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A novel surgical technique in feminizing genital reconstruction: a prospective cohort case series

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ABSTRACT

Background: Disorders of Sexual Development (DSD) encompass congenital conditions with inconsistencies between sex chromosomes, gonads, and sexual anatomy. This study aimed to evaluate the outcomes of a genitoplasty technique in DSD patients, focusing on functionality and cosmetics.

Methods: A prospective cohort case series was conducted at a university hospital in Riyadh, Saudi Arabia. Ten DSD patients underwent genitoplasty procedures with excision of the urogenital membrane and use of the local flap of labia minora for reconstruction. Data, including demographic information, operative findings, and immediate and delayed postoperative information, were collected.

Results: The single-stage genitoplasty technique demonstrated improved delayed cosmetic outcomes. Patients undergoing single-stage genitoplasty with excision of urogenital membrane and use of local flap of labia minora for reconstruction. This approach yielded superior long-term aesthetic outcomes, eliminating the need for additional surgeries.

Conclusion: The new novel technique showed delayed satisfactory cosmetic outcomes compared to the previous one. Additionally, the new technique significantly reduced the need for subsequent interventions, such as second genitoplasty. These findings support the potential benefits of the new novel technique in improving long-term outcomes for individuals with DSD.

Keywords: Genitoplasty, feminizing reconstruction, prospective cohort, pediatric surgery.

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Background

Disorders of Sexual Development (DSD) encompass a diverse range of congenital conditions that result in inconsistencies between an individual's sex chromosomes, gonads, and sexual anatomy. Surgical intervention plays a crucial role in addressing the anatomical, functional, and psychosocial aspects associated with DSD, particularly in feminizing genital reconstruction [1,2]. Historically, surgical techniques for feminizing genital reconstruction in DSD have evolved alongside advances in understanding anatomy and surgical practices. These techniques aim to create genitalia that align with an individual's gender identity and enhance their physical and psychological well-being. Common surgical procedures include clitoroplasty, vaginoplasty, and other tailored approaches [1-4]. The surgical management that most commonly used previous technique that was compared with our new one is where the urogenital membrane is refashioned to resemble the labia minora, while the labia majora is constructed using the remnant of skin from the scrotum. However, the new technique involves the complete excision of the urogenital membrane and the creation of a 270-degree superior-based flap, to which the skin is then attached.

However, the long-term outcomes and the need for subsequent surgeries in DSD patients undergoing feminizing genital reconstruction require further investigation. While surgical techniques have improved, there is still limited data on the effectiveness of these procedures and the occurrence of complications. Understanding the rate of additional surgeries needed to achieve satisfactory cosmetic results is essential to optimize patient outcomes and minimize the need for further interventions [5].

To address these gaps, this prospective cohort case series aims to evaluate the outcomes of surgical techniques in DSD patients, specifically focusing on functionality, cosmetic results, the occurrence of complications, and delayed outcomes. By assessing the need for additional surgeries and identifying factors that contribute to successful outcomes, this study aims to improve the care provided during feminizing genital reconstruction for individuals with DSD. Ultimately, the goal is to enhance both the physical and psychological well-being of DSD

patients and provide evidence-based guidance for future surgical interventions.

Methodology and Materials

Study design and setting

This study employed an observational, single-center prospective design. The study focused on 10 infants who were born with DSD and underwent surgical reconstruction (Figures 1-3). The infants were assigned a female gender based on imaging, chromosomal analysis, and genetic testing. The study was conducted at a university hospital. Data collection of the previous technique was spanned from 2016 to 2019, and patients were enrolled for the new one from 2019 to 2022. The length of follow-up postoperative to measure the delayed cosmetic outcome ranges between 6 months to 1 year.

Data collection

Patients meeting the selection criteria were identified through electronic medical records and were involved in the study. Data were collected and included demographic information: assigned gender, confirmation by imaging, analysis, and genetic testing age at the time of gender assignment, family history, and associated anomalies. Operative information: age at the time of surgery, clitroplasty, excision of the urogenital membrane, and use of local flap of labia minora for reconstruction. Immediate postoperative information: proper pain management and use of morphine infusion, admission to the Pediatric Intensive Care Unit (PICU), and presence of wound discharge. Delayed postoperative information (6 months to 1 year postoperative): cosmetic outcome during follow-ups, urethral function, and the need for future surgical intervention.

The study obtained institutional research ethics board approval from the medical ethics committee at King Saud University in Riyadh, Saudi Arabia.

Statistical analysis

The data were collected, reviewed, and then fed to Statistical Package for Social Sciences version 21 (SPSS: An IBM Company). All statistical methods used were two tailed with an alpha level of 0.05 considering significance if the *p*-value less than or equal to 0.05. Descriptive analysis was done by prescribing frequency distribution and percentage for study variables including cases age, gender, and other demographic data. Also, operative and post-operative data and the surgery outcome indicators were compared between the study groups (new *vs.* previous technique) Pearson chi-square test for significance and exact probability test if there were small frequency distributions. An exact logistic regression model was used to identify the most significant predictors of favorable outcomes (patient satisfaction).

Results

A total of 10 cases were included in the study, with 5 cases undergoing the previous technique and 5 cases undergoing the new technique. All cases were assigned a female gender at birth and confirmed through imaging, analysis, and genetic studies. Among the new technique group, 2 cases (40%) had a family history, compared to 1 case (20%) in the previous technique group (p = 0.490). Urogenital anomalies were observed in 3 cases (60%) of the new technique group, while only 1 case (20%) in the previous technique group had associated anomalies (p = 0.197) (Table 1).

Table 2 presents the operative data for the two study groups. The average age at the time of surgery was 24

	GROUP					
DEMOGRAPHIC DATA	NEW TECHNIQUE		PREVIOUS	P-VALUE		
	NO	%	NO	%]	
Gender						
Female	5	100.0	5	100.0	_	
Gender confirmed by image	aging, analysi	s, and genetic s	studies			
Yes	5	100.0	5	100.0	_	
Age at the time of gende	r assignment	(months)				
At birth	5	100.0	5	100.0	_	
Family history						
Yes	2	40.0	1	20.0	0.490	
No	3	60.0	4	80.0		
Associated urogenital anomalies						
Yes	3	60.0	1	20.0	0.197	
No	2	40.0	4	80.0		

Table 1. Demographic characteristics of study cases by previous and new techniques.

P: Exact probability test.

Table 2. Operation data among the study groups (previous vs. new technique).

	GROUP					
OPERATION DATA	NEW TECH	INIQUE	PREVIO	P-VALUE		
	NO	%	NO	%		
Sexual reconstruction surgery (Genitopla	asty)					
Yes	3	60.0	4	80.0	0.490	
No	2	40.0	1	20.0		
Urogenital membrane excised totally						
Yes	5	100.0	0	0.0	0.002*	
No	0	0.0	5	100.0		
Local flap with labia minora used for reconstruction						
Yes	5	100.0	1	20.0	0.010*	
No	0	0.0	4	80.0		
Age at surgery time (months)						
Range	5-156		5-180		0.481#	
Mean ± SD	54.2 ± 62.5		53.0 ± 74.6			
Median	24		15			

P: Exact probability test.

months for the new technique cases and 15 months for the previous technique cases (p=0.481). Sexual reconstruction surgery (Genitoplasty) was performed in 3 cases (60%) of the new technique group compared to 4 cases (80%) in the previous technique group (p=0.490). Additionally, all cases in the new technique group underwent excision of the urogenital membrane, while none of the cases in the previous technique group required this procedure (p=0.002). Moreover, all cases in the new technique group underwent the use of a local flap of labia minora for reconstruction, whereas only 1 case (20%) in the previous technique group had this procedure (p=0.010).

Post-operative data are presented in Table 3. All cases in the new technique group had proper pain management, they required morphine infusion after surgery with PICU admission to ensure a local flap healing process, compared to 2 cases (40%) and 1 case (20%), respectively, in the previous technique group (p = 0.038). Both groups had clean wounds without oozing or discharge.

Table 4 outlines the clinical outcomes of the study cases based on the technique used. Immediate satisfactory cosmetic outcomes were observed in all cases in both the previous and new technique groups, as well as normal urethral function. Delayed satisfactory cosmetic outcomes were reported in all cases of the new technique group, compared to 2 cases (40%) in the previous technique group (p = 0.038). Additionally, none of the cases in the new technique group required a second genitoplasty, whereas 3 cases (60%) in the previous technique group needed a second genitoplasty (p = 0.038). Refer to Table 2 for age at surgery details.

Table 5 presents the results of the exact logistic regression model for predictors of good satisfaction post-surgery. The new technique showed a significant effect on patient satisfaction, with new technique cases having a 10% higher likelihood of favorable satisfactory outcomes (OR = 1.10; 95% CI: 1.0-12.6).

Discussion

The present study aimed to assess the outcomes of surgical techniques in feminizing genital reconstruction for patients with DSD and compare the results with the existing literature. Our findings provide valuable insights into the effectiveness of the new technique compared to the previous one.

In our study, which focused on patients with DSD, both the previous and new techniques demonstrated immediate satisfactory cosmetic outcomes and normal urethral function, consistent with findings from previous studies [6,7]. However, it is noteworthy that all cases in the new technique group experienced delayed satisfactory cosmetic outcomes compared to a subset of cases in the previous technique group (p = 0.038). This delay in achieving optimal cosmetic outcomes in the new technique group may be attributed to the complexity of the procedure and the need for longer-term healing and remodeling. Regarding the surgical approach, our study revealed that the new technique involved the excision of the urogenital membrane, and the use of a local flap of labia minora for reconstruction, while such procedures were not required in the previous technique group. These differences in surgical approach likely contributed to the variations in outcomes observed between the two groups. The comprehensive excision of

^{#:} Mann-Whitney test.

^{*} p < 0.05 (significant).

Table 3. Post-operative data among the study groups (previous vs. new technique).

	GROUP				
POST-OPERATIVE DATA	NEW TECHNIQUE		PREVIOUS TECHNIQUE		P-VALUE
	NO	%	NO	%	
Proper analgesia					
Morphine infusion	5	100.0	2	40.0	0.038*
paracetamol	0	0.0	3	60.0	
PICU admission					
Yes	5	100.0	1	20.0	0.010*
No	0	0.0	4	80.0	
Clean wound without oozing or discharge					
Yes	5	100.0	5	100.0	-
No	0	0.0	0	0.0	

P: Exact probability test.

Table 4. Clinical outcome among study cases by the undergone technique.

	GROUP				
OUTCOME	NEW TECHI	NIQUE	PREVIOUS TECHNIQUE		P-VALUE
	NO	%	NO	%]
Immediate satisfactory cosmetic outcome					
Yes	5	100.0	5	100.0	-
No	0	0.0	0	0.0	
Delayed satisfactory cosmetic outcome					
Yes	5	100.0	2	40.0	0.038*
No	0	0.0	3	60.0	
Initial planning for second genioplasty					
Yes	0	0.0	3	60.0	0.038*
No	5	100.0	2	40.0	
Normal urethral function					
Yes	5	100.0	5	100.0	-
No	0	0.0	0	0.0	

P: Exact probability test.

the urogenital membrane in the new technique may have provided improved aesthetic results, although it should be noted that the need for such extensive excision should be carefully considered on a case-by-case basis [7-9].

Furthermore, our study highlighted the need for additional interventions following feminizing genital reconstruction. None of the cases in the new technique group required a second genitoplasty, while a significant proportion of cases in the old technique group necessitated additional surgeries (p = 0.038). This suggests that the new

Table 5. Exact logistic regression model for predictors of good satisfaction of undergone surgery.

FACTORS	ODDS	SE	P > Z	95% CI	
PACTORS	RATIO		P > Z	LL	UL
New versus previous technique	1.10	0.05	0.049*	1.00	12.65
Age at the time of surgery (months)	0.99	0.01	0.502	0.97	1.02

SE: Standard error CI: Confidence interval LL: Lower limit UL: Upper limit p < 0.05 (significant).

^{*} p < 0.05 (significant).

^{*} p < 0.05 (significant).







Figure 1. (A-C): Showed cases of ambiguous genitalia upon presentation and prior surgical intervention.

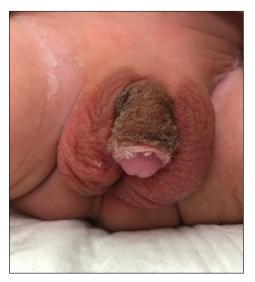




Figure 2. (A and B): Illustrate case no. 1 of the new novel technique, showcasing the excision of the excision of urogenital membrane and use of the local flap of labia minora for reconstruction, before and after surgery.





Figure 3. (A and B): Illustrate case no. 2 of the new technique, showcasing the excision of urogenital membrane and use of local flap of labia minora for reconstruction, before and after surgery.

technique may offer better long-term results, potentially reducing the need for subsequent interventions.

Comparing our findings to the existing literature, several studies have reported favorable outcomes in feminizing genital reconstruction surgery for patients with DSD [9,1,10]. However, direct comparisons between different

surgical techniques are limited in the literature, making it challenging to draw definitive conclusions regarding the superiority of one technique over another. Therefore, our study contributes to the growing body of evidence by specifically evaluating the outcomes of the new technique and comparing them to the traditional approach.

It is essential to acknowledge the limitations of our study. The sample size was relatively small, and the study was conducted at a single center, which may limit the generalizability of our findings. Additionally, the follow-up period for assessing long-term outcomes was relatively short. Future research should involve larger multicenter studies with longer follow-up periods to provide more robust evidence regarding the outcomes and potential advantages of the new technique.

In summary, our study highlights the promising outcomes of the new surgical technique in feminizing genital reconstruction for patients with DSD. The new technique demonstrated immediate satisfactory cosmetic outcomes and normal urethral function, although delayed cosmetic outcomes were observed compared to the previous technique. The need for subsequent interventions, such as second genitoplasty, was significantly reduced in the new technique group. These findings contribute to the existing literature and warrant further investigation through larger-scale studies to optimize surgical approaches and improve long-term outcomes for individuals with DSD.

Conclusion

Our study found that both the previous and new techniques in feminizing genital reconstruction for patients with DSD achieved immediate satisfactory cosmetic outcomes and normal urethral function. However, the new technique showed delayed satisfactory cosmetic outcomes reported by the parents during follow-up in the clinic compared to the previous one. Additionally, the new technique significantly reduced the need for subsequent interventions, such as second genitoplasty. These findings support the potential benefits of the new novel technique in improving long-term outcomes for individuals with DSD.

What is new?

This study evaluates a novel genitoplasty technique for DSD, demonstrating improved long-term cosmetic outcomes and reducing the need for further surgeries. A promising advancement in feminizing genital reconstruction.

Conflict of interest

Not necessary for this manuscript.

Funding

None.

Consent for publication

Not necessary for this manuscript.s

Ethical approval

IRB Approval of Research Project No. E-23-8298.

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References

- Sturma RM, Durbin-Johnson B, Kurzrock EA. Congenital adrenal hyperplasia: current surgical management in United States Academic Medical Centers. J Urol. 2015 May;193(5 Suppl):1796–801. https://doi.org/10.1016/j. juro.2014.11.008
- Lee PA, Nordenström A, Houk CP, Ahmed SF, Auchus R, Baratz A, et al. Global disorders of sex development update since 2006: perceptions, approach and care. Horm Res Paediatr. 2016;85(3):158–80. https://doi.org/10.1159/000442975
- Lee PA, Houk CP, Ahmed SF, Hughes IA; International Consensus Conference on Intersex organized by the Lawson Wilkins Pediatric Endocrine Society and the European Society for Paediatric Endocrinology. Consensus statement on management of intersex disorders. Pediatrics. 2006 Aug;118(2):e488–500. https://doi.org/10.1542/ peds.2006-0738
- Meyer-Bahlburg HF, Dolezal C, Baker SW, Carlson AD, Obeid JS, New MI. Prenatal androgenization affects gender-related behavior but not gender identity in 5-12-yearold girls with congenital adrenal hyperplasia. Arch Sex Behav. 2006 Oct;35(5):667–84. https://doi.org/10.1007/ s10508-006-9068-9
- Yang J, Felsen D, and Poppas DP. Nerve sparing ventral clitoroplasty: analysis of clitoral sensitivity and viability. J Urol. 2007;178(4):1598–601.
- Sircili MH, de Mendonca BB, Denes FT, Madureira G, Bachega TA, e Silva FA. Anatomical and functional outcomes of feminizing genitoplasty for ambiguous genitalia in patients with virilizing congenital adrenal hyperplasia. Clinics (São Paulo). 2006 Jun;61(3):209–14. https://doi. org/10.1590/S1807-59322006000300005
- Roll MF, Kneppo C, Roth H, Bettendorf M, Waag KL, Holland-Cunz S. Feminising genitoplasty: one-stage genital reconstruction in congenital adrenal hyperplasia: 30 years' experience. Eur J Pediatr Surg. 2006 Oct [cited 2023 Oct 25];16(5):329–33. https://doi.org/10.1055/s-2006-924602
- Waterloos M, Claeys T, Sempels M, Van Laecke E, Hoebeke P, Spinoit AF. Genitoplasty in newborn females with adrenogenital syndrome: focus on the reconstruction technique and its outcomes. J Pediatr Urol. 2018 Apr [cited 2023 Oct 27];14(2):198–9. https://doi. org/10.1016/j.jpurol.2018.02.011
- Ekenze SO, Chikani UN, Ezomike UO, Okafor DC. Outcome of feminizing genital reconstruction in female sex assigned disorder of sex development in a low-income country. J Pediatr Urol. 2019 May;15(3):244–50. https:// doi.org/10.1016/j.jpurol.2019.02.021
- Crépaux V, Legendre G. Nymphoplastie: motivations et satisfaction postopératoire. Gynecol Obstet Fertil Senol. 2019 Jun;47(6):489–96. https://doi.org/10.1016/j. gofs.2019.04.005

Summary of the case

	·		
1	Patient (gender, age)	lewborn female babies, gender confimred by testing	
2	Final diagnosis	DSD and underwent surgical reconstruction	
3	Symptoms	mbigous genitalia	
4	Medications	one	
5	Clinical procedure	Senitoplasty	
6	Specialty	Pediatric Surgery	