasty was 4.6 years (0.5–12). Patients had an inadequate perineal body requiring reoperative perineoplasty due to: anterior mislocation of the anus (n = 11, 73.3%), prior perineal wound dehiscence with perineal body breakdown (n = 2, 13.4%), acquired rectovaginal fistula (n = 1, 6.6%), and posterior mislocated introitus with invasion of the perineal body (n = 1, 6.6%). During the preoperative evaluation, additional gynecologic abnormalities were identified that required concomitant surgical intervention including: introital stenosis (n = 4, 26.6%), retained vaginal septum (n = 3, 20%) and remnant recto vestibular fistula (n = 2, 13.3%).

Conclusions: Patients with a previously repaired ARM may present with gynecologic concerns that require subsequent surgical intervention. The most common finding was an inadequate perineal body, but other findings included introital stenosis, retained vaginal septum and remnant recto vestibular fistula. Multidisciplinary evaluation to assess and identify abnormalities and coordinate timing and surgical approach is crucial to assure optimal patient outcomes.

Type of study: Case series with no comparison group. *Level of evidence:* IV.

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Anorectal malformations (ARM) occur in 1 in 5000 live births and comprise a wide spectrum of diseases involving the distal anus and rectum as well as the urinary and genital tracts [1] In females, the most common ARM is an imperforate anus with a rectovestibular fistula followed by an imperforate anus with a rectoperineal fistula and then a cloacal anomaly [2]. The vast majority of female patients with ARM

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are primarily repaired by a posterior sagittal anorectoplasty (PSARP) in the first year of life.

After a posterior sagittal anorectoplasty (PSARP), several gynecologic complications have been described such a persistent urogenital sinus, acquired vaginal atresia or stricture, urethrovaginal fistula, rectovaginal fistula and inadequate or shortened perineal body [3]. An inadequate perineal body may impact fecal continence due to anterior anal mislocation outside of the muscle complex, as well as sexual function or obstetrical mode of delivery because of the proximity between the rectum and vagina.

This study aimed to describe our experience with perineal reconstruction in patients with prior repair of an ARM – the multidisciplinary evaluation of a female patient with prior PSARP, indications for reoperative perineoplasty, concomitant gynecologic findings, the surgery or surgeries performed, and subsequent outcomes.

1. Methods

A single site retrospective chart review from May 2014 to May 2016 was performed. All female patients with previously repaired ARM referred to our center underwent a multidisciplinary evaluation. Patients that required reoperative perineoplasty after multidisciplinary evaluation met inclusion criteria. Patients with previous anorectal malformation repair not requiring perineal body reconstruction after evaluation were excluded. The type of original malformation, indication for reoperation, concomitant gynecologic abnormalities, procedure(s) performed, postoperative complications, and the length of follow up was recorded.

1.1. Diagnostic examination under anesthesia (EUA)/vaginoscopy/cystoscopy

All female patients referred to our center for evaluation after previous repair of an ARM underwent a multidisciplinary assessment including examination under anesthesia with rectal examination and electrical stimulation of the anal sphincters, vaginoscopy, and cystoscopy (vaginoscopy and cystoscopy were done if needed) by colorectal surgery, gynecology, and urology. This evaluation allowed for comprehensive anatomic assessment and collaborative discussion about indications(s) and timing of subsequent surgical intervention(s).

(Table 1). They also underwent a complete evaluation of her spine (ultrasound of the spine in the newborn period or MRI if older) and Sacral Ratio measurements in order to assess their potential for continence. All of the patients were evaluated preoperatively as well as 1 month postoperatively. The indication for perineoplasty was individualized for each patient. If we found a mislocated anus (outside of the sphincter complex) after the electrical stimulation and the patient was incontinent we decided to perform a reconstructive surgery. In patients

Table 1Diagnostic examination under anesthesia (EUA)/vaginoscopy/cystoscopy.

Diagnostic EUA/Vaginoscopy/Cystoscopy

All female patients with previously repaired ARM underwent multidisciplinary evaluation which included:

- **2.1.1** EUA anus/perineum with electrical stimulation of the anal sphincters to assess for:
- Anal mislocation in relation to the sphincter complex
- Anal stricture
- Rectal prolapse
- Perineal body
- **2.1.2** Cystoscopy (only if urologic concerns) in the assessment of:
 - Urethral and bladder anomalies, ectopic ureters
- 2.1.3 Vaginoscopy in the assessment of:
- Remnants of rectovaginal or rectovestibular fistulae
- Residual vaginal longitudinal septum
- Assessment of the vaginal introitus
- Evaluating for stricture and need for introitoplasty/dilations
- Identification of a cervix or cervices

After 1 month post redo-surgery we examine the patients in the office. If the anatomy cannot be assessed properly in the office we repeat the EUA, and vaginoscopy or cystoscopy if needed.

with good potential for bowel control we performed the surgery in order to improve their continence. In those with Tethered cord and bad sacrum (patients 3, 11 and 15) and therefore bad potential for bowel control our first aim wasn't to improve the continent but to improve their quality of life as they have rectal prolapse, anal stenosis and the enema took a long time to get out.

1.2. Perineoplasty with reconstruction of the perineal body: Surgical procedure

All patients requiring perineal body reconstruction were repaired via a posterior sagittal approach and underwent a posterior perineoplasty with reconstruction of the perineal body. The rectum was dissected, mobilized, and separated from the perineal body and posterior vaginal wall. Once separated, the rectum was placed within the sphincter complex, identified intraoperatively with the electrical stimulator. The perineal body was repaired with absorbable interrupted stitches from the fourchette to the anterior boundary of the muscle complex. Then the posterior part of the sphincter muscles is approximated to the posterior rectal wall. After restoration of the muscle complex, an anoplasty was performed with interrupted absorbable sutures. Patients with additional gynecologic abnormalities identified at the time of multidisciplinary evaluation underwent concomitant, corrective procedures. (Fig. 1a, b, c).

1.3. Postoperative management

Postoperatively, patients were kept on clears liquids for 4 to 7 days (depending on wound healing, perineal body wound length and concomitant procedures done at the time of the perineoplasty) in attempt to avoid disruption of the perineoplasty then, diet was advanced to regular. Patients were discharged to home if the perineal wound healing was deemed satisfactory. At 1 month postoperatively, we assessed the anatomic/surgical results with an examination in clinic or EUA (if we can't assess properly the postoperative results in clinic) in all of the patients undergoing a perineoplasty (with or without cystoscopy and vaginoscopy, as indicated). Long term functional follow up was done by phone at 3 months, 6 months and then yearly after the perineoplasty. Recommended long term gynecologic follow up as the patients move through puberty and beyond include pelvic ultrasound to assess mullerian structures after thelarche, pelvic examination to assess introitus prior to vaginal penetrative intercourse, and either preconcpetual or early pregnancy determination of obstetrical mode of delivery.

2. Results

Twenty-eight female patients were evaluated at our center between May 2014 and May 2016 for potential complications after primary ARM repair at another institution. 13/28 patients were excluded as they did not require perineal body repair, their findings at the time of multidisciplinary evaluation included posterior anal mislocation (5), rectal prolapse (3), or anal stricture (5). 15/28 patients met inclusion criteria, requiring repair of the perineal body as determined at the time of the multidisciplinary evaluation (Table 2). Of these 15 patients, 13 (86.6%) had undergone primary repair of a rectovestibular fistula with PSARP and 2 (13.4%) had undergone primary repair of a cloacal malformation with posterior sagittal anorectovaginourethroplasty (PSARPVUP). One cloacal patient was identified with a short common channel (<3 cm) and the other with an unknown common channel length.

The most common indication for reoperative perineoplasty was an inadequate perineal body with anterior mislocation of the anus outside of the sphincter; no sphincter muscles were identified at the anterior aspect of the anus during the electrical stimulation of the sphincters at time of EUA (n = 11, 73.3%) (Fig. 2). Less common indications were perineal wound dehiscence with perineal body breakdown occurring in the

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