GYNECOLOGY

Surgery is not superior to dilation for the management of vaginal agenesis in Mayer-Rokitansky-Küster-Hauser syndrome: a multicenter comparative observational study in 131 patients

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BACKGROUND: Vaginal agenesis in Mayer-Rokitansky-Küster-Hauser syndrome can be managed either by various surgeries or dilation. The choice still depends on surgeons’ preferences rather than on quality comparative studies and validated protocols.

OBJECTIVE: We sought to compare dilation and surgical management of vaginal agenesis in Mayer-Rokitansky-Küster-Hauser syndrome, in terms of quality of life, anatomical results, and complications in a large multicenter population.

STUDY DESIGN: Our multicenter study included 131 patients >18 years, at least 1 year after completing vaginal agenesis management. All had an independent gynecological evaluation including a standardized pelvic exam, and completed the World Health Organization Quality of Life instrument (general quality of life) as well as the Female Sexual Function Index and Female Sexual Distress Scale-Revised (sexual quality of life) scales. Groups were: surgery (N = 84), dilation therapy (N = 26), and intercourse (N = 20). One patient was secondarily excluded because of incomplete surgical data. For statistics, data were compared using analysis of variance, Student, Kruskal-Wallis, Wilcoxon, and Student exact test.

RESULTS: Mean age was 26.5 ± 5.5 years at inclusion. In all groups, World Health Organization Quality of Life scores were not different between patients and the general population except for lower psychosocial health and social relationship scores (which were not different between groups). Global Female Sexual Function Index scores were significantly lower in the surgery and dilation therapy groups (median 26 range [2.8–34.8] and 24.7 [2.6–34.4], respectively) than the intercourse group (30.2 [7.8–34.8], P = .044), which had a higher score only in the satisfaction dimension (P = .004). However, the scores in the other dimensions of Female Sexual Function Index were not different between groups. The Female Sexual Distress Scale-Revised median scores were, respectively, 17 [0–52], 20 [0–47], and 10 [10–40] in the surgery, dilation therapy, and intercourse groups (P = .38), with sexual distress in 71% of patients. Median vaginal depth was shorter in dilation therapy group (9.6 cm [5.5–12]) compared to surgery group (11 cm [6–15]) and intercourse group (11 cm [6–12.5]) (P = .039), but remained within normal ranges. One bias in the surgery group was the high number of sigmoid vaginoplasties (57/84, 68%), but no differences were observed between surgeries. Only 4 patients achieved vaginas <6.5 cm. Delay between management and first intercourse was 6 months (not significant). Seventy patients (53%) had dyspareunia (not significant), and 17 patients all from the surgery group had an abnormal pelvic exam. In the surgery group, 34 patients (40.5%) had complications, requiring 20 secondary surgeries in 17 patients, and 35 (42%) needed postoperative dilation. In the dilation therapy group, 13 (50%) needed maintenance dilation.

CONCLUSION: Surgery is not superior to therapeutic or intercourse dilation, bears complications, and should therefore be only a second-line treatment. Psychological counseling is mandatory at diagnosis and during therapeutic management.

Key words: Female Sexual Distress Scale-Revised, Female Sexual Function Index, Mayer-Rokitansky-Küster-Hauser syndrome, multicenter study, quality-of-life studies, sexual distress, vaginal aplasia, vaginal dilation, vaginoplasty, World Health Organization Quality of Life instrument.

Introduction

Mayer-Rokitansky-Küster-Hauser (MRKH) syndrome is a rare disease, affecting 1/4500 of 46,XX women. The major issue in these patients is vaginal agenesis (VA), which affects sexuality and alters quality of life (QOL).

Classic VA management is to create a cavity that allows satisfying painless penetrative intercourse. Vaginoplasty can be achieved either surgically or by progressive dilation of the vaginal dimple. Most surgical or nonsurgical techniques are reported to provide good anatomical and functional results of at least 70%.

Most recently, there has been a trend toward first-line dilation therapy, supported by experienced teams and national recommendations, but in many countries, surgery is still performed soon after diagnosis despite the high morbidity of surgical vaginoplasty.

However, there is no evidence of the superiority of one technique over another, and thus the choice depends on surgeons’ preferences. Most studies report one surgical technique, sometimes compared to dilation therapy and/or to a control population, and hold methodological flaws. Patients with VA and disorders of sexual development such as androgen insensitivity syndrome (AIS) are frequently included. Success is often reported as possible coitus. Satisfaction and QOL were only recently...
evaluated with standardized validated questionnaires. Only reviews, metaanalysis, or small series actually compared several different techniques, and the correlation between vaginal length and satisfaction has not been studied yet.

We conducted a national multicenter cross-sectional study to assess the general and sexual health status of women with MRKH syndrome who had a vaginoplasty, and compare the results and complications of the different surgical and nonsurgical techniques, in order to identify the optimal management of VA in MRKH syndrome patients.

**Materials and Methods**

**Patients**

Patients included were exclusively MRKH syndrome patients age ≥18 years, whose VA management started ≥1995, for at least 1 year. They had to be French-speaking, with social security coverage active for at least 1 year. Patients with severe chronic disease or illiteracy were excluded.

Recruitment was conducted by the National Reference Center for Rare Gynecological Diseases from October 2012 through April 2015. Physicians experienced in MRKH syndrome from 16 centers agreed to contact their patients who met inclusion criteria. A clinical research associate then called patients by telephone. Some patients contacted us directly after receiving information by a national MRKH syndrome peer support group.

**Methods**

All patients had a complete medical evaluation by a single experienced independent gynecologist (M.B.).

- Medical history included diagnosis and announcement circumstances, process of VA management (surgical or nonsurgical technique of vaginoplasty, complications, maintenance dilation), gynecological history (age at first intercourse, dyspareunia, medical follow-up), and counseling.
- A standardized pelvic examination was performed with patient consent, inserting 2 digits, then using dilators (Amielle, Owen-Mumford Ltd) of increasing diameter (2–3.5 cm). Length and width of vagina were defined by painless maximal insertion of the dilator.
- Validated QOL questionnaires were orally explained, then filled in by patients at the end of the medical evaluation.

The main evaluation criterion of the study was the global QOL assessed by the World Health Organization Quality of Life instrument (WHOQOL-BREF) questionnaire.

Secondary endpoints included anatomical characteristics of vagina and assessment of sexual QOL. Vagina was considered to be normal if ≥3-cm width and 9-cm length, and within normal ranges when >6.5 cm. Vagina was considered abnormal if pelvic exam identified stenosis, trigger, or if vagina was <6.5 cm. Sexual QOL was evaluated by the Female Sexual Function Index (FSFI) and the Female Sexual Distress Scale-Revised (FSDS-R) questionnaires.

Patients were compared according to the type of VA management: surgery, dilation therapy, or dilation by intercourse without medical management.

**Statistics**

All statistical analyses were undertaken using R 2.11.1 software (https://cran.r-project.org/). Statistical tests were 2-sided and P values <.05 were considered statistically significant. Characteristics of patients and details of therapeutic course were described overall, and according to type of VA management. Mean ± SD or median [range] was reported for quantitative variables and frequencies (%) for qualitative variables. The management technique groups were compared using $\chi^2$ test (or Fisher test if appropriate) for qualitative variables and analysis of variance (or nonparametric Kruskal-Wallis test) for quantitative variables. When a significant overall difference was found, the t test or Wilcoxon test was applied to each pair of groups, and corresponding P values were adjusted using Benjamini-Hochberg method to take into account the multiple comparisons. Same methods were used for the comparison of types of surgery. Mean QOL scores were compared to theoretical mean score on French general population using t test (overall and separately on each age group). Scores on the 2 separate items were compared to theoretical percentages on French general population using $\chi^2$ test for given probabilities data.

**Ethics**

All patients signed an informed consent. The protocol was approved by the ethics committee Comité pour la Protection des Personnes Ile de France VI and by the French Agency for Security of Health Products in 2012. The study is registered at ClinicalTrials.gov under number NCT01911884.
Results
Among 397 eligible patients, 138 were included and 131 finally analyzed. One patient was secondarily excluded because the type of surgery she had could not be identified (Figure 1).

Patients’ characteristics are shown in Table 1.

Therapeutic course
Mean age at inclusion was significantly lower in the dilation group: 23.8 ± 4.7 years vs 27.1 ± 5.2 years and 27.8 ± 6.6 years in the surgery and intercourse groups, respectively (P = .01). Age at management was not significantly different: 18.6 ± 2.4 years vs 17.8 ± 3.4 years in the surgery and dilation groups.

Dilation therapy
Forty patients had first-line dilation therapy, once a day, with a median frequency of 5.5 [1–14] times/wk, a median of 6 months [0–44], and 26 (65%) of them completed dilation without requiring surgery. Half of them required maintenance dilation (median frequency of once a week [1–7]). Seven patients (27%) had mild complications (pain, bleeding, urinary or vaginal infections).

In all, 13/40 patients (33%) switched to surgery, 10.7 months [0.9–91.3] after ending dilation therapy, because of pain, failure, or refusal.

Surgery
A total of 84 patients underwent surgery, including the 13 who failed dilation. Sigmoid vaginoplasty was performed in 57/84 (68%) patients, and the others had various surgeries (Figure 2). Surgery was performed in all centers: 2 major centers managed 52 patients, while the other centers managed between 1–4 patients.

In all, 35 (42%) patients wore a postoperative mold for 3 months [0.2–30], and 52 (62%) needed maintenance dilation. A total of 34 patients (40.5%) had complications (Table 2), requiring 1–4 additional surgeries in 17. Abnormal pelvic exam was found in 17/84 patients (20%), and 49 (58%) had dyspareunia.

Complication rates were not different among the different techniques, except for dyspareunia, which was significantly more frequent after sigmoid vaginoplasty (70%, P = .014).

Intercourse
Twenty (15%) patients expanded their vagina with intercourse only, sometimes before MRKH syndrome diagnosis.

Global QOL
Only the global WHOQOL-BREF score (item 1) was not different between MRKH syndrome patients and the general population (P = .13), and higher than in chronic disease. On the contrary, satisfaction toward health score (item 2) was significantly lower than in the general population (P < .01), as were the psychological and the social dimensions scores (Table 3).

There were no significant differences in the general QOL depending of the type of management (Tables 4 and 5). Scores were slightly lower in a subset of 4 patients with vaginas <6.5 cm, without reaching significance (Table 6).

Anatomical results
Median vaginal length was 10.2 cm [5.5–15]. Vaginal length was <9 cm but within normal ranges in 26 patients. Only 4 patients had a vagina <6.5 cm.

Vaginal length was significantly shorter after dilation therapy (9.3 cm [5.5–12]) vs 11 cm [6–15] after surgery and 11 cm [6–12.5] after intercourse (P = .039), without difference between the surgery and intercourse groups. All surgical techniques provided normal-range vaginas, but Davidov and Dupuytren techniques provided shorter vaginas (Table 7).

Median vaginal width was 3 cm [2–4] in all patients, with no differences among groups.

Pelvic exam was abnormal in 17 patients, all in the surgery group (20%),
including introital stenosis, vaginal shrinking, or trigger. After sigmoid vaginoplasty, the graft was visible at the introit in 23 patients (40%).

**TABLE 1**

<table>
<thead>
<tr>
<th>Patients characteristics</th>
<th>At inclusion</th>
<th>At diagnosis</th>
<th>At puberty</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, y</td>
<td>26.5 ± 5.5</td>
<td>16.5 ± 2.9</td>
<td>11.3 ± 1.5</td>
</tr>
</tbody>
</table>

**Diagnostic circumstances**

- Primary amenorrhea: 111 (84.7%)
- Other: 20 (15.3%) Including dyspareunia, apareunia, chronic pain, incidental, systematic examination

**Family history**

- MRKH: 10 (7.8%): 7 Sisters, 3 cousins, 1 aunt
- MRKH-associated malformations: 11 (8.6%)

**Associated malformations**

- Total: 73 (57.7%)
  - Renal: 35 (48%) Including 18 solitary kidney
  - Skeletal: 50 (68.5%) Including 46 scoliosis
  - ENT: 10 (13.7%)
  - Cardiac: 4 (5.5%)
  - Other: 18 (25.3%)

**Counseling**

- At diagnosis: 26 (19.9%)
- Follow-up: 54 (41.2%) (Mean no. of sessions: 3)

**Gynecological follow-up**

- 57 (43.9%)

**FIGURE 2**

Surgical techniques for vaginoplasties

Sigmoid vaginoplasty was main technique, but number of other techniques reflect disparity of management throughout country.


**Sexual QOL**

Of patients, 95% were sexually active, starting at a mean age of 18 ± 2.6 years, 2.2 ± 3.7 years after diagnosis, 0.6 ± 3.1 years after starting vaginal management, with no significant differences between the surgery and the dilation therapy groups. Median time to achieve comfortable intercourse was 10 months [1–18] in the intercourse group (data available only for 10 patients).

Three patients did not answer the FSFI questionnaire. Global FSFI scores in MRKH syndrome patients were not different to the general population. There were no significant differences in the sexual QOL between the dilation therapy and the surgery groups, neither between the surgical subgroups. The intercourse group had a significantly higher FSFI score of 30.2 [7.8–34.8] (P = .044), related to a higher score in the satisfaction dimension (Table 8).

FSFI scores were significantly lower when the vagina was < 6.5 cm (19.7 [2.6–34.8] vs 26.1 [7.8–22.3], P = .04). Of the patients with normal-sized vaginas, 48% had FSFI scores < 26.

Sexual distress (FSDS-R score ≥ 11) was very high, affecting 70% of all MRKH syndrome patients, irrespective of the type of management (72.3% in the surgery group, 76.9% in the dilation therapy group, and 50% in the intercourse group, P = .10) (Table 9). FSDS-R scores were higher for patients with vaginal length < 6.5 cm (30.5 [20–49] vs 17 [0–52]), but insignificantly (P = .11).

Discordance between FSFI and FSDS-R scores occurred in 38% of the patients.

**Comment**

The principal finding of our study is that surgery was not superior to dilation in terms of global and sexual QOL, or anatomical results, and bore more complications. A majority of patients achieved a normal-sized vagina regardless of the type of management. Sexual distress was high in most patients, irrespective of vaginal length when within normal ranges.

This is a unique MRKH syndrome cohort, one of the largest reported. The establishment of national rare diseases centers, and the contribution of the French MRKH support group were key points to this multicenter recruitment, demonstrating a change of practice in the management and the study of rare
### TABLE 2

**Complications of surgery**

<table>
<thead>
<tr>
<th>Surgical technique</th>
<th>Vecchietti n = 6</th>
<th>Sigmoid n = 57</th>
<th>McIndoe n = 5</th>
<th>Davidov n = 8</th>
<th>Dupuytren n = 8</th>
<th>All n = 84</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patients</td>
<td>2 (33%)</td>
<td>24 (42%)</td>
<td>2 (40%)</td>
<td>2 (25%)</td>
<td>4 (50%)</td>
<td>34 (40%)</td>
</tr>
<tr>
<td>Complications</td>
<td>2</td>
<td>29</td>
<td>2</td>
<td>2</td>
<td>5</td>
<td>39</td>
</tr>
<tr>
<td>Hemorrhage/pelvic hematoma/rectal hemorrhage</td>
<td>9</td>
<td>1</td>
<td>10</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sepsis/urinary tract infection/peritonitis</td>
<td>7</td>
<td>7</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Thromboembolism</td>
<td>2</td>
<td>2</td>
<td></td>
<td></td>
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<tr>
<td>Bowel adhesions</td>
<td>1</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Lower limb compartment syndrome</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vaginal stenosis</td>
<td>1</td>
<td>4</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>8</td>
</tr>
<tr>
<td>Myofascial syndrome</td>
<td>3</td>
<td>1</td>
<td></td>
<td>1</td>
<td></td>
<td>4</td>
</tr>
<tr>
<td>Vaginal vault disruption</td>
<td>1</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rectal wound</td>
<td>1</td>
<td>1</td>
<td></td>
<td>1</td>
<td></td>
<td>1</td>
</tr>
<tr>
<td>Vaginal prolapse</td>
<td>2</td>
<td></td>
<td></td>
<td>2</td>
<td></td>
<td>2</td>
</tr>
<tr>
<td>Vaginal healing complications</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>3</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>


### TABLE 3

**General quality of life comparing Mayer-Rokitansky-Küster-Hauser syndrome with other populations**

<table>
<thead>
<tr>
<th>WHOOQL-BREF</th>
<th>MRKH patients</th>
<th>French general population</th>
<th>$P^a$</th>
<th>French women with chronic diseases</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical health</td>
<td>78.2 ± 13.5</td>
<td>79.5</td>
<td>.28</td>
<td>64.7 ± 0.3</td>
</tr>
<tr>
<td>Psychological health</td>
<td>63.9 ± 16.6</td>
<td>66.9</td>
<td>.03</td>
<td>62.5 ± 0.3</td>
</tr>
<tr>
<td>Social relationships</td>
<td>69.9 ± 18.6</td>
<td>77.6</td>
<td>&lt;.001</td>
<td>71.3 ± 0.3</td>
</tr>
<tr>
<td>Environment</td>
<td>73 ± 14.6</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
</tr>
<tr>
<td>Item 1: Global health</td>
<td></td>
<td></td>
<td>.13</td>
<td></td>
</tr>
<tr>
<td>• 1—3: Very bad to neither bad nor good</td>
<td>26 (19.9%)</td>
<td>27%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• 4: Good</td>
<td>80 (61.1%)</td>
<td>58%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• 5: Very good</td>
<td>25 (19.1%)</td>
<td>15%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Item 2: Satisfaction toward health</td>
<td></td>
<td></td>
<td>.001</td>
<td></td>
</tr>
<tr>
<td>• 1—3: Not satisfied at all to neither satisfied nor nonsatisfied</td>
<td>51 (38.9%)</td>
<td>25%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• 4: Satisfied</td>
<td>66 (50.4%)</td>
<td>61%</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• 5: Very satisfied</td>
<td>14 (10.7%)</td>
<td>14%</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*MRKH, Mayer-Rokitansky-Küster-Hauser syndrome; N/A, not available; WHOOQL-BREF, World Health Organization Quality of Life instrument.

$^a$ MRKH patients vs French general population.

various techniques reflecting different types of management throughout the country. In addition, 40 patients of this cohort underwent an individual psychological assessment to evaluate the impact of MRKH syndrome diagnosis announcement, which will be reported elsewhere.

The strength of our study was its methodology. We chose to exclude other cases of VA, as MRKH syndrome vaginal dimples may be different from those of complete AIS patient in terms of embryologic development. Moreover, a 46,XY karyotype may have a significant impact on QOL, independently from the absence of vagina. One study reported that vaginal aplasia had a higher psychological impact in MRKH syndrome patients compared to complete AIS patients. The accuracy of our results was supported by the use of validated QOL questionnaires and the standardized pelvic exam performed by an independent evaluator. Older studies reported good sexual QOL by asking questions such as “are you happy with your sex life,” which could be understood as “can you have sex” by the patients, and answers might depend on the relationship between the patient and her physician. Meeting all the patients in person with a supportive attitude probably helped get good-quality responses, but could also explain the unexpected low rate of participation among eligible patients (34.7%) compared to the literature. Many patients travelled for the evaluation, and meeting a new physician could be disruptive.

The groups were inhomogeneous, with more patients in the surgery group compared to the dilation therapy group, and mean age in the latter was significantly younger. This reflects the recent change in VA management, with more patients going through dilation first. Interestingly, there was a higher than expected number of patients who spontaneously created their vaginas with intercourse.

Global QOL questionnaires measure the impact of a disease and its treatment on physical, psychological, and social well-being, compared to the general population. We chose the WHOQOL-BREF questionnaire as it was validated in French, and used in other MRKH syndrome studies. The general QOL of our patients was similar to the general population as reported by others. The same results were found in the physical dimension, as VA has no visible impact from the outside. In the psychological dimension, QOL was significantly lower in MRKH syndrome patients, raising the issue of counseling, which was low in our population. The significantly lower scores in the social dimension were related to the question about satisfaction with sex life, obviously impaired in MRKH syndrome especially in the youngest patients. These were, however, satisfied with their social and friendly relationships. It was demonstrated that age and chronic illness could affect each dimension of the WHOQOL-BREF questionnaire. We chose to compare our patients to the general population, as it seemed to us that MRKH syndrome is not a chronic illness, but rather an intimate deficiency.

The vast majority of our patients were sexually active. Age at first intercourse was slightly older than in the French general population (18.2 vs 17.6 years),

<table>
<thead>
<tr>
<th>TABLE 4</th>
<th>General quality of life according to type of management</th>
</tr>
</thead>
<tbody>
<tr>
<td>WHOQOL-BREF dimensions</td>
<td>MRKH patients</td>
</tr>
<tr>
<td>Physical health</td>
<td>78.2 ± 13.5</td>
</tr>
<tr>
<td>Psychological health</td>
<td>63.9 ± 16.6</td>
</tr>
<tr>
<td>Social relationships</td>
<td>69.9 ± 18.6</td>
</tr>
<tr>
<td>Environment</td>
<td>73 ± 14.6</td>
</tr>
</tbody>
</table>

MRKH, Mayer-Rokitansky-Küster-Hauser syndrome; WHOQOL-BREF, World Health Organization Quality of Life instrument.

<table>
<thead>
<tr>
<th>TABLE 5</th>
<th>General quality of life according to type of surgery</th>
</tr>
</thead>
<tbody>
<tr>
<td>WHOQOL-BREF dimensions</td>
<td>Vecchietti n = 6</td>
</tr>
<tr>
<td>Physical health</td>
<td>75 [67.9–92.9]</td>
</tr>
<tr>
<td>Psychological health</td>
<td>64.6 [62.5–83.3]</td>
</tr>
<tr>
<td>Social relationships</td>
<td>70.8 [66.7–91.7]</td>
</tr>
<tr>
<td>Environment</td>
<td>76.6 [68.7–87.5]</td>
</tr>
</tbody>
</table>

WHOQOL-BREF, World Health Organization Quality of Life instrument.
but younger than in other European countries.\textsuperscript{15}

We chose to use 2 different scales to achieve a complete evaluation of sexual QOL as satisfaction and distress can coexist.\textsuperscript{12} which occurred in our series.

Sexual QOL is considered very good when FSFI score is $>30$, good between 23–29, and impaired $<23$ points.\textsuperscript{16} The choice for a cut-point score over which sexual QOL is good is questionable, and varies in the MRKH syndrome literature between 23–26.5. We chose the score of 26 as a cut-point, as French validated FSFI mean score for French women is 25.7 when aged 30–39 years, and 24.9 when $<30$ years,\textsuperscript{17} in order not to underestimate a bad sexual QOL. It was reported that sexual satisfaction improved significantly 5 years after diagnosis.\textsuperscript{18} In our cohort, mean time between diagnosis and inclusion was 10 years, which may explain the high proportion of good FSFI scores.

Our results confirm that FSFI scores are similar in MRKH syndrome patients and the general population, without difference whether the patients were operated or had dilation therapy.\textsuperscript{11,19} In the meta-analysis of Callens et al,\textsuperscript{11} scores of patients who dilated were lower than of operated patients, but not significantly.

FSFI scores of operated patients in our cohort were similar to patients who had sigmoid vaginoplasty\textsuperscript{12} and Vecchietti,\textsuperscript{20} but not for Davidov procedure\textsuperscript{3} (when FSFI scores were available). The small number of patients for each technique (except sigmoid vaginoplasty) did not allow an accurate comparison to the literature, but in our cohort, no technique was superior to another in terms of sexual QOL.

FSFI scores were significantly higher in the intercourse group, related to a higher score in the satisfaction domain. This underlines the need for a least invasive management that respects the physical and psychological sexual maturity of the patients.

Despite an average to good sexual satisfaction, sexual distress was very high (70%) in our patients, lower in the intercourse group (50%), but independent of the type of management, as previously reported in a smaller cohort.\textsuperscript{12} Fear of dyspareunia was the major cause of distress. It could not be compared to the general population as no norms exist.

Finally, the only patients with significantly altered sexual (but not global) QOL was the subset of 4 patients with vaginas $<6.5$ cm, which may be considered as the minimal goal to achieve.\textsuperscript{11} A normal-sized vagina was not the key point for a good sexual QOL as half of the patients in this case had FSFI scores $<26$. One report of a happy MRKH syndrome patient with 3-cm vagina\textsuperscript{21} underlines that sexual well-being is a complex issue with many variables.

The definition of functional success is variable in the literature, and authors chose a cut-point vagina length, the capacity to coital intercourse, or specific items depending on the technique reported.\textsuperscript{11} In our series, Vecchietti, sigmoid vaginoplasty, and McIndoe seemed superior to Davidov and Dupuytren techniques in terms of vaginal length. One bias was the high number of sigmoid vaginoplasties, which was until recently the main technique used in our country, historically performed in our cohort by highly experimented surgeons of 2 major centers for MRKH syndrome.

Dilation therapy provided significantly shorter vaginas than surgery as reported by the meta-analysis of Callens et al,\textsuperscript{11} but within normal ranges compared to the general population.\textsuperscript{7}

### TABLE 6

<table>
<thead>
<tr>
<th>WHOQOL-BREF dimensions</th>
<th>Vaginal length $&lt;6.5$ cm $n = 4$</th>
<th>Vaginal length $&gt;6.5$ cm $n = 121$</th>
<th>$P$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical health</td>
<td>66.1 [46.4–85.7]</td>
<td>78.6 [25–100]</td>
<td>.10</td>
</tr>
<tr>
<td>Psychological health</td>
<td>62.5 [54.2–75]</td>
<td>66.7 [12.5–95.8]</td>
<td>.85</td>
</tr>
<tr>
<td>Social relationships</td>
<td>58.3 [25–83.3]</td>
<td>75 [0–100]</td>
<td>.16</td>
</tr>
<tr>
<td>Environment</td>
<td>59.4 [43.8–81.3]</td>
<td>75 [21.9–100]</td>
<td>.11</td>
</tr>
</tbody>
</table>


### TABLE 7

<table>
<thead>
<tr>
<th>Surgical technique</th>
<th>Vaginal length, cm</th>
<th>FSFI score</th>
<th>FSDS-R score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sigmoid, $n = 57$</td>
<td>11 [6–15]</td>
<td>25.7 [2.8–34]</td>
<td>20.5 [0–50]</td>
</tr>
<tr>
<td>Davidov, $n = 8$</td>
<td>8.5 [7–11]</td>
<td>23.0 [4.8–33.6]</td>
<td>8 [0–41]</td>
</tr>
<tr>
<td>Dupuytren, $n = 8$</td>
<td>9.3 [8–12]</td>
<td>27.9 [15.3–34]</td>
<td>14.5 [1–32]</td>
</tr>
</tbody>
</table>

Our results were significantly superior to others’ (9.6 vs 5.98 and 6.7 cm), but dilation programs were different, and one should take in account the plasticity of normal vaginas.

While complications after dilation therapy were mild, as no patient presented with urethral dilation, which is the sole serious risk of dilation therapy, complication rate after surgery was elevated (40%), with high morbidity as half of patients needed secondary procedures. Lethal complications were also reported. Sigmoid vaginoplasty drove significantly more dyspareunia and vaginal stenosis, and should probably be abandoned, particularly as it is a major surgery, with specific severe intestinal risks such as inflammatory disease or neoplasia, and as it may compromise uterine transplantation. Vecchietti procedure was the least morbid with only 16% dyspareunia, and may be the technique of choice when dilation fails, as it preserves normal vulvar mucosa.

At the end of the road, all surgical and nonsurgical techniques provided normal-sized vaginas when compared to the general population, with a significantly higher morbidity for surgery. Still, despite the good results of dilation therapy reported since the 1930s, surgeons are still looking for the best surgical technique, as reflected in the literature with 174 surgical studies reported vs 30 on vaginal dilation. Dilation therapy has a bad reputation, while experienced teams report high success rates. Initial size of the vaginal dimple was reported not to be an issue, access to penetrative intercourse was not faster after surgery in our cohort, and cost-effectiveness is higher than surgery.

However, in our study, failure rate was high (33%), similar to other reports, but higher than the 5% failure rate of Edmonds et al, whose patients were slightly older at management. Classic reasons for failure are motivational, as if surgery would quickly fix the problem (ie, VA), which is clearly not the main issue in light of our results.

As global and sexual QOL are not related to vaginal size, whether to create a vagina and when to do it should be an intimate self-decision, according to physical and psychological maturity to engage sexuality. Dilation should be the first-line therapy as it is patient-centered care, and superior to surgery in terms of risk-benefit balance. Efforts should be made to improve results with the development of validated dilation programs and therapeutic education by multidisciplinary experienced teams, in alliance with the patients.

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**TABLE 8**

Female Sexual Function Index scores according to type of management

<table>
<thead>
<tr>
<th>Type of management</th>
<th>Global FSFI score</th>
<th>Desire</th>
<th>Excitation</th>
<th>Lubrication</th>
<th>Orgasm</th>
<th>Satisfaction</th>
<th>Pain</th>
</tr>
</thead>
<tbody>
<tr>
<td>All patients</td>
<td>26 [2.6–34.80]</td>
<td>4.2 [1.2–6]</td>
<td>4.5 [0–6]</td>
<td>4.8 [0–6]</td>
<td>4.4 [0–6]</td>
<td>5.2 [0.8–6]</td>
<td>3.6 [0–6]</td>
</tr>
<tr>
<td>Surgery (1)</td>
<td>26 [2.8–34.8]</td>
<td>4.2 [1.2–6]</td>
<td>4.5 [0–6]</td>
<td>4.8 [0–6]</td>
<td>4.2 [0–6]</td>
<td>4.8 [0.8–6]</td>
<td>3.2 [0–6]</td>
</tr>
<tr>
<td>Dilution (2)</td>
<td>24.7 [2.6–34.4]</td>
<td>4.5 [1.8–5.4]</td>
<td>4.4 [0–5.7]</td>
<td>4.7 [0–6]</td>
<td>3.6 [0–6]</td>
<td>4.8 [0.8–6]</td>
<td>3.6 [0–6]</td>
</tr>
</tbody>
</table>

\[P = .044^{a}\]
\[P = .17\]
\[P = .57\]
\[P = .57\]
\[P = .33\]
\[P = .04^{b}\]
\[P = .22\]


FSFI: Female Sexual Function Index.

\(^{a}\) (1) vs (2): \(P = .85\) (1) vs (3): \(P = .048\) (2) vs (3): \(P = .048\); \(^{b}\) (1) vs (2): \(P = .39\) (1) vs (3): \(P = .007\) (2) vs (3): \(P = .002\).


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**TABLE 9**

Female Sexual Distress Scale-Revised scores according to type of management

<table>
<thead>
<tr>
<th>Type of management</th>
<th>FSDS-R score</th>
</tr>
</thead>
<tbody>
<tr>
<td>All patients</td>
<td>18 [0–52]</td>
</tr>
<tr>
<td>Surgery, n = 83</td>
<td>17 [0–52]</td>
</tr>
<tr>
<td>Dilution, n = 26</td>
<td>20 [0–47]</td>
</tr>
<tr>
<td>Intercourse, n = 20</td>
<td>10 [10–40]</td>
</tr>
</tbody>
</table>

\[P = .38\]

FSDS-R: Female Sexual Distress Scale-Revised.

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