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

Alaa Cheikhelard, MD[1]; Maud Bidet, MD[1]; Amandine Baptiste, MSc; Magali Viaud, MSc; Christine Fagot, MD; Naziha Khen-Dunlop, MD, PhD; Christine Louis-Sylvestre, MD; Sabine Sarnacki, MD, PhD; Philippe Touraine, MD, PhD; Caroline Elie, MD; Yves Aigrain, MD, PhD; Michel Polak, MD, PhD; for the French MRKH Study Group

BACKGROUND: Vaginal agenesis in Mayer-Rokitansky-Küster-Hauser syndrome can be managed either by various surgeries or dilation. The choice still depends on surgeon's 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 dilatation 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,[1–4] but in many countries, surgery is still performed soon after diagnosis[5] 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

Cite this article as: Cheikhelard A, Bidet M, Baptiste A, et al. Surgery is not superior to dilation for the management of vaginal agenesis in MRKH syndrome: a multicenter comparative observational study in 131 patients. Am J Obstet Gynecol 2018;219:281.e1-9.

0002-9378/$36.00

https://doi.org/10.1016/j.ajog.2018.07.015

AJOG at a Glance

Why was this study conducted?

To compare surgery or dilation in the management of vaginal aplasia in women with Mayer-Rokitansky-Küster-Hauser syndrome.

Key findings

Global and sexual quality-of-life scores were similar between groups. Vaginal depth was shorter in the dilation group than the surgical group, but was still within normal range. Complications were more frequent and severe after surgical vaginoplasty.

What does this add to what is known?

Based on a risk-benefit assessment, this independent study confirms that dilation is superior to surgery, and should be the first-line treatment for vaginal aplasia in women with Mayer-Rokitansky-Küster-Hauser syndrome.

evaluated with standardized validated questionnaires. Only reviews, metanalyses, or small series actually compared several different techniques,[6] 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 benefits. 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,[7] 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 χ^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 χ^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.