Should Progressive Perineal Dilation be Considered First Line Therapy for Vaginal Agenesis?

Patricio C. Gargollo, Glenn M. Cannon, Jr., David A. Diamond, Phaedra Thomas, Vicki Burke and Marc R. Laufer*

From the Departments of Urology and Gynecology (PT, VB, MRL), Children's Hospital Boston, Harvard Medical School, Boston, Massachusetts

Purpose: In women with vaginal agenesis progressive perineal dilation provides a minimally invasive method to create a functional vagina without the attendant risks or complications of traditional surgical options. We report our 12-year experience with this technique.

Materials and Methods: Patients with vaginal agenesis treated at our institution were analyzed retrospectively and followed prospectively using case report forms and semistructured interviews. Patients diagnosed with vaginal agenesis were counseled on vaginal reconstruction options. Those electing progressive perineal dilation were instructed on the proper use of vaginal dilators by one of us (MRL) and advised to dilate 2 or 3 times daily for 20 minutes. All patients received physician, nursing and social work education and counseling. Parameters reviewed included primary diagnosis, start and end of vaginal dilation, dilation frequency, dilator size, sexual activity and whether the patient experienced pain or bleeding with dilation or sexual activity. Functional success was defined as the ability to achieve sexual intercourse, vaginal acceptance of the largest dilator without discomfort or a vaginal length of 7 cm. Univariate and multivariate analysis was performed to identify factors associated with successful neovaginal creation.

Results: From 1996 to 2008 we enrolled 69 females with vaginal agenesis in a progressive perineal dilation program. The primary diagnosis was Mayer-Rokitansky-Küster-Hauser syndrome in 64 patients. Mean age at the start of vaginal dilation was 17.5 years (range 14 to 35) Mean followup was 19 months (range 0 to 100). Four patients (5.7%) were lost to followup. In 7 of the remaining 65 patients (12%) treatment failed due to noncompliance and 50 (88%) achieved functional success at a median of 18.7 months. Patients who dilated frequently (once daily or greater) achieved a functional neovagina at a median of 4.3 ± 2.4 months. Functional success correlated positively with frequent (once daily or greater) dilation and the initiation of sexual activity. Complications were minor. Three patients reported infrequent pain and 2 reported a single episode of bleeding with dilation. A total of 18 sexually active patients reported satisfactory intercourse without dyspareunia.

Conclusions: Progressive perineal dilation for neovaginal creation is a valuable, minimally invasive therapy to create a functional vagina with a high success rate and a much lower complication rate than that in published surgical series. Given these findings, progressive perineal dilation should be offered as first line therapy in adolescents with a congenitally absent vagina.

Key Words: vagina, abnormalities, dilatation, coitus
**Vaginal agenesis** is an uncommon condition with an estimated incidence of 1/5,000 to 1/10,000 live female births. Implicated etiologies are MRKH, androgen insensitivity syndrome (including Morris syndrome) and certain intersex disorders. Regardless of cause, creating a functional neovagina in these patients remains a significant challenge and a controversial subject. While a number of techniques have been described and continue to be used, long-term outcomes and diagnosis related success rates remain indeterminate. Of the techniques used, including split-thickness skin grafts, bowel vaginoplasty, myocutaneous grafts and progressive dilation, pediatric urologists and pediatric surgeons prefer bowel vaginoplasty, as reflected in the urological literature. While short-term results are favorable, bowel vaginoplasty remains a major surgical procedure that often requires postoperative dilation and carries significant long-term complications, including mucous production, vaginal stenosis, vaginal prolapse, diversion colitis, bowel obstruction and rarely carcinoma.

The primary end point in these patients should be creating a vagina that is satisfactory for sexual intercourse and provides adequate cosmesis of the external genitalia, while minimizing short-term and long-term patient morbidity. Progressive dilation is recommended by the American College of Obstetricians and Gynecologists as the first choice in neo-vaginal creation, particularly in patients with Mayer-Rokitansky syndrome. Successful neovaginal creation by PPD obviates the need for major surgery. We reviewed our 12-year experience with this technique.

**MATERIALS AND METHODS**

Patients with vaginal agenesis treated at our institution were analyzed retrospectively and followed prospectively using case report forms and semistructured interviews. Institutional review board approval was obtained. Initial patient evaluation was performed by a single practitioner (MRL), and included vaginal examination and structured interview with a nurse specialist (PT or VB). A review of all techniques available for neovaginal creation was discussed with each patient. Patients electing PPD received spoken and written instructions on the proper technique. During the initial visit the physician instructed the patient on the correct use of dilators with mirror. Syracuse vaginal dilators (Syracuse Medical Devices, Syracuse, New York) are used in our practice. The patient is shown the vulvar anatomy and how to place the tip of the smallest dilator at the introital dimple between the anus and the urethra.

When starting the home dilation regimen, patients were advised to take a warm bath for at least 10 minutes before dilator use and then assume a semireclining position with the knees flexed and apply gentle pressure to the vaginal dimple for 20 minutes 3 times daily. A small amount of water based lubricant, such as K-Y® Jelly or Surgilube® Lubricating Jelly, could be used on the tip of the vaginal dilator as needed. Patients were seen at followup visits 4 to 8 weeks apart with the nurse and physician. Vaginal length and width were assessed by a single practitioner (MRL) in all patients. Patients were instructed to proceed to the next dilator length and width depending on progress. Emotional and psychological support from the team clinical psychologist, nurse specialist or social worker was made available as needed before, during and/or after treatment.

Parameters assessed were age, vaginal length and width at dilation start and end, total dilation time, dilation frequency, dilator size and any complications during or after dilation, such as bleeding or pain. Sexually active patients were asked whether they noticed bleeding or dyspareunia with intercourse. Functional success was defined as the ability to achieve satisfactory intercourse, vaginal acceptance of the largest dilator without discomfort or a 7 cm vaginal length. Patients who attained functional success but were not routinely sexually active were instructed to dilate on a maintenance regimen consisting of the largest dilator (Syracuse 3 or 4, measuring 15.2 × 3.2 and 15.2 × 3.5 cm) 2 or 3 times weekly.

Statistical analysis was performed using SPSS® 15.0. Statistical significance was determined with the independent sample t tests with equal variance not assumed and 1-way ANOVA with the Bonferroni post hoc test.

**RESULTS**

From 1996 to 2008 we enrolled 69 females with vaginal agenesis in a PPD program. The primary diagnosis was MRKH in 64 patients (93%), VATER in 2, androgen insensitivity syndrome in 1 and Goltz syndrome in 1. Mean ± SD age at the start of vaginal dilation was 17.48 ± 3.27 years (range 14 to 35). Mean followup was 18.89 ± 21.86 months (range 0 to 100) (table 1). Four patients (5.7%) were lost to followup. Of the remaining 65 patients treatment failed in 7 (12.1%) due to noncompliance and 50 (88%) achieved functional success at a median of 18.7 months. Eight patients (11.6%) remained in the program (table 2). Of the 50 patients who achieved functional success 29 achieved a vaginal length of 7 cm or greater.

There was no statistically significant difference in starting age at dilation and mean dilating time in patients with vs without functional success (p = 0.85 and 0.54, respectively, table 3). There was a statistically significant difference between these 2 groups in vaginal length at the start and end of PPD, and in

<table>
<thead>
<tr>
<th>Table 1</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parameter</td>
</tr>
<tr>
<td>Dilation start</td>
</tr>
<tr>
<td>Dilation end</td>
</tr>
<tr>
<td>Change</td>
</tr>
</tbody>
</table>
the change in vaginal length (table 3). There was no statistically significant difference in vaginal length at the start or end of PPD in successful patients who were vs were not sexually active. Patients who performed dilation frequently required a statistically significant shorter time to create the neovagina and achieved a greater change in vaginal length and a higher functional success rate ($p < 0.001$, figs. 1 and 2).

A total of 18 patients who were sexually active reported satisfactory intercourse without dyspareunia. They noticed that the vagina was adequate for sexual intercourse. There were no major complications in any patient. Three patients reported infrequent pain and 2 reported a single episode of bleeding with dilation.

**DISCUSSION**

Various surgical and nonsurgical methods have been proposed for neovaginal creation in patients with vaginal agenesis, including perineal dilation techniques (Frank, Ingram and Vecchietti procedures), bowel vaginoplasty (Baldwin), inlay skin flaps (McIndoe), peritoneal vaginoplasty (Davydov procedure), and local cutaneous (Williams operation) and myocutaneous flaps. Laparoscopic modifications of these techniques have also been described. All techniques have inherent advantages and disadvantages, and the eventual choice of procedure is often biased by surgeon comfort and experience with certain techniques. Ultimately the responsibility falls to the physician to present all options available for vaginal substitution along with the attendant risks and benefits. Into this must be factored significant psychological stress associated with this condition, in that these patients may not only be capable of having sexual intercourse, but also may face infertility.

Currently the American College of Obstetricians and Gynecologists recommends PPD as first line therapy for vaginal agenesis. PPD was first described in 1832 by Amussat and modified in 1938 by Frank. PPD is a patient driven technique that is easy to perform, inexpensive, associated with minimal morbidity and highly successful. It may be performed on an outpatient basis and does not require the sometimes lengthy hospitalization of conventional surgical methods. The neovagina created after PPD is histologically composed of vaginal tissue. If PPD fails, the option still exists to proceed to surgical neovaginal creation. Furthermore, a large percent of patients who undergo surgical neovaginal creation subsequently require dilation therapy, including up to 73% when skin grafts were used. Therefore, proper understanding of dilator use is beneficial regardless of the outcome.

Perineal dilation can be divided into passive and active techniques. Active PPD techniques include the Frank method, which was used in this study, in which patients actively apply pressure to the perineum. Passive techniques include the Ingram modification of the Frank PPD technique and the Vecchietti procedure. The latter involves placing an acrylic olive in the vaginal dimple, which is attached by 2 traction sutures through the potential neovaginal space and secured to an anterior abdominal wall traction apparatus. These sutures are tightened sequentially, pulling the olive deeper into the perineal space and, thus, creating the neovagina. Laparoscopic modifications of this technique have been implemented with good success. Using this technique some investigators reported a vaginal length of 10 to 12 cm in 7 or 8 days, although this was not confirmed at other centers. Furthermore, the traditional Vecchietti instrumentation is currently not Food and Drug Administration approved, and is not available in the Unites States. In the Ingram modification of the Frank procedure patients sit on a bicycle seat equipped with a dilator, using their weight to apply perineal pressure and allowing them to participate in simultaneous productive activities. The criticism of this technique is that many young women find that sitting on the bicycle seat dilator is too uncomfortable or awkward. Despite this, reports of the Ingram modification show that a neovaginal depth of 10 cm can be achieved within 4 to 6 months, and functional and anatomical success was reported in up to 92% of patients.

**Outcomes**

Surgical or nonsurgical neovaginal creation is elective and best performed when the patient is emotionally mature and can be involved in the decision making process. At some centers groups have sur-

**Table 2**

<table>
<thead>
<tr>
<th>Parameter</th>
<th>No. Success (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vaginal length (cm):</td>
<td></td>
</tr>
<tr>
<td>Greater than 5</td>
<td>16 (28.1)</td>
</tr>
<tr>
<td>Greater than 7</td>
<td>16 (28.1)</td>
</tr>
<tr>
<td>Sexually active</td>
<td>18 (31.6)</td>
</tr>
</tbody>
</table>

**Table 3**

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Mean Failure</th>
<th>Mean Functional Success</th>
<th>$p$ Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at PPD start (yrs)</td>
<td>18.0</td>
<td>17.6</td>
<td>0.85</td>
</tr>
<tr>
<td>Dilatation time (mos)</td>
<td>19.0</td>
<td>11.0</td>
<td>0.54</td>
</tr>
<tr>
<td>Vaginal length (cm):</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dilatation start</td>
<td>0.1</td>
<td>0.9</td>
<td>0.03</td>
</tr>
<tr>
<td>Dilatation end</td>
<td>2.6</td>
<td>6.7</td>
<td>0.001</td>
</tr>
<tr>
<td>Change</td>
<td>2.4</td>
<td>5.7</td>
<td>0.002</td>
</tr>
</tbody>
</table>
gically created a neovagina in patients as young as 1 year old. While the argument was made that this procedure is technically easier in younger children, others advocate that reconstruction in adolescence improves the outcome and carries a lower complication rate. The motivation for reconstruction is often based on the desire for women to appear and feel normal, not only in terms of sexual anatomy, but also in terms of sexual activity and experiences. A major limitation when evaluating outcomes after neovaginal creation regardless of technique is the lack of a standardized definition of success. The outcome of these various procedures is usually gauged in terms of anatomical and functional success. Anatomical success refers to an adequately sized vagina and functional success refers to satisfactory intercourse. Studies, including ours, define success as attaining a vaginal length of at least 7 cm. Others define anatomical success as a 5 to 10 cm vagina. Reference values for normal vaginal length were previously described to be 7 to 13 cm (mean 9.25 ± 1.56 cm). Therefore, although 7 cm is at the lower end of normal, it may be an appropriate value. Correlations exist between vaginal depth and satisfactory outcomes. However, functional success of the operation as perceived by the patient does not seem to be proportional to the length of the neovagina.

The time required for neovaginal creation by PPD varies. Most groups report an interval of 4 to 6 months. We noted a slightly higher overall time with patients achieving anatomical or functional success with dilation at a mean of 11 months and failed PPD in those who dilated a mean of 19 months (p = 0.54, table 3). Patients in whom PPD failed were more likely to perform dilation infrequently and, hence, the overall increased dilation time. Grouping our patients by functional and anatomical success revealed that anatomical success was possible in a relatively short interval (median 7 months, mean 11) (table 4). The explanation for increased overall time for successful neovaginal creation after PPD relates to 1 patient in the functional success group who intermittently dilated for a total of 8.3 months. Patients who performed frequent dilation had statistically significant shorter neovaginal creation time (A) and total vaginal length change (B) (each p <0.001).

Table 4

<table>
<thead>
<tr>
<th></th>
<th>Mean ± SD Mos PPD (range)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lost to followup</td>
<td>9.0 ± 7.0 (1–14)</td>
</tr>
<tr>
<td>Failure</td>
<td>27.9 ± 29.9 (2–81)</td>
</tr>
<tr>
<td>Success:</td>
<td></td>
</tr>
<tr>
<td>Vaginal length greater than 5 cm</td>
<td>11.6 ± 12.5 (1–43)</td>
</tr>
<tr>
<td>Vaginal length greater than 7 cm</td>
<td>14.4 ± 14.2 (1–50)</td>
</tr>
<tr>
<td>Sexually active</td>
<td>31.5 ± 29.0 (1–100)</td>
</tr>
<tr>
<td>Still in program</td>
<td>4.6 ± 4.3 (0–11)</td>
</tr>
<tr>
<td>Overall</td>
<td>18.5 ± 21.9 (0–100)</td>
</tr>
</tbody>
</table>

Figure 2. Patients who performed frequent dilation, defined as 4—frequent (at least once daily), 3—moderately frequent (every other day, or 2 or 3 times weekly), 2—in frequent (1 to less than 1 time weekly) and 1—rarely, had statistically significantly higher functional success rate (p <0.001).
years, which increased the mean in this group to 31.5 months (table 4). If only patients with anatomical success are considered, time to successful neovagina creation was shorter at a median of 7 months. What seems clear is that the more frequently the patient dilates, the shorter period is required to create a neovagina and the greater the change in neovagina length with time (fig. 1). In our cohort patients who dilated at least once daily created a functional neovagina at a mean of 4.3 ± 2.4 months (fig. 1, A). In contrast, patients who dilated once or less than once weekly created a functional neovagina at a mean of 41 ± 23.4 months (p < 0.001).

Correlation analysis by other groups showed that patients with a longer starting vaginal length have a greater trend toward success but this was not statistically significant. It was our experience that mean beginning vaginal length was marginally significantly larger in patients with functional success vs those with failure (0.85 vs 0.14 cm, p = 0.03, table 3). However, median vaginal length in our successful patients was 0.5 cm, which refutes the claim by some groups that successful PPD can only be achieved in patients with a longer starting vaginal length. Others stated that PPD will fail in patients with only a vaginal dimple. This was not our experience. Roberts et al found that patients younger than 18 years at the start of PPD had a statistically significant dilation failure rate. We did not note this in our patients, although our entire cohort was slightly older (mean age 17.5 years).

Some criticize that the Frank PPD method is unsuitable for neovaginal creation, given that patients are unable to generate enough perineal pressure for the necessary protracted time daily and may become bored with manually applying pressure for an adequate time. This was not our experience.

How do the outcomes of bowel and split-thickness vaginoplasty compare to the PPD outcome? Bowel vaginoplasty was first reported in 1904 by Baldwin. Later reports established this as a viable alternative to neovaginal construction with good results. The advantage of bowel vaginoplasty is a well lubricated segment that grows with the patient and provides a cosmetically acceptable result in most cases. The concept that patients who undergo bowel neovaginal reconstruction do not need dilation postoperatively is not supported in the literature. After rectosigmoid vaginoplasty patients often require extended dilator use. Recently Kim et al reported that 8.3% of their patients required intermittent vaginal stent use for less than 1 year and 6% required it for longer than 1 year. Hensle and Reiley reported that 10% of their patients required chronic home dilation after bowel vaginoplasty. In a subsequent study Hensle et al noted that 5.6% of their patients required routine dilation and estrogen suppositories. O’Connor et al reported that 20% of their patients required routine home dilation after bowel vaginoplasty. and Khen-Dunlop et al observed the need for postoperative dilation in 26%.

Extended dilator use in the form of a vaginal mold is also required in patients who undergo split-thickness skin graft neovaginal creation (McIndoe technique). After initial use of the vaginal mold immediately after surgery for up to 2 weeks patients are instructed to wear the mold continuously for 3 to 6 months. Given that there is a relatively high rate of graft contracture and neovaginal stenosis when using this technique, a fair number of these patients need lifelong dilation to keep the neovagina patent. Klingele et al reported that 73% of their patients required the vaginal mold for greater than 6 months and 40% commented that wearing it was a nuisance.

Complications
Complications of any surgical techniques for neovaginal creation are vesicovaginal or rectovaginal fistula, rectal or bladder perforation, vaginal prolapse, graft contracture and keloid scar formation in cases with skin grafting. In a study of the outcomes of McIndoe vaginoplasty 28% of patients thought that the skin graft harvest site scar was disfiguring. Another study of 71 patients who underwent McIndoe vaginoplasty revealed an overall 14% complication rate. Recent reports of bowel neovaginal outcomes also had a significant complication rate of 16% to 26% (table 5). Hensle et al reported long-term results (mean followup 8.8 years) in a cohort of 57 patients who underwent bowel vaginoplasty using various bowel segments. The overall complication rate was 19.3%, including vaginal stenosis requiring surgery in 14% and prolapse requiring surgery in 3.5% (table 5). This is in contrast to the negligible and minor complication rate in PPD series, including our series.

Excessive mucus was also reported with bowel vaginoplasty. The impression in the literature is that the quantity of mucus decreases with time. However, in a cohort of patients with bowel vaginoplasty followed in the long term Kim et al noted that 8.3% reported malodorous and excessive mucous production. Hensle et al observed excessive vaginal mucous requiring home douching in 94% of patients and sanitary pad use in 55% after bowel vaginoplasty.

Some groups reported that PPD is associated with neovaginal prolapse and because of this, they advocate that this technique not be used. It is neither our experience nor that in published studies in more than 300 patients that this is a significant problem.
Table 5

<table>
<thead>
<tr>
<th>References</th>
<th>Technique</th>
<th>Tissue</th>
<th>No. Pts</th>
<th>No. Complication (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Khen-Dunlop et al²</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid</td>
<td>23</td>
<td>11 Total (43), 1 compartment syndrome (4.3), 6 vaginal stenosis, requiring chronic dilation (26), 2 vaginal prolapse (8.7)</td>
</tr>
<tr>
<td>Hensle et al¹</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid, ileum, cecum</td>
<td>57</td>
<td>11 Total (19.3), 8 vaginal stenosis requiring repeat surgery (14), 2 vaginal prolapse requiring surgery (3.5), 1 diversion colitis (1.7)</td>
</tr>
<tr>
<td>Dargent D: 2004; 32: 1023</td>
<td>Laparoscopic Davydov</td>
<td>Peritoneum</td>
<td>28</td>
<td>1 Vesicovaginal fistula (3.6), 4 vaginal stenosis requiring repeat surgery (14.3), 2 Intraop ureter + bladder injury (7.1)</td>
</tr>
<tr>
<td>O’Connor et al¹⁴</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid, ileum</td>
<td>10</td>
<td>12 Total (20)</td>
</tr>
<tr>
<td>Klingele et al⁹</td>
<td>McIndoe</td>
<td>Split-thickness skin</td>
<td>71</td>
<td>10 Total (14.1), 2 vaginal prolapse requiring surgery (2.8), 1 rectovaginal fistula (1.4), 1 vesicovaginal fistula (1.4), 1 vaginal stenosis requiring repeat surgery (1.4)</td>
</tr>
<tr>
<td>Kim et al⁷</td>
<td>Bowel vaginoplasty</td>
<td>Rectosigmoid</td>
<td>36</td>
<td>8 Total (22.2), 2 vaginal stenosis requiring repeat surgery (5.5), 2 urethral stenosis requiring surgery (5.5), 3 vaginal prolapse (8.3), rectovaginal fistula (2.7), 3 excessive mucus (8.3), 3 malodorous (8.3)</td>
</tr>
<tr>
<td>Seccia et al²⁹</td>
<td>McIndoe</td>
<td>Split-thickness skin</td>
<td>32</td>
<td>3 Partial graft take (9.3), 3 vaginal stenosis (9.3), 1 donor site keloid scar (3.1)</td>
</tr>
<tr>
<td>Freitas Filho et al²⁷</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid</td>
<td>10</td>
<td>2 Total (20)</td>
</tr>
<tr>
<td>Syed et al²⁶</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid</td>
<td>18</td>
<td>3 Diversion colitis (16.7)</td>
</tr>
<tr>
<td>Hensle and Reiley²²</td>
<td>Bowel vaginoplasty</td>
<td>Sigmoid, ileum, cecum</td>
<td>31</td>
<td>8 Total (26), 3 vaginal stenosis requiring chronic dilation (9.7), 3 vaginal stenosis requiring repeat surgery (9.7), 4 excessive mucus (15.4), 2 vaginal prolapse (6.5)</td>
</tr>
</tbody>
</table>

after PPD.¹³,²⁰,²⁸ The prolapse rate after bowel vaginoplasty is 3% to 8% (table 5).

**Malignancy**

Biopsies from the neovaginal wall of patients with McIndoe vaginoplasty revealed pseudomucinous metaplasia in some.²⁹ Furthermore, carcinoma was reported in bowel and skin graft neovaginas.³⁰ To our knowledge no malignancies were reported in neovaginas created with PPD and some groups argue that malignancy is related to transplanted tissue. Although implications for the bowel neovagina are not clear, bowel segments isolated from the fecal stream undergo significant mucosal changes.³¹

**Multidisciplinary Team**

It is imperative that patients who undergo PPD be instructed on the proper use of dilators and treated by a multidisciplinary team. Given the stigma associated with genital anomalies and the potential drawbacks in vaginal reconstruction approaches, specialist emotional and psychological support must be integrated along with medical care. This support is particularly important since patients with vaginal agenesis perceive that they have genital anomalies and often may have sexual anxiety and depression.¹⁷ This coupled with the reproductive repercussions of androgen insensitivity syndrome and MRKH makes access to mental health providers imperative in these patients. We and others²⁰ believe that treatment by a multidisciplinary team is imperative. We have established a diverse group of nursing, medical and psychological practitioners for our patients. Furthermore, patients are encouraged to participate in support groups and yearly conferences (http://www.youngwomenshealth.org/vaginalagensesis.html and http://www.youngwomenshealth.org/mrkh_conf.html).

**Psychosexual Adjustment and Function**

PPD is not without drawbacks. Some groups stated that the main disadvantage is that compliance is low because of the time required to create the neovagina.³² In an interview of 6 patients Boyle et al reported that some women thought that the regimen of PPD was distasteful.³³ Jasonni et al stated that the psychological outlook of an individual appears to have an important role in performing self-dilation.²⁸ In a small pilot study of 10 patients undergoing PPD Liao et al used the Multi-Dimensional Sexuality Questionnaire and found that these women had lower scores on sexual esteem, sexual assertiveness and sexual satisfaction, and higher scores on sexual anxiety, sexual depression and fear of sexual relationships compared with those of the standardized sample.³⁴ Although we did not use a standard questionnaire to query our patients on psychosexual adjustment, sexual desire or function, patients were receptive to the technique and for the most part
completed the regimen successfully and without difficulty. Three reasons for discontinuing treatment were cited by the 7 patients in whom therapy failed. One reason was that they did not have the privacy necessary for the dilation regimen, which was reported by 5 of the 7 patients with failed treatment. Four of these patients attended college and had roommates, and another shared a room with a sibling. The remaining patient stated that she was not interested in pursuing neovaginal creation. In the final patient PPD failed after a fairly extensive case of perineal lichen sclerosus, which made dilation too painful.

It is difficult to assess the psychological and emotional ramifications of müllerian tract structural abnormalities in patients. Furthermore, to our knowledge the impact of surgical correction on individual psychosocial relationships and psychosexual function is unknown. Mobus et al surveyed 44 women after operative correction for vaginal agenesis and found that 61% had increased self-esteem. They also found that women with a functionally unsatisfactory result had worse scores on self-esteem, attractiveness, self-confidence and body accentuation-sensibility scales. They concluded that a successful operation leads to the reestablishment of female self-confidence, self-esteem and sexual life.

Followup Care

Women who have created a neovagina by PPD require routine gynecological care with annual pelvic examination. They should be made aware that they are at the same risk for sexually transmitted disease. Furthermore, a vaginal speculum examination should be performed in all patients with a bowel or skin graft neovagina to evaluate malignancy, colitis or ulceration. To our knowledge no data exist on the need or lack of need for routine Papanicolaou testing in women with a neovagina.

While functional and anatomical success has been used as an outcome measure in these patients, the nuances of sexual and psychosexual function are much more complex than simple anatomical assessment. This remains a limitation of our study as well as of most studies of reconstructive techniques in patients with vaginal agenesis. Thus, future studies must emphasize objective outcomes in prospective fashion using validated questionnaires for patients and partners, in addition to clinical examinations and patient interviews.

CONCLUSIONS

Using dilators to manage congenital vaginal agenesis is appropriate and successful in most patients. A high success rate can be expected with PPD in mature, motivated patients who wish to avoid surgery. Briefly, PPD is a patient driven technique that is easy to perform, inexpensive, associated with minimal morbidity and highly successful. Failed PPD therapy for neovaginal creation does not preclude subsequent surgical reconstruction. Because of these factors, PPD should be considered as first line therapy for congenital vaginal agenesis.

REFERENCES

PROGRESSIVE PERINEAL DILATION FOR VAGINAL AGENESIS


EDITORIAL COMMENTS

These authors make a compelling case for PPD to create a neovagina in postpubertal girls born with vaginal agenesis. The key success factor is a combination of motivated patients and a dedicated multidisciplinary team of health care providers. They achieved 88% functional success at a median of 18.7 months of followup. The more frequently the patient self-dilated, the shorter the time required to create a neovagina. Their results contradict those of Liao et al, who concluded that vaginal dilation can have a negative emotional impact (reference 34 in article).

The authors criticize bowel vaginoplasty based on select references in the literature. I have no experience with PPD but I defend vaginal construction using a bowel segment since my patients have much longer followup than those in the cited reports. Since my first series in 1987 and my subsequent report in 1998, I now have 31 patients with a bowel vagina, of whom most are now in the late fourth and fifth decades of life. None has complained of any significant problems with mucus. In fact, the 2 patients with conversion from McIndoe skin vaginoplasty to a sigmoid neovagina are the most vocal advocates of the procedure. My impression from my patients is that the bowel vagina is truly low maintenance. In my first report an inverted V-shaped perineal skin flap was raised and incorporated in the construction of the posterior wall of the bowel vagina to avoid a circumferential suture line. The only 2 patients in whom introital stenosis developed were those in whom I omitted using a perineal flap.

MRKH is now being diagnosed at an earlier age. My last 3 patients with MRKH were toddlers. All concerned, including parents, pediatricians, psychologists and myself, favored early vaginoplasty to enable the child to grow with almost normal anatomy, rather than with the awareness of abnormal genital anatomy. One may argue that the child did not participate in the decision but such is the nature of reconstructive pediatric surgery for most congenital anomalies.

In my opinion the psychological benefits of early reconstructive surgery outweigh the disadvantages of possible revision at a later date. Finally, the treatment choice must be individualized based on patient age, patient or parent preference and motivation, cultural/religious factors and surgeon experience.

Moneer K. Hanna
Department of Urology
Presbyterian New York Weill-Cornell Medical Center
Great Neck, New York

REFERENCES
The authors asked the question, “Should progressive perineal dilation be considered first line therapy in cases of vaginal agenesis?” Their series encompasses a 12-year retrospective review of patients with vaginal agenesis, the majority of whom are females with müllerian failure (MRKH syndrome). Progressive perineal dilation or PPD was the treatment of choice. In 7 (12%) of the 65 patients PPD failed secondary to noncompliance and 8 patients remain in the program.

Total success rate was 88% with 50 of the 65 patients achieving functional success during a mean of 18.7 months. They define functional success as attaining a vaginal length of at least 7 cm. However, only 29 of the 50 patients achieved a vaginal length of 7 cm or greater, which is 58% of the patient population. A total of 18 individuals were sexually active and another 16 achieved a vaginal length of greater than 7 cm (table 2 in article), which is 68% of the total population and not 88% as reported.

PPD is a valuable tool for dealing with vaginal agenesis secondary to MRKH. It works best in individuals who are highly motivated and committed to the procedure (reference 13 in article). It also works best in those who have some definable vaginal length to start with and, in our experience, a starting vaginal length of 3 cm or greater offers a greater chance of success (references 13 and 22 in article). PPD requires a good deal of commitment and a long time (18.7-month average in this series) to achieve success. It must be part of the discussion with patients who present with müllerian failure. Other treatment options include vaginal substitution surgery (colovaginoplasty), which has its drawbacks but has been an effective alternative for many patients with this syndrome (reference 1 in article), and newer alternatives such as autologous buccal mucosa vaginal replacement.¹

PPD is best performed by a well organized, highly effective, multispecialty team to supply not only the expertise, but also the ancillary support necessary to achieve this kind of success. The answer to the authors’ question, “Should progressive perineal dilation be considered first line therapy in cases of vaginal agenesis?” should be maybe. There are individuals for whom PPD will not be effective and they should be counseled on other forms of therapy before going through the long and arduous task of prolonged perineal discomfort and frustration.

Terry W. Hensle
Department of Urology
Children’s Hospital of New York
New York, New York

REFERENCE


REPLY BY AUTHORS

Most pediatric urology reconstructive procedures for congenital anomalies are currently performed without patient assent. However, these repairs serve a clinical purpose. Unfortunately the clinical indication for creating bowel neovaginas in toddlers is not provided. Patients with MRKH have normal appearing external genitalia and the diagnosis is almost always delayed until the patient presents with amenorrhea. Therefore, the assertion that these patients will “grow...with the awareness of abnormal genital anatomy” is unfounded. Furthermore, studies suggest that bowel segments isolated from the fecal stream may have a higher rate of mucosal atypia and perhaps carcinoma (reference 31 in article). Creating a bowel neovagina in a toddler would create a defunctionalized loop of diverted bowel for a much longer time than in an older patient. Thus, it is not medically necessary to create a neovagina until the child can participate in the decision.

This is clearly an area of heated controversy and to our knowledge there are no data regarding the ideal timing for vaginal reconstruction. However, there are data regarding the complication rate of PPD for neovagina creation vs that of other methods. Tillem et al reported an 11% reoperative rate which is in line with complication rates for bowel neovaginas published in the literature (reference 2 in comment). Comparing contemporary series of PPD, it is clear that PPD is less invasive, less morbid and more successful for neovagina creation in the appropriate patient. We agree that key factors in our success were a combination of motivated patients and a dedicated multidisciplinary team of health care providers.
We clearly define functional success as the ability to have satisfactory intercourse, vaginal acceptance of the largest dilator (15.2 × 3.5 cm) without discomfort or a vaginal length of 7 cm. Of our patients 18 were sexually active, 16 achieved a vaginal length of greater than 7 cm and 16 were able to tolerate the largest sized dilator (18 + 16 + 16 = 50, (50/57) × 100 = 87.7%). The denominator in the percentage calculation is 57 and not 65 because 8 patients were still in the program. The 29 patients who achieved a vaginal length of 7 cm or greater includes sexually active patients.

The median starting vaginal length in our successful cases was 0.5 cm, which refutes the claim that successful PPD can only be achieved in patients with longer starting vaginal lengths. Although we agree that PPD requires patient commitment, it does not take a “long time.” The time required for neovagina creation by PPD varies with most studies reporting 4 to 6 months (reference 20 in article). We noted slightly longer mean times of 11 months in patients who achieved anatomical or functional success and 19 months in those in whom PPD failed (p = 0.54, table 3 in article). Patients in whom PPD failed were more likely to engage in infrequent dilation and, hence, the overall increased dilation time.

A breakdown of our patients by functional and anatomical success (table 4 in article) reveals that anatomical success is possible in a relatively short time (median 7 months, mean 11). One patient in the functional success group intermittently dilated for 8.3 years, raising the mean time for successful neovagina creation in this group of patients. If only the patients with anatomical success are considered, the time to successful neovagina creation is shorter (median 7 months). Clearly the more frequently the patient is able to dilate the less time it will take to create a neovagina (fig. 1 in article) and the greater the change in neovagina length with time (fig. 2 in article). Those patients who dilated at least once a day were able to create a functional neovagina at a mean of 4.3 ± 2.4 months.

We agree that the time required for PPD needs to be discussed with parents and patients, as should all benefits and complications of the various techniques for neovagina creation. We also agree that if patients are not motivated they will likely fail. Therefore, we advocate a multidisciplinary approach with followup visits every 1 to 2 months. Failure is rarely due to pain, discomfort or patient unreliability with treatment, and usually relates to privacy issues (roommates in college etc).