Bilateral Pudendal Thigh Flap In The Creation of A Neovagina and Restoration of Utero-Neovaginal Continuity – A Case Report

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\begin{abstract}
The pudendal thigh flap is a sensate flap based on the posterior labial artery, which has shown great versatility in the reconstruction of genital anomalies as well as genital oncologic reconstruction. This report showcases the creation of a neovagina with the restoration of uterovaginal continuity in a 12-year-old girl who had vaginal atresia. She presented with concealed menstruation and cyclical abdominal pain and had a creation of a neovagina with utero-vaginal continuity using a bilateral pudendal thigh fascio-cutaneous flap. Post-operatively, she attained normal menstrual flow and a sensate neovagina with a satisfactory vaginal canal depth. Partial wound dehiscence of the flap donor site was managed with dressing and healing by secondary intention. Even in new centres, the pudendal thigh flap remains a versatile flap for managing vaginal atresia.

\textbf{Keywords} \\
Vaginal atresia, Pudendal thigh flap, Neovagina, Uterovaginal continuity.
\end{abstract}

\section*{Introduction}
Conditions requiring vaginal reconstruction can be from congenital anomalies or acquired conditions. Congenital anomalies vary from imperforate hymen, vaginal atresia, vaginal aplasia, congenital adrenal hyperplasia to other intersex states. Acquired gynatresia have mostly followed the insertion of caustic materials into the vagina [1].

Several techniques of vaginal reconstruction for both congenital and acquired cases have been described. Reconstruction aims to create a functionally and cosmetically satisfactory neovagina using a simple, reliable technique applicable to most patients [2]. Notwithstanding the surgical technique utilised, a neovagina is created as a pocket between the urinary bladder and the rectum.

Frank's nonoperative dilatation is tedious and faced with the interminable chore of vaginal stenting to maintain the dimension of the new vagina [3]. The existing dimple at the vulva should also be at least 3-4cm deep to use this option. McIndoe described the usage of split-thickness inlay grafts, which, however, also required continuous stenting. The Vecchietti procedure, the Williams vaginoplasty and its modification by Creatsas, the Davydov technique, and bowel colpoplasty are also used for vaginal reconstruction [4].

The Vecchietti procedure elongates the vaginal dimple by dragging it from the inside of the abdominal cavity. A small acrylic ball placed in the vaginal dimple is gradually pulled up by sutures that
run from the ball to an apparatus attached to the abdominal wall, passing intra- or extraperitoneally. The tension on the suture needs frequent adjustment that can be painful to the patient while also requiring regular dilation.

In the Davvdov technique, the neovagina is created using the patient's peritoneum. Closely related bowel colpoplasty utilises the bowel, thus requiring bowel resection and anastomosis. While the latter provides immediate supple tissue, discharge from the bowel may be quite discomforting for the patient.

William vaginoplasty and its Creatsas modification use a perineal skin flap to create a perineal pouch that serves as a neovagina. Post-operative urinary stream distortion and hair growth in the neovagina are potential complications.

When used, a musculocutaneous flap based on the gracilis muscle has a high risk of flap necrosis coupled with a worrisome scar in the thigh.

In more recent times, the usage of sensate flaps has been encouraged. Two of such flaps are the medial upper thigh flap and the bilateral flap of the labia majora and minora. While the latter provides a soft, pliable, durable, and sensate vagina, the labia majora and minora are sacrificed and the vulva distorted. The medial upper thigh flap, described by Wang et al. [5], is based on a suprafascial vascular plexus, and a partially sensate flap of about 20cm x 9cm can be raised without using the gracilis as the carrier. However, the blood supply is precarious, and there is a need for a secondary procedure to divide the pedicle. Nevertheless, it has found good usage in the coverage of significant perineoscrotal defects [6].

The pudendal thigh flap offers a sensate skin flap with a reliable and robust blood supply in the reconstruction of the vagina. The versatility of the flap, even when used as an island, has been well attested to. Monstrey et al. [7] reported survival of the flap in all his 31 pudendal thigh flaps, with excellent functional outcomes. The technique is simple, safe, and reliable, requiring no stents or dilators. The neovagina is sensate and has a natural angle. The donor site in the groin can be closed primarily with an inconspicuous scar [8].

**Case Report**

A 12-year-old girl, accompanied by her mother, presented at the Afe Babalola University Multi-System Hospital, a 400-bed tertiary hospital, located in Ado-Ekiti, South-West Nigeria. Her complaints were of an abnormally large clitoris since birth and recurrent cyclical low abdominal pain lasting about 5 days every month for the preceding eight months. Her developmental milestones had been as expected, and she had developed breast buds two years earlier. There was no history of a similar condition in any family member, no significant past medical history, and no previous surgery. She had never been sexually active. Essential findings on physical examination were symmetrical breasts in Tanner stage 4, absent labia majora and minora, hypertrophic clitoris with a rugae-like fold inferior and posterior, the external urethral opening was located almost at its tip of the hypertrophic clitoris (Figure 1). Abdominopelvic ultrasound showed well-placed bilateral ovaries and no testicular tissue. The left hand/wrist radiograph placed the patient's bone age between 15 and 16 years. Pelvic MRI showed a uterus with an intact endometrial plate, which was continuous with a short vagina segment that ended blindly (Figures 2a and 2b). The MRI also confirmed the presence of bilateral ovarian tissues and hypertrophied clitoris.

![Figure 1: Pre-op picture showing absent labia majora and minora, hypertrophic clitoris, with a rugae-like fold inferior and posterior it.](image1)

![Figure 2: Pelvic MRI showed (a) a uterus with an intact endometrial plate, contiguous with a short vagina segment that ended blindly and (b) the presence of bilateral ovarian tissues.](image2)
catheterised, 2% lignocaine in 1:100,000 adrenaline was infiltrated into the perineal body, and by blunt and sharp dissection, taking care to avoid injury to the bladder and urethra anteriorly and the rectum posteriorly, a tunnel was made down to about 6 cm from the perineum where the blind end of the proximal vagina was encountered and opened into. Three stay stitches were placed at 12, 4, and 7 o' clock positions on the proximal vagina (Figure 3a). The cervix was felt digitally. The technique of flap elevation was like the Singapore flap described by Wee and Joseph [9]. Based on the internal pudendal artery, a flap measuring about 10 cm by 4 cm was marked on each side and centred on the inguinal crease (Figure 3b). The incision, beginning at the anterior margin, was deepened through the skin and subcutaneous tissue to the deep fascia. Each flap was elevated deep to the deep fascia. As the elevation continues posteriorly, the deep fascia was sutured to the skin to prevent the former from inadvertently shearing off the deep fascia. Care was taken to elevate the flap along with the epimysium over the proximal part of the adductor muscle. At the flap base, the incision was made only skin deep down to the subcutaneous tissue. The dissection was carried more posteriorly in that plane to ensure ease of medial transposition of the flap.

The flaps from both sides were tunnelled under the adjoining skin between the flap and the neovagina, to have both flaps in the midline without any tension. The flaps were tubularised starting from the medial edge (Figure 3c) and then the lateral edge (Figure 3d) into a neovaginal tube. The neovaginal tube was then invaginated at its anterior edge into the newly created space between the urethra/ bladder and the rectum and then sutured to the proximal end of the vagina (Figure 3e). The posterior end of the neovagina was inserted into the neo-vulva. The donor sites were closed primarily and without tension over a drain (Figure 3f). Some adduction of the lower limbs facilitated this. A stent made of gauze and wrapped with petroleum-based gauze (Bactigras®) was inserted into the neovagina. Dressings were applied.

Figure 4: Mild donor site wound dehiscence was noticed on the 7th-day post-op and managed with honey dressing.
The most common presentation of vaginal atresia is primary amenorrhoea in a girl with a normal XX genotype associated with cyclical abdominal pain. The lack of an introitus which is replaced by a vaginal dimple, is pathognomonic of this condition [13]. Our patient presented with primary amenorrhoea and cyclical abdominal pain. However, there was no vaginal dimple, presenting with ruggae-like folds inferior and posterior to the hypertrophied clitoris. Diagnosis is confirmed by ultrasound and magnetic resonance imaging, revealing normal uterus and cervix, normal ovaries, normal fallopian tubes, and a well-formed proximal vaginal with or without haematocolpos, as was observed in our patient.

As stated earlier, the modalities for managing patients with vaginal atresia vary widely, including various surgical procedures. In this study, the pudendal thigh flap was used to create the neovagina and restore uterovaginal continuity in a 12-year-old girl, with its ability to overcome the disadvantages associated with graft vaginoplasty and other techniques [11]. Most studies have presented the use of pudendal thigh flap in older age groups. In a study of 20 cases by Humayra et al. [9] the patients were between 15 and 20 years. We undertook the surgery on our patient at age 12 because of the cyclical abdominal pain that she presented with. However, the left hand/wrist radiograph had put her age at between 15 and 16 years. This age discrepancy may be a technology limitation or a notably poor attitude towards an appropriate registry of births in our locality. There was no untoward postoperative complication except mild wound dehiscence at the donor site. The patient was also exceedingly difficult to handle; change of dressing was a serious issue. We, therefore, had to stent the neovagina for four weeks using a vacuolated tube since a vaginal examination was not so acceptable to the patient. We had faced that challenge because of her young age and poor emotional maturity and motivation to maintain a neovagina. A study on the management of vaginal agenesis observed that vaginal reconstruction should be deferred until adolescence or adulthood when the patient presents in childhood [15]. This ensures that the patient reaches physical and psychological maturity and awareness of her sensuality that can be immensely helpful in maintaining the neovagina.

Post-operatively, the patient was kept in bed with the urethral catheter in situ, on a soft diet, broad-spectrum antibiotics, and alternate-day wash out of the neovagina with saline for two weeks. She had partial dehiscence of the donor site (Figure 4), which did well with honey dressing and was discharged home on sitz bath on the 14th-day post-op for follow-up in the clinic. Stenting of the neovagina was sustained using a vacuolated tube (Figure 5a & b). On follow-up at the 6th-week post-op, she had already had a regular menstrual blood flow, and the neovaginal depth and width were 9cm and 4cm, respectively, with intact sensation. Also, the neovagina could accommodate two fingers and the cervix could be felt. However, hair growth was noticed in the neovagina by the 6th week after the operation.

Discussion

Vaginal atresia, often referred to as distal vaginal atresia, is a congenital anomaly that occurs when the urogenital sinus fails to develop and contribute to the formation of the distal part of the vagina, while the Mullerian structures are unaffected. The ovaries, fallopian tubes, uterus, and proximal vagina are therefore unaffected. Associated anogenital malformations are also unusual presentations. It is a rare condition [11,12] occurring in 1:5000 births [13], and it presents in various ways. The treatment approaches are therefore varied on presentation and driven mainly by the presenting symptoms. In contrast, vaginal agenesis is the absence of the proximal portion of the vagina in an otherwise phenotypically, chromosomally, and hormonally intact female, and is usually associated with various syndromes like Mayer-Rokitansky-Kuster-Hauser syndrome, Bardet–Biedl syndrome, Kaufman–McKusick syndrome, Fraser syndrome, Winters syndrome and abnormal development of the anus and rectum [14].

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Six weeks post-op, the depth of the neovaginal is 9cm with a width of 4cm being the length of the vacuolated tube, which the patient could easily insert into the neovagina and the width of two fingers inserted on vaginal examination. The depth and width achieved are comparable with the mean depth of 9.5 cm and the mean width of 3.8cm in the Anwar study [11]. In their study, Giraldo and Gonzalez [8] reported a mean depth of 9.5cm and a width of 3cm from a total of 40 flaps elevated for 20 vaginoplasties one year postoperatively. Ajmal and Yusuf [16] also reported a mean vaginal length and width of 9.2cm and 4.3 cm, respectively, one-year post-op and concluded that this method of vaginoplasty is simple, safe, and reliable and has shown satisfactory functional and cosmetic results.

Most authors believe that there is no need for stenting or dilatation postoperatively; we, however, chose to stent the neovagina to prevent ring constriction at the anastomotic site of the proximal
vagina and the neovagina tube. Wamalwa and Khainga [17] also used vaginal packs in the early postoperative period and later dilators to prevent neo-vaginal canal stenosis.

The patient in our study had a normal menstrual flow three weeks postoperatively, confirming a functional uterovaginal continuity. Selvaggi et al. [18] equally reported that uterovaginal continuity was established in both patients in their study with vaginal aplasia and a functioning uterus, resulting in normal menstruation.

The donor site in our patient healed well with a scar that is fast becoming inconspicuous. This is so despite the initial partial wound dehiscence noted on the 7th-day post-op. Other well-noted advantages of the pudendal thigh flap are that it can be done as a single stage, the donor scar is inconspicuously hidden in the groin, and the reconstructed vagina has a natural angle for intercourse and is sensate [1,7,10]. Our patient alluded to the preservation of sensation in the neovagina even though she is not sexually active so as to report the observance of erotic sensations.

Notwithstanding the advantages, reported disadvantages with the use of pudendal thigh flaps include the presence of some sebaceous vaginal secretions and hair in the vaginal lining [18]. We notice some hair growth in the neovagina of our patient as early as the 6th week postoperatively. Hair growth has been reported as aesthetically displeasing to patients, even though it is not a functional problem [8,11,19]. Laser or other methods of depilation are available to manage hair growth and can be done pre- or post-operatively. Some studies have described progressive hair atrophy in the proximal two-thirds of the vagina in long-term follow-up [19]. We hope to follow up with the patient for hair atrophy later.

**Conclusion**

Notwithstanding the above, the pudendal thigh flap remains versatile for constructing a neovagina and maintaining uterovaginal continuity, even in facilities that do not routinely carry out such procedures.

**References**