# Long-Term Psychosexual and Anatomical Outcome after Vaginal Dilation or Vaginoplasty: A Comparative Study

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**ABSTRACT**

***Introduction.*** In patients with disorders of sex development requiring creation of a neovagina, a number of techniques are available, including surgical vaginoplasty and self-dilation therapy. Vaginal dilation therapy has been recommended as a first-line treatment because of its less invasive character and high success rate. However, no data exist on long-term psychosexual functioning after vaginal dilation as compared with that after vaginal surgery.

***Aims.*** The aim of this study is to compare the psychosexual and anatomical outcome of women with congenital vaginal hypoplasia followed in the same clinical setting after vaginoplasty with that after vaginal dilation.

***Methods.*** The sexual quality of life of 35 women at least 2 years after vaginoplasty (N = 15), vaginal dilation therapy (N = 8), or coital dilation/no treatment (N = 12) was investigated and compared with the Dutch test validation population (as control).

***Main Outcome Measures.*** Psychosexual functioning was assessed with the Female Sexual Function Index, the Female Sexual Distress Scale-Revised, and a semi-structured interview. A gynecological examination was performed to determine the anatomical outcome after both vaginal treatment regimens.

***Results.*** After either treatment, 26% of these women had a shortened vaginal length of less than 6.6 cm, i.e., more than two standard deviations below the published mean value (9.6 ± 1.5 cm). Irrespective of the treatment, 47% of the patients had (a) sexual dysfunction(s) and experienced sexual distress. However, after vaginoplasty, patients reported significantly more problems with lubrication ($P = 0.025$) than after self-dilation therapy.

***Conclusion.*** Both psychological and physical factors are predisposing for sexual difficulties. To optimize psychosexual comfort, the clinical management of women with vaginal hypoplasia needs to be multidisciplinary and individually tailored. With high success rates reported, vaginal dilation should remain the cornerstone of treatment.

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***Key Words.*** Vaginal Hypoplasia; Vaginal Agenesis; Disorders of Sex Development; Vaginoplasty; Vaginal Dilation; Psychosexual Functioning; Neovagina

## Introduction

Vaginal hypoplasia or vaginal aplasia is an uncommon congenital anomaly with an estimated incidence of 1/5,000 to 1/10,000 live female births; it involves the complete or partial absence of the vagina, uterus, or both [1,2]. Implicated etiologies are Mayer–Rokitansky–Küster–Hauser (MRKH) syndrome, Complete Androgen Insensitivity Syndrome (CAIS), and certain other disorders of sex development (DSD) [3]. Although healthcare providers agree on the importance of the construction of a good functional vaginal substitute that is durable, requires a minimum of postoperative maintenance manipulation, and provides long-term sexual satisfaction, there is substantial disagreement as to which technique best accomplishes these goals [4,5].

Specific surgical vaginoplasty procedures, including split-thickness skin grafts (e.g., McIndoe procedure [6]), local flaps [7], bowel and peritoneal vaginoplasty (e.g., Davydov procedure [8]), or the use of a traction and pressure device (e.g., Vecchietti procedure [9]) can be complex, and the timing of surgery remains controversial. Complications, such as scarring, vaginal stenosis, vaginal prolapse, dry vagina, or excessive vaginal discharge have been described [10]. Therefore, the American College of Obstetricians and Gynecologists recommends a nonsurgical treatment option as first-line therapy—with the use of graduated vaginal dilators involving intermittent pressure on the vaginal introitus (Frank or Ingram method [11,12])—due to the absence of surgical risk and preservation of vaginal tissue [13]. However, while both approaches yield high anatomical success rates [14,15]—defined as an adequate vaginal depth between 5 and 10 cm [2,16]—functional results remain unclear. Functional outcomes refer to sexual functioning, but studies are difficult to compare due to the heterogeneity of reports. Some state these outcomes as satisfactory, adequate, or unsatisfactory [17], whereas others make use of standardized sexual function tools [15,16,18–20] and a more detailed assessment of psychosexual well-being [21], including the effects of this lifelong condition on fertility and bodily integrity [22,23].

## Aims

While most studies evaluate a specific treatment (either surgical or nonsurgical) to create a neovagina, the primary aim of this retrospective study is to compare in a standardized manner the psychosexual functioning—and the relationship with psychological adjustment—of these women seen in the same clinical setting after vaginoplasty with that after vaginal dilation. This approach may lead to evidence-based practice guidelines and further clinical implications for the management of women with vaginal hypoplasia or vaginal aplasia to optimize their psychosexual and psychosocial comfort.

## Methods

### *Participants and Procedure*

The study was conducted as a long-term follow-up audit of DSD patients referred for management of vaginal agenesis to the University Hospital Ghent, Belgium; Erasmus Medical Center Rotterdam, the Netherlands; or Radboud University Nijmegen Medical Center, the Netherlands. Randomization to the two vaginal substitution treatments was not conducted when these patients initially received care, because operative vaginoplasty was in these institutions the standard procedure in the past. Exclusion criteria were: age <18 years and >60 years, recent diagnosis (<6 months), gonadal dysgenesis (because of the heterogeneous clinical picture), and intellectual disability. After exclusion, 57 eligible participants were contacted in Rotterdam and Nijmegen, 24 participants in Ghent, inviting them to attend a clinic visit with a gynecologist and psychologist, who were not previously involved in the care of these women. Twenty-four patients in Rotterdam and Nijmegen (42%) and 11 patients in Ghent participated (46%) between February 2007 and January 2010. A flowchart of the study design can be found in Figure 1. All patients gave written informed consent, and the study was approved by the Medical Ethics Committee of the different institutions. Each research participant was asked to complete standardized questionnaires assessing psychosexual functioning and was invited for a gynecological check-up.

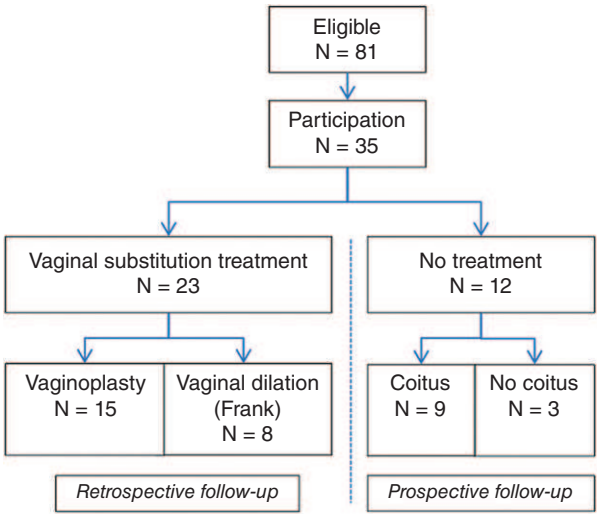


**Figure 1** Study design.

### *Assessment of Psychosexual Functioning*

A sexual dysfunction, according to the Diagnostic and Statistical Manual of Mental Disorders (Fourth edition, text revision) (DSM-IV-TR), is diagnosed by increased sexual distress and decreased sexual function. Sexual distress was assessed by the Female Sexual Distress Scale-Revised (FSDS-R) [24], validated for the Dutch-speaking population [25]. Sexual function was assessed by the Female Sexual Function Index (FSFI) questionnaire [26] (Dutch translation with excellent psychometric properties) [25]. This short 19-item questionnaire assesses adult female sexual quality of life in the 4-week period before completing the survey [27,28], and its score is unbiased regarding age, education, and economic status. The items are assigned to six separate domains of female sexual function: desire, arousal, orgasm, sexual pain, vaginal lubrication, and global sexual and relationship satisfaction. All items in the FSFI have a five-point basic response scale (1–5) denoting variations in frequency, intensity or degree of satisfaction. In addition, some items carry a zero-category coding for "no sexual activity" or "did not attempt intercourse" in the last 4-week period. Instead of interpreting zero answers as extreme degrees of dysfunction [29], women were also asked how they were sexually functioning on the six domains beyond the 4-week period (adjusted FSFI). In case women had not had a sexual partner, the domain score for satisfaction was solely based on item 16. However, the original FSFI scores were used when comparing women with vaginal agenesis to the test validation population and clinical cutoff scores [30]. Additionally, a semi-structured interview delivered in-depth information about psychosexual and psychosocial adjustment.

### *Gynecological Evaluation*

The gynecological evaluation consisted of a (i) medical–somatic anamnesis and (ii) a gynecological exam including visual inspection (clitoral size, labia majora, labia minora, pigmentation, meatus externus urethrae, hair growth, labial scarring, and perineum length), speculum examination (assessment vagina, internal hair growth, granulated tissue, atrophy, and cervix), and pelvic examination (accessibility by number of fingers, vaginal length and width [Hegar], strictures, pelvic floor tone, and vaginal discharge). Vaginal length, defined as the distance from the posterior fourchette to the most proximal part of the blind-ending vagina, was compared with normal reference values previously established [29]. Both patients and gynecologists scored the *cosmetic appearance* of the external genitalia (vagina, clitoris labia majora, and labia minora) on a rating scale (going from 1 = extremely poor to 10 = excellent). In addition, an adapted version of the body image scale [31] was used to assess patient *satisfaction* with the external genitalia and total body image (based on 31 sex-specific and nonsex-specific body characteristics) (scale 1 = very satisfied to 5 = very dissatisfied). Medical notes were reviewed to confirm the diagnosis and treatment procedures.

### *Statistical Analysis*

Differences between proportions were tested by chi-square tests or Fisher exact tests, as appropriate. The Mann–Whitney *U*-test was used to compare sexual functioning in women after vaginoplasty with that after vaginal dilation. Student's *t*-tests were used to compare women with vaginal agenesis with the test validation population (as control). Spearman correlations were used to assess associations; the Wilcoxon signed-rank test for intergroup comparisons. $P < 0.05$ was considered statistically significant. Two-tailed statistical tests were chosen to reduce the risk of type I errors. Sensitivity and specificity were further analyzed through the receiver operating characteristic (ROC) curve. Analysis was carried out by the statistical software package SPSS 19.0 (SPSS Inc., Chicago, IL, USA), and the authors received help from a statistical expert.

**Table 1** Treatment details and diagnoses

| | | Diagnosis | | |
|---|---|---|---|---|
| | | Disorder of androgen action or synthesis* | MRKH | Total |
| None | No coitus | 3 | 0 | 3 |
| | Coitus | 9 | 0 | 9 |
| Dilators | Frank method | 8 | 0 | 8 |
| Surgery | Intestinal† | 2 | 0 | 2 |
| | Skin grafts | 4 | 7 | 11 |
| | Vecchietti | 2 | 0 | 2 |
| Total | | 28 | 7 | 35 |

*Nineteen women had a diagnosis of Complete Androgen Insensitivity Syndrome (CAIS). Other diagnoses comprised 17β3 hydroxysteroid dehydrogenase deficiency in four, Leydig cell hypoplasia in three, and 5α reductase deficiency and 17,20 lyase deficiency each in one woman.
†In one woman the Vechietti failed and she then had an intestinal vaginoplasty.
MRKH = Mayer–Rokitansky–Küster–Hauser

## Results

The median age of the 35 participants was 26 years (18–48 years); 21 participants (60%) were in a stable relationship. The various treatment regimens are summarized in Table 1. Operative vaginoplasty was, in our institutions, the standard procedure in the past. The choice of surgical procedure relied greatly on the surgeon's preference and experience. In the last decade, clinical practice has changed, and women are offered a choice between dilation therapy and surgery, with a suggestion to start with dilation first. Six out of 15 (40%) of the vaginoplasty patients had tried dilation therapy before having surgery but failed to reach sufficient vaginal length. Fourteen out of 15 women who had undergone vaginoplasty followed a postoperative dilation program (mean 3.7 months, range 1–12 months) to maintain their vaginal patency. Psychosexual counseling has only become a vast part of any of the treatment regimens in the last decade. Mean duration of follow-up for the 23 medically treated patients was 6 years (0.5–23 years). Twelve patients received no treatment, because health providers and/or more likely patients themselves judged this was not necessary (yet). Nine of them were sexually active at the time of examination. It is likely that these women increased their vaginal length by regular coitus alone. Because this can be seen as a natural dilation method and because of the small numbers, these women were considered as part of the dilation group in the next analyses. The other three women who had no treatment were the youngest ($M = 18.7$ years, standard deviation [SD] = 0.6) and did not yet engage in coitus. Because this group is too limited, they were not considered as a separate group but were only included in analyses when the total group of women with vaginal agenesis was considered, irrespective of treatment.

The individual diagnoses of the participants are summarized in Table 1. Except for MRKH, all diagnoses were confirmed by gene mutation analyses. All 28 women with a disorder of androgen action or synthesis received bilateral gonadectomy. Twenty out of 28 women were on regular hormone replacement therapy (HRT), one woman refused this, and seven women were not compliant with this treatment.

### Anatomical Outcome

Only 27 out of 35 women participated in the gynecological check-up (Table 2). Four out of eight women did not participate because they lacked the time to do so. Four other women refused because of

**Table 2** Anatomical outcome grouped according to type of intervention for the vagina

| | None—no coitus | None—dilation by coitus | Dilators | Surgery | St data |
|---|---|---|---|---|---|
| N | 3 | 9 | 8 | 15 | 50 |
| Median age (yrs) | 19 (18–19) | 28 (20–48) | 24 (18–36) | 29 (20–42) | |
| Follow-up (yrs) | NA | NA | 5 (0.5–17) | 6 (2–23) | |
| Vag length (cm) | 7.3 (3.7) (N = 3) | 8.9 (2.6) (N = 9) | 7.3 (1.3) (N = 6) | 9.1 (2.7) (N = 9) | 9.6 (N = 50) |
| Complications | None | None | None | Moderate strictures (1–2 cm): N = 2; internal hair growth and vaginal prolapse: N = 1 | |
| *Cosmesis* | | | | | |
| Patient | 7.0 (0.9) | 7.3 (1.2) | 7.7 (1.0) | 7.0 (2.1) | |
| Gynecologist | 8.5 (1.3) | 8.1 (0.5) | 8.4 (1.1) | 6.9 (1.7) | |
| *Satisfaction* | | | | | |
| Genitals | 3.3 (0.3) | 2.7 (0.7) | 2.5 (0.4) | 2.8 (0.7) | |
| Body | 2.7 (0.1) | 2.3 (0.5) | 2.4 (0.3) | 2.5 (0.6) | |

Data shown as mean (standard deviation), the standardization data for vaginal length were based on [32].
Variables: follow-up since last treatment, complications at follow-up, cosmetic appearance of the genitals according to the patient and gynecologist (scale 1–10), patient satisfaction with genital and total body image (scale 1–5)
Used abbreviations: St data = standardization data; vag = vaginal; yrs = years; NA = not appropriate

repeated and shameful examination of the genitalia, including medical photography, experienced in the past. Only two women with MRKH participated.

### *Vaginal Length*

Overall mean (SD) vaginal length was 8.5 cm (±2.5), which was significantly shorter than the published mean value (9.6 ± 1.5 cm) ($P = 0.027$). The mean vaginal length of women (9.1 ± 2.7 cm) who had surgery did not differ from the reference value and was on average greater than in women who had no treatment (8.5 ± 2.8 cm) or followed a medical vaginal dilation program (7.3 ± 1.3 cm), although no significant differences were found (Table 2). Coital dilation led to a mean vaginal length of 8.9 cm (±2.6), compared with 7.3 cm (±1.3) when dilators were used (not significant [ns], $P = 0.21$). Seven out of 27 (26%) of the women had a shortened vaginal length of less than 6.6 cm, i.e., more than two standard deviations below the mean, irrespective of treatment [32] (22% after vaginoplasty, 20% after vaginal dilation). Two out of the three women, who had no treatment, had a short vagina. Women with a short vaginal length, in both treatment groups alike, had a higher incidence of sexual dysfunction compared with those with a normal vaginal length (75% vs. 14%, respectively); however, because of the small numbers, this did not reach statistical significance ($P = 0.088$). No significant association was found between vaginal length and having had intercourse in the last 4 weeks (8.2 cm vs. 8.7 cm, $P = 0.723$). Significant correlations were found between vaginal length and arousal ($r = 0.471$, $P = 0.015$) and between vaginal length and orgasm ($r = 0.409$, $P = 0.047$). Because the vaginal length of only two women with MRKH could be measured ($M = 8$ cm), no meaningful comparison with the 25 women with a disorder in androgen action or synthesis could be made ($M = 8.5$ cm). Specific data for the 19 women with CAIS are provided in Table 4. Overall mean (range) vaginal length was 7.8 cm (4.5–12 cm), which was significantly shorter than the published mean value (9.6 ± 1.5 cm) ($P = 0.004$). Five out of 17 had a shortened vaginal length of less than 6.6 cm, irrespective of treatment. The four women, who underwent surgery, had on average a larger vaginal length (8.8 ± 2.9 cm) than the 12 women who dilated coitally (8.1 ± 2.2 cm) or with dilators (7.3 ± 1.4 cm), although not significant.

### *Complications at Follow-Up*

At gynecological examination, three out of nine women (30%) in the vaginoplasty group displayed complications. No resurgery was required at the time of follow-up. However, six out of 15 women had had multiple operations in the past because of vaginal insufficiency or complications (strictures, excessive mucus production). Median time between the first and second surgery was 1.5 years (2 months–10 years). No meaningful comparison of the complication rate after different types of surgery could be made because of the small numbers.

### *Cosmetic Outcome, Genital, and Body Image Satisfaction*

The patient and gynecologist scored different parts of the external genitalia (vagina, clitoris, labia majora, and labia minora) on a 10-point scale (Table 2). Gynecologists, in contrast with patients themselves, scored the genital appearance of women who had surgery significantly lower than women who dilated (regular coitus and Frank dilation combined) ($M = 6.86$ vs. 8.2, $P = 0.01$).

Participants were also asked to rate satisfaction with their genital and total body image on a 1–5 scale, with higher scores indicating more dissatisfaction. Women who had surgery reported more dissatisfaction with their genital and total body image than women who dilated (regular coitus and Frank dilation combined) ($M = 2.78$ vs. 2.6, $P = 0.642$ for genitals; $M = 2.47$ vs. 2.31, $P = 0.650$ for total body), but no statistical difference was reached. A positive association was found between genital and total body image satisfaction (Spearman's rho = 0.409, $P = 0.015$).

## Psychosexual Outcome

Five women out of 35 (14%) had not been sexually active within 4 weeks before completing the surveys; two of them had undergone vaginal surgery and three did not have treatment. Sexual activity could include caressing, foreplay, masturbation, and/or vaginal intercourse. A further 13 women (37%) specifically had no penile-vaginal intercourse (four women did not have treatment, four women had undergone vaginal surgery, and five women had used dilators). Reasons for having no intercourse were varied: two women reported this was physically impossible, three women were still virgin, and one woman was involved in a homosexual relationship; in seven women, no specific reasons were given.

No difference in prevalence of sexual difficulties was found between women who also participated

in the gynecological examination and those who only filled out psychological questionnaires ($P = 0.596$, ns).

### *FSFI*

As a group, and irrespective of treatment, participants fall below the cutoff score of 26.55 on the total FSFI score when compared with the Dutch test validation population, which implies they are significantly at risk for sexual difficulties [25] (Table 3). Sixty-five percent (11/17) of the participants had a total FSFI score below the cutoff score. Lower scores were evident in all of the subscales equally: desire, arousal, lubrication, orgasm, satisfaction, and pain. On the basis of a clinical cutoff score of 26.55 or less, we determined that 100% of cases in the study sample (N = 6/6) were correctly classified as sexually nondysfunctional and 72.7% (N = 8/11) were correctly classified as sexually dysfunctional. The area under the ROC curve—as a combined measure of sensitivity and specificity—was 0.864 [95% confidence interval 0.686]; which indicated the ability of the FSFI instrument to correctly demonstrate the presence or absence of sexual problems in this study sample. Women who had undergone treatment (surgery or vaginal dilators) did not have better FSFI indicators of sexual function than those who were untreated (regular coitus). When the adjusted FSFI was used, significantly more problems with lubrication were reported after vaginoplasty than after vaginal dilation/coitus ($P = 0.025$) (Table 3, right panel). Within the vaginal dilation group, no significant differences were observed between the women who followed the Frank dilation program and women who had regular coitus, except for desire (Table 3). Women who had a lower FSFI score were significantly more dissatisfied with their genital appearance ($R = -0.553$, $P = 0.021$).

No differences were found between MRKH patients and patients with a disorder of androgen action or synthesis on both the original FSFI and adjusted FSFI. Within the CAIS group, all but two women were sexually active. A further five women did not have sexual intercourse. In five out of nine women in whom the total FSFI score could be calculated, a score below the clinical cutoff of 26.55 was found, indicating that they are at risk for developing a sexual dysfunction (Table 4).

### *FSDS-R*

Both treatment groups had a mean score above the cutoff value of 11 on the FSDS-R and experienced significantly more sexual distress than the control

**Table 3** Psychosexual outcome grouped according to type of intervention for the vagina

| | St data* (N = 108) | Total patient data (N = 35) | No coitus (N = 3) | Surgery (N = 15) | Dilation (N = 17) | Dilation by coitus (N = 9) | Dilators (N = 8) |
|---|---|---|---|---|---|---|---|
| FSFI total | 31.2 (3.9) | 21.3 (9.6)† N = 17 | 6.2 (NA) N = 1 | 21.9 (9.5)† N = 8 | 22.7 (9.3)† N = 8 | 19.2 (4.9) N = 2 | 23.8 (10.5) N = 6 |
| Adjusted total FSFI | | 24.2 (5.0) N = 22 | 24.8 (NA) N = 1 | 23.9 (5.7) N = 11 | 24.6 (4.8) N = 10 | 27.6 (4.8) N = 5 | 21.6 (2.5) N = 5 |
| Desire | 4.0 (0.8) | 3.5 (1.0)† N = 35 | 3.4 (0.4) N = 3 | 3.8 (1.2)† N = 15 | 3.3 (0.9)† N = 17 | 2.8 (0.8)‡ N = 8 | 3.8 (0.6) N = 9 |
| Adjusted desire | | 3.6 (1.0) N = 32 | 3.6 (0.0) N = 2 | 3.9 (1.2) N = 14 | 3.3 (0.9) N = 16 | 3.8 (0.6) N = 8 | 2.8 (0.8) N = 8 |
| Arousal | 5.3 (0.8) | 3.4 (1.8)† N = 34 | 3.0 (2.8) N = 3 | 3.9 (1.9)† N = 14 | 4.3 (1.5)† N = 17 | 4.3 (1.0) N = 8 | 4.3 (2.0) N = 9 |
| Adjusted arousal | | 4.5 (1.1) N = 31 | 4.5 (1.3) N = 2 | 4.3 (1.3) N = 13 | 4.6 (1.0) N = 16 | 4.8 (1.2) N = 8 | 4.3 (1.1) N = 8 |
| Lubrication | 5.7 (1.0) | 3.7 (2.0)† N = 33 | 2.2 (2.8) N = 3 | 3.0 (2.2)† N = 14 | 4.5 (1.6)† N = 17 | 4.7 (1.4) N = 8 | 4.3 (1.8) N = 9 |
| Adjusted lubrication | | 4.2 (1.6) N = 31 | 3.3 (3.0) N = 2 | 3.5 (1.8) N = 13 | 4.8 (1.1)§ N = 16 | 4.8 (0.9) N = 8 | 4.8 (1.3) N = 8 |
| Orgasm | 5.1 (1.1) | 3.9 (1.8)† N = 32 | 2.8 (4.0) N = 2 | 3.6 (1.7)† N = 14 | 4.3 (1.7)† N = 16 | 4.3 (1.6) N = 8 | 4.3 (2.0) N = 8 |
| Adjusted orgasm | | 4.4 (1.3) N = 29 | 5.6 (NA) N = 1 | 4.0 (1.3) N = 13 | 4.6 (1.4) N = 15 | 4.9 (1.0) N = 7 | 4.3 (1.6) N = 8 |
| Satisfaction | 5.4 (0.8) | 4.5 (1.5)† N = 23 | 3.2 (NA) N = 1 | 4.6 (1.4)† N = 10 | 4.5 (1.7)† N = 12 | 4.4 (1.8) N = 4 | 4.5 (1.7) N = 8 |
| Adjusted satisfaction | | 4.5 (1.1) N = 32 | 3.6 (1.7) N = 2 | 4.5 (1.2) N = 14 | 4.6 (1.0) N = 16 | 4.9 (1.0) N = 8 | 4.4 (1.0) N = 8 |
| Pain | 5.7 (0.8) | 2.9 (2.6)† N = 27 | 0.0 (0.0) N = 3 | 3.3 (2.7)† N = 3 | 3.2 (2.4)† N = 11 | 2.1 (2.5) N = 5 | 4.2 (2.2) N = 6 |
| Adjusted pain | | 3.6 (2.4) N = 24 | 0.0 (0.0) N = 2 | 4.1 (2.2) N = 12 | 3.7 (2.3) N = 10 | 5.0 (0.7) N = 5 | 2.4 (2.5) N = 5 |
| FSDS-R distress | 5.1 (6.4) | 12.2 (10.3)† N = 33 | 7.7 (1.2) N = 3 | 13.9 (12.1)† N = 14 | 11.5 (9.5)† N = 16 | 9.4 (8.4) N = 7 | 13.1 (10.5) N = 9 |

Data shown as mean (standard deviation, adjusted scores go beyond a 4-week period), *Standardization data for FSFI and FSDS-R were used from [23]
†Different from the standardization data, $P < 0.01$
‡Different from the dilators group, $P < 0.05$
§Different from the surgery group, $P < 0.05$
Used abbreviations: FSFI = Female Sexual Function Index (six domains of the FSFI, range 0–6), total FSFI (combined scores of the six domains, 2–36); FSDS-R = Female Sexual Distress Scale-Revised (0–52); NA = not appropriate; St data = Standardization data

**Table 4** Functional and anatomical outcome within the CAIS and MRKH group

| | CAIS | MRKH |
|---|---|---|
| Desire | 3.3 (0.9) (N = 19) | 3.9 (0.8) (N = 7) |
| Arousal | 3.7 (1.8) (N = 19) | 4.5 (1.2) (N = 6) |
| Lubrication | 3.7 (2.1) (N = 19) | 3.1 (2.1) (N = 7) |
| Orgasm | 3.8 (1.9) (N = 17) | 3.6 (1.4) (N = 7) |
| Satisfaction | 4.1 (1.8) (N = 11) | 4.8 (1.3) (N = 6) |
| Pain | 2.3 (2.5) (N = 15) | 4.9 (1.6) (N = 7) |
| Total FSFI | 20.7 (10.6) (N = 9) | 25.0 (8.3) (N = 5) |
| FSDS-R | 12.8 (9.6) (N = 18) | 11.7 (11.9) (N = 6) |
| Vaginal length | 7.8 (2.2) (N = 17) | 8.0 (2.8) (N = 2) |

Data shown as mean (standard deviation)
Used abbreviations: Female Sexual Function Index (six domains of the FSFI, range 0–6) and total FSFI (combined scores of the six domain, 2–36); FSDS-R = Female Sexual Distress Scale-Revised (0–52)

group (Table 3). No significant difference was found between the vaginoplasty and dilation group (57% vs. 50%, $P = 0.667$). Within the CAIS group, 10/18 women had a score above 11 on the FSDS-R, which indicates that they experience sexual distress (Table 4).

A significant but not clinically relevant association was found between sexual distress and satisfaction with genital appearance ($R = 0.389$, $P = 0.025$) but not with total body image satisfaction ($R = 0.165$, $P = 0.360$).

### *Combined Score*

A score below 26.55 on the FSFI together with a score above 11 on the FSDS-R implies a sexual dysfunction according to the DSM-IV-TR [30]. In general, 47% of the women (8/17 of whom the total original FSFI and FSDS-R score could be determined) had a sexual dysfunction vs. 6.5% of the Dutch test validation population ($P < 0.001$). There was no statistically significant difference between the different treatment groups (43% [3/7] after vaginoplasty, 50% [4/8] after vaginal dilation/coitus, $P = 0.595$).

### *Psychosexual Counseling*

Forty-two percent (13/31) of the women with vaginal hypoplasia received psychosexual counseling before or during either treatment. Only half of the women who attended counseling (7/13) thought this was useful. Some were already psychologically well adjusted and needed little in the way of counseling. Others had specific issues that they felt needed to be explored more such as feelings about infertility and other diagnosis related obstacles. Interestingly, 83% of the women within the dilation group were satisfied with the usefulness of the psychological care they received, compared with only 20% of the women within the vaginoplasty group ($P = 0.048$).

The influence of specific patient characteristics on the obtained results was further explored. Women involved in a current relationship experienced less dyspareunia than women who had no partner (4.0 vs. 1.5, $P = 0.014$). Women above the median age of 26 years had more problems with lubrication (2.8 vs. 4.4, $P = 0.027$). No associations were found between HRT use and sexual counseling on the one hand, and the prevalence of sexual problems on the other hand.

## Discussion

Our study findings show that vaginal hypoplasia or vaginal aplasia, despite treatment, is associated with compromised sexual wellness (difficult lubrication and dyspareunia), as was found in previous studies [14,15,18,33–36]. Eighty-six percent of our sample was sexually active, but only 49% had vaginal intercourse. Although a problematic sexual functioning was reported in both treatment groups, women who had undergone vaginal surgery had more complications at follow-up and experienced on average more sexual problems, specifically with respect to self-reported lubrication. This difference was not influenced by the underlying diagnosis, nor by compliance with estrogen therapy. Age was however positively correlated with lubrication problems, and women who had had vaginal surgery were older than the Frank dilation group (presumably reflecting a change in clinical practice). Other suggested interfering factors are the type of vaginoplasty procedure and the lack of cervical mucus [27]. Lubrication during intercourse is thought to be the result of several processes including transudation of plasma through the vaginal epithelium, secretions from the uterus, and the vestibular and Bartholin's glands [1]. Although the vaginal lining may be normal in the neovagina created by dilators and the Vecchietti and Davydov techniques—in contrast to other surgical procedures—it is not known whether the blood supply of the neovagina and its capacity to produce the transudate is adequate. The lack of vestibular and Bartholin's glands, which are a source of lubricating secretions during arousal, may also be relevant. Whether or not these glands are present in women with vaginal hypoplasia should be the topic of future studies [1].

Vaginal dilation has been put forward as a first-choice treatment, because it is a patient-driven technique that is easy to perform, cost-effective,

safe, and can be highly successful [2,5,13,37]. Sixty-seven percent of the Frank dilation patients in this study acquired a normal-sized vagina within 3–12 months. It also obviates the need for postsurgery dilation therapy, which was required in 93% of women who underwent surgery. Moreover, six out of 15 women (40%) in the vaginoplasty group needed resurgery, showing that these procedures carry significant long-term complications, including increased mucus production, vaginal prolapse, and strictures. The women who had had vaginal surgery acknowledged that it was not the "quick fix" (and also less emotionally involved) procedure that it initially appeared to be. However, success of dilation therapy as a first-line treatment depends on a large time investment and motivation of the patient. Forty percent of the vaginoplasty patients had tried dilation therapy first but without success. The reported problems can be summarized as persistent discomfort and pain or lack of privacy; the regimen was regarded as shameful and "distasteful" [38], or reinforced a feeling of being different. The profound emotional impact of this diagnosis inevitably evokes feelings of depression, anger, and loss of self-esteem. A poor organization of the therapy with little psychological input at that time may also reflect the lack of enthusiasm for the dilation technique; moreover, most of these patients were young (range 14–17 years). Roberts et al. [39] found that patients younger than 18 years at the start of dilation treatment had a statistically significant dilation failure rate. Our study confirms previous reports that the compliance and patient satisfaction are generally low in vaginal dilation programs [40]. Every unsuccessful attempt will decrease the motivation of the young patient and lead to emotional instability, which highlights that adequate sexual and psychological support is an integral part of the management [10].

The description of long-term outcome taking into account different pathogenesis of vaginal hypoplasia is difficult because of several DSD enclosed. However, we provided specific data for 19 women with CAIS, allowing to further examine the effects of androgen deficiency on sexual function. Androgens are thought to influence sexual function in females by their affects on sexual motivation and desire [40]. When compared with the MRKH and the Dutch control group, desire and arousal scores in the CAIS group were lower, but 79% and 84% of the CAIS sample yielded scores within normal limits for desire and arousal, respectively. Vaginal hypoplasia and various psychological factors undoubtedly also impact sexual functioning in women with CAIS [40].

We recognize several weaknesses of this study. First, there was a potential selection bias, because participants in this study were recruited exclusively from a clinical sample. Further studies should also recruit from other samples such as peer-support groups. Additionally, data on nonresponders should be gathered. Fewer than half of the eligible women approached actually participated in the study, indicating that physical aspects of female sexuality are still a very sensitive subject. Second, due to the rarity of the condition, the actual number of patients did not lend itself to robust regression analyses. Because the sample of patients who followed a vaginal dilation program was rather small to draw definite conclusions, we included patients who created a sufficient vagina by coitus alone in this group. Although coitus can be considered as a form of dilation (with the penis as only dilator size) this might have influenced the results. Meanwhile, it has been demonstrated that the dimensions of the neovaginal increase at coitus is comparable in magnitude with the normal vagina. In young women with an understanding and cooperative sexual partner, the possibility of coital dilation should be taken into consideration as one of the available therapeutic procedures. Additionally, those women who had undergone treatments for vaginal hypoplasia had similar sexual function scores to women who had not undergone any treatment. It is likely that those offered treatment for vaginal hypoplasia were a group with more severe vaginal hypoplasia, so no definite conclusions can be made on the impact of vaginal hypoplasia treatment on the incidence of sexual difficulty. These results do suggest, however, that any treatment for vaginal hypoplasia may be of limited usefulness without concomitant psychological expertise to address other aspects of self-perception. Third, because randomization to the two treatments was not conducted, we measured several important confounders, such as age, HRT, genital appearance, satisfaction with total body image, and psychosexual counseling, which could influence sexual functioning. However, the long-term assessment of vaginal reconstruction methods remains difficult, and complications may be troublesome many years after the primary procedure. Patient satisfaction will be influenced as well by the procedure as the clinical (e.g., hormonal treatments) and psychosocial implications (e.g., infertility) of the underlying condition. Lastly, this was a retrospective study with a cross-sectional design, which does not permit interpretation of

causal relationships. No information was available on vaginal length or psychosexual functioning and expectations of the participants before treatment. Prospective, longitudinal studies with a focus on diagnosis-related success rates (e.g., CAIS cases with distal third vaginal remnant vs. cases with vaginal aplasia or MRKH) should be undertaken. Comparison with normative data from other gynecological conditions may also be worthwhile.

## Conclusion

The findings in this study have implications for clinical management as they suggest that long-term psychosexual outcome after vaginal dilation is at least equivalent to that of vaginoplasty. It appears reasonable to consider self-dilation as the first therapeutic procedure. However, if it wants to reach a high success rate, gradual self-dilation has to be supported by an expert multidisciplinary team that integrates endocrinology, gynecology, sexology, and clinical psychology expertise. Failed dilation therapy for neovaginal creation does not preclude subsequent surgical reconstruction. Equal priority should be given to quality of life outcomes, including psychosexual treatments, as is currently given to the traditional clinical concerns such as anatomical outcome [40]. Psychological counseling as both a primary and adjuvant treatment has a clear role in discussing any aspects of this lifelong condition. Further studies must clarify the multiple obstacles for women in different age groups and life stages and emphasize objective outcomes in a prospective way using validated questionnaires for patients and partners, in addition to clinical examinations and patient interviews.

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