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ORIGINAL ARTICLE

Reduction clitoroplasty for congenital adrenal hyperplasia: our experience

Borko Stojanovic[1,2], Marta Bizic[1,2], Marko Bencic[1,2], Miroslav L. Djordjevic[1,2]

[1]University Children's Hospital, Belgrade, Serbia
[2]School of Medicine, University of Belgrade, Serbia

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Correspondence to:
Borko Stojanovic, MD, PhD
University Children's Hospital, Department of Urology
School of Medicine, University of Belgrade
Tirsova 10, 11000 Belgrade, Serbia
E-mail: stojanovicborko@gmail.com

Summary

Introduction: Feminizing genitoplasty for congenital adrenal hyperplasia (CAH) includes clitoroplasty, vaginoplasty, urethroplasty and labioplasty. The aim is to achieve normal female anatomy without compromising sexual function. We present our technique and outcomes of reduction clitoroplasty.

Methods: A total of 21 patients with CAH, aged from 13 months to 24 years (median 16 months), underwent feminizing genital reconstruction in our center between January 2011 and December 2020. Surgical treatment included reduction clitoroplasty and labioplasty, with or without urethroplasty and vaginoplasty. Clitoroplasty was performed by complete disassembly of the clitoris into glans with neurovascular bundle and cavernosal bodies. Glans cap was separated from the tips of the cavernosal bodies, avoiding injury of the arteries that run lateroventrally. Cavernosal bodies were completely removed, followed by glans reconstruction and reassembly of all entities, to attain normal clitoral morphology. For hypothesis testing, we used the Mann-Whitney test and Fisher's exact test.

Results: Follow-up ranged from 44 to 162 months (median 84 months). An excellent aesthetic outcome with normal appearance of the genitalia was achieved in all cases. Five patients (24%) reached puberty, and all have regular menstrual cycle. Postoperative complications were observed in 5 (24%) cases. No correlation was found between complications and age at the time of surgery (p=0.131) or duration of surgery (p=0.136).

Conclusion: Reduction clitoroplasty by clitoral disassembly presents a good choice for clitoromegaly repair in patients with CAH. This approach leaves the neurovascular bundle intact and completely preserves the glans cap and urethral plate blood supply. Long-term follow-up is necessary for the evaluation of sensation and sexual function.

Keywords: Congenital adrenal hyperplasia, feminizing genitoplasty, clitoroplasty

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INTRODUCTION

Congenital adrenal hyperplasia (CAH) is diagnosed in one of 10000 newborns, and its treatment requires multidisciplinary approach. The role of surgery is to achieve anatomy and function of female external genitalia, enabling normal psychosocial and psychosexual development. Feminizing genital reconstruction in virilized girls with CAH involves clitoroplasty, labioplasty, urethroplasty and vaginoplasty. The type of surgical reconstruction depends on the severity of the virilization and may include several stages. Appreciation for female genital anatomy and individualized approach are necessary for successful outcome (1-3).

Clitoral hypertrophy is the important feature of CAH, and different treatment approaches have been used throughout history (4). The total amputation of the clitoris used to be the single option because the clitoris was considered unnecessary for female sexual maturation (5). As understanding of clitoral anatomy and sexual function has advanced, more refined surgical techniques have been developed since the initial introduction of reduction clitoroplasty in the 1960s (6,7). Preservation of the neurovascular bundle has become the main goal, and therefore less invasive procedures have been proposed lately. However, loss of sensation, aesthetic mutilation and ultimately compromising sexual function are still relevant issues, making clitoral reconstructive surgery quite challenging. Additionally, data about long-term effects of these procedures are not available (4).

Due to experience in penile disassembly procedure for various congenital and acquired penile anomalies, reduction clitoroplasty based on clitoral disassembly was introduced in our center as a standard part of feminizing genitoplasty. In this study we evaluated surgical outcomes of the procedure for clitoromegaly in CAH.

MATERIALS AND METHODS

A retrospective, single-center study included 21 patients with CAH, aged from 13 months to 24 years (median 16 months), who underwent primary surgical repair between January 2011 and December 2020. The study protocol was approved by the Institutional Ethics Committee at University Children's Hospital (number 12/262, date 11/2024). Patients who underwent re-do clitoroplasty after primary repair elsewhere were excluded from the study. Complete multidisciplinary evaluation is carried out preoperatively, and all patients underwent echosonography and voiding cystourethrography (VCUG). Hormonal therapy was administered preoperatively. Complete one-stage genital reconstruction was performed in 17 patients, and included reduction of hypertrophied clitoris, labioplasty, urethroplasty and vaginoplasty. In 4 cases with severe urogenital sinus, a two-stage surgery was planned, leaving vaginal reconstruction for later age.

Surgical technique

Surgery starts with subcoronal circumferential incision and clitoral skin degloving **(Figures 1A and B)**.

Wide suspensory ligament is dissected in the right plane and preserved, to maintain its role - to prevent the clitoris from straightening, while keeping its stability during sexual activity. Urethral plate is mobilized with Buck's fascia. Dissection is continued towards glans cap, and neurovascular bundle is dissected under Buck's fascia in order to preserve its' structures. Paired clitoral neurovascular bundle originates from pudendal neurovascular bundle, ascending to the upper part of the clitoral body where the crura unite. Glans cap is separated from the tips of the cavernosal bodies, avoiding the injury of the arteries running lateroventrally, as well as dorsal clitoral nerves.

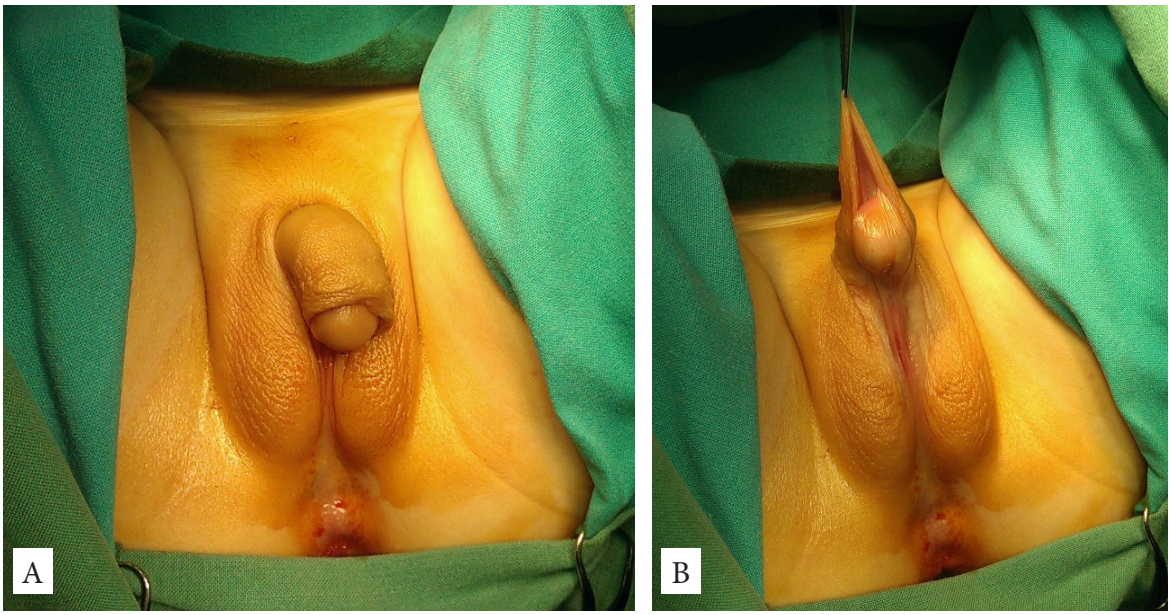

Figure 1. Preoperative appearance of virilized genitalia in an 18-month-old female infant. A. Clitoromegaly with abundant skin. B. Genitalia have hypospadiac aspect. Authors obtained written informed permission to publish these images in scientific/medical journal.

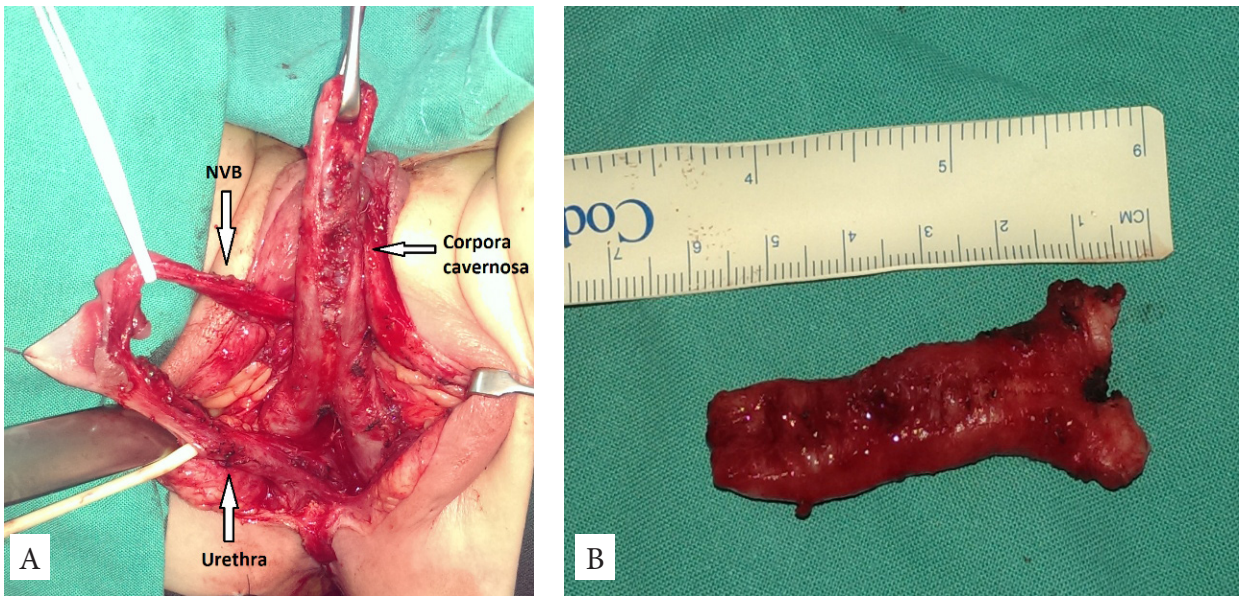

Figure 2. Complete disassembly of the clitoris is done. A. Enlarged cavernosal bodies are totally separated from the glans cap, neurovascular bundle and urethra. B. A 6-cm long cavernosal bodies are removed. Authors obtained written informed permission to publish these images in scientific/medical journal.

The dorsal clitoral nerves pass in large fibers to enter the deep layers of the glans, whose innervation is rich, particularly in its dorsal part. Finally, complete disassembly of the clitoris into glans with neurovascular bundle, urethral plate and corpora cavernosa is done **(Figure 2A)**. This allows adequate removal of corpora cavernosa by complete excision below bifurcation, and suturing the remaining parts limited to the attachment only **(Figure 2B)**.

The glans is then resected, if needed, to achieve normal size, with preserving its' dorsal part that has rich innervation. Reconstruction of the glans and reassembly of all entities is then performed, in order to attain normal clitoral morphology **(Figure 3)**. Labioplasty is then performed, when labia minora and majora are created **(Figure 4)**.

Reconstruction of urogenital sinus follows. A simple cut-back procedure in mild cases, vaginoplasty with U-flap in more severe, or complete urogenital sinus mobilization with complex urethral and vaginal reconstruction in most severe cases and may be performed in one- or two-stages.. Foley catheter is left indwelling for 7 days. Postoperative vaginal dilations are performed in most patients **(Figure 5)**.

Statistical analysis

Depending on the type of variables and the normality of the distribution, data are described as n (%), arithmetic

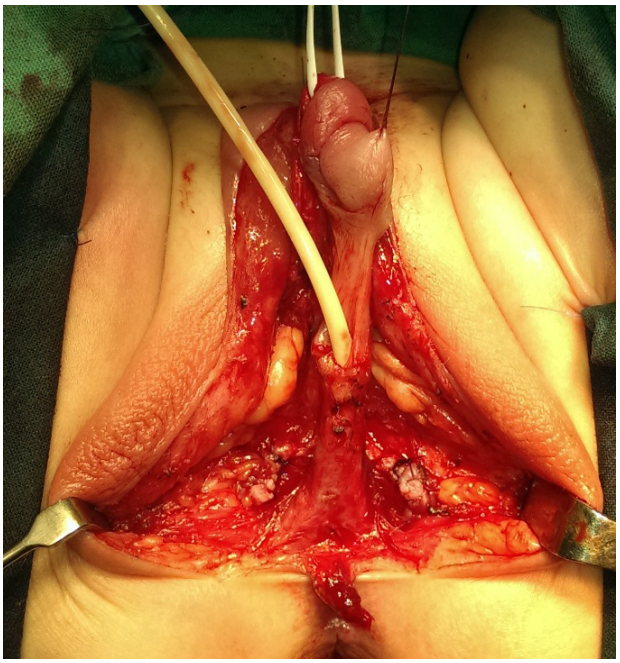

Figure 3. Reassembly of all entities is anatomically planned. Authors obtained written informed permission to publish these images in scientific/medical journal.

Figure 4. Appearance after surgery. Labioplasty, urethroplasty and introitoplasty are performed. Authors obtained written informed permission to publish these images in scientific/medical journal.

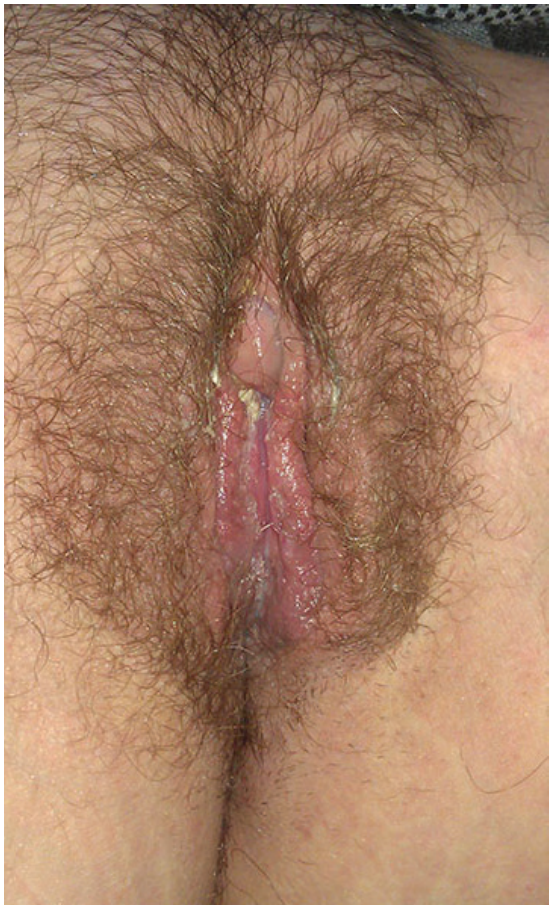

Figure 5. Good aesthetic outcome 10 years after surgery. Authors obtained written informed permission to publish these images in scientific/medical journal.

mean ± standard deviation, or median (min-max). For hypothesis testing, we used the Mann-Whitney test and Fisher's exact test. Statistical hypotheses were tested at a significance level (alpha level) of 0.05.

RESULTS

Follow-up ranged from 44 to 162 months (median 84 months). The median hospital stay was 3 days, ranged from 2 to 7 days. There was no significant intraoperative or early postoperative bleeding. Median length of the removed cavernosal bodies was 4cm, ranging from 3.5cm to 8cm **(Table 1)**.

Good aesthetic outcome with female appearance of vulva was achieved in 20 (95%) patients. Complications were observed in 5 (24%) patients. One case resulted in asymmetry of the glans and labia minora, and partial necrosis of the clitoral skin, which required minor revision 6 months later. One patient presented with stenosis of the vaginal introitus 3 years after surgery and underwent successful repair. Three patients (14%) had postoperative recurrent urinary tract infections and transient urinary leakage, and there were no cases of incontinence. Five patients (24%) reached puberty, and all have regular menstrual cycle. Sensitivity of the reduced clitoris seemed preserved in all cases, according to self-reports. One adult patient reported good sensitivity, penetrative sexual intercourse and satisfaction with surgical outcome. Still, patients' age do not allow complete functional and psychosexual assessment, which is expected after puberty.

There was no correlation between complications and age at the time of surgery (p=0.131) or duration of surgery (p=0.136). Complications occurred in 2 (12%) cases with a low confluence and 3 (60%) with a high confluence, which was not a statistically significant difference (p=0.063). Complications were more frequent among participants who underwent clitoroplasty and labioplasty (100%) compared to those who underwent complete one-stage reconstruction (6%) (p=0.001). Due to the limited sample size and low incidence of potential outcomes of interest - revision surgery (10%), unsatisfactory aesthetic result (5%), transfusions (0%), and clitoral sensitivity disturbance (0%), it was not possible to perform an analysis regarding these outcomes. A limitation of the study is that multivariable analyses could not be performed due to the limited sample size.

DISCUSSION

The treatment for genital ambiguity in CAH is still a subject of debates. Current guidelines suggest a multi-disciplinary approach and a shared decision model, including gender assignment and genital surgery in accordance with national regulations. The type of feminizing genitoplasty depends on the degree of virilization, and current standard of care is to perform reconstructive surgery at an early age rather than wait for adolescence (1,8). A survey among pediatric surgeons and urologists showed that early surgery, before the age of two years, is preferred by 78% of the surgeons and most of them would include clitoroplasty, vaginoplasty and labioplasty (9).

Clitoral reconstruction has evolved from clitoral amputation to less invasive procedures including plication,

Table 1. Summary of case parameters

	Age at surgery (months)	Follow-up period (months)	Hospital stay (days)	Duration of surgery (minutes)	Length of removed cavernosal bodies (cm)
Mean	30.3	91.6	3.2	143.3	4.1
SD	57.4	36.8	1.2	29.8	1.1
Median (range)	16 (13-280)	84 (44-162)	3 (2-7)	140 (90-200)	4 (3.5-8)

* SD – Standard deviation

concealing and reduction (2-7). Recent improvements are based on new insights in clitoral anatomical features and components, including location of the clitoral nerves along the shaft of the clitoris and glans, clitoral body and ligaments (10). Reduction clitoroplasty where part of the shaft is excised with glans preservation is currently the most accepted and used technique. However, this may result in various problems and poor long-term outcomes, including loss of sensation, glans atrophy and sexual dysfunction (11).

Yankovic et al. reported partial excision of the corpora cavernosa as the most frequent surgical technique used for the clitoroplasty (9). A corporal preserving approach is preferred in 20% of surgeons, including surgeons that bury the corpora and those who perform a split-dismembered clitoroplasty. The self-reported outcomes from the participants that perform clitoral surgery are very good in 57%, good in 26% and 16% poor (9). Gupta et al. evaluated long-term cosmetic, functional and psychosexual outcomes in 50 patients with CAH who underwent feminizing genitoplasty (12). They report clitoral reduction with preservation of the glans, without description of the surgical procedure. After the mean follow-up of 6 years, they reported poor cosmetic outcome in 3 out of 50 cases, due to atrophic clitoris in 2 and clitoromegaly in 1 case. The case of recurrent clitoromegaly was explained by non-compliance to hormonal therapy for a period of 3 years, and required re-do surgery. Functional and psychosexual outcomes were satisfactory in the majority of cases (12). One study reports 100% success in terms of size of the clitoris and cosmesis after partial excision of the cavernosal bodies in 82 cases, but without data regarding clitoral sensation or patient satisfaction after surgery (13).

Ventral approach has also been used to preserve neurovascular bundle, removing cavernosal bodies with tunica albuginea, but only distal to bifurcation (14). The authors suggest that by leaving some erectile tissue distal to the bifurcation, clitoral erection is maintained, as well as allowing support and elevation of the clitoris beneath the pubis. However, the long-term results of these partial excisions are not available. A recent comprehensive retrospective study reports favorable long-term outcomes in 40 patients with CAH who underwent genitourinary reconstruction. They also used ventral approach with partial excision and preservation of the bundle. In a median follow-up of 7 years (1-19 years), clitoral glans size was normal and hidden by labia minora in the vestibule in 37 (92.5%) patients. The clitoris was atrophic in one (2.5%), and too large in two cases (5%). (15) Still, concerns related to remaining erectile tissue remain, as the potential need for additional procedures, long-term effects on psychosocial and sexual function, and cosmetic and functional outcomes (16).

We report good surgical outcomes and high level of patients' satisfaction in 21 cases who underwent feminizing genitoplasty, with a mean follow-up of 7 years. Our technique includes clitoral disassembly, which enables complete removal of erectile tissue to the attachments of cavernosal bodies and preservation of all neurovascular elements, allowing anatomical reassembly of entities. In this way, pain and difficulties during sexual intercourse in the future due to remaining erectile tissue is definitely avoided. This approach is based on vast experience in penile disassembly technique as well as clitoral reconstruction in many congenital anomalies and gender affirmation surgeries (10,17). In a long-term follow-up, in this cohort there were no complications such as clitoral atrophy, clitoromegaly recurrence or patients' reports on psychosexual impairment. The statistical analysis revealed no correlation between complications and age at the time of surgery or duration of surgery, as well as significant difference in complication rate regarding level of confluence. However, analysis of other outcome parameters was not possible due to a small number of cases.

The limitations of our study are inconsistent functional and psychosexual evaluation, which can be expected when all patients reach late adolescence or adulthood. The importance of transitional care, especially in these severe cases, is crucial. Also, a limited sample size did not allow for multivariable statistical analysis.

CONCLUSION

Reduction clitoroplasty based on clitoral disassembly in CAH enables complete removal of cavernosal bodies with preservation of the neurovascular bundle, glans cap and urethral plate blood supply. This approach has good long-term surgical outcomes, without compromising the sensation and potential sexual function. Transitional care into adulthood is necessary for complete evaluation.

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Author Contributions: BS: preparing the draft of the manuscript; the acquisition, analysis, and interpretation of data; MBiz: the acquisition, analysis, and interpretation of data; MBen: analysis and interpretation of data; supervision of content; MDj: the conception and design of the work; supervision of content

Ethical approval: The study protocol was approved by the Institutional Ethics Committee at University Children's Hospital (number 12/262, date 11/2024).

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REDUKCIONA KLITOROPLASTIKA KOD KONGENITALNE ADRENALNE HIPERPLAZIJE: NAŠA ISKUSTVA

Borko Stojanović[1,2], Marta Bižić[1,2], Marko Benčić[1,2], Miroslav L. Đorđević[1,2]

Sažetak

Uvod: Feminizirajuća genitoplastika kod kongenitalne adrenalne hiperplazije (KAH) uključuje klitoroplastiku, vaginoplastiku, uretroplastiku i labioplastiku. Cilj je postići izgled i normalnu anatomiju ženskih genitalija, bez oštećenja seksualne funkcije. Predstavljamo našu tehniku i rezultate redukcione klitoroplastike.

Metode: Feminizirajuća genitoplastika je u našem centru urađena kod 21 pacijenta sa KAH, uzrasta od 13 meseci do 24 godine (medijana 16 meseci), u periodu od januara 2011. do decembra 2020. godine. Operacija je uključivala redukcionu klitoroplastiku i labioplastiku, sa ili bez uretroplastike i vaginoplastike. Klitoroplastika je podrazumevala kompletno rastavljanje klitorisa na glans sa neurovaskularnom peteljkom i kavernozna tela. Glans je odvojen od vrhova kavernoznih tela uz prezervaciju arterija. Kavernozna tela su kompletno uklonjena, a potom je učinjena rekonstrukcija glansa i svih struktura, kako bi se postigla normalna morfologija klitorisa. Od metoda za testiranje statističkih hipoteza koristili smo Mann-Whitney test i Fisherov test tačne verovatnoće.

Rezultati: Period praćenja je od 44 do 162 meseca (medijana 84 meseca). Odličan estetski rezultat je postignut u svim slučajevima. Pet pacijenata (24%) je prošlo adolescenciju i ima uredne menstrualne cikluse. Komplikacije operacije su se javile u 5 (24%) slučajeva. Sa komplikacijama nisu povezani: uzrast u vreme operacije (p=0,131) i trajanje operacije (p=0,136).

Zaključak: Redukciona klitoroplastika zasnovana na kompletnom rastavljanju klitorisa daje dobre rezultate kod pacijenata sa KAH. Ovom tehnikom se čuva neurovaskularna peteljka, kao i vaskularizacija glansa i uretralne ploče. Dugoročno praćenje je neophodno za kompletnu evaluaciju osetljivosti i seksualne funkcije.

Ključne reči: kongenitalna adrenalna hiperplazija, feminizirajuća genitoplastika, klitoroplastika

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