QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

Case Report

Anatomical and sexual functions at post neovagina operation on Mayer Rokitansky Kuster Hauser Syndrome (MRKH) patients with sigmoid, amnion, and conventional methods in Dr. Soetomo Hospital

Uning Marlina[1,2*], Hari Parathon[2], Azami Demas[3], Jimmy Yanuar[4], Iwan Kristian[5]

1) Faculty of Medicine, Muhammadiyah Surabaya University
2,3) Urogynecology Division, Department/ SMF of Gynecologic Obstetrics, Medical Faculty of Airlangga University – RSUD Dr. Soetomo, Surabaya
4) Fertility Division, Department/SMF of Gynecologic Obstetrics, Medical Faculty of Airlangga University – RSUD Dr. Soetomo, Surabaya
5) Department/ SMF of Digestive Surgery, Medical Faculty of Airlangga University – RSUD Dr. Soetomo, Surabaya

ARTICLE INFO

Submitted : November 2019
Accepted : December 2019
Published : January 2020

Keywords:

Neovagina, MRKH, sigmoid, amnion, anatomic function

Correspondence:
uningmarlina@gmail.com

ABSTRACT

The Mayer–Rokitansky–Kuster–Hauser (MRKH) syndrome is congenital malformation due to utero-vaginal agenesis. For many years, Dr Soetomo Hospital has been applying McIndoe technique using biomaterial amnion. Recently, in collaboration with digestive surgery, neovagina operation using sigmoid was performed. However, no data are available on the complications, anatomic, and functional results of the neovagina operation using sigmoid and amnion. MRKH patients who have performed neovagina operations from January 2011 to December 2014 were involved in this case review. The anatomical function was measured based on minimal vaginal length of more than 6 cm and width of 3 cm, while sexual performance is measured by Female Sexual Function Index (FSFI), which is above 23, and Female Sexual Distress Score Revised (FSDSR), whose score is < 11. In 4 years period, there were 6 cases of MRKH underwent neovagina (1 used sigmoid and 5 used amnion) at Dr Soetomo Hospital. All of them had a satisfying anatomical and sexual function. Even though neovagina operation significantly reduces sexual distress, but it does not necessarily mean alleviate as seen by the high FSDSR score. Neovagina using amnion has faster operation time and cheaper with the same length of hospital stay than sigmoid neovagina. Sigmoid neovagina has a better vaginal length, lubrication and no need dilatation after operation, but it has higher complication risk. The Neovagina technique should be used at Soetomo Hospital depends on patient wishes and circumstances. For those who have strong financial support and are not willing to do manual dilatation, they should use the sigmoid neovagina, while those who are able to do manual dilatation, then the amnion neovagina is a good choice.

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

INTRODUCTION

Genital organs have both reproduction and sexuality functions. But, some patients experience developmental disorders of genitalia organs, such as Mayer-Rokitansky-Küster-Hauser (MRKH) patients who do not have fully performed vagina and uterus, but their ovaries are well functioning. These patients are usually female phenotypes, in which the women have large breasts and their pubic and armpit hair grow normally, but they do not have menstrual periods, and after the examination, they do not have vagina or uterus. The women will be very difficult to get pregnant and give birth, except with surrogacy. However, these women are still possible to enjoy sexual life, like other women, if their vaginal agenesis is reconstructed using various techniques, either surgical neovagina and non-surgical neovagina (Chandler, Machan, Cooperberg, Harris, & Chang, 2009).

The main goal of surgical neovagina for MRKH patients is not for reproductive function, but for sexual function. The operation can be said to be successful if the neovagina results are given not only adequate vaginal length, good lubrication with minimal maintenance such as the need for regular dilatation but also able to satisfy in sexual intercourse (Fedele et al., 2010).

For years, the urogynecology division in Dr. Soetomo Hospital has used surgical neovagina techniques with amniotic biomaterials. Amnion as biomaterial has several advantages in the form of the ability to prevent cicatrix and encourage epithelialization to reduce the possibility of vaginal narrowing (Rennie et al., 2012). At the end of 2014, neovagina operation is performed, using sigmoid colon, in collaboration with the department of digestive surgery. Sigmoid is a very good material for neovagina allographs because sigmoid is easy to mobilize and thick, but it can also produce lubricants and the inner part is like the original vaginal rugae (S. Robert Kovac, Carl W Zimmerman, 2012).

The report of this case is intended to evaluate the results of neovagina operation, neo vaginal operation using amnion in comparison with sigmoid allograft from anatomical aspect, sexual function, length period ready for sexual intercourse after surgery, length of operation, length of stay in hospital, operation costs and complications that may arise on MRKH patients at Dr. Soetomo Hospital between 2011 and 2014. These results will be then used in the service of choosing the type of operation that suits the needs of patients by the capability of operation at Dr Soetomo Hospital.

METHODS

This case report was taken from MRKH patients at Dr. Soetomo Hospital from January 2011 to December 2014 who had performed neovagina operation. These patients were then evaluated for neovagina anatomy by measuring neovagina length and also for their sexual function using Female Sexual Function Index (FSFI) and sexual psychological aspects by Female Sexual Distress Score Revised (FSDSR). In addition, anamnestic data was collected regarding the waiting time between the operation and its first use for intercourse, the calculation of treatment cost and length of stay in the hospital, as well as surgical complications from medical record data.

Neovagina operation can be said to be anatomically successful if the vaginal length is at least 6 cm with a minimum width of 3 cm. (Fedele et al. 2010) Meanwhile, neovagina operation can be said to be well-functioned sexually if the FSFI score is above and the FSDSR score is less than 11.

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

RESULTS

From 2011 to 2014 (3 years), seven MRKH patients had neovagina operation in Dr Soetomo Hospital (one patient was excluded from this case report due to the lost follow-up, thus only six cases in total). One patient used sigmoid (AL), while the others used an amniotic method.

Table 1. Result of MRKH Suporting Examination

	Vagina	Testosterone	Karyotyping
SIGMOID 1	3 cm	2	46 xx
AMNION 1	2 cm	< 2.0	Not performed
AMNION 2	2 cm	< 2	Not performed
CONVENTIONAL 1	1.5 cm	1.9	46 xx
CONVENTIONAL 2	1.5 cm	0.162	Not performed
CONVENTIONAL 3	2 cm	< 2	Not performed

DISCUSSION

Age for performing Neovagina operation

Most neovagina surgeries are performed at women in their 20s due to several reasons. Firstly, some techniques, like those using amnion require postoperative dilatation either by manual or continuous intercourse. The study found that vaginal length in women who were sexually active was significantly longer than those who were not sexually active or who had been sexually active (6.3 cm vs. 3.6 cm vs. 3.2 cm, $p<0.05$) (Liao, Conway, Ismail-Pratt, Bikoo, & Creighton, 2011). Manual dilatation requires psychological maturity and strong motivation, which allows the manual dilatation can be done continuously. Manual dilatation is sometimes a source of stress for patients, because they have to do it routinely in a place that requires privacy, so it is very tiring. Secondly, in order to minimize the risk of recurring narrowing due to non-compliance with manual dilatation, neovagina operation should be performed prior to sexual activity. The patients who experience neovagina narrowing postoperatively are mostly due to not manually dilated for so long or absent from for sexual intercourse. For this reason, most doctors recommend delaying the vaginal operation when the patient approaches to sexual active. Previous studies have reported that neovagina using sigmoid do not require post-surgical dilatation can indeed be done in pediatric patients (the youngest is at the age of 2 years old), but it is because the development of an imperfect vulva is often problematic in its anastomosis (Lima et al., 2010).

The patient number amnion 2, neovagina 1 dan 2, firstly came to the gynecology center with the complaints of primary amenorrhea and they have been diagnosed with MRKH when they were still in senior high school. But, the surgery to construct the vagina was delayed until they were ready to have active sexual intercourse. Meanwhile, the patient number sigmoid 1, amnion 1 and neovagina 3 were first diagnosed when they were married due to their difficulty in penetrating during intercourse. This is in line with previous studies, in which the majority of MRKH patients come with their complaints of primary amenorrhea or intercourse difficulties (Fotopoulou, Neumann, Klapp, Lichtenegger, & Sehouli, 2008)

Table 2. Timetable when the Respondents Perform the Operation

Patient Name/ RM No.	Age at first visit	Age at operation	At operation
SIGMOID 1	28 yo	28 yo	3 years of marriage
AMNION 1	18 yo	18 yo	1 year of marriage
AMNION 2	27 yo	27 yo	5 months of marriage
CONVENTIONAL 1	17 yo	23 yo	1month before marriage
CONVENTIONAL 2	16 yo	26 yo	1 week of marriage
CONVENTIONAL 3	16 yo	32 yo	1 month before marriage

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

Anatomical Function of MRKH Patients After Neovagina operation

Neovagina operation is said to be anatomically successful if a minimum vaginal length is 6 cm and its minimum width is two fingers at evaluation of 6 months postoperative (Fedele et al., 2010). Neovagina operation patients who use amnion method (cases amnion 1 and 2) have shorter vaginal length (averagely 6.8 cm) compared to those using sigmoid colon media (case no. 1). However, all patients have minimum vaginal length of 6 cm with a width of 3 cm; thus it can be said to be anatomically successful. Although neovagina patients with amnion method have shorter vaginal length than those using the sigmoid colon method, patients with the longest evaluation, namely four years after neovagina operation using amniotic methods, have not shown any signs of narrowing.

Patients with the amniotic method, if compared to those with sigmoid colon method, have problems with less lubrication. Patient amnion 2 and neovaginal 2 in early period after surgery require jelly lubrication before their intercourse, but they are no longer using it now. In addition, the form of neovagina rugae in post-operation using sigmoid method is even more than those in amniotic method. This is in line with previous studies of MRKH vaginoplasty patients using amniotic media that require artificial lubrication at the beginning of sexual intercourse (Fotopoulou et al., 2008). In contrast to the amniotic method, the research on patients using sigmoid colon method will excrete the excessive vaginal discharge in the first 8 weeks postoperative which will then decrease (Rawat et al., 2010).

Table 3. Evaluation on Post-CONVENTIONAL operation

Patient Name/ RM No.	Evaluation	Length	Width
SIGMOID 1	3 months	12 cm	3 cm
AMNION 1	3 months	7 cm	3 cm
AMNION 2	4 months	7 cm	3 cm
CONVENTIONAL 1	6 months	6 cm	4 cm
CONVENTIONAL 2	2 months	7 cm	3 cm
CONVENTIONAL 3	3 months	7 cm	3 cm

Sexual Function of Neovagina Patients based on FSFI (Female Sexual Function Index)

Patients of neovagina operation with amniotic and sigmoid method had relatively the same sexual function (FSFI of sigmoid method: 29.8, average amniotic method: 27.06) and both were in good category (above 23). Patients number sigmoid, amnion 1 and neovagina 3 who have sexual intercourse before and after neovagina operation show that neovagina operation improves their sexual function from bad to good (FSFI is less than 23 to be more than 23, p: 0.00652). The improving appearance of sexual intercourse is also felt by their spouses. From two out of three couples of patients who couples who have the surgery after marriage) state that they feel more satisfied when having sexual intercourse after the operation.

Fotopoulou's study has compared the sexual function after neovagina operation using amniotic method with those using sigmoid method and normal women, and the results are not different (total scores on those using amnion is 30.0, sigmoid is 28.1, and normal women is 30.2) (Fotopoulou et al., 2008).

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

Table 4. Total FSFI (Female Sexual Function Index) Score

Patient Name/ RM No.	Total score	
	Pre	Post
SIGMOID 1	15.1	29.8
AMNION 1	16.2	28.9
AMNION 2	15.1	26.2
CONVENTIONAL 1	-	26.9
CONVENTIONAL 2	-	27.1
CONVENTIONAL 3	-	26.2

Sexual Distress Level on Neovagina Patients based on FSDS-R

Although FSFI score of all neovagina patients was good (above 23), their FSDS-R scores were not. Although there was a significant decrease in sexual stress level before and after surgery p: 0.015 (patient sigmoid, amnion 1, amnion 2), the average FSDS-R score in patients with amniotic methods was still high, which was 14.2 (above 11), along with one patient whose score was 11 (20%). The discrepancy between FSFI and FSDS-R scores is in accordance with Carrad's research. FSDS-R score in patients with the post-sigmoid operation and manual dilatation shows scores above 11 which indicate distress in intercourse (sigmoid method shows 21.35, in which scores below 11 are 15%, compared to manual dilatation which was 18.40, and scores below 11 are 11%). The discrepancy between FSFI and FSDs-R scores in the study was found in 39% of cases. This is because FSFI assessed the sexual function aspects, while FSDR assessed the psychological aspects of intercourse.

From Liao's research, it was found that women with neovagina MRKH have lower sexual esteem (50 %), higher sexual depression (205 %), higher sexual anxiety (172 %) and higher fear of sexual relationship (146 %) (Liao et al., 2011). Therefore, although neovaginal operation significantly improves the sexual function on MRKH patients, many patients still experience stress due to their physical imperfections and sadness since they cannot give birth.

Table 5. Distress Level due to Sexual Problems on Neovagina by FSDS-R (Female Sexual Distress Scale-Revised)

Patient Name/ RM No.	Pre-operation	Post-operation
SIGMOID 1	28	11
AMNION 1	30	14
AMNION 2	25	14
CONVENTIONAL 1	-	18
CONVENTIONAL 2	-	11
CONVENTIONAL 3	-	14

Waiting Period for Intercourse can be performed after Neovagina operation

There were variations in the results between one study and another, regarding the waiting time of first intercourse after surgery and the length of time to do a manual dilatator after neovagina operation with the amniotic method. In Fotopoulou's research, neovagina patients who use amniotic method have 5.28 months for their average waiting time, between the operation and intercourse (about 2 to 7 months), and the use of dilator after operation is averagely 4.1 months (about 3-5 months) (Fotopoulou et al., 2008). In this research, the respondents are prohibited to do intercourse until 6-8 weeks after the operation and they are suggested to do dilatation in 3 to 5 months after the operation. Furthermore, in Sarwar's research, the patients, after the operation with amniotic method, are asked to use molding for three months and the next three months is only at night (Chan, Levin, Ford, Stanton, & Pfeifer, 2017). The intercourse was permitted after three months operation. Averagely, the patients had intercourse in three months after the operation. Patients who used sigmoid colon media in Imperato's study, averagely, had their first intercourse in four months after the

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

operation (about 2 months to 4 years)(Imparato, Alfei, Aspesi, Meus, & Spinillo, 2007).

The patients claimed that they did not have to wait too long between the operation and intercourse (averagely, 1.2 months). Once the surgical wound healed, they immediately had intercourse. One of the factors that influence it is that the operation is done before marriage, shortly after marriage or already married so that the patients and their spouses want to immediately enjoy the results of the operation. However, some patients claimed to continue using dilatator after the operation (averagely, 2.2 months), despite having intercourse, because of their fears of shrinking and less intercourse. Two of the patients with amniotic method complained about the painful intercourse at the beginning, so they rarely used it for sexual intercourse and chose to use dilatation. Patients with sigmoid method experienced the narrowing of vaginal introitus at the beginning of post-operation, in which the vaginal width is 2-finger narrowing. In this case, the patients had intercourse in 1 month after the operation, because they waited for a surgical wound in the abdomen to heal. These patients used their finger to widen their introitus, which in the second month of evaluation after the operation, it showed satisfactory results (2-finger widening).

Table 6. Waiting Time between Operation and Intercourse and the Length of Manual Dilatation

Patient Name/ RM No.	Waiting time between operation and	Use of dilatator
SIGMOID 1	1 month	-
AMNION 1	1 month	3 months
AMNION 2	1 month	2 months
CONVENTIONAL 1	1 month	3 months
CONVENTIONAL 2	1 month	1 month
CONVENTIONAL 3	2 months	2 months

Length of Stay in Hospital After Neovagina operation

From the data, it was found that the length of stay in the hospital was 14-15 days of treatment. The length of stay in hospital for patients with sigmoid colon method in Imparato's study was 8.3 days (about 5 to 23 days), (Imparato et al., 2007) whereas in El Sayed's study, it was 11 days (about 7 to 22 days). Meanwhile, the length of stay in hospital for the patients with amniotic method from Fotopoulou's study was 10.8 days (about 8 to 14 days) (Fotopoulou et al., 2008). One of the considerations for no difference in the length of stay in hospital between patients with amnion and sigmoid colon media is because both of them use molding with approximately the same insertion time of 10-14 days.

Table 7. Length of Stay in Hospital for Patients with Neovagina operation

Patient Name/ MR No.	Length of stay in hospital (day)
SIGMOID 1	15
AMNION 1	15
AMNION 2	15
CONVENTIONAL 1	15
CONVENTIONAL 2	14
CONVENTIONAL 3	14

Length of neovagina operation

The length of operation using amniotic media is shorter, which more or less is 54 minutes averagely, whereas those using the sigmoid colon method are about four times (3.5 hours or 210 minutes). Meanwhile neovaginal operation conventional seems faster compared to other two method.This result is in line with the results of other studies on more complex operation with colon media, by involving the expertise from department of digestive surgery that requires longer length of operation. The operation with

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

amniotic method in RSDS is still around one hour. In Fotopoulou's research, it took 24.7 minutes for amniotic method (about 20-33 minutes), (Fotopoulou et al., 2008) whereas in Sarwar's study, its surgical length is less than one hour, which is between 20 to 45 minutes9. Meanwhile, the operation using sigmoid colon method, in Imperato's research, is 145 minutes (about 95 to 250 minutes), whereas in Yang's study is 279, ± 32 minutes, and in El Sayed's study, it is 184 minutes (about 160 to 210 minutes) (Imparato et al., 2007).

Table 8. Length of Neovagina operation

Name/ MR No.	Length of Operation
SIGMOID 1	3 hours 30 minutes (210 minutes)
AMNION 1	1 hour 35 minutes (95 minutes)
AMNION 2	30 minutes
CONVENTIONAL 1	1 hour (60 minutes)
CONVENTIONAL 2	45 minutes
CONVENTIONAL 3	40 minutes

Costs for neovagina operation

The costs for operation and total costs for the surgical technique are more expensive if using sigmoid colon method. This is reasonable because it involves another division (surgery). The costs for non-surgical treatment does not differ much between those who use amniotic media and sigmoid colon media.

Table 9. Costs for Neovagina operation and Treatment in BPJS era

No	Remarks	Sigmoid 1	Amnion 1	Amnion 2
1.	Operation cost	13.850 M 9.5 M	12 M	12 M
2.	Non-surgical treatment cost	4.373 M (class 3)	5.838 Mt (class 2)	5.838 M
	Total cost	27.723 M	17.838 M	17.838 M

Table 10. Complication on Neovagina Patients

Patient Name/ RM No.	Complication
SIGMOID 1	Rupture buli iatrogenic
AMNION 1	Painful → MST, Copar, Constipation → laxadine
AMNION 2	-
CONVENTIONAL 1	-
CONVENTIONAL 2	--
CONVENTIONAL 3	-

Complications from neovagina operation

A serious complication due to neovagina operation is rupture buli iatrogenic, on the patients with the sigmoid method, which has been corrected during operation. On another side, a painful complication is postoperatively experienced by patients with amniotic method but manageable by giving strong analgesics.

One patient out of six patients who used amnion in Fotopoulou's study had complications following a postoperative urinary tract infection, whereas another patient had a rectovaginal fistula due to the shortening of the vaginal canal and continued with neovagina operation using sigmoid. Another study using amnion by Sarwar had evaluated for three months postoperatively. There was 11% of the patients experiencing narrowing complications due to non-compliance with manual post-operative dilatation. Postoperative complications of the sigmoid method can be in the form of the narrowing of vaginal introitus, which in Imperato's study, as much as 8% was successfully overcome by using dilatation, and 16% of the patients in Moundoni's study also experienced a narrowing in introitus and treated with V or Y incision.

CONCLUSION

At Dr. Soetomo Hospital, it was found that anatomical and sexual functions after neovaginal operation either conventional using amniotic and sigmoid colon media were not different. Thus, which surgical technique the

QANUN MEDIKA
JURNAL KEDOKTERAN FKUM SURABAYA
http://journal.um-surabaya.ac.id/index.php/qanunmedika

sigmoid method, depends on the needs and circumstances of the patients. Conventional and amniotic media provides the advantage of shorter surgical times and lower surgical costs. Neovagina operation with colon media does not require postoperative dilatation with better vaginal length and rugae than using amniotic media, but it has a greater risk of operation.

REFERENCES

Chan, J. L., Levin, P. J., Ford, B. P., Stanton, D. C., & Pfeifer, S. M. (2017). Vaginoplasty with an Autologous Buccal Mucosa Fenestrated Graft in Two Patients with Vaginal Agenesis: A Multidisciplinary Approach and Literature Review. *Journal of Minimally Invasive Gynecology*, *24*(4), 670–676. https://doi.org/10.1016/j.jmig.2016.12.030

Chandler, T. M., Machan, L. S., Cooperberg, P. L., Harris, A. C., & Chang, S. D. (2009). Müllerian duct anomalies: from diagnosis to intervention. *The British Journal of Radiology*, *82*(984), 1034–1042. https://doi.org/10.1259/bjr/99354802

Fedele, L., Frontino, G., Restelli, E., Ciappina, N., Motta, F., & Bianchi, S. (2010). Creation of a neovagina by Davydov's laparoscopic modified technique in patients with Rokitansky syndrome. *American Journal of Obstetrics and Gynecology*, *202*(1), 33.e1-6. https://doi.org/10.1016/j.ajog.2009.08.035

Fotopoulou, C., Neumann, U., Klapp, C., Lichtenegger, W., & Sehouli, J. (2008). Long-term effects of neovaginal reconstruction with sigmoid loop technique on sexual function and self image in patients with gynecologic malignancies: Results of a prospective study. *Gynecologic Oncology*, *111*(3), 400–406. https://doi.org/10.1016/j.ygyno.2008.09.018

Imparato, E., Alfei, A., Aspesi, G., Meus, A. L., & Spinillo, A. (2007). Long-term results of sigmoid vaginoplasty in a consecutive series of 62 patients. *International Urogynecology Journal*, *18*(12), 1465–1469. https://doi.org/10.1007/s00192-007-0358-0

Liao, L.-M., Conway, G. S., Ismail-Pratt, I., Bikoo, M., & Creighton, S. M. (2011). Emotional and sexual wellness and quality of life in women with Rokitansky syndrome. *American Journal of Obstetrics and Gynecology*, *205*(2), 117.e1-117.e6. https://doi.org/10.1016/j.ajog.2011.03.013

Lima, M., Ruggeri, G., Randi, B., Dòmini, M., Gargano, T., La Pergola, E., & Gregori, G. (2010). Vaginal replacement in the pediatric age group: a 34-year experience of intestinal vaginoplasty in children and young girls. *Journal of Pediatric Surgery*, *45*(10), 2087–2091. https://doi.org/10.1016/j.jpedsurg.2010.05.016

Rawat, J., Ahmed, I., Pandey, A., Khan, T., Singh, S., Wakhlu, A., & Kureel, S. (2010). Vaginal agenesis: Experience with sigmoid colon neovaginoplasty. *Journal of Indian Association of Pediatric Surgeons*, *15*(1), 19. https://doi.org/10.4103/0971-9261.69136

Rennie, K., Gruslin, A., Hengstschläger, M., Pei, D., Cai, J., Nikaido, T., & Bani-Yaghoub, M. (2012). Applications of Amniotic Membrane and Fluid in Stem Cell Biology and Regenerative Medicine. *Stem Cells International*, *2012*, 1–13. https://doi.org/10.1155/2012/721538

S. Robert Kovac, Carl W Zimmerman. (2012). *Advances in Reconstructive Vaginal Surgery* (2nd ed.). Lippincott Williams & Wilkins.