Pudendal Thigh Flap in Vaginal Agenesis

WALID MOSTAFA, M.D.
The Departments of Plastic and Reconstructive Surgery Unit, Tanta Faculty of Medicine, Tanta, Egypt

ABSTRACT

Objective: To evaluate vaginal reconstruction using pudendal-thigh flaps.

Methods: The pudendal-thigh flaps lateral to labia majora were elevated and brought to the midline through tunnels formed under labia majora. The two flaps were then sutured together to create a new vagina, then invaginated into a created rectovesical space.

Results: In eight cases aged 16 to 23 years with congenital absence of vagina a new vagina was constructed with neurovascular pudendal-thigh flaps. One patient developed partial necrosis of a unilateral flap and was repaired by small split thickness skin graft. In other patients all flaps survived and the neovagina was spacious and quite deep.

Conclusion: Vaginal reconstruction using pudendal thigh flaps is one of the best methods of vaginoplasty. The technique is simple, the flap is sensate and reliable, and the donor site is hidden with little secondary deformity.

INTRODUCTION

Vaginal agenesis is a rare congenital condition and is often associated with the Mayer-Rokitansky-Kuster-Hauser syndrome. Its incidence approximates 1 in 4,000 to 10,000 female births [1]. It consists of primary amenorrhea with normal female phenotype, genotype, ovarian function and endocrine status. The uterus is either completely absent or is represented by two rudimentary horns. In 8% of cases, a functional uterus with or without cervix is present, resulting in concealed menstruation and hematometra [2].

The condition carries a serious sexual, psychological, and even social burden [3,4]. The aims of reconstruction are to provide a cosmetically satisfactory introitus, a conduit for normal menstruation, and also to facilitate pain-free, enjoyable sexual intercourse [5]. Only recently, sexual function data has been included in outcome studies [6].

The ideal procedure to reconstruct the vagina should pose little risk to the patient, be a single-stage procedure with minimal donor-site morbidity, and create a vaginal canal in the correct axis and of adequate size that allows intercourse without the need for long-term postoperative dilatation [7,8].

Numerous procedures and techniques, both surgical and non-surgical, have been proposed over the years for the treatment of vaginal agenesis [9]: graduated dilators [10], split-skin-covered molds placed into the rectovesical space [11], colon interposition vaginoplasty [12], abdominal pull through technique [13], myocutaneous flaps [14], vulvovaginoplasty using labia minora [15] (eventually with or without tissue expanders) [16], more recently, the infragluteal skin flap or lotus flap [17,18] and the pudendal thigh fasciocutaneous flap [19] have been described.

The pudendal thigh flap is a sensate fasciocutaneous flap based on the terminal branches of the superficial perineal artery, which is a continuation of the internal pudendal artery [20]. Several authors [2,21-25] have reported on the use of this flap in a bilateral manner to reconstruct vaginal atresia.

The technique is simple, reliable, and safe, with no need to use stents or dilators postoperatively. The angle of inclination of the vagina is physiological, the donor site can be closed primarily, and the neovagina is sensate, with the same erogenous potential as the perineum and upper thigh [8].

We report on eight patients with congenital aplasia of the vagina reconstructed with bilateral pudendal thigh flaps.

PATIENTS AND METHODS

Between January 2007 and January 2011 a new vagina was constructed in eight patients, aged 16 to 23 years, having Mayer-Rokitansky-Kuster-Hauser syndrome with congenital absence of vagina, using neurovascular pudendal-thigh flaps. The uterus was absent in seven cases while it was
bicorneate in one case, with a huge hematocolpus in one hemi uterus and a rudimentary small one.

Surgical technique:

The patient is placed in a lithotomy position (Fig. 1), with the legs in stirrups. The bladder was catheterized.

A 2.5- to 3.0-cm U shaped incision was made in the vestibular mucosa and submucosal compact tissue across the region of the dimple, 1cm under the urethral meatus, and extended transversely to the labia minora.

A rectovesical space was formed by means of blunt digital dissection between the bladder and rectum. A Foley catheter in the bladder and a finger or Hegar dilator in the rectum facilitated the development of this plane to avoid visceral injuries. A relatively bloodless cleavage plane was found, except for a critical zone over the anterior rectal wall, about 2 to 3cm distant from the approach incision. Absolute hemostasis was obtained with the use of electrocautery and packing (Fig. 2a-2b).

Bilateral pudendal thigh flaps measuring 15 x 6cm were marked. Each flap was essentially horn-shaped, with the flare of the horn at its base. It was planned lateral to the hair-bearing area of the labia major and centered on the crease of the groin. The base of the flap was marked transversely at the level of the posterior end of the introitus, it was 6cm in width, allowing direct closure of the donor site without much undermining, while the tip was placed in the femoral triangle with a length reaching up to 15cm (Fig. 3).

The incision, beginning at the tip of the flap, was deepened through skin and subcutaneous tissue down to deep fascia on two sides, except for the posterior margin of the flap. The subfascial plane was developed, raising the epimysium of the adductor muscles with the deep fascia (Fig. 4), the deep fascia was tacked to the skin flap to prevent shearing. The deep fascia created an additional barrier, which prevented inadvertent damage to the neurovascular structures of the flap.

The flap was elevated until the posterior skin margin is reached. The margin was incised through dermis to subcutaneous tissue to a depth of 1.0 to 1.5cm, and then undermined in a plane parallel to the skin for a distance of about 4cm posteriorly to enable the flap to be transferred through 70 to 90 degrees, to meet its counterpart in the midline, and to allow the posterior skin margin of the flap to be sutured to the labia minora.

The transfer was effected by elevating the labia off the pubic rami and the perineal membrane, in order to tunnel the flaps. The labia were not denervated as the posterior labial nerves enter the labial fat pad far behind posteriorly. The clitoral nerves were also in no danger, because they do not pass through the superficial perineal pouch, but instead, course through the deep perineal pouch to reach the clitoris.

The flaps were tunneled under the labia medially with de-epithelialization of the buried part of the flap (the most posterior part) (Figs. 5, 6). The posterior suture line where both flaps meet was completed first, with the flaps everted through the introitus (Fig. 7). After the tip was reached, the anterior suture line was then begun (Fig. 8).

The tip of the cul-de-sac was then invaginated (Fig. 9) and anchored by a Vicryl 2/0 suture to the depth of the created rectovesical space. In one case, the tips of both flaps were not sutured together as the free margins were used to establish uterinevaginal continuity.

The opening of the vagina was sutured to the mucocutaneous edge of the labia minora. Drains were inserted into the cavity containing the vagina, as well as flap donor sites (Fig. 10a-10b). A condom packed with sterile cotton was introduced into the new vaginal pouch and secured by (T) bandage. The patient was kept in bed with thighs adducted for 48 hours. Urinary catheter drainage was continued for 5 days. Lower part of the flap was used to monitor its circulation postoperatively.

In one case, in which a bicorneate uterus was found with huge hematocolpus in the right hemi uterus, uterovaginal continuity was established with the right hemi uterus through laparotomy. An incision 1cm in diameter was made into the uterine fundus, allowing suction of the uterine cavity, and to help in identifying the upper limit of the atretic tissue of the cervix using 4-mm dilator. The atretic tissue was excised and anastomosis was done between the uterus and the new vagina over a T tube to provide constant drainage of the menstrual blood and a possible chance for pregnancy.

All cases were encouraged to come for regular follow-up visits every month for at least one year to assess the results as regards the length, width, appearance, sensation and function of the new vagina (Figs. 11a-11b, 12a-12b, 13a-13b).
RESULTS

Over the period between January 2007 and January 2011, the pudendal thigh flap was used to construct the vagina in eight cases with Mayer-Rokitansky-Kuster-Houser syndrome having congenital vaginal aplasia, in Plastic Surgery Unit, Tanta University Hospitals. The age ranged between 16 to 23 years, four of them were recently married having no previous idea about the nature of their abnormality.

Postoperatively, the mean length of the created vagina was 10cm and the mean width was 4cm. There was only one case of unilateral distal flap necrosis that was repaired by small split thickness graft over a packed condom. The mean hospitalization time was 7 days.

The length and width of the created vagina were assessed during the follow up visits. Minimal shrinkage occurred in the case of distal flap necrosis as the vagina reached 7cm in length and 3cm in width however it responded well to frequent dilatation and wearing cotton filled condom over night for 2 months.

All cases showed mild prolapse of the reconstructed vagina in the early postoperative period which improved completely over the following month. All the flaps also showed preserved sensation specially near the base. In three cases, hair growth was observed in the proximal neovagina area (the base of the flap), which was successfully managed with laser treatment.

The functionality of the neovagina was assessed during the follow-up visits of the married patients, they were encouraged to use small amounts of emollients or lubricating gel during the sexual intercourse and asked to evaluate their sexual life as satisfactory, adequate or unsatisfactory, and also to report any problems or dyspareunia. Sexual life was assessed to be satisfactory in two patients and unsatisfactory in the other two. In the case of uterovaginal anastomosis, unfortunately, stenosis and adhesions resulted in recurrence of abdominal colic and huge collected hematocolpus which necessitates hysterectomy one year later.

Fig. (1): Congenital vaginal agenesis in Mayer-Rokitansky-Kuster-Houser Syndrome.

Fig. (2A-B): Creation of the rectovesical space.
Fig. (3): Marking of bilateral pudendal thigh flap.

Fig. (4): Elevation of the pudendal thigh flap.

Fig. (5): De-epithelialization of the proximal 4-5 cm of the flap.

Fig. (6): Flaps are medially tunneled under the labia majora.

Fig. (7): Flaps are sutured in the midline posteriorly.

Fig. (8): Flaps are sutured anteriorly.

Fig. (9): Sutured flaps were invaginated as a skin lined tube into the previously created rectovesical space.
Fig. (10A-B): The skin lined tube was sutured to the edges of the vaginal opening and the remaining wounds were also sutured after insertion of suction drains.

Fig. (12A-B): Postoperative results.

Fig. (13A-B): Postoperative results.
DISCUSSION

Vaginal aplasia is a rare congenital anomaly of the female genital tract. The commonest cause is the Mayer-Rokitansky-Kuster-Hauser syndrome (MRKH syndrome) which occurs in one in 4000-10000 female births. It is considered to be genetic; however, the exact etiology is still unknown. The patient has normal female phenotype, 46,XX karyotype, the ovaries have normal endocrine function, the fallopian tubes are normally developed and the uterus is absent or rudimentary [2]. Other malformations associated with MRKH syndrome involve the upper urinary tract, the skeleton, the auditory system and the heart [26].

The second common cause is complete androgen insensitivity syndrome (CAIS), which affects one in 13,000 to 40,000 live births. It is attributed to a mutation in the androgen receptor gene which renders the body insensitive to testosterone and hence results in a female phenotype with female external genitalia, having 46,XY karyotype, testicular gonads, absent Mullerian structures, and a short vagina [6].

Diagnosis of the syndrome can be emotionally disturbing for the young girl, who realizes her inability to have normal sexual intercourse and pregnancy, leading to significant consequences in self-image and sexual identity as well as serious social problems considering engagement and marriage. Treating patients with vaginal aplasia requires psychological support, together with the creation of a neovagina to give the patient the prospect of a normal sexual life [9].

The aim of vaginoplasty should be the creation without excessive morbidity of a neovagina that will be satisfying in appearance, function and feeling [27]. The definition of “success” in earlier studies was either vaguely defined or mainly focused on the anatomical success [28,29]. Later studies included sexual function as an integral element of success [6].

Both surgical and non-surgical techniques have been suggested during the past century for neovagina creation. There is a lack of long term outcome data on these options and no comparative studies between them [2].

Frank technique [10] and its modification by Ingram [30] are among the most widespread performed non-surgical techniques [9]. The Frank technique involves the use of dilators, progressively increasing in diameter and length, which are placed in the vaginal dimple by the patient herself [10]. Ingram introduced a bicycle seat stool where the patient could sit and use her trunk weight to hold in place the vaginal dilators and create pressure to the vaginal dimple [30]. Although non operative techniques are associated with less morbidity than operative ones, the use of vaginal dilators is not pleasant for the adolescent patient and, furthermore, it can be applied only when a vaginal dimple of at least 3-4cm is present [9].

One of the most widespread surgical techniques performed for the creation of a neovagina was the McIndoe operation [31], in which a space is surgically created between the patient’s rectum and urethra, then a mould covered with a full-, or a split-thickness skin graft is placed in it. It has the disadvantage of shrinking of neovagina in the upper third with complete and partial vaginal obliteration [32]. Furthermore, patients are obliged to use dilators postoperatively for about a year, even if they have regular sexual activity [12].

The use of enteric segments for vaginoplasty was reported as early as 1907 by Baldwin [33]. In 1975, this technique was improved by Kun who was the first to use stapler devices for these procedures [34], the technique has then been widely used [35,36,37]. It provides adequate vaginal length, lack of shrinkage and natural lubrication. However, patients may complain of excessive mucous production, diversion colitis of the bowel vagina, prolapse [36,37] as well as frequent occurrence of necrosis that may occur due to tension on the mesenteral pedicle [12].

The Vecchietti procedure [38] dilates the vagina by passive traction on an ovoid bead or mold placed in contact with the vestibule and attached to the abdominal wall by traction wires that are threaded retroperitoneally [39]. It has numerous variations [40].

Main Disadvantages of this technique include pain resulting from the sustained traction that may necessitate hospital stay throughout the whole traction process, and instruments that are not FDA approved and may be bought from Italy or constructed as “home-made” [41].

Regional musculocutaneous flaps as gracilis myocutaneous flap was tried in 1976 by McCraw [14], then the inferiorly based rectus abdominis myocutaneous flap for both vaginal and pelvic floor reconstruction in 1988 by Tobin and Day [42]. These large tubed flaps are too stiff to create a pliable vagina [43], and therefore they are mostly
used for immediate vaginal reconstruction after radical perineal surgery [44]. They are also reported to have a high incidence of necrosis [19].

Williams in 1964 [15] and Capraro in 1972 [45] constructed a canal by tubing the skin between the labia without dissecting a vaginal canal. Some authors tried tissue expansion of the labial pocket to mobilize sufficient tissues to create the vagina [16]. A disadvantage of this technique, however, is the unphysiologic angle of the vagina, with a vaginal axis assuming a 'Kangaroo pouch' position which may prove awkward during intercourse [19]. Furthermore, opening of this neovagina is positioned too far anteriorly and vagina might not become of adequate size [27].

More recent skin flaps include the infragluteal skin flap or lotus flap [17,18], free flaps from the scapular region [46] and the deep inferior epigastric perforator flap [47].

In 1989, Wee and Joseph described a technique using neurovascular pudendal-thigh skin flaps to (re-)construct the vagina in congenital or acquired conditions. They operated on three patients with good results [19]. In 1992 Woods et al., reported similar results in eight cases using the same technique with minor modification [48]. Thereafter, several authors have reported on the use of this flap in a bilateral manner to reconstruct vaginal atresia [21,24,49]. Monstrey [8] demonstrated its versatility in congenital atresia, oncologic resection, complex rectovaginal fistulas and posterior urethral defects. Selvaggi [2] tried using this flap to restore uterovaginal continuity in two cases with vaginal atresia. Ugboro [50], used the flap in the treatment of acquired gynatresia from caustic pessaries.

In this study, the pudendal thigh flap was used to construct the vagina in eight cases with Mayer-Rokitansky-Kuster-Houser syndrome having congenital vaginal aplasia, in Plastic Surgery Unit, Tanta University Hospitals.

The age ranged between 16 to 23 years, four of these cases were recently married. We believe that these cases were not discovered earlier because of the cultural taboos limiting early diagnosis specially in rural areas from which these cases came from.

Nakhal and Creighton [6] stated that the majority of these conditions present in adolescence, which enables the patient to be involved in decisions about the type and timing of treatment. When presentation is earlier in childhood, it is accepted to defer both non-surgical and surgical methods of vaginal creation until adolescence or even adulthood, when the patient reaches physical and psychological maturity [26,51]. This allows for proper decision making and also increases the compliance with vaginal dilation therapy whether used as primary treatment or post-operative adjuvant treatment to prevent vaginal stenosis [6]. We believe that vaginal reconstruction should be performed when the patient is emotionally mature and motivated to maintain a neovagina once it is created.

In present study, the mean length of the created vagina was 10cm and the mean width was 4cm. Ganatra and Ansari [49] used the same technique in five patients, they reported easy entry of two fingers in the neo-vaginal pouch, and the vaginal length was 8cm.

There was only one case of distal flap necrosis in our study that was repaired by small split thickness graft. In their original study, Joseph and Wee [19] discussed the blood supply of the pudendal thigh flap being based on the terminal branches of the superficial perineal artery, which is a continuation of the internal pudendal artery. These branches, named posterior labial arteries, anastomose with branches of the deep external pudendal artery as well as the medial femoral circumflex artery and the anterior branch of the obturator artery over the proximal part of the adductor muscles. They emphasized that the vascular bases of the flap is to capture the adjacent territory of the deep external pudendal artery, hence the posterior labial arteries extend to the femoral triangle and the flap is nourished by this direct cutaneous system of arteries. They recommended that the deep fascia and the epimysium over the proximal part of the adductor muscles underlying the flap should be included in the flap, to prevent injury of the direct cutaneous arteries when the flap is elevated.

In the study of Monstrey et al. [8], all 31 pudendal thigh flaps survived completely. They suggested that the flap should be designed more medially and recommended delay procedure to avoid flap necrosis.

All the flaps in the present study showed preserved sensation specially near the base. Joseph and Wee [19] illustrated that the posterior part of the pudendal thigh flap retains its innervation from the posterior labial branches of the pudendal nerve, as well as from the perineal rami of the posterior cutaneous nerve of the thigh, while the anterior part of the flap near the medial corner of the femoral triangle, supplied by nerve twigs from the gen-
itofemoral and ilioinguinal nerves, may be denervated in the process of elevation. Hence, sensation would be retained only in the lower part of the reconstructed vagina. They also confirmed that the elevation of the labia off the pubic rami and the perineal membrane in order to tunnel the flaps, does not denervate the labia, as the posterior labial nerves enter the labial fat pad far behind posteriorly and the clitoral nerves do not pass through the superficial perineal pouch, but course instead through the deep perineal pouch to reach the clitoris.

Monstrey et al. [8] in their study, stated that their flaps were sensate having the same erogenous potential as the upper thigh and perineum. However, the five cases of Ganatra and Ansari [49], complained of heaviness and numbness of the new vagina.

In this study, sexual life was evaluated to be satisfactory in two married patients and unsatisfactory in the other two. We believe that technically good reconstruction as well as stable psychological status and supportive husbands all integrate to achieve satisfactory sexual life.

In one married case in this study, uterovaginal continuity was established with the bigger hemi uterus of a bicornoate uterus, to provide constant drainage of the menstrual blood and a possible chance for pregnancy. Unfortunately, recurrence of abdominal colic and huge collected hematocolpus necessitates hysterectomy one year later.

In 8% of cases of total or partial vaginal aplasia, a functional uterus with or without cervix is present, resulting in concealed menstruation and hematometra. In such cases, two surgical options are available: hysterectomy, or reconstruction of the vagina and connection of the uterus with the neovagina [2].

Creation of a uterovaginal connection was first reported by Warthon in 1938 [52]. The first pregnancy after this procedure was reported by Zarou et al., in 1973 [53].

Although some successful cases have been reported [54,55,56] which were mainly incomplete vaginal atresia cases, restoration of continuity between a functioning uterus and the vagina in cases of cervical or vaginal atresia is daunting. As a consequence, hysterectomy has been recommended as the most straightforward treatment option [57,58].

In three cases, hair growth was observed in the neovagina area, which was successfully managed with laser treatment. Giraldo et al. [59] noticed that hair growth was more a cosmetically unpleasant problem than a functional one. He also found metaplasia and nearly complete atrophy of the hair in the posterior two third of the reconstructed vagina Monstrey et al. [8], advocated preoperative laser depilation to prevent postoperative intravaginal hair growth.

**Conclusion:**

The pudendal thigh flap is a very suitable flap for vaginal reconstruction in cases of congenital vaginal atresia; having reliable blood supply, preserving the erogenous innervations of the perineum and upper thigh thus providing a sensate sexual function. The reconstructed vagina has a natural physiological angle and a correct anatomic axis to facilitate intercourse. It is ideally performed in one stage with no need for obturator or stent to maintain patency and the donor site is hidden in the groin crease.

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