

**Cosmetic Outcome of Feminizing Genitoplasty in DSD (Disorders of Sex Development)**

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**Abstract**

**Background:** Disorders of sex development (DSD) include a diverse array of congenital disorders characterized by the abnormal growth of anatomical sex, chromosomes, gonads, & the brain, all of which are essential to the process of sexual differentiation.

**Objectives:** To evaluate the cosmetic outcome of feminizing genitoplasty in DSD.

**Patients and methods:** This interventional, non-controlled study was conducted on 50 virilizing females at Qena University Hospitals over one year. The study included detailed history, genital examination, Prader staging, serum electrolytes, karyotyping, hormonal profiles, abdominal ultrasound, genitography, cystoscopy, and surgeries (clitroplasty, vaginoplasty, labioplasty). Postoperative care involved antibiotics, analgesics, oral feeding, and vaginal calibration. Outcomes were assessed using Creighton and Lean scores.

**Results:** 38% of patients were under one year old; 92% had congenital adrenal hyperplasia, and 100% had 21-hydroxylase deficiency. Cosmetic outcomes were favorable, with 82% achieving good results. Postoperative complications were low: stress incontinence(8%), dysuria(6%), difficult micturition(4%), and wound infections(4%). Older age was significantly associated with poorer cosmetic outcomes( $p=0.006$ ) and increased stress incontinence( $p=0.041$ ), but not with dysuria, difficult micturition, or wound infections( $p>0.05$ ).

**Conclusion:** The number of patients with a decreased aldosterone level in the research cases was 41 (82%). The number of patients with an increased renin level in the research population was 41 (82%). The number of patients with a decreased cortisol level in the research population was 49 (98%). The number of patients with 17-OH-progesterone  $>20,000$  ng/dl in the research population was 41 (82%). The number of patients with increased testosterone levels in the research cases was 50 (100%).

**Keywords:** Cosmetic outcome; Feminizing Genitoplasty; DSD.

**DOI:** 10.21608/SVUIJM.2024.306501.1938

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**Received:** 1 July, 2024.

**Revised:** 1 August, 2024.

**Accepted:** 18 August, 2024.

**Published:** 3 October, 2024.

**Cite this article as:** Asmaa Gaber Rizk, Hamdy Muhammad Hussein, Sara Mohamed Habib Abdel-Hameid, Nezar Abdelraouf Abo Halawa.(2024). Cosmetic Outcome of Feminizing Genitoplasty in DSD (Disorders of Sex Development). *SVU-International Journal of Medical Sciences*. Vol.7, Issue 2, pp: 603-616.

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## Introduction

Sexual differentiation disorders, formerly referred to as intersex disorders, are among the most difficult conditions pediatric endocrinologists and surgeons must manage. In neonates, ambiguous genitalia necessitate a quick and precise diagnosis (Ahmed et al., 2021). The occurrence of urogenital anomalies in females is comparatively uncommon. seventy percent of cases involve persistent urogenital sinusitis & virilization resulting from congenital adrenal hyperplasia (CAH). CAH occurs at a rate of 1 in 15,000 live births (Witchel et al., 2022).

The internal gonads are ovaries, and the genetic sex of these cases is female; however, the external genitalia have been virilized to resemble male characteristics. The present treatment approaches consist of hormonal replacement therapy & early feminization surgery (Sandberg et al., 2015).

Urogenital sinus anomalies (UGS) are characterized by a constant association between the vagina & the lower urinary tract. This connection typically takes place in the middle portion of the urethra. Both structures terminate in a common channel that exits in a single opening at the perineum (Smith et al., 2017). The vagina lacks descent to the perineum & is attenuated. The older formulations categorize this as a "low" confluence for connections that are further distal or a "high" confluence for those that are closer together (Chaudhry et al., 2017). Recent descriptions classify the UGS anomaly as a spectrum issue, as opposed to a simple high or low value. As a result, evaluation and management must be tailored to the Vice Dean Research Office (Smith et al., 2017; Chaudhry et al., 2017).

The ovaries, uterus, and fallopian tubes are all normal; the

gonads are symmetrical and intra-abdominal and never descend into the labio-scrotal folds (Schweigmann et al., 2018). This research aimed to assess the cosmetic outcome of feminizing genitoplasty in patients with DSD.

## Patients and Methods

This was an interventional, non-controlled study conducted on 50 virilizing females at Qena University Hospitals. The study's duration was one year.

**Inclusion criteria:** virilizing females

**Exclusion criteria:** male DSD

## Methods

**All cases were exposed to the following:** A full history was taken, and in all cases, an external genital examination and Prader staging were done. The Prader scores were determined through a standardized genital examination, assessing stretched phallic length, urethral meatus, gonad type, chordee degree, labia/scrotum appearance, & the presence of the vagina and uterus. Serum electrolytes were measured at diagnosis, and during follow-up, karyotyping was done to confirm being 46XX. **A hormonal profile in the form of:** cortisol, ACTH, 17OH progesterone, dehydroepiandrosterone, androstenedione, 11-deoxycortisol, testosterone, and plasma remain activity were assayed initially and during follow-up, an abdominal ultrasonography, routine laboratory investigations, and **genitography and cystoscopy:** Genitography under sedation was used to delineate internal anatomy, starting with positioning the patient in the correct lateral position. Contrast material was injected under fluoroscopic control, revealing the confluence, urethral and vaginal bifurcation, and assessing urethral length and vaginal caliber. Cystoscopy was performed before surgery, using a

9Fr. or 11Fr. scope and water flow to appreciate the vaginal orifice.

**Surgical technique**

**Position:** The infant was placed in a supine position while undergoing a pan-endoscopic assessment, with their legs attached to an inverted U-shaped bar. **Clitroplasty:** Following the insertion of a 4/0 traction suture into the clitoris' glans, a circular incision

was made. Degloved and incised dorsal skin margins were produced in preparation for the labia minora. Cisternal reduction was accomplished through either dissecting the neurovascular bundle & removing the corpora or by employing the Kogan technique for protecting all neurovascular elements (Acimi, 2008) (Figs. 1-5).



**Fig. 1.** Preoperative photograph of a two-month old patient with salt losing CAH.



**Fig.2.** Degloving of dorasal skin of the enlarged clitoris. The clitoral skin flaps are used to construct labia minora.



**Fig .3.** Clitoral reduction, and UGS mobilization.



**Fig. 4.** UGS mobilization.



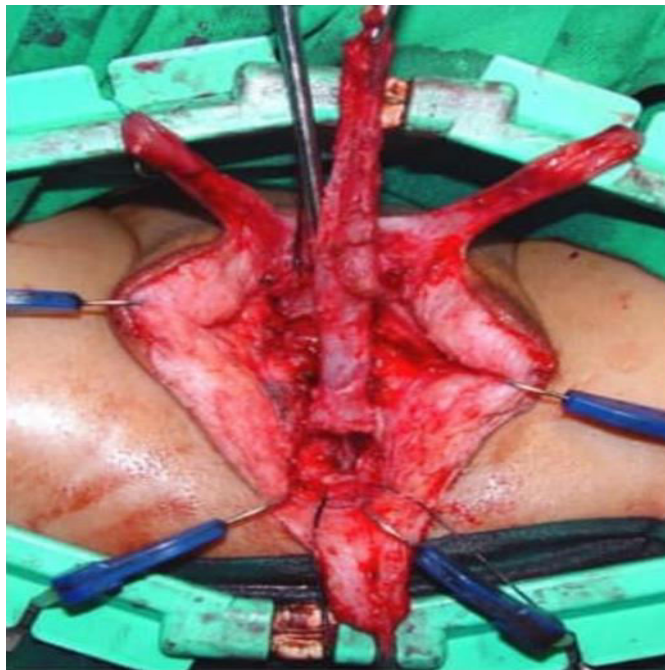
**Fig .5.** The UGS is incised posteriorly and incision is extended into posterior vaginal wall. The sinus is sutured to posterior aspect of clitoris.

**Vaginoplasty:** By preparing a posterior inverted U-shaped skin flap with its apex in contact with the posterior margin of the UGS opening, the sinus was enclosed through an incision. Above the levatorani muscles & extending to the upper border of the symphysis pubis & peritoneal reflection, the dissection was observed. An incision was made in the posterior

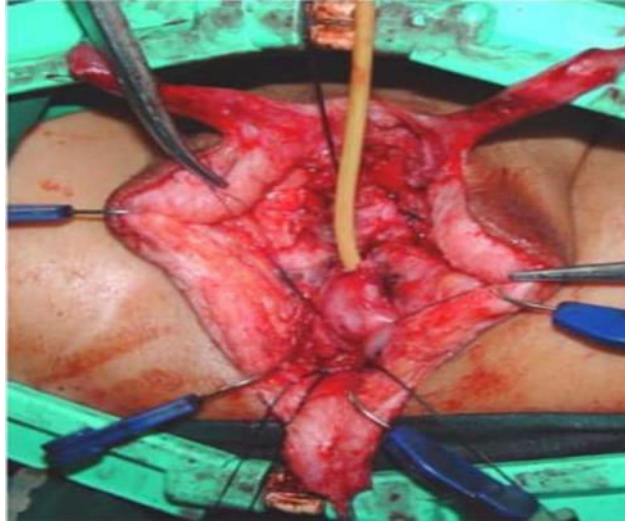
region of the UGS, & it was further extended into the posterior vaginal wall. When the vagina failed to reach the perineum, 6/0 Vicryl sutures were used to close the posterior wall of the vagina, which had been separated from the urethra. A gauze containing vaseline was utilized as a vaginal pack (Davies & Creighton, 2007). (Figs. 6-10).



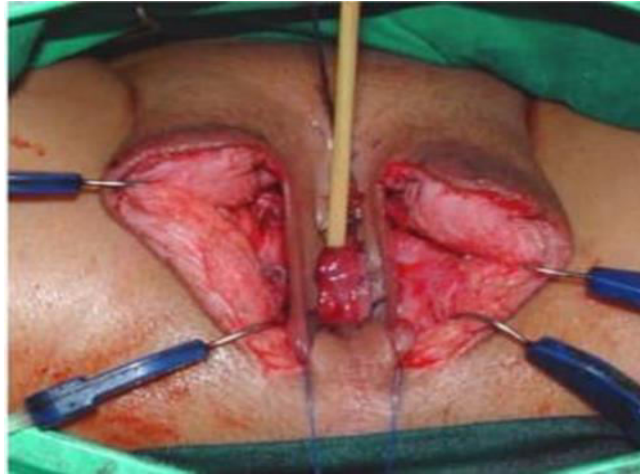
**Fig. 6.** Skin incision is made for degloving of clitoral skin and for preparation of a posterior inverted U shaped perineal skin flap.



**Fig. 7.** Kogan's reduction clitoroplasty and total urogenital mobilization is completed. A very long UGS is shown. The posterior wall of the high vagina is opened just proximal to its junction with urethra. A catheter is shown in vagina.



**Fig. 8.** The distal part of the UGS is split leaving its proximal part to increase urethral length. The split part of UGS is inverted and sutured to the short anterior and lateral vaginal walls.



**Fig. 9.** The inverted U shaped perineal skin flap is used to construct posterior vaginal wall. The long clitoral skin flaps are used to construct the labia minora.



**Fig. 10.** The labial scrotal folds are transposed to create labia majora. A vaginal vasline pack is left for 48 hours.

**Labioplasty:** Following the stitching of the flaps to the vagina, the skin of the labia majora was moved downwards until it reached the bottom of the inverted u-shaped flap. A urinary catheter was retained for a duration of 2–3 days throughout the after-surgery phase (Özer et al., 2018).

**Postoperative management:** cases were administered cephalosporin for a duration of 5 days following surgery. They were also given analgesics and allowed to ingest food orally within the first 24 hours after the operation. Additionally, vaginal calibration & regular dilation were performed if stenosis developed.

#### **Outcome measures**

The postoperative aesthetic and anatomical outcomes were assessed using the genital proportions & symmetry criteria developed by Creighton et al. (2007) and Lean et al. (2005). The study assessed the cosmetic and functional outcomes of a woman's genital appearance, including size, position, vaginal introitus, labial appearance, genital skin quality, & pubic hair distribution. Results were

categorized as good, satisfactory, or poor. Functional results were evaluated based on bowel control, urinary voiding pattern, and vaginal size. Data analysis was done using SPSS software.

**Ethical consideration:** The protocol was submitted for approval by the Research Ethics Committee. Informed consent was obtained from the cases before enrollment in the study. All data was kept confidential.

**Ethical approval code:** SVU-MED-SUR011-2-2020-2-30

#### **Statistical analysis**

Statistical analysis was performed on all collected, tabulated, and assessed data using SPSS 26.0 for Windows (SPSS Inc., Chicago, IL, USA). Qualitative data were described in terms of percentages and numbers. Utilizing range (minimum and maximum), mean, standard deviation, & median, quantitative data were described. The Pearson correlation coefficient, which measures the intensity of the relationship among two variables, was utilized in the tests.

#### **Results**

**Table 1. Age and Sex development disorder among the study population**

Variables	Study population (n = 50)
<b>Age</b>	
Less than one year	19 ( 38% )
Between one year and three years	16 ( 32% )
Older than 3 years	15 ( 30% )
<b>Sex development disorder</b>	
Congenital adrenal hyperplasia	46 ( 92% )
Complete androgen insensitivity syndrome (CAIS)	4 ( 8% )

(Table.1) shows that the number of patients with less than one year of age in the research population

was 19 (38%). The number of patients with congenital adrenal hyperplasia in the research population was 46 (92%).

**Table 2. Clitoris characteristics among the study population**

Variables	Study population (n = 50)
<b>Clitoris size</b>	
Normal	41 ( 82% )
Small	3 ( 6% )
Absent	3 ( 6% )
Large	3 ( 6% )
<b>Clitoris position</b>	
Normal	46 ( 92% )
Absent	4 ( 8% )

(Table.2) showed that the number of patients with normal Clitoris size in the research individuals was 41 (82%), and the number of

patients with normal Clitoris position in the research individuals was 46 (92%).

**Table 3. Labia majora and labia minora among the study population**

Variables	Study population (n = 50)
<b>Labia Majora</b>	
Normal	43 ( 86% )
Redundant	5 ( 10% )
Scrotalized	2 ( 4% )
<b>Labia Minora</b>	
Normal	43 ( 86% )
Poor	4 ( 8% )
Absent	3 ( 6% )

(Table.3) showed that the number of patients with Normal Labia Majora in the research population was

43 (86%). The number of patients with normal labia minora in the research was 43 (86%).

**Table 4. Overall cosmetic outcome among the study population**

Variables	Study population (n = 50)
<b>Overall cosmetic outcome</b>	
Good	41 ( 82% )
Satisfactory	6 ( 12% )
Poor	3 ( 6% )

(Table.4) showed that the number of patients with a good overall

cosmetic outcome in the study population was 41 (82%).



**Table 5. Postoperative complications among the study population**

Variables	Study population (n = 50)
Stress incontinence	4 ( 8% )
Dysuria	3 ( 6% )
Difficult micturition	2 ( 4% )
Wound infection	2 ( 4% )

(Table.5) showed that the number of patients with stress incontinence among the research individuals was 4 (8%). The number of patients with dysuria in the research

was 3 (6%). The number of patients with difficult micturition in the research was 2 (4%). The number of patients with wound infections among the research individuals was 2 (4%).

**Table 6. Congenital adrenal hyperplasia characters among the congenital adrenal hyperplasia population**

Variables	Congenital adrenal hyperplasia population (n = 46)
<b>21-hydroxylase deficiency</b>	
- Yes	46 (100%)
- No	0 (0%)
<b>CAH type</b>	
- Salt wasting (SW)	41 (89.13%)
- less severe simple virilizing (SV)	5 (10.87%)
<b>Prader's score</b>	
- II	2 (4.35%)
- III	9 (19.57%)
- IV	20 (43.48%)
- V	15 (32.61%)

(Table.6) showed that the number of patients with 21-hydroxylase deficiency in the research population was 46 (100%). The number of patients with salt-wasting

(SW) CAH type in the research population was 41 (89.13%). The number of patients with (II) Prader's score among the research individuals was 2 (4.35%).

**Table 7. Pearson's correlation coefficients (r) between Age (Older than 3 years) and postoperative outcomes**

Variables	Age (Older than 3 years)	
	Pearson's correlation coefficients (r)	P
<b>Overall cosmetic outcome (Poor group)</b>		
	<b>0.386</b>	<b>0.006</b>
<b>Stress incontinence</b>		
	<b>0.290</b>	<b>0.041</b>
<b>Dysuria</b>		
	<b>0.202</b>	<b>0.159</b>
<b>Difficult micturition</b>		
	<b>0.089</b>	<b>0.538</b>
<b>Wound infection</b>		
	<b>0.089</b>	<b>0.538</b>

(Table.7) presents Pearson's correlation coefficients (r) between age (older than 3 years) and various postoperative outcomes. Significant positive correlations were found between age and overall cosmetic outcome ( $r = 0.386$ ,  $p = 0.006$ ) and stress incontinence ( $r = 0.290$ ,  $p = 0.041$ ), indicating that older age is associated with poorer cosmetic outcomes and increased stress incontinence. No significant correlations were observed between age and dysuria, difficult micturition, or wound infection, with p-values of 0.159, 0.538, and 0.538, respectively.

### Discussion

Disorders of sex development (DSD) include a diverse array of congenital conditions characterized by incongruent development of the anatomical sex, chromosomes, gonads, & brain, all of which are essential to the process of sexual differentiation. (Fisher et al., 2016).

Our results showed that age and sex development disorders were similar among the research individuals. The number of cases with less than one year of age in the research was 19

(38%). The number of cases of congenital adrenal hyperplasia in the research individuals was 46 (92%).

Our results agree with those of Ekenze et al. (2019), who discovered that the patients presented at a median age of twelve months (ranging from 2 days to 30 years), with fifteen (60%) cases having been raised as females prior to presentation and ten (40%) cases having been regarded as males. Our results showed similar characteristics between the research individuals. The number of cases with normal criteria in the research population was 41 (82%). The number of cases with a normal clitoris position in the research individuals was 46 (92%).

Cisternal reduction and vulvar-vaginal reconstruction were performed in our case in accordance with the patient's gender assignment. Following the patient's intention to enter into marriage as a woman, she underwent progesterone-based hormonal therapy, bilateral orchidectomy, and a vaginal introitus widening operation to restore vaginal function. To prevent the need for surgery to create a neovagina, the vaginal introitus widening procedure

was conducted utilizing a finger and speculum.

In the meantime, clitoral reduction was performed in a nerve-sparing fashion to preserve sexual satisfaction. Sexual satisfaction and the overall outcome subsequent to the operation were considered satisfactory. (Moegni et al., 2022).

Our results showed labia majora and labia minora between the research individuals. The number of cases of normal labia majora in the research individual was 43 (86%). The number of cases of normal labia minora in the research individual was 43 (86%).

Our findings align with those of Brodie et al. (2019), who reported a mean age of 14.4 years with a range of ten to nineteen years. The mean weight was 60.8 kilograms, & the mean height was 159.6 cm. The lengths of the right and left labia minora varied in (n = 19/44) cases (43%). The widths of the right & left labia minora varied between one & twenty-two millimeters between extended (n = 20/33; 61%) & unstretched (n = 24/44; 55%) labia. There wasn't an association among the age or height of the case & the size or morphology of the labia minora.

The results of our research demonstrated the cosmetic outcome for the entire study population. The proportion of cases in the research of individuals who achieved good cosmetic outcomes was forty-one (82%).

Our results agree with Al-Dessoukey et al. (2009) who found that 11 patients with low & intermediate vaginal confluence did vaginal back cutting with excellent cosmetic & functional outcomes in all 49 patients with intermediate-high confluence and did urogenital sinus (UGS) mobilization with very good cosmetic results in 42 patients (86%).

Our results showed postoperative complications between the research individuals. The number of cases of stress incontinence in the research individuals was 4 (8%). The number of cases of dysuria in the research individuals was 3 (6%). The number of cases with difficult micturition in the research individuals was 2 (4%). The number of patients with wound infections among the research individuals was 2 (4%).

Outcomes are urinary complications as regards voiding function and continence, which is a long-term outcome. Intermittent urinary difficulties, dysuria, & frequency were observed in two cases (6.7%); their uroflowmetry and ascending cystourethrography demonstrated hyperactive bladder & detrusor instability. One case had urinary incontinence & dribbling & is currently being investigated for further management (Marei et al., 2014).

In 16.66% of cases, wound infections occurred; these were treated only with wound care. This percentage was greater than that of Badawy et al. (2012), Bernabé et al. (2018), and Farkas et al. (2001), whose wound infection rates were 3.33 percent, 0%, & 2%, respectively. This finding primarily shows the necessity for enhanced infection control measures in the operating room & following surgery.

Palmer et al. (2012) observed that cases who were continent prior to the surgery didn't have any complaints of urinary incontinence in the initial weeks following surgery. This finding is consistent with the results reported by Badawy et al. (2012), who documented stress urinary incontinence that endured for a duration of six months.

Our results showed congenital adrenal hyperplasia among the congenital adrenal hyperplasia

population. The number of patients with 21-hydroxylase deficiency in the study population was 46 (100%). The number of cases of salt wasting (SW) CAH type in the research individuals was 41 (89.13%). The number of cases with (II) Prader's score among the research individuals was 2 (4.35%).

Our findings are consistent with those of **Muhammed et al. (2023)**, who discovered that chromosomal analysis identified 95.83% XX and 4.16% XY. As per Prader's classification, 74.07% were Prader 3/4 individuals. with regards to Prader's categorization One case (4.16 percent) was Prader 1, five cases (20.83 percent) were Prader 2, seven cases (29.16 percent) were Prader 3, ten cases (41.66%) were Prader 4, and one case (4.16 percent) was Prader 5. In 83.33% of cases, the urogenital sinus length was less than three centimeters, while in 16.66% it was greater than three centimeters. Surgical interventions: 70.83 percent underwent labioplasty, clitoroplasty, monsplasty, or a Fortunoff flap as part of partial urogenital mobilization; 16.66 percent underwent cutback vaginoplasty; & 12.5 percent staged repair without vaginal replacement. 8.33 percent of patients experienced minor hemorrhage after surgery, 4.14 percent experienced seroma, 16.66 percent experienced wound infection, & 4.16 percent experienced minor labia majora wound dehiscence; urinary issues & no clitoral loss were reported. Satisfaction of patients and their families: 87.5 percent reported positive results, while 12.5 percent were satisfactory.

Pearson's association coefficients (r) among age (greater than three years) & outcomes after surgery were calculated in the present research.

Pearson's correlation coefficients (r) between age (older than

3 years) and overall cosmetic outcome (poor group) were 0.386, with a strong positive relationship between the two variables. Pearson's correlation coefficients (r) between age (older than 3 years) and stress incontinence were 0.290, with a strong positive relationship between the two variables.

Our findings are consistent with those of **Baskin et al. (2020)**, who discovered that the Prader score & cosmesis rating for parents did not correlate ( $r = 0.18$ ,  $p = 0.22$ ;  $r = 0.07$ ,  $p = 0.67$  for the father). At baseline, there was an association ( $r = 0.45$ ,  $p = 0.01$ ) among the Prader score & cosmesis for surgeons, but this association did not remain at 6- or 12-month monitoring. Furthermore, no correlation was observed among the Prader score & complications at either the 6-month or 12-month marks.

Additional analysis was conducted to determine whether cosmesis ratings differed between the initial evaluation & the follow-up. There is a statistically significant decrease in the possibility of mothers who provided a lower cosmesis rating at baseline following up at six months and twelve months ( $p = 0.03$ ). The trend indicates that fathers who provide a lower cosmesis rating at baseline aren't significantly less likely to follow up ( $p = 0.06$ ). A lack of correlation ( $p = 0.45$ ) is observed among the cosmesis score & the probability of surgeon monitoring.

### Conclusion

The number of patients with a decreased aldosterone level in the research population was 41 (82%). The number of patients with an increased renin level in the research population was 41 (82%). The number of patients with a decreased cortisol level in the research population was 49 (98%). The number of patients with 17-OH-progesterone >20,000 ng/dl in the research population was 41 (82%). The

number of patients with an increased testosterone level in the research population was 50 (100%).

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