Bilateral Pudendal Thigh Fasciocutaneous Flap Vaginoplasty in Mayer Rokitansky Kuster Hauser Sydnrome: A Step-by-Step Approach

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The surgical management in a case of vaginal agenesis is technically challenging and requires a strong demand for expertise. The objective of this report was to record the hospital's first experience in vaginal reconstruction using Bilateral Pudendal Thigh Fasciocutaneous Flap. Presented here is a case of 23-year old, female who consulted for primary amenorrhea who plans of getting married soon. Physical examination revealed normal secondary sexual characteristics with absent vagina. Imaging revealed an absent uterus with normal ovaries and distal vagina that ends in a blind pouch. Patient underwent vaginal reconstruction with no post-operative complications. Follow-up revealed incision site infection but eventually resolved with oral and topical antibiotics. Four weeks post-operatively, good wound healing was noted, vagina was 6 centimeters in length, admits two fingers on internal examination. Overall, the authors' first neovaginoplasty using Bilateral Pudendal Thigh Fasciocutaneous Flap was a technically safe procedure with good cosmetic outcome.

Key words: Bilateral pudendal thigh fasciocutaneous flap vaginoplasty, Mayer Rokitansky Kuster Hauser sydnrome

Introduction

Vaginoplasty and flap procedures are indicated in cases of congenital disorders presenting with abnormal genitalia such as Mullerian anomalies and in adults after a surgery for malignancy or trauma. The goal of surgical vaginal creation is to create a potential space between the bladder and rectum that provides a normal looking vaginal opening and to facilitate a pain-free sexual intercourse using a technique which is simple, reliable, and applicable to most of the patients.[1] Mullerian anomalies include vaginal agenesis which is a rare congenital condition and may be an isolated anomaly, or as part of more complex anomalies, like Mayer Rokitansky Kuster Hauser (MRKH) syndrome with an incidence of 1 per 4,500 to 5,000 females. It is characterized by absence of the vagina, uterus or both in women with normal secondary sexual characteristics.[2] The vaginal dimple or pouch seen in MRKH usually measures around 1-4 cm. The presence of functional ovaries and fallopian tube is noted, thus, affected individuals will have pubertal growth and development as well as ovulation. They present with primary amenorrhea, at times, periodic abdominal pain and difficulty in sexual intercourse. Thus, diagnosed patients with MRKH suffers from sexual and psychological distress.

Although this condition is psychologically devastating, its anatomical defects can be surgically reconstructed. Following the diagnosis, surgical intervention allows the patients to have a normal sexual function. On the reproductive side, adoption and assisted techniques can be used.

There are various methods that can be used in the creation of the neovagina. It could be surgical or non-surgical. Non-surgical options include the use of dilators and either intermittent pressure by the patient (Frank method) or passive pressure applied by bicycle seat beneath the perineum (Ingram method). Surgical procedures include simple and multi-staged procedures, which only differ in the type of lining that is used.[1] Some make use of the large bowel, small bowel (Baldwin Procedure),

amniotic membrane or split skin graft (McIndoe) in the creation of neovagina, interposition of intestinal segments, such as ileum or sigmoid, and more recently the pudendal thigh fasciocutaneous flaps. In general, non-operative techniques require a great deal of patient motivation and participation, with functional success achieved after many months of effort. Operative techniques hold the distinct advantage of being faster, but most require hospitalization and significant risk for perioperative morbidity, such as infection and graft rejection.[3]

The case involved a young woman who planned to get married soon. The goal of surgery was the creation of a functional sensate copulatory vagina. Bilateral pudendal thigh fasciocutaneous flap was the procedure of choice because of its mentioned advantages and the availability of a plastic surgeon working together with the reproductive endocrinologist. This procedure was done on a few cases in other regions in the country but present case is the first ever known to be carried out in the region. The objective of this report was to document the hospital's experience in creating a neovagina using bilateral Pudendal Thigh Flap Vaginoplasty in a patient with Mayer Rokitansky Kuster Hauser syndrome.

The Case

A 23 year old, female, Filipino, presented to the Obstetrics and Gynecology Outpatient Department with primary amenorrhea unremarkable family and past medical history. She noticed thelarche at 11 years and developed pubic hair at 13 years old . There was no growth stunting. On examination, she was 147 cm tall, 42.5 kg and had normal appearing secondary sexual characteristics with presence of pubic hair and axillary hair (Tanner Stage 4). The breast examination showed Tanner Stage 4. No dysmorphic features were noted. Vaginal examination showed a blind vagina with a vaginal dimple measuring 2 cm in depth (Figure 1). Uterus was not palpable.

Transrectal ultrasonographic examination revealed an absent uterus (Figure 2) and a tubular muscular structure measuring 2.1cm x 2.0cm x 2.0cm posterior to the urinary bladder and anterior to the rectum (probable uterine anlage/rudimentary

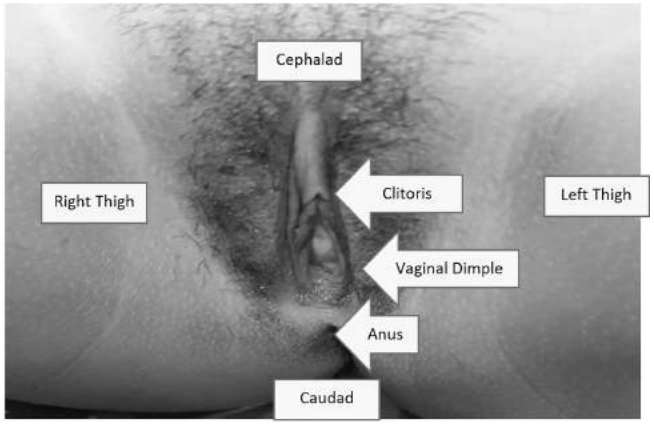

Figure 1. Absent vaginal opening upon examination.

uterus), with normal right ovary with right adnexal cyst probably paraovarian cyst measuring 0.7cm x 0.8cm x 0.6cm (Figure 4a-4b) and normal left ovary containing follicles (Figure 5). Other findings revealed that the distal vagina measured 1.7cm x 1.7cm and ends in a blind pouch. Bilateral kidneys were normal. The urethra, urinary bladder mucosa and rectum were intact (Figure 3). Sonologic findings were suggestive of Mayer Rokitansky Kuster Hauser Syndrome Type 1. The hormonal profile was within normal limits: FSH of 4.36 mIU/mL, compatible with females during follicular phase/ luteal phase, and serum testosterone of 0.43 mIU/mL. The patient has no problems regarding her physical appearance, since the secondary characteristics were all normal. The concern at that time was the absence of menstruation and a vagina, and how she would be able to have a satisfying sexual intercourse since she already planned for a marriage with her longtime partner. In order to immediately address this need, the initial plan was to do a McIndoe Procedure. Patient was then referred for co-management with Plastic Surgery service for possible skin grafting for the McIndoe procedure. Pre-operative interdepartmental conference was held to discuss the case.

During the multidisciplinary conference attended by Reconstructive and Plastic Surgery consultants, Surgery residents, Reproductive and Endocrinology specialist, Ultrasonologists and Obstetrics and Gynecology consultants and residents, it was agreed upon that the procedures to be performed would: creation of a neovagina

to be done by the Reproductive Endocrinology specialist to be followed by bilateral pudendal thigh fasciocutaneous flap vaginoplasty by Plastic Surgery service.

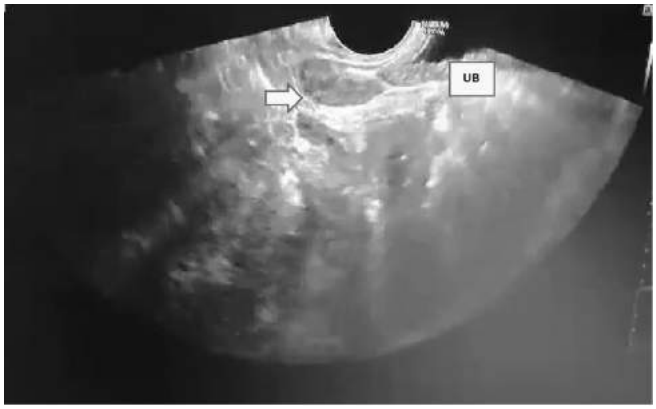

Figure 2. Transrectal sonogram showing the tubular muscular structure. UB- urinary bladder.

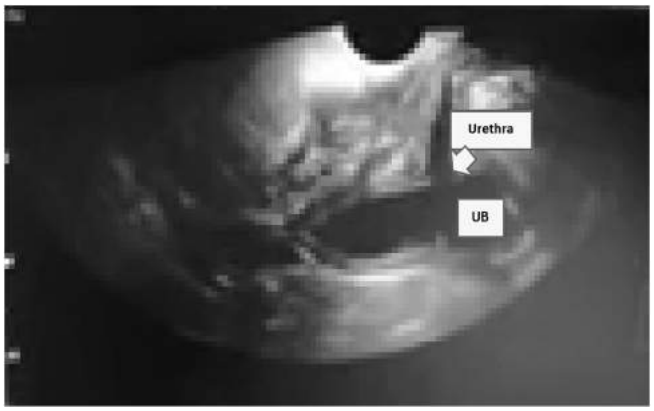

Figure 3. Transrectal sonogram showing the urinary bladder and urethra.

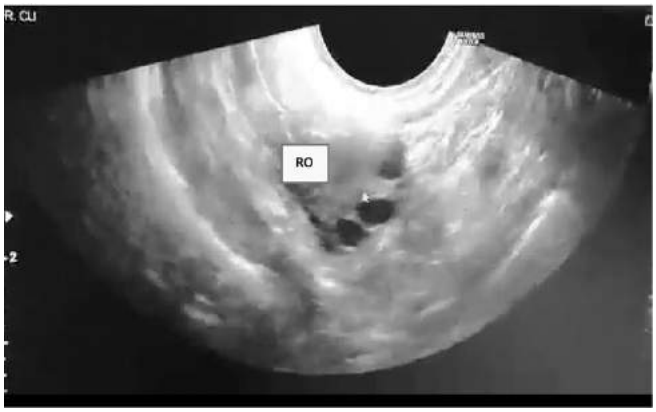

Figure 4a. Transrectal sonogram showing the right ovary with follicles. RO-right ovary.

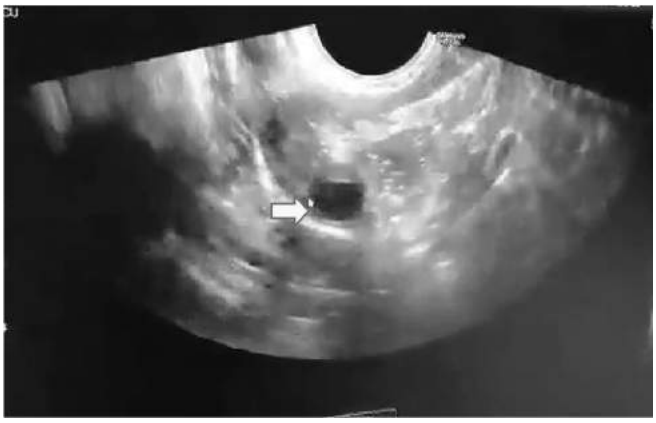

Figure 4b. Transrectal sonogram showing the right paraovarian cyst.

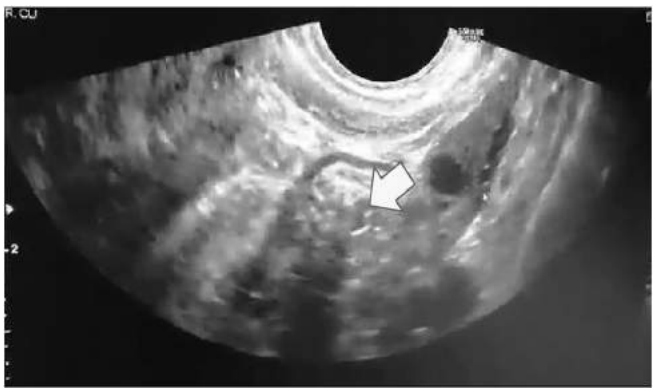

Figure 5. Transrectal sonogram showing the left ovary.

Patient was admitted as a case of G0, Mayer Rokitansky Kuster Hauser syndrome. Preoperative work-up was done. Laboratory examination included complete blood count, blood typing, thyroid stimulating hormone, serum electrolytes (sodium and potassium), renal function tests and all showed normal results. Chest X-ray was also normal. Patient was then scheduled for bilateral pudendal thigh fasciocutaneous flap vaginoplasty under combined spinal and epidural anesthesia.

Patient was placed in a dorsal lithotomy position. Prophylactic antibiotic was given. Creation of neovagina under ultrasound guidance was first done by the REI specialist and urogynecologist with the help of an OB-sonologist. The vaginal dimple was identified (Figure 6). Labia was retracted with Allis clamps and transverse incision was made in the area where the vaginal opening should be. A vesicorectal space was created by blunt and

occasional sharp dissection to create a space about 2 cm in diameter between the rectum and the bladder with pelvic ultrasound guidance, all the time ensuring hemostasis (Figure 7). A Vicryl 0 suture was placed at the apex of the dissection, just 1 cm from the muscular uterine remnant identified on ultrasound, which would be used for suspension of the flap. Meticulous hemostasis of the neovaginal space was done before turning over to Plastic Surgery service, who then took over to perform the pudendal thigh fasciocutaneous flap vaginoplasty. The creation of the vesicorectal space took about 30 minutes.

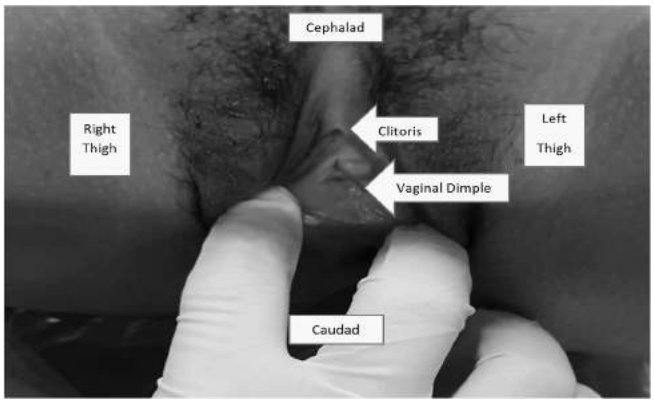

Figure 6. Pre-operative identification of the vaginal dimple.

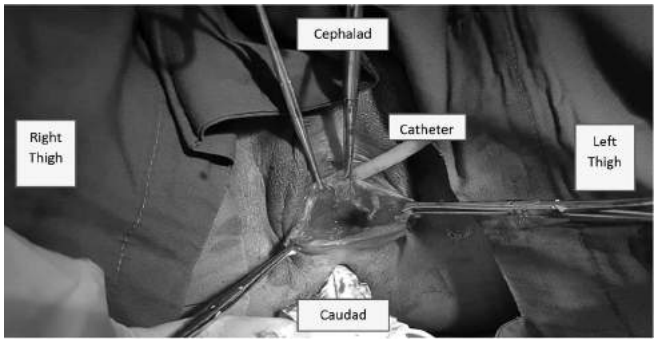

Figure 7. Open space between bladder and rectum after blunt and sharp dissection of the vesicorectal space.

The flap was marked with a length and breadth of 12cm x 5cm, base at the level of introitus extending from lateral to hair bearing part of labia majora across groin crease to medial thigh (Figure 8). It was arterialized throughout by the posterior labial artery and deep external pudendal arteries. Incision was made around planned flaps until fascia was undermined. The labia were lifted off the pubic rami and perineal membranes and flaps from both sides were tunnelled under labia (Figures 9 & 10). This was safe for the posterior labial nerves as they had entered the labial fat far posteriorly. Clitoral nerves were also in no danger because they did not pass through the superficial perineal pouch and coursed through the deep perineal pouch to reach the clitoris. Bilateral flaps were sutured to each other to create neovagina and superior portion was anchored to the apex of the neovaginal tunnel using the previously placed Vicryl 0 suture . Inferior portion of the flaps were sutured to labia after de-epithelialization. Wound closure was done using vicryl 3-0 . Bilateral Jackson Pratt drain was left in place. Skin closure subcuticular with vicryl 3-0 and nylon 3-0 (Figure 11). The whole procedure took about 3 hours, with an estimated blood loss of 200 cc.

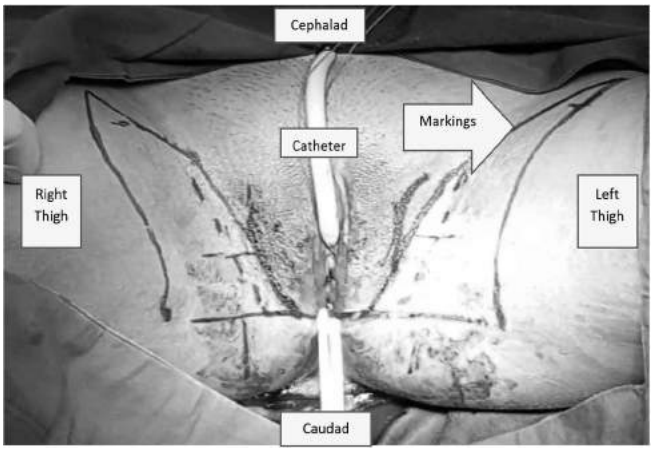

Figure 8. Surface marking of the pudendal thigh flap.

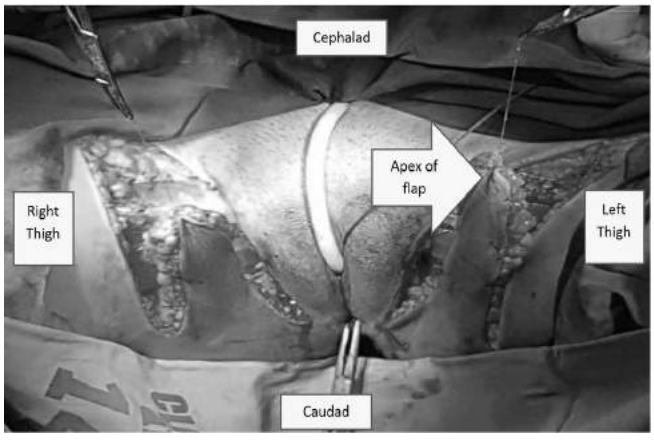

Figure 9. Raising of the flap from the apex.

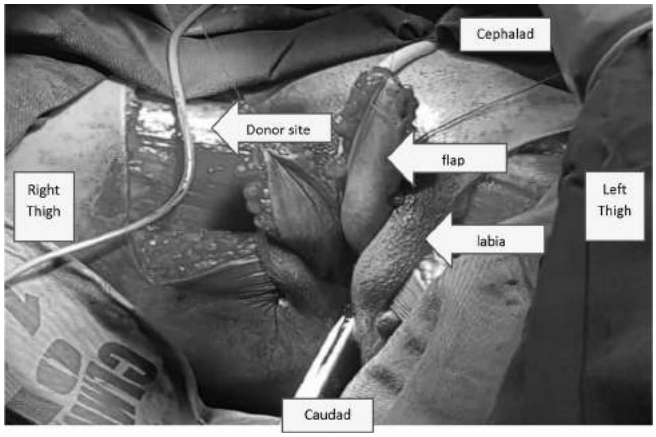

Figure 10. Pudendal flaps turned under the labia.

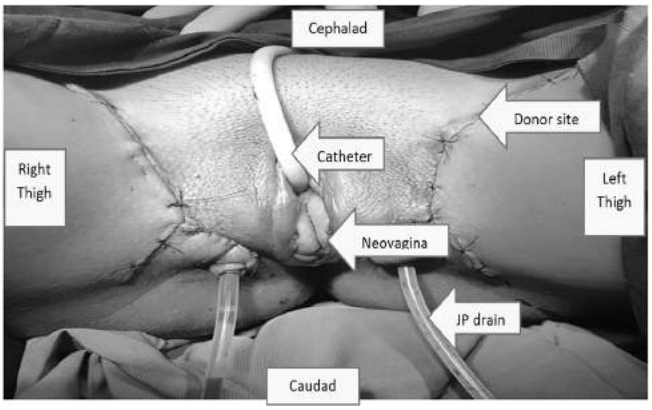

Figure 11. Immediately post-operative picture with JP drains.

Post-operatively the patient was kept in bed for 48 hours. Urinary catheter was maintained for 48 hours. The vaginal canal was washed with normal saline everyday and broad spectrum antibiotics were given for 1 week. JP drains were removed at post-operative day 3 (Figure 12) and patient was discharged at post-operative day 5.

Postoperative follow-up after 2 weeks revealed healed operative site with minimal pus discharges with vaginal length of 6 cm and admits 2 fingers (Figure 13). Patient was given antibiotics for 7 days. Follow-up visit 4 weeks post-operatively, revealed no signs of infection and no contractions. The vagina admits 2 fingers and is 6 cm in length (Figures 14a & 14b]). Sensation was noted upon entrance of the finger used during internal examination about 6/10. The patient started using her neovagina 12 weeks post-surgery with good sexual satisfaction.

Figure 12. 3rd day post operative. JP drains removed.

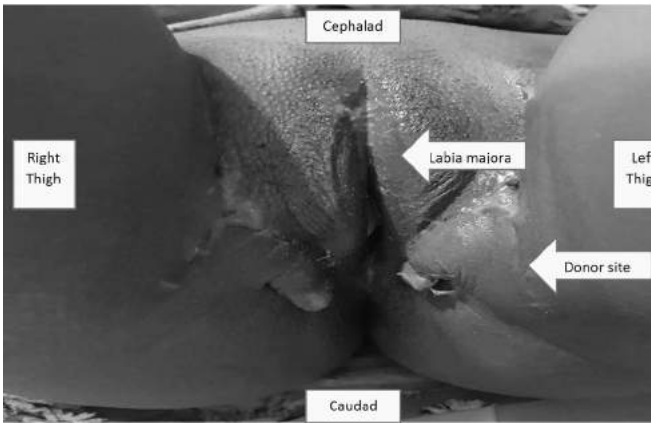

Figure 13. Postoperative picture after 2 weeks.

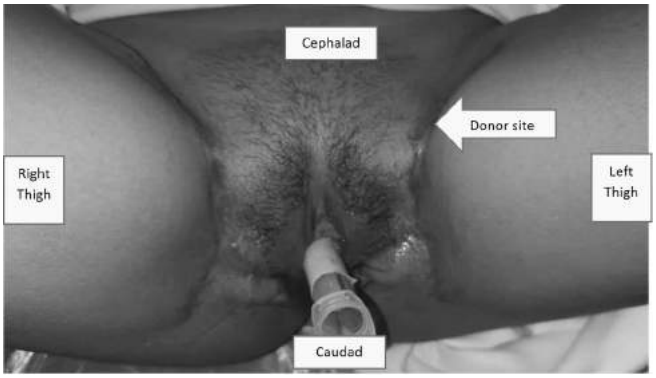

Figure 14a. Post-operative picture after 4 weeks.

Discussion

Vaginal agenesis is a rare condition with devastating repercussions on fertility and sexual function. It usually occurs in conjunction with an absent uterus, for example, Mayer Rokitansky

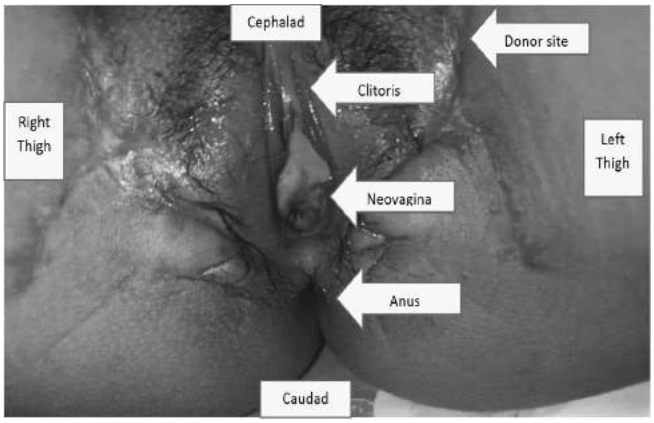

Figure 14b. Post-operative picture after 4 weeks, with vaginal length of 6 cm, admits 2 fingers.

Kuster Hauser syndrome and Complete Androgen Insensitivity syndrome. In addition, genital tract anomalies are associated with other conditions that may involve the urinary and gastrointestinal system such as anomalies from the cloacal and anorectal area.[4] These patients suffer distortion of body image, anxiety, depression, interpersonal sensitivity, and face a lot of psychological distress such as interpersonal alienation at diagnosis.[5,6,7] Treatment options may either be non-surgical or surgical which includes creating a neovagina, which can be offered to patients when they are emotionally mature and ready to commence sexual activity. Thus, interventions on these cases need to be tailored based on individual needs, motivation and options available.[4,7] In this case, the patient chose to undergo surgical intervention, as she is contemplating marriage with her partner.

Mayer Rokitansky Kuster Hauser syndrome (MRKH syndrome) or vaginal agenesis has an incidence of 1 per 4,500 to 5,000 females. Mullerian Agenesis is caused by embryologic underdevelopment of the mullerian ducts, with resultant agenesis or atresia of the vagina, uterus, or both.[8] The female reproductive tract develops from a pair of Müllerian ducts. The following structures are derived from Müllerian ducts: uterus, cervix, fallopian tube and the upper two-thirds of the vagina. The ovaries and lower third of the vagina have different embryological origins. Ovaries are derived from germ cells that migrate from the primitive yolk sac while lower one third of vagina is derived from sino-vaginal bulb. Normal development of the Müllerian ducts occurs in three phases: organogenesis, fusion and septal resorption. During first phase i.e. organogenesis, bilateral Müllerian ducts are formed. Failure of this phase results in agenesis or hypoplasia of uterus or unicornuate uterus. During second phase, fusion of Müllerian ducts leads to formation of uterus with a central septum. Failure of this step results in a bicornuate or didelphys uterus. Septal resorption is the third phase during which resorption of the central septum occurs. Failure of this stage results in a septate or arcuate uterus.[9]

Patients with MRKH are 46, XX females who undergo normal puberty since they have normal ovarian function. They demonstrate normal breast and pubic hair development. Hymenal ring and vaginal dimple are usually present because these structures are derived from the urogenital sinus.[10] In the present case, the patient had normal female secondary characteristics, as evidenced by the presence of pubic hair, axillary hair, and normal breast examination. Upon pelvic examination, only a vaginal dimple with a depth of 2 cm was noted.

MRKH syndrome, which represents 5–10% of congenital anomalies, may be considered as a result of a failed development between the fifth and the sixth week of pregnancy and of a consequent fusion on the median line of Mullerian ducts. The condition is rare, and it is known to be the second most common cause of primary amenorrhea, next to gonadal dysgenesis. As in this case, the patient does not have any affected relative. Aside from the embryological cause of MRKH, other factors include genetic causes, fetal/ maternal mutation of GALT enzyme, and the involvement of HOXA gene and WNT-4 gene. HOXA-10 gene is associated with development of uterus, HOXA-11 gene with fallopian tube and cervix, HOXA-13 gene with vagina. HOXA gene is also found to be associated with development of kidney, bone and vascular structures, thus explaining the association of MRKH syndrome with other anomalies.[9]

It is subdivided into two types: Type I (Isolated) or Rokitansky sequence, and type II or MURCS association (Mullerian duct aplasia, Renal dysplasia, and cervical somite anomalies).[11] The former type (Mayer-Rokitansky-Kuster-Hauser syndrome type I) has a prevalence of 1-9/100 000 and is most often diagnosed among adolescents with primary

amenorrhea as the first symptom in otherwise normal development of secondary sexual characteristics and normal external genitalia.[12] The latter type is more frequently associated with renal, vertebral, and to a lesser extent, auditory and cardiac defects.[19] The patient in this case belongs to type I showing normal bilateral kidneys with no skeletal abnormalities and no hearing abnormalities.

There are various differential diagnoses that one can think of based on primary amenorrhea alone, but it can be sorted out based on the clinical information and diagnostic evaluation. Imperforate hymen and transverse vaginal septum can also present with primary amenorrhea but these anomalies would have an intact uterus. The presence of a normal endometrium in these cases would present with monthly hypogastric pains coinciding with the onset of menstrual flow. In this case, the patient had an absent/ rudimentary uterus with inactive endometrium noted on ultrasound and does not experience hypogastric pain.

Another differential is that of an isolated vaginal atresia, but this one also has uterus with intact endometrium. Fertility can be gained by surgical vaginal reconstruction. Unlike in MRKH, where there is absent or only rudimentary uterus with non-functional endometrium, and fertility is not gained even after creating a neovagina.

It is usually easy to differentiate these individuals from those with androgen resistance by the presence of normal pubic hair pattern. Because women with congenital absence of the uterus are endocrinologically normal females, and those with androgen resistance are endocrinologically male, with male testosterone levels and an XY karyotype, the differential diagnosis is easily made.[13] As in the present case, there is no feature suggestive of hyperandrogenism. The levels of testosterone and FSH were checked and were within normal range for females. Karyotyping was not done due to financial constraints.

Initial evaluation of patients diagnosed with MRKH includes testing for the hormonal status including FSH, testosterone and karyotyping. Radiologic studies such as translabial, transrectal or transabdominal two dimensional or three-dimensional ultrasonongraphy can be done to assess for the presence of midline uterus, ovaries, adnexae, and vagina. Magnetic resonance imaging can be used to assess presence of rudimentary mullerian structures which is present 90% of the time.[8,14] The case being presented here did have a normal endocrine function of the hypothalamic-pituitary-ovarian axis. The presence of ovarian follicles is a proof of a normal ovulatory function. MRI was not done due to financial incapacity.

Laparoscopy is applied in case of doubtful diagnosis after the realization of non-invasive exams. This can define the exact morphology and anomalies of the uterus, tubes, and ovaries. It is also used to evaluate patients complaining of pelvic pain, which can be due to ovulation or endometriosis.[8]

Young women diagnosed with MRKH suffer from extreme anxiety and very high psychological distress when they are told they have no uterus and vagina. Thus counselling is recommended to both patient and family members, before and throughout the treatment.[11] Fertility options for having children should also be addressed which include adoption and assisted reproductive techniques with gestational surrogacy.[8] In this case, the patient was counselled regarding her chances to get pregnant by using her own eggs but will be needing a surrogate. Another option offered was adoption.

Treatment options can either be non-surgical by vaginal elongation or surgical. But the chosen method should be tailored to individual needs, motivation of the patient and options available.[11] As in this case, the patient chose to undergo a surgical procedure.

Non-surgical vaginal dilatation is widely considered as the first line treatment. It has physically low complication rate and offers a success rate of 75-85%. The most commonly used method is the Frank's method, which was discovered in 1983. It is done by introducing a vaginal mold as a dilator device by the physician for 20 minutes a day, progressively increasing the width and length of the dilator. The method requires the presence of a short vaginal dimple to start, to reach a functional depth and width. Some commonly cited barriers were cramping and fatigue of the patient, lack of comfort, privacy issues and lack of time to dilate daily.[15]

Different surgical techniques for vaginal reconstruction is currently available, all with the same principle, to create a potential space between the bladder and rectum.[15] A number of surgical

techniques to create a neovagina has been created and the decision of which surgical method to offer is often based on the surgeon's personal experience and preference. Referral to centers with expertise should be sought and recommended to the patient because the primary surgery is the most likely to succeed. Multiple studies have shown that subsequent surgeries increased the chance of operative morbidity with injury to surrounding organs and poor functional outcome. Interestingly, outcomes of surgical treatment were not changed by the previous use of non-surgical techniques or attempted intercourse.[15]

The most common surgical procedure used to create a neovagina has been the modified Abbe–McIndoe operation. This procedure involves the dissection of a space between the rectum and bladder, placement of a stent covered with a split-thickness skin graft into the space, and the diligent use of vaginal dilation postoperatively. Other procedures for the creation of the neovagina are the Vecchietti procedure and other laparoscopic modifications of operations previously performed by laparotomy. The laparoscopic Vecchietti procedure is a modification of the open technique in which a neovagina is created using an external traction device that is affixed temporarily to the abdominal wall. The anatomical success is 98% and functional success is 97%. The potential complications for Vechietti procedure are visceral damage during laparoscopy and an increased risk of stress urinary incontinence.[16]

Another procedure, the Davydov procedure, was developed as a three-stage operation that requires dissection of the rectovesicular space with abdominal mobilization of a segment of the peritoneum and subsequent attachment of the peritoneum to the introitus. Post-operative vaginal moulds for 6 weeks are needed and regular vaginal dilators are used until commencement of regular sexual activity. The procedure offers good anatomical and functional success. No major complications are reported other than growth of granulation tissue at the vaginal vault. The laparoscopic approach has the added benefit of clear visualisation of the anatomy, a shorter hospital stay and less postoperative pain.[16]

As Baldwin procedure, which popularized the use of various portions of the bowel such as Ileum and colon to reconstruct a vagina, bears increased mortality and morbidity associated with intraabdominal surgery, along with other disadvantages associated with the use of ileum including bleeding with coital trauma, excessive mucus secretion, periumbilical pain associated with coitus and tendency to prolapse. It is generally abandoned in favor of other safer options.[16]

Currently, flaps have been used for vaginal reconstruction. McGrawet al. used gracilis myocutaneous flap for vaginal reconstruction. Gordon, et al. described the use of distal rectus abdominis myocutaneous flap. The problems on these flaps are the extent of donor scar, the difficulty of flap positioning into the pelvic cavity and the risk of incisional hernia. Chen, et al. used the axial subcutaneous pedicle flap from the inferior abdominal wall in 30 patients. The study had better outcomes in terms of flap volume with no incidence of hernia noted.[17]

The pudendal thigh fasciocutaneous flap, first described by Hagerty et. al and Wee and Joseph in 1989, is a one-stage operation, a sensate flap based on the terminal branches of superficial perineal artery. The reconstructed vagina had the natural physiologic angle. In addition to that, its sensation is the same as the innervations of the erogenous zones of the perineum and the upper thigh through the posterior branches of the pudendal nerve and through the perineal rami of the posterior cutaneous nerve of the thigh. Furthermore, it has the same erotic sensation as the perineum and the upper part of the thigh. The donor site can be closed primarily with scar hidden in the groin crease.[17] It looks very ideal, as it has a robust blood supply and chances of necrosis are almost negligible. The technique is simple, safe and reliable, no stents or dilators required. Some disadvantages with the use of this flap is that it is technically more difficult than McIndoe procedure and requires more time. Having hair in the neovagina is among the few problems but can be dealt with laser therapy or depilatory creams.[1] The index patient has a sexual partner and plans to get married very soon. This flap offers a more satisfying sexual sensation and a more aesthetic appearance as scar is hidden in groin crease, also there is no need to use dilator or stent, thus the choice of bilateral pudendal thigh fasciocutaneous flap.

Several studies internationally were done with the use of bilateral pudendal thigh fasciocutaneous flaps for vaginal reconstruction on MRKH. These

studies concluded that the use of this flap has good cosmetic results and longer mean vaginal length and width one year post-operatively, with no stenting or dilatation required, and satisfying sexual intercourse with partner. No post-operative complications were noted.[1]

The flap can also be used in a unilateral fashion. A published study has reported about the use of unilateral pudendal thigh fasciocutaneous flap in a case of a patient with vaginal defect resembling type IIA Cordiero classification defined as defects involving the upper one-third and two-thirds of the vagina, and may result from resection of cervical and uterine malignancies. She had normal uterus and introitus. Patient underwent multiple procedures, but all had failed. After having pudendal thigh fasciocutaneous flap, she had normal menses with no local complication, and the flap offered great mobility.[17]

The pudendal thigh fasciocutaneous flap can also be used in several pathologies such as congenital vaginal atresia and after an oncologic resection such as vulvar intraepithelial neoplasia and recurrent vulvar squamous cell carcinoma as cited by Monstrey, et al. This flap can also be utilized in a unilateral fashion in patients with recurrent or complex vaginal fistulas and in patients with a defect of the posterior urethra in a heavily scarred perineum. The flap offers good anatomical and functional results.[17]

Conclusion

It is a recognized fact that in creating a neovagina, McIndoe operation is commonly used as it is a simple procedure that offers more comfort with lesser morbidity. However, the use of bilateral pudendal thigh fasciocutaneous flap for vaginal reconstruction that is made possible in the hands of an expert plastic surgeon working together with the reproductive medicine specialist, is a safe procedure of choice. It is a one-stage operation that is well-tolerated by the patient, with minimal postoperative pain and short hospital stay. A good length of the neovagina was achieved post-operatively, with remarkable anatomic and cosmetic results.

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