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## Research Article

# Staged feminizing genitoplasty with total urogenital sinus mobilization

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## Abstract

**Background:** The diagnosis and treatment of individuals with Disorders of Sexual Development is a complex process and requires a multidisciplinary approach. Each individual should be evaluated objectively and the treatment process of each individual has its own unique differences. The aim of this study is to discuss the effectiveness of staged feminizing genitoplasty operations using the total urogenital mobilization technique.

**Material and methods:** Hospital records of patients who underwent vaginoplasty using the TUM technique between 2014 and 2019 by a single surgeon in a pediatric surgery center of a tertiary pediatric hospital were retrospectively reviewed.

**Results:** All ten patients who underwent TUM had 46 XX chromosomes. The TUM procedure for vaginoplasty was performed in all patients by the author. A clitorolabioplasty had been previously performed in six patients with congenital adrenal hyperplasia. The follow-up period of the patients was 1-6 years. No serious complications developed in any of the patients. Neither infection nor tissue necrosis occurred. A second surgical procedure was not required for urinary fistula, vaginal stenosis, urinary incontinence, or early suture dehiscence.

**Conclusion:** This study shows that vaginoplasty can be performed in a single session in infancy for patients with low confluence and a short Urogenital Sinus (UGS). Apart from this, postponing vaginoplasty surgeries to the adolescence period and performing UGS mobilization surgery seem to carry successful results.

## Introduction

The diagnosis and treatment of individuals with Disorders of Sexual Development (DSD) is a complex process and requires a multidisciplinary approach. In this complex situation, medical, psycho-social and ethical situations should be considered. Each individual should be evaluated objectively. Therefore, the treatment process of each individual has its own unique differences.

There have been intense debates on this subject, from nomenclature to the time of surgery and the surgical technique; however, there remains no consensus [1,2]. While there are those who recommend an early and single-session surgery [3,4] and those who recommend that the surgery be performed gradually [5,6], studies have advocated for postponing all

surgeries until the patients reach an age where they can decide on their own [7]. In terms of surgical technique, different techniques have been suggested, from flap vaginoplasty to total urogenital sinus mobilization (TUM) [8].

In this study, staged feminizing genitoplasty operations with the TUM technique are presented and their results are discussed in the light of previous studies.

## Materials and methods

Hospital records were retrospectively reviewed for patients who underwent vaginoplasty with a perineal approach using the TUM technique by the author between 2014 and 2019 in the Pediatric Surgery Clinic of Ankara Children's Hematology and Oncology Training Hospital (later on moved to Ankara City

Hospital Children's Hospital). The information collected from the patient's records was including demographics, radiological imaging, the type of operation, and the follow-up period.

The patients were questioned regarding the age of menarche of the mother and any sisters. The patients also received a physical examination to determine their Tanner classification and to perform measurements of ovarian volume, uterine volume and endometrial thickness by ultrasound imaging, which were performed to estimate the age of menarche. A time for the operation was planned for individuals just before the predicted menarche or menstruation.

## Technique

Before the operation, all patients underwent a cystoscopy in the form of Urogenital Sinus (UGS) endoscopy under general anesthesia. At the cystoscopy, a urinary catheter to the bladder and a Fogarty catheter to the vagina was placed. Vaginoplasties were performed with the TUM technique that was previously described by Ludwikowski, et al. and UGS was preserved as vaginal flep [9] (Figures 1-3). A small gauze dressing was placed on the genitalia and removed the next day.

The urinary catheter was retained for 10 days and taken off just before hospital discharge. Pain was controlled with analgesics. Patients were called for examinations at the clinic in the 2<sup>nd</sup>, 4<sup>th</sup> and 12<sup>th</sup> weeks and 6<sup>th</sup> and 12<sup>th</sup> months postoperative



**Figure 1:** Operative preparing. An Foley urinary catheter and vaginal Fogarty catheter (green) were placed.



**Figure 2:** Total mobilizasyon of urogenital sinus. The urinary and the vaginal catheter are still held together.



**Figure 3:** Postoperative view of the patient.

period. In the 12<sup>th</sup> week, all patients were examined and vaginal calibration was performed with Hegar dilatator under sedation.

Median age, median UGS length, and median follow-up duration were used for the statistical method.

Ethical approval was obtained from the Ankara City Hospital Ethical Committee (Number: E221-519).

## Results

Ten patients who underwent TUM were included in the study, other patients who were operated with different techniques are excluded. Ages of patients were between 6-16 years (median: 12 years). TUM surgery was performed on nine patients with a diagnosis of CAH, while in one patient with a diagnosis of Mc Kusick-Kaufmann syndrome, who had numerous previous operations in another center.

Six patients were previously operated by the author for clitorolabioplasty in their early childhood as the two-stage vaginoplasty plan, since the UGS was long and they were waiting for vaginoplasty surgery. Due to failed vaginoplasty on two patients with CAH, the TUM was performed.

Cystoscopic evaluation showed that the shortest UGS length was 2.5 cm, while the longest was 4.5 cm (median: 3.5 cm). The data of the patients is shown in Table 1.

Of the five patients without preoperative menarche; score of Tanner Breast were 3-4, volume of uterus was 11-18 cm<sup>3</sup> (median: 16.1 cm<sup>3</sup>), endometrial thickness 4-9 mm (median: 5 mm) and ovarian volumes were 3-6 cm<sup>3</sup> (median: 4 cm<sup>3</sup>). These patients began to menstruate within 1-5 months (median: 3.6 months).

Only one complication, mild vaginal-entrance stenosis developed in one patient. She was diagnosed by Mc Kusick Kaufmann Syndrome and had multiple surgeries before. Her stenosis was solved by minor cut-back surgery. Infection and tissue necrosis did not occur. A second surgical procedure was not required for urinary fistula, urinary incontinence or early suture dehiscence.

There was no vaginal stenosis at the postoperative 3<sup>rd</sup> month examination and vaginal calibration under sedation.

**Table 1:** Data of the patients.

Patient No	Age at operation (years)	Diagnosis	Previous Operation	Length of UGS (cm)	Follow-up (years)	Menarche after operation (months)	Complication
1	6	CAH	None	2,5	2	-	-
2	13	CAH	CLP	3	6	+	-
3	12	CAH	CLP	3,5	2	2	-
4	12	CAH	CLP	4,5	4	4	-
5	11	CAH	CLP	4	5	5	-
6	12	CAH	CLP	4	6	3	-
7	11	CAH	CLP	3,5	3	4	-
8	14	CAH	CLVP	3	4	+	-
9	15	CAH	CLVP*	3,5	5	+	-
10	16	McKKS	Numerous vaginoplasties*	4	5	+	Mild stenosis

CAH: Congenital Adrenal Hyperplasia; McKKS: Mc Kusick Kaufman Syndrome; CLP: Clitorolaboplasty Operation; CLVP: Clitorolabiovaginoplasty Operation; UGS: Urogenital Sinus; +: Menstruating; \*: at another center

The follow-up period of the patients was 1–6 years (median: 4 years). No urinary infections and no urinary incontinence occurred in any of the patients. None of the patients required a vaginal dilatation.

## Discussion

In a feminizing genitoplasty, there are several aims, including creating a genital appearance compatible with the patient's gender, providing an anatomical and urinary tract that will not cause incontinence and ensuring a genital structure that will fulfil sexual and reproductive functions during adult life [10].

There is no consensus on the timing and the surgical technique to perform these operations [2]. For example, at the time of diagnosis, a single-session operation can be performed during infancy.

However, if the operation involves the genital appearance, a genitoplasty can either be performed or the operation can be completely postponed.

The most important anatomical factor that will affect vaginoplasty is the confluence of the urethra and vagina, which may be low or high, and the length of the UGS [11]. This will also affect the surgical technique and results.

Many reports have been published advocating early surgery. These have argued that this will eliminate parental stress and the procedure can be finalized with a single surgery [3,4]. It is an important advantage that the surgery is completed before the patient suffers psychological trauma, as with in hypospadias surgery. Moreover, surgery is easier with the benefit of maternal estrogen during the infantile period [12].

The biggest argument of those who advocate a vaginoplasty in late childhood is that vaginoplasties performed at an early age mostly require revisions later. There are studies that have reported a high rate of complications and revisions after early operations [5,6]. Considering these reports, it has been recommended that a clitorolaboplasty be performed, and if necessary for cosmetic reasons, this should be performed at an

early age in a virialized patient and then a prepubertal–pubertal vaginoplasty operation can be performed at a later date.

It has been suggested that pubertal estrogen will facilitate the operation. Moreover, it has been claimed that postponing a vaginoplasty to a later period, and allowing hydrometrocolpos, will ensure sufficient dilatation that facilitates the vaginoplasty operation [13]. In the present study, the general approach was a staged feminizing genitoplasty.

Estrogen is known to play an important role in coagulation, homeostasis and the overall inflammatory process [14]. Various animal experiments have shown that estrogen increases the blood supply and elasticity of vaginal tissues and accelerates wound healing [15,16]. Alternatively, it has been reported that preoperative vaginal estrogen administration in the postmenopausal period increases the synthesis of mature collagen and decreases the degrading enzyme activity. It also strengthens the vaginal tissue by increasing both the integrity of the connective tissue and the chances of surgical success [17]. In this study, patients were followed up and waited for vaginoplasty until adolescence since the facilitating effect of estrogen was expected at a later time. During this period, some parameters were recorded, such as questioning the menarche age of the mother and other sisters. Also, during this period, patients received a physical examination to determine their Tanner classification and measurements of ovarian size, uterine volume and endometrial thickness were determined using ultrasound imaging [18,19].

Many techniques have been described for vaginoplasty. A U-flap vaginoplasty, described by Fortunoff, et al. can be considered an important step in these surgeries [20]. Hendren, et al. later described a pull-through vaginoplasty [18]. Passerini' technique and anterior sagittal transanorectal approach (ASTRA), are among the techniques described [21,22]. TUM operations were described by Pena to correct the cloacal anomaly, while Ludwikowski et al. suggested using these for vaginoplasty [9,23]. Rink, et al. also suggested a partial urogenital sinus mobilization and showed that the sinus mucosa excised by Pena could be used for repair [24]. Intestinal substitutions can also be used for vaginoplasty. Undoubtedly, a combination of these techniques can also be applied.



Bailez, et al. reported that TUM achieves good results with a combination of different techniques in a study involving 55 patients [11]. Ludwikowski, et al. reported adapting TUM for DSD and stated that it would be suitable for cases where the UGS is  $\geq 3$  cm and that it would reduce potential urethral innervation damage and surgical times since a urethral and vaginal separation is not needed [9]. It can be considered that a non-intensive dissection and U flap vaginoplasty would be sufficient with a UGS  $\leq 2-2.5$  cm.

Although Palmer et al. have reported that a TUM or partial urogenital mobilization [PUM] did not increase urinary incontinence in patients [25]. Stites, et al. have reported they prefer a PUM instead of a TUM because of severe urinary incontinence after a TUM. There were no incontinence complaints in any of presented patients. Although there are some concerns about continence, it is clear that urogenital sinus mobilization is a positive step in feminizing genitoplasty.

TUM looks promising with its early results and has become increasingly popular in recent years. Ludwikowski and Gonzales also advocate the feminizing genitoplasty, which can be performed in a single session with TUM [26]. In the same article, they drew attention to the word "total" in TUM and emphasized the integrity concept by saying "'total' refers to an en bloc dissection and not to the extent of the proximal dissection". The survey conducted by Yankovic et al. found a significant portion of the participating surgeons declared that a PUM or TUM gave better results in their personal experience [27-30].

Although knowledge concerning the surgical technique has increased, there is no consensus on performing surgery in a single session or in stages. In this study, six patients underwent clitorolabioplasty in infancy and vaginoplasty with the TUM technique during adolescence. The patients were followed up without any problems. On the other hand, a re-vaginoplasty was performed with the TUM technique in two patients who underwent a single-session clitorolabiovaginoplasty in childhood and whose vaginoplasty was unsuccessful. These results support the view that a staged vaginoplasty and TUM technique give good results.

Carrying out in a single center by a single surgeon with a single surgical technique is the strength of this study. On the other hand, not being a comparative study with other techniques is the limitation of this study.

## Conclusion

This study shows that vaginoplasty can be performed in a single session at an early age in patients with low confluence and short UGS, but in patients with long UGS, postponing vaginoplasty operations until puberty and performing TUM technique will yield successful results.

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## Ethical consideration

Ethical approval was obtained from the Ankara City Hospital Ethical Committee (Number: E221-519).

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