



Case series

Modified Passerini-Glazel feminizing genitoplasty outcomes in adults: Two rare cases[☆]

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ABSTRACT

Introduction: Modified Passerini-Glazel feminizing genitoplasty is typically performed in children with atypical genitalia. In our article, we have performed the procedure in adults with genital anomalies.

Case presentation: The first case was a 22 years old woman who was planning to get married. She presented with a chief complaint of having no vaginal canal. Gynecological examination showed no vaginal opening. The common channel was visible, and the size of the perineal body was 3 cm. The patient underwent vaginal reconstruction using a modified Passerini-Glazel technique without amnion graft.

The second case was a 20 years old girl planning to get married, with a chief complaint of small vaginal introitus. Gynecology examination showed small minor labia with an introitus size of 1 cm. The patients underwent vaginal reconstruction and labioplasty using a modified Passerini-Glazel technique with an amnion graft. Both patients have undergone anal atresia surgery in childhood.

Discussion: There was no difference in outcome between using amnion graft and without amnion graft following the modified Passerini-Glazel feminizing genitoplasty procedure. The first patient had been pregnant and had successful delivery by elective cesarean section. Moreover, the second patient had no complaint of sexual disorders after the procedure. No cases of dysuria, urinary tract infection, leukorrhea, hematocolpos, or malodorous vaginal discharge were reported in both cases.

Conclusion: Modified Passerini-Glazel feminizing genitoplasty is a safe and effective procedure. Daily vaginal dilation in the postoperative period was unnecessary, and it allowed for an excellent cosmetic result.

1. Introduction and importance

Congenital anomalies of the urogenital tract are the leading associated anomalies in patients with ARM (anorectal malformation). Previous studies have shown the incidence of UGA (urogenital anomalies) associated with high or intermediate forms of ARM to be approximately 50–60% and with low anomalies to be 15–20%. Usually, the ARM management gets priority, and the gynecologist's evaluation is not always considered essential; although, in many cases, the genital tract disorder is the primary cause of morbidity and mortality, especially if menarche occurs [1].

Feminizing genitoplasty aims to construct external genitalia with a female appearance, produce a functional vagina for future intercourse, menses, and childbirth, and preserve the delicate genital nerve supply. Feminizing genitoplasty is usually performed in patients with genitalia ambiguity and most commonly in patients with congenital adrenal

hyperplasia [2]. In 1989, Passerini-Glazel presented a one-stage procedure for the correction of severely masculinized female pseudohermaphrodites in whom a high, short vagina was detached from the urogenital sinus (UGS) and then exteriorized through a cylinder of tissue made of "penile" skin and UGS, which was inserted into the perineum and anastomosed to the native vagina. The use of all available tissue was thought to avoid introital vaginal stenosis, a complication that plagued genitoplasty techniques in the past [3]. In literature, feminizing genitoplasty was performed at a mean age of 22 months [2]. However, in our cases, the patients presented to us because they planned to get married (no vaginal opening).

The original Passerini-Glazel feminizing genitoplasty procedure begins with clitoral reduction. Modifications include vaginal dissection and disconnection from the urethrovaginal sinus as the initial stage of the procedure; large dissection of the neurovascular bundle on both dorsal and lateral faces of the clitoris; plication of the skin around the

[☆] No patient or author details are included in the figures.

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reduced clitoris; and suturing the lateral edge of the proximal portion of the mucocutaneous plate with the labia majora medial edge to a plane deeper than the subcutaneous tissue. These modifications reduce bleeding and operating time, preserve clitoral sensitivity, form the clitoral prepuce, and create labia minora [4].

These case series are interesting because the Passerini-Glazel feminizing genitoplasty procedure was performed in adults, whereas in previous cases, this procedure was usually performed in childhood since genitalia anomaly is often associated with anal atresia.

This case report has been reported in line with the SCARE 2020 criteria [5].

2. Case presentation

These patients came to the Urogynecology Outpatient clinic in Cipto Mangunkusumo Central general hospital, Jakarta.

2.1. Case I

A 22 years old Javanese woman presented with a complaint of not having a vaginal opening. She was unemployed and a senior high school graduate. The patient did not have any complaints during menarche because she thought it was normal menstruation and micturition. She had complaints when she was married that she was unable to have sexual intercourse. She had a regular menstrual cycle that exits from the common channel with no dysmenorrhea. When voiding, the menstrual blood exits alongside urine. Sometimes she felt dysuria after menstruation. The patient had a history of anal atresia and was treated surgically with posterior sagittal anorectoplasty (PSARP) by a surgeon 20 years ago. Defecation was normal after PSARP. She had no family history with the same condition.

2.2. Case II

A 20 years old Javanese woman presented with a complaint of having a small vaginal opening. She was an entrepreneur and a senior high school graduate. She complained that she was unable to have sexual intercourse and no complaint during menarche. She had an irregular menstrual cycle (2–3 months) without dysmenorrhea, normal urinary frequency, and no dysuria. During menstruation, she found that her urine becomes brownish in appearance. She had no complaint of defecation. She had a posterior sagittal anorectoplasty (PSARP) history due to anal atresia at 3 years old. She had no family history with the same condition.

3. Clinical findings and investigations

3.1. Case I

Gynecological examination showed no vaginal opening. Labia majora and the common channel were visible. The size of the perineal body was 3 cm. The anal canal was visible without blood/inflammation (Fig. 1).

On rectal toucher, anal sphincter tone was normal, and the ampulla was not collapsed. A scar was palpable on 12 o'clock direction on the anal mucosa, and no cystic mass was found at the anterior mucous. The uterus was in normal size and shape without adnexal mass.

3.2. Case II

Gynecologic examination showed a small labia minor with an introitus size of 1 cm. Labia majora was normal. The common channel was visible. Vaginal length based on bimanual sondage/sound was 7 cm. There were 2 orifices which were the urethra and vagina in the introitus (Fig. 2).

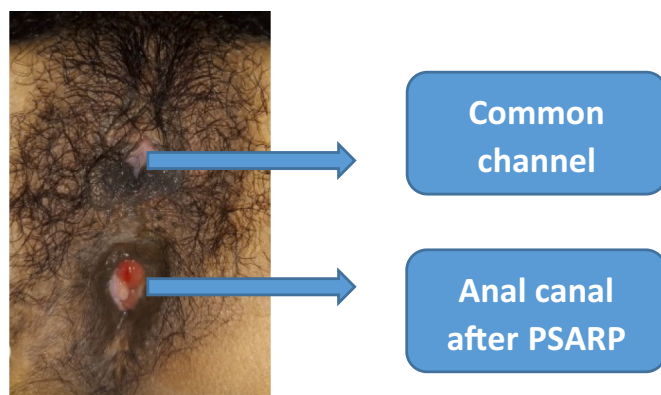


Fig. 1. Labia majora and the common channel were visible. No vaginal opening.

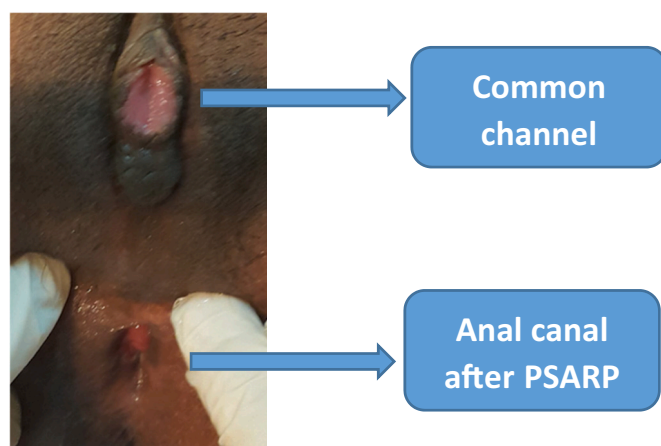


Fig. 2. The common channel was visible anal orifice was visible (post-PSARP).

4. Diagnostic assessment

4.1. Case I

Ultrasonography showed an arcuate uterus and homogenous myometrium. The vagina was not visible; the distal part of the cervix was visible. A small pocket of the vagina connected to the urethra by a small fistula was present. The normal vaginal passage was filled with an anorectal part. It is concluded that there was an arcuate uterus, vagina agenesis (predominantly distal part) with a common channel (Fig. 3).

The patient was diagnosed with urogenital sinus malformation anal atresia post-posterior sagittal anorectoplasty (PSARP) and post colostomy. From the performed vaginoscopy, after the hysteroscopy sheet was inserted to the external ostium, there were 2 canals at the cranial and posterior sheet. At the cranial, the canal continues to the urethra and bladder. At the posterior, the canal continues to the vagina.

4.2. Case II

Ultrasonography examination showed hematocolpos and right hydrosalpinx or hematosalpinx with common channel (Fig. 4).

5. Therapeutic intervention and outcomes

Both of these patients' preoperative investigations showed normal laboratory and thorax radiology results. This procedure was performed by an experienced urogynecologist.

The patient underwent vaginal reconstruction by using the modified



Fig. 3. Ultrasonography examination: arcuate uterus, vagina agenesis with common channel.

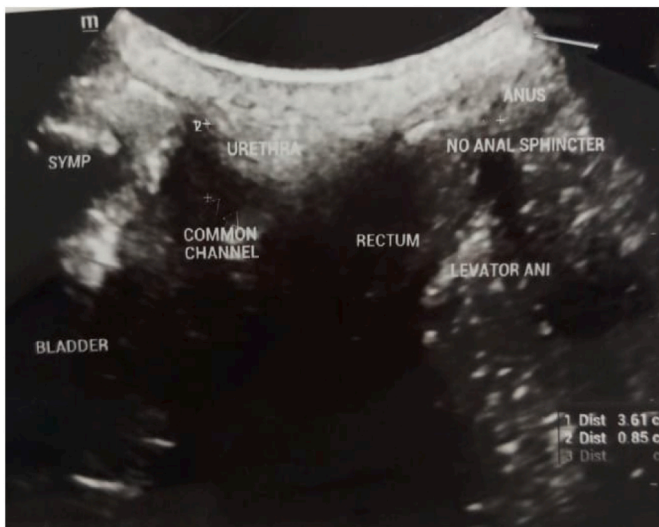


Fig. 4. Ultrasonography examination.

Passerini-Glazel technique (Fig. 5). A perineal incision was performed vertically until 1 cm above the anus. The vaginal mucosal was separated from the surrounding tissue at 3–9 o'clock from the vaginal canal. The vaginal mucosal was penetrated from the posterior part of the vagina as high as the external urethral ostium. A posterior urethral flapping suture was performed from the vaginal mucosal. Lower vaginal mucosal was incised until the external urethral ostium to cover the lateral vaginal wall (right and left vaginal wall). The upper part vaginal mucosal was sutured with the lower part of vaginal mucosal. A lateral incision was performed to make the perineal body. Posterior vaginal mucosa was sutured at the edge of the perineal tip, continue with insertion of vaginal silicone without amnion until one week after surgery. Vaginal closure was performed subsequently.

In the second case, we performed vaginal reconstruction and labioplasty by using the procedure of Passerini-Glazel feminizing genitoplasty modified using amnion graft (Fig. 6).

The catheter was inserted intravaginally as a guide. A transversal incision was performed at the orifice of the common channel, continued by medial incision about 3 cm to anal. Incision at the posterior common channel was performed as high as the normal vagina. A urethral catheter

was inserted, and cutting was performed at the posterior common channel. The proximal vaginal mucosa was identified and stitched distally with the bulbocavernosus muscle using PGA 2.0. The anterior common channel mucosal was split into two equal parts, pulled posteriorly, and made into the lateral and posterior vaginal mucosal. The distal vaginal mucosal was sutured interruptedly with the common channel mucosa. After all the vaginal mucosal was sutured, the distal mucosal was sutured with the labia minora skin. After that, a mold was inserted with an amnion graft to the vagina (Fig. 6).

6. Follow up and outcome

Post-operative evaluations in these patients were conducted every month for one year. We evaluated the clinical symptoms, total vaginal length, and sexual function.

The first patient had been pregnant after 8 months of procedure and had undergone delivery by elective cesarean section due to obstetric indication (Fig. 7). Furthermore, the second patient had no complaint of sexual disorders after the procedure. Daily vaginal dilation in the post-operative period was not necessary. No complaints of dysuria, urinary tract infection, leukorrhea, hematocolpos, or malodorous vaginal discharge were reported (Fig. 8).

7. Discussion

Patients may present with gynecologic concerns after previous posterior sagittal anorectoplasty (PSARP) to repair an anorectal malformation (ARM). Common findings include an inadequate or shortened perineal body and introital stenosis, retained vaginal septum, and remnant rectovestibular fistula. An inadequate or shortened perineal body may impact fecal continence, sexual function, and recommendations regarding obstetrical mode of delivery [6]. Both of our patients with an inadequate or short perineal body required preoperative perineoplasty.

The modified Passerini-Glazel feminizing genitoplasty was performed. There was no different outcome between using an amnion graft or not. The first patient who underwent the modified Passerini-Glazel technique had been pregnant and had successfully undergone delivery by elective cesarean section. Furthermore, the second patient had no complaints of sexual disorders after the procedure. The perineal body of both patients was 3 cm after the procedure. No complaints of dysuria, urinary tract infection, leukorrhea, hematocolpos, or malodorous vaginal discharge were reported. Lesma A et al. reported the outcome of modified Passerini-Glazel feminizing genitoplasty in which vaginal dissection and disconnection from the urethrovaginal sinus were performed at the first time of the procedure. This procedure was subsequently performed by dissecting the neurovascular bundle on both dorsal and lateral faces of the clitoris, stretching the skin surrounding the reduced clitoris, and performing a suture to approach the lateral edge of the proximal portion of the mucocutaneous plate and labia majora medial edge. Lesma A et al. stated that modifications of one-stage Passerini-Glazel feminizing genitoplasty in children are associated with reduced bleeding and operating time. All patients were reported to have complete day and night continence with no urgency or frequency. In addition, these modifications resulted in an excellent cosmetic appearance. The genital examination showed that the mean clitoris index was $15,2 \text{ mm}^2$ in patients with surgical reparation at a mean age of 2 years who were re-examined at a mean age of 10 years. Women who underwent surgery at a mean age of 13 years and were followed up at a mean age of 16,5 years had a clitoris index of 18 mm^2 [4].

Bocciardi A et al. reported that ambiguous genitalia in infancy was corrected completely using the Passerini-Glazel technique procedure. The procedure was also safe and had an excellent cosmetic outcome. Vaginal stenosis was commonly reported, but the reparation could be conducted using a simple introitoplasty procedure before the onset of menarche [7]. Since these procedures were performed in childhood, our

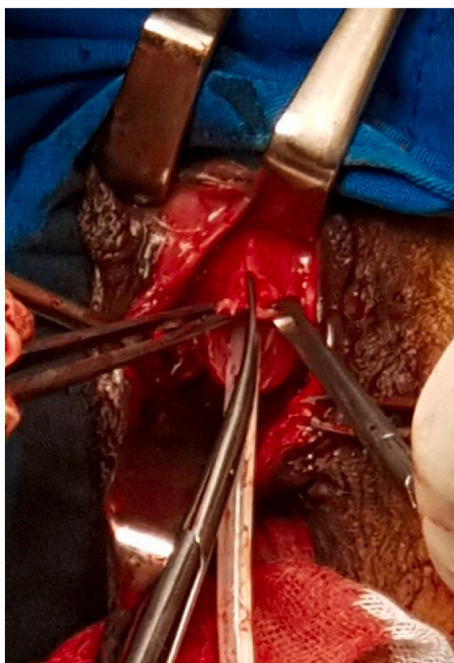


Fig. 5. Modified Passerini-Glazel without amnion graft molding (using silicone catheter).



Fig. 8. 3rd months after the procedure (Case II).

patients also had good results in cosmetics and sexual function in 6 months after the procedure.

Passerini-Glazel feminizing genitoplasty was effective in adult common channel cases with good outcomes, including sexual function and pregnancy.

8. Conclusion

Passerini-Glazel feminizing genitoplasty is often performed in the management of children genital anomalies. In our cases, the technique was performed in adult patients with genitalia anomalies with a good outcome. Daily vaginal dilation in the postoperative period was not necessary.

Provenance and peer review

Not commissioned, externally peer review.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Not required.

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Guarantor

Tyas Priyatini, Roziana.

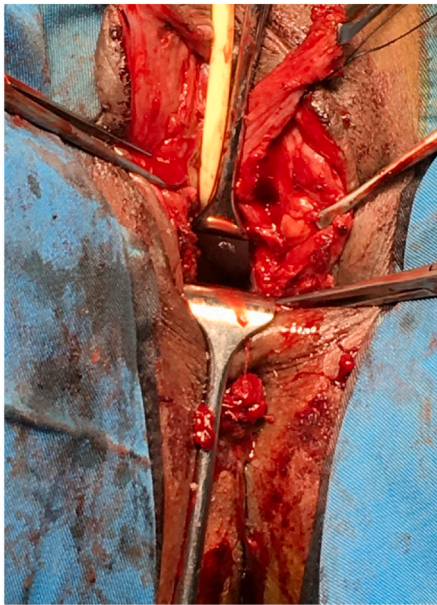


Fig. 6. Modified Passerini-Glazel with amnion graft molding.



Fig. 7. 2nd weeks after the procedure (Case I).

Research registration number

None.

CRediT authorship contribution statement

Tyas Priyatini: concept, operator, data analysis, drafting and revising, final approval
 Roziana: data collection, data analysis, writing the paper.

Declaration of competing interest

None declared.

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