Well-being and sexual function outcomes in women with vaginal agenesis

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Objective: To assess long-term quality of life and sexual satisfaction outcomes of women treated for vaginal agenesis.

Design: An audit project using questionnaires to assess quality of life (World Health Organization Quality of Life Bref [WHOQoL-Bref]) and sexual satisfaction (Golombok Rust Inventory of Sexual Satisfaction [GRISS]).

Setting: Gynecology Department of Royal Children’s Hospital.

Patient(s): Women with vaginal agenesis.

Intervention(s): Medical data were extracted from notes. Women were contacted at their review consultation or were offered the opportunity for review and participation in the audit.

Main Outcome Measure(s): WHOQoL-Bref and GRISS were measured. Correlation with medical details and treatment method was undertaken.

Result(s): Age range was 16–71 years (median, 23 years). WHOQoL-Bref (n = 28) scores (mean ± SD, 4.2 ± 0.8), were similar to the mean Australian population score. The GRISS questionnaire (n = 20) revealed that 75% of women had overall satisfactory scores. WHOQoL scores had a high correlation with GRISS scores. Time since diagnosis correlated to GRISS scores with 12 of 13 diagnosed >5 years earlier having satisfactory results. Women without a hymen had more problems with vaginismus.

Conclusion(s): General outcomes are good for these women, although vaginismus was an issue. There was a correlation between lower health satisfaction scores, feeling less feminine and feeling down, but not GRISS. (Fertil Steril® 2010; 93: 6–12. ©2010 by American Society for Reproductive Medicine.)

Key Words: Vaginal agenesis, MRKH, dilator therapy, sexual function, quality of life, hymen, outcome

Vaginal agenesis is an uncommon congenital anomaly, occurring in approximately 1 in 5,000 females (1); it involves the complete or partial absence of the uterus, vagina, or both. When it is an isolated anomaly, it is commonly referred to as the Mayer-Rokitansky-Kuster-Hauser syndrome. Vaginal agenesis usually presents in post-pubertal girls as primary amenorrhea, XX karyotype, and normally functioning ovaries. The condition can be treated in several ways, including nonsurgically with the use of graduated vaginal dilators involving intermittent pressure on the vaginal introitus—that is, the Frank method (2, 3)—or surgically with approaches such as the McIndoe (4, 5), Sheare’s (6), modified Williams (7), bowel segment vaginoplasty (8), Vecchietti (9, 10), and Davydov techniques (11).

The reported outcomes of the different approaches are generally good, although most techniques have some complications (12–15) and failures (2, 16–19). Success is measured in terms of the anatomic success in creating a vagina and also in functional terms. Anatomic success refers to depth and width of the neovagina. Vaginal lengths vary (20), and although an objective physical examination might reveal an unsatisfactory anatomic result, this does not necessarily correlate with the patient’s interpretation of successful outcome (21, 22). In the study by David et al. (23), it was discovered that no male partner who was uninformed about the operation became aware as a result of intercourse that the vagina had been surgically created. In addition, most women with Mayer-Rokitansky-Kuster-Hauser syndrome reported after dilator therapy that their vaginal size was “more or less normal” (24).

Functional outcomes refer to sexual function, with a trend away from simply reporting these outcomes as satisfactory, adequate, or unsatisfactory (6, 7, 18, 25, 26) to the use of more detailed assessment of psychosexual wellbeing (22, 23, 27, 28) and standardized sexual function tools (7, 13–15, 19, 24–29).

Beyond the physical issue of vaginal agenesis, there are many social and psychological issues also affecting these women, including the impact on their infertility if a functioning uterus is not present. Despite the numerous psychological and psychosexual obstacles to be overcome, research has suggested that women with vaginal agenesis can expect to have a near normal quality of life (30). In general, study findings show that the acute response to the diagnosis is accompanied by diminished self-esteem in almost all patients and some reports of suicidal thoughts at that time (22).

This study was undertaken to assess the outcomes of the women with vaginal agenesis who received care in Melbourne. The emphasis was on the quality of life and sexual function using standardized questionnaires. An effort to correlate these results with a number of other features related to the time since diagnosis and correlating the sexual function with anatomical findings.
MATERIALS AND METHODS

The study was conducted as a long-term follow-up audit of patients referred for management of vaginal agenesis over the past 20 years, at either the Royal Children’s Hospital or the private rooms of a pediatric and adolescent gynecologist in Melbourne, Australia. There were 70 patients with vaginal agenesis or atresia identified from the database of the Royal Children’s Hospital Department of Gynecology and the private practice between 1990 and 2009. Patients <17 years old, diagnosed within the last 6 months and those with an intellectual disability were excluded. Participants were included regardless of presence or absence of a uterus. Patients who were treated either surgically or with dilators were considered eligible. If the patient had not yet commenced treatment, they were not asked to complete some components of the questionnaires.

After exclusions, 61 eligible participants were mailed a letter informing them of the audit study, inviting them to attend a clinic visit with a gynecologist where they would complete the questionnaires. Alternatively, the option of a phone interview or mail-out questionnaire was offered. Information regarding age of diagnosis and presence or absence of a hymen was gathered from their medical notes. Hymens were considered normal if they were present or were a normal variation, such as sepate or annular. Women in whom there was only a small fragment of abnormal hymenal tissue were grouped with those without a hymen.

The World Health Organization (WHO) Quality of Life Bref (WHOQoL-Bref) (31) was administered to all women to assess general well-being. The WHOQoL-Bref is a 26-question, validated and standardized questionnaire that covers four domains of quality of life—physical, psychological, social, and environment—and asks patients to rate their overall quality of life and satisfaction with physical health on a scale 1–5. Quality-of-life scores of the women treated for vaginal agenesis were then compared with the Australian population average as published in the WHOQoL User Manual, which was based on a study of a stratified community sample of 396 people (32).

The Golombok and Rust Inventory of Sexual Satisfaction (GRISS) questionnaire (33) was administered to the women who were sexually active. The GRISS questionnaire is a validated and standardized measure of sexual satisfaction, comprising 28 questions and covering seven domains of sexual dysfunction: infrequency, anorgasmia, vaginismus, dissatisfaction, avoidance, noncommunication, and nonsensuality. Sexual satisfaction scores ≤4 are considered to be satisfactory according to the GRISS scale. In addition, all patients were given a series of other questions to answer regarding their reactions to diagnosis, feelings, future plans, and relationships. Institutional approval was granted from the RCH Human Research Ethics Committee (CA28095).

RESULTS

A response was received from 34 women, with six women declining participation. Two women had moved overseas, and 25 did not respond or could not be located because of address changes.

Of the 28 participants in the study, all completed the WHOQoL-Bref, and 20 completed the GRISS questionnaire. (Seven of the eight patients who did not complete the GRISS were not yet sexually active, and the other felt uncomfortable responding to the questions.) Of the patients who participated in the GRISS questionnaire, four had been treated surgically with either the Sheare’s procedure (n = 3) or the McIndoe procedure (n = 1), and 16 had created a neovagina surgically using dilators.

The age range was 16 to 71 years, with a median age of 23 years. Ten (36%) of the 28 women were single, 13 (46%) were in an intimate relationship, two (7%) were in a relationship without sex, and three (11%) were married.

The mean score of the WHOQoL-Bref questionnaire was 4.2 ± 0.8 (mean ± SD), compared with the mean quality of life score in the Australian population of 4.3 ± 0.8. Results across all four domains in this questionnaire were highly similar to those seen in the average Australian population (Fig. 1).

One of the 28 patients in this study described feeling suicidal upon diagnosis. The other reactions were largely negative emotions, displayed in all but two women. Patients were also asked which aspect of their condition they found to be the most distressing, to which 79% of women responded “unable to carry a pregnancy.” In addition, four patients said they did not feel as though they could identify themselves as a normal woman, these patients scored significantly worse on overall quality of life and satisfaction with health respectively (P < 0.04 and P < 0.01, Mann-Whitney test) on the WHO Quality of Life questionnaire.

The average overall transformed GRISS score was 3.1 ± 0.4 (mean ± SE), with 75% of the women having satisfactory scores (GRISS scores ≤4) (Fig. 2). Mean scores were 4 or less across all domains except vaginismus (mean ± SD, 5.0 ± 2.3) and infrequency (mean ± SD, 5.1 ± 2.1). When the “infrequency” was analyzed further, it was related to avoidance rather than vaginismus (P = 0.005). Scores for the GRISS dissatisfaction domain were less than 4 for 19 of 20 patients (95%), as were noncommunication subscale scores.

Overall dissatisfaction with physical health was correlated with feeling down often or always (P = 0.02), thinking about their condition most days or all the time (P = 0.02), and not feeling feminine (P = 0.04). Interestingly, GRISS scores were not influenced by these three factors.

Overall quality-of-life scores and overall sexual satisfaction (i.e., GRISS) scores were strongly correlated (P < 0.0001). Likewise, satisfaction with physical health was also linked with GRISS score (P = 0.02). Poor quality-of-life scores were correlated with feeling down often or always (P = 0.04) and not feeling feminine (P = 0.04).

A correlation was found between women with no hymen (or an abnormal variant) and poor GRISS scores on the vaginismus domain (P = 0.045, unpaired t test). The mean vaginismus subscale score for women with a hymen (or a normal variation) was 4.7 ± 2.2 (mean ± SD), compared with 7.0 ± 1.0 (mean ± SD) for women without a hymen.

GRISS scores were significantly better in the group diagnosed >5 years ago (mean ± SD, 2.5 ± 1.6) compared with those who had
be diagnosed ≤5 years ago (4.3 ± 2.1; P=0.04). Twelve of 13 patients (93%) diagnosed >5 years ago showed satisfactory GRISS scores. There was no difference in GRISS scores in those who had met others or attended a psychotherapeutic group. Most patients (67%) thought sexual partners would not be able to tell that they had a gynecologic condition if they did not tell them, and 82% did not believe sex was less enjoyable for their partner because of their condition.

DISCUSSION
The outcomes of the WHOQoL-BREF questionnaire showed overall quality of life to be comparable to that of the average Australian population according to the WHOQoL user’s manual. Patient scores were highly similar to the average Australian population across all four domains: physical, psychological, social, and environmental. It is not possible to know whether the nonresponders and those refusing participation lead to biased results. Nevertheless, this work concurs with the majority of works on this subject, in that women with vaginal agenesis can expect a quality of life that is almost indistinguishable from that of the average population (30), although most studies do not report the use of quality-of-life questionnaires (7, 13, 19, 24–26). Despite this lack of assessment, the importance of the psychosocial support is acknowledged by all studies.

Hecker and McGuire reported that 5 of 23 patients were suicidal at the time of diagnosis (22), whereas in this study, one of 28 patients described feeling suicidal. The other emotions experienced upon diagnosis were negative in all but two women. The inability to carry a pregnancy was an ongoing source of distress for 79% of the women, an issue also reported by others (15, 27). This finding reinforces the importance of ensuring that the woman receives adequate psychological support and care and that the topic of pregnancy and options for reproduction in terms of surrogacy are discussed during consultations.

The issue of feeling like a woman or feeling feminine is a concern that has been raised by others (15, 30) and has been shown to have a negative effect on self esteem. In this study, 4 of 28 patients said they did not feel as though they could identify themselves as a normal woman. These patients scored significantly worse on overall quality of life and satisfaction with health in the WHO Quality of Life questionnaire.

The importance of general adjustment and psychological well-being has been recognized as being an important predictor of positive sexual functioning (27, 30). The majority of studies measuring sexual function report good outcomes overall. In this study, the sexual satisfaction of women with vaginal agenesis was good, with 75% of women having satisfactory scores in the GRISS.

Women who received a diagnosis >5 years ago scored significantly (P=0.04) better than those who had received a diagnosis within the last 5 years. This result could be due to a number of factors: more comfort with diagnosis, more experience with intercourse, stable partner and trust in relationship, and decreased anxiety. Whatever the reason (or combination of reasons), this information gives hope that time indeed heals and that they will mostly likely be able to enjoy a satisfactory sexual relationship with their partner despite their gynecologic condition.

The use of a standardized questionnaire makes it possible to see which areas were particularly problematic, showing infrequency and vaginismus to be the main contributors to the lowering of overall scores. Those reporting on function for sigmoid vaginoplasty do not report problems with lubrication, although dyspareunia associated with stenosis can be a problem requiring revision of the perineal anastomosis (15, 18). Secretions requiring the use of daily pads are reported in this population (15). For studies reporting the standardized sexual function outcome after the use of dilators, the Davydov and Vecchietti techniques (all of which result in the creation of a neovagina lined by epithelium that is identical to normal vaginal mucosa) (22, 26), a consistent finding has been that lubrication (13, 19, 22, 34) continues to be a problem, at least in some individuals. In the GRISS, questions regarding lubrication are categorized as dyspareunia, and our study also supports this finding. It has been suggested by Nadarajah et al. (19) that this may relate to the lack of cervical mucus. However, lubrication during intercourse is thought to be the result of several processes including transudation of plasma through the vaginal epithelium; secretions form the uterus and the vestibular and Bartholin’s glands. Although the epithelium may be normal in the neovagina created by dilators and the Creatsas, the Davydov, and Vecchietti techniques, it is not known whether the blood supply of the neovagina and its capacity to produce the transudate is normal. The lack of vestibular and Bartholin’s glands, which are also a source of lubricating secretions during arousal, may also be critical. The presence or absence of these glands has not been previously reported in women with vaginal agenesis, but we have noted that some of our women with vaginal agenesis have hymens, whereas others have no hymen or only a remnant of abnormal hymenal tissue. In those with a normal hymen, one would expect the vestibular and Bartholin’s glands to be present, whereas those without a hymen do not appear to have any duct associated with Bartholin’s glands and presumably do not have vestibular glands. Our results showed a correlation between those women with absent hymens and those who had poorer lubrication (dyspareunia) scores compared with those with a normal hymen (P=0.045), suggesting that this may be an important factor. This is clearly an issue that could be resolved if others also note the presence or absence of the hymen in women with vaginal agenesis.

CONCLUSIONS
Overall, quality of life and sexual satisfaction was generally good in this population of women with vaginal agenesis. This study suggests that nonsurgical creation of a neovagina using vaginal dilatation is a safe and effective method of treatment, with sexual satisfaction scores being almost identical to those who have been treated.
Outcomes in women with vaginal agenesis


