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MRK증후군 환자에서 변형 Singapore피판술을 이용한 질 재건

김도훈 · 변재경 · 문구현 · 방사익 · 오갑성 · 임소영

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Vaginal Reconstruction with Modified Singapore Flap in MRK Syndrome Patients

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Purpose: Mayer-Rokitansky-Kuster syndrome (MRK) is second common cause of primary amenorrhea. It is a syndrome of vaginal aplasia and Mullerian duct anomaly. Vaginal aplasia varies from agenesis of whole vagina to aplasia of upper 2/3. For reconstructing vagina, various methods are introduced. Gracilis myocutaneous flap was the first attempt in that the flap is used in vaginal reconstruction. Various flap-based vaginal reconstruction methods have been introduced. Modified Singapore flap (pedicled neurovascular pudendal thigh fasciocutaneous flap) is one of those methods that used posterior labial artery as pedicle, and pudendal nerve branch as sensory root. As its donor lies on inguinal crease that is easily hidden and there are benefits on sexual intercourse by early sensory recovery, authors think that modified Singapore flap is effective for young MRK syndrome patients.

Methods: Eight patients underwent surgery between 2008 and 2010. The flap was designed on both groin area with external pudendal artery branch as a pedicle. All flaps were fixated in pelvic cavity with absorbable suture, and additional compression on neovaginal wall was supplied by polyvinyl alcohol sponge (Merocel®).

Results: All patients were successfully reconstructed without flap related complications such as congestion or partial flap loss. The average size of the flap (each side) was 69.34 cm^2. Polyvinyl alcohol sponge (Merocel®) was inserted into neovagina for 5 days on every patient. One case of rectal laceration was occurred while making pelvic pocket by OBGY team. Other complications such as lumen narrowing, wound contracture or vaginal prolapsed were not reported during 8 months of follow up.

Conclusion: Modified Singapore flap is a sensate fasciocutaneous flap that is thinner than other myocutaneous flap such as VRAM, and more durable over skin graft. Therefore this is a good choice for vaginal reconstruction in MRK syndrome. And known complications of Modified Singapore flap could be reduced with careful procedure and mild compression techniques.

Key Words: Vaginal reconstruction, MRK syndrome, Modified Singapore flap

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I. INTRODUCTION

Mayer-Rokitansky-Kuster syndrome is a complex of vaginal aplasia and Mullerian duct anomaly. The spectrum of vaginal aplasia varies from agenesis to absent of upper 2/3.[1] The first vaginal reconstruction was reported by Frank et al.[2] who used vaginal stent for serial dilatation of vaginal pocket. Although the reconstruction technique reported by Frank et al. was non-surgical method, other various surgical reconstruction techniques have been introduced by many surgeons. On 1949, McIndoe et al. reported McIndoe operation which used split thickness skin graft for vaginal reconstruction.[3] Disadvantages of McIndoe operation were the fragility of skin graft and contracture of the neo-lumen. To overcome these disadvantages of split thickness skin graft-based reconstruction, flap-based reconstruction by gracilis myocutaneous flap was introduced in 1976 by McCraw et al.[4] But the other problems of flap-based reconstruction such as flap tip necrosis and congestion were observed.

There are several aspects which must be considered to be ideal vaginal reconstruction, such as single stage reconstruction, flap stability, function, angle, sensory gain, and minimal donor site morbidity.[5] Wee and Joseph reported pedicled neurovascular pedendal thigh fasciocutaneous flap,[6] which was based on posterior labial artery as a pedicle and including pudendal nerve branch

to gain sensation. It was named Singapore flap. In 1992, Woods and his colleague reported the modified Singapore flap.[7] It was performed by releasing the labia majora and allowing them to retract anteriorly, thereby facilitating rotation of the flap. But significant studies showed that vertical rectus abdominis myocutaneous flap (VRAM) based vaginal reconstruction technique is superior than modified Singapore flap in overall complication rate and flap related complication rate.[8] But it was in general pelvic reconstruction, not limited in MRK reconstruction. Authors have thought that the modified Singapore flap can be more effectively applied to young women with MRK syndrome than VRAM. Because MRK syndrome patients have no pelvic dead space like pelvic cancer patients. Also the large abdominal scar which is followed by VRAM, seems to be critical to young patients. So authors reported the experiences of vaginal reconstruction of 8 MRK syndrome patients with our surgical tips for preventing complications.

II. MATERIALS AND METHODS

A. Patients

From 2008 to 2010, 8 MRK syndrome patients underwent modified Singapore flap. Their ages ranged from 13 to 29, and their initial symptom was primary amenorrhea. All of patients were diagnosed by OBGY physician with chromosomal study and magnetic resonance image. The results of chromosomal studies were 46, XX for all patients. Internal organ anomaly other than uterus was found in 1 case as ectopic crossed kidney. All patients had normal looking distal vaginal introitus including labium, but the inner vagina was formed as fibrous band on magnetic resonance image. There was no anomaly on ovary, but in all of 8 cases, uterus anomaly were observed. 3 cases were absent of uterus, 1 case was hypoplastic uterus, 1 case was bicornuated uterus, and 3 cases were unicornuated uterus (Table I).

B. Surgical methods

The surgery was performed under general anesthesia in lithotomy position. Initial step was making pocket in pelvic cavity by gynecologist. Through the cruciform incision on introitus, space between bladder, urethra and rectum was dissected. After making pocket, plastic surgeons started with designing Singapore flap in appropriate size on both groin creases. The medial margin of the flap was laid on the lateral margin of the hair bearing portion of the labia majora and the flap base was placed on posterior end level of vaginal introitus. The unilateral flap width was 7~9 cm and the length was 9.5~13 cm. The flap elevation was started from distal, medial margin under the plane of adductor muscle fascia without muscle component. The elevation was formed from anterior to posterior by cutting lateral edge. Around the flap base area, about 2 cm of subcutaneous dissection was performed peripherally to make flap mobility better. The similar size and shape of flap was elevated from the other side of the groin. Labia majora was released and through here, elevated flaps on each groin were moved to center where the vagina pocket was made. Both flaps were sutured together from medial margin with 4-0 Vicryl® to distal and lateral margin by side to side manner. With these sutures, two flaps were changed to single inside-out pouch. The pouch was pushed into the pelvic pocket where gynecologist was made (this is different from dead space in pelvic exenteration surgery), and turned into neovagina. One anchoring suture was made between neovaginal outer tissue and pelvic cavity to prevent prolapse of neovagina. Occasionally, the skin of the Singapore flap under labia majora tunnel was de-epithelized instead of labial releasing cut, and

Table I. Patient Demographics

Patient	Age(yr)	Sex	Karyotype	Uterus	Combined anomaly
1	20	F	46, XX	Absent	None
2	27	F	46, XX	Bicornuate	None
3	23	F	46, XX	Absent	None
4	13	F	46, XX	Unicornuate	None
5	29	F	46, XX	Absent	Ectopic crossed kidney
6	29	F	46, XX	Hypoplastic	None
7	25	F	46, XX	Unicornuate	None
8	15	F	46, XX	Unicornuate	None

Fig. 1. Flap elevation and transposition into cavity. Both labium are undermined and let flap pass under through it.

Fig. 2. Flap inset. Both flaps are joined as a tube and pushed into cavity.

the margin of the neovagina skin and the margin of the tunnel of labia majora were sutured with 4-0 Vicryl® (Figs. 1, 2). Donor sites on both groin creases were primarily closed with 4-0 Vicryl® and 4-0 Nylon®. Hemovac® drains were applied to vaginal pocket and both groin donor sites. In the lumen of neovagina, polyvinyl alcohol sponge (Merocel®) was inserted with lubricant and inflated by saline to make mild compression the space between pelvic cavity and neovaginal outer wall (Fig. 3).

III. RESULTS

The average size of flaps was 69.34 cm^2, length was

Fig. 3. Post operation status with the polyvinyl alcohol sponge (Merocel®). Both donor sites are closed primarily with Hemo-Vac drain. Foley catheter is applied.

Fig. 4. 80 days after the operation. Scar widening and tightening is tolerable.

11.66 cm, and depth was 6.6 cm. Merocel® was packed for 5 days on every patient. Average hospital days were 25.37 days, and follow up periods were ranged from 2 months to 15 months (Table II). There was no flap related complication, such as congestion, partial flap tip necrosis and total flap necrosis, during both admission period and follow up period. Other surgical complication was occurred in 1 case. It was rectal laceration which occurred during pocket dissection in the pelvic cavity by gynecologist. It was managed with primary closure and applying one week of rectal tube. Lumen narrowing was observed on 3rd months of follow up period in one case, which was managed with dilation by trimmed syringe for several weeks. This was maybe due to secondary healing of the partial dehiscence of the flap margins. Although the donor site scar was recognizable, it was located in hidden area and patients were tolerable to scar (Fig. 4).

Table II. Flap Information

Patient	Flap size (cm) / unilateral	Depth	Hospital day / Complication
1	5.5 × 12.5	6.5	20 days
2	5 × 14	6	30 days Rectal laceration
3	6 × 12.5	6.5	22 days
4	7 × 12.5	7	24 days
5	6.5 × 12	6.5	21 days
6	5 × 13	7	21 days
7	5 × 13	7	21 days
8	4.5 × 13	6.5	21 days

IV. DISCUSSION

The aims of this study were to report the safety and effectiveness of modified Singapore flap in MRK syndrome patients. The modified Singapore flap was first described by Woods on 1992. The modification is performed by releasing the labia majora, allowing them to retract somewhat anteriorly, and rotating the flap into position. 7 Authors experienced a few cases of other vagina reconstruction methods, McInode operation, before modified Singapore flap but the result was disappointing because of several complications such as graft loss, vagina stricture (lumen narrowing), and discomfort from long-term use of stent, and etc. To solve those problems, authors have tried modified Singapore flap. All flaps were survived without flap related complications, and the depth and angle of neovaginas were successful. As it was described by others, this operation was relatively fast and blood loss was minimal.[5-7] Donor site scar is also an advantage of this technique because it lies along the crease of groin. The other advantages of this technique are absent of stent usage after reconstruction. Previous techniques need post-operative stent in the neovagina lumen to prevent contracture of walls,[3] but modified Singapore technique doesn't need stent as the walls of neovagina are flap. Park et al. have suggested that the flap length has to be less than 12 cm in order to avoid the exposure of large vessels of femoral area,[5] but authors have faced to no problems with vessel exposure while elevating flap up to 13 cm.

Extrusion of flap (prolapsed vagina) is being continuously reported problem.[5,8,9] To prevent this complication, authors tried two simple maneuvers. The one was anchoring suture between flap and pelvic cavity, and the other was Merocel® packing in neovagina. As the neovagina is always placed against the gravity, authors think that adhesion between the wall of neovagina and pelvic cavity is important in preventing vaginal prolapse in initial postoperative period. So we tried the polyvinyl alcohol sponge (Merocel®) packing to give gentle compression to the wall of neovagina, which can minimize the dead space and help adhesion between pelvic wall and flap wall in initial postoperative period. It is hard to proved statistically as numbers of cases were small, but further trial can prove effect of Merocel® or other mild compression, against vaginal prolapse.

Woods have reported a large series of vaginal reconstruction study in 2004. In that study, vertical rectus abdominis myocutaneous flap (VRAM) was reported to be superior to modified Singapore flap and gracilis myocutaneous flap, because of its ability to obliterate the pelvic dead space and was selected as a primary option for vaginal reconstruction. In that study, most cases were pelvic cancer patients and there must have been large dead space. But in MRK syndrome patients, there is no large pelvic dead space but artificial pelvic tunnel. It means that we only need resurface the wall delicately with thinner flap instead of space occupying bulky flap. Additionally, modified Singapore flap is superior in donor site scar as it doesn't involve abdominal area. With combining tips as described above, authors could avoid complications which had previously reported.

V. CONCLUSION

Although the modified Singapore flap is less effective in obliterate the pelvic cavity than the VRAM based vagina reconstruction, it can be used effectively in MRK syndrome patients as they don't need pelvic obliteration, but need fine resurfacing of neovaginal pocket. The

advantage of donor site scar and sensory gain from modified Singapore flap is important in young female patients like MRK syndrome patients. Although more cases have to be analyzed, modified Singapore flap can be the first choice of young vaginal aplasia patients.

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