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#### Keywords

Sexual function; Voiding status; Long-term follow-up; Feminizing genitoplasty

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# Sexual function and voiding status following one stage feminizing genitoplasty

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## Summary

#### Background

The data on long-term outcome of feminizing genitoplasty (FG) with regard to the sexual function and voiding status is limited and nearly always retrospective, reviewing only small numbers of patients. Furthermore, most articles only looked at the success as an adequately open vagina with a feminized appearance. Success should include normal erotic sensation, lubrication, sexual satisfaction, orgasm, and intercourse without discomfort. These latter factors are just beginning to be studied

# Objective

To investigate sexual function and voiding status in women who underwent vaginal reconstruction in childhood.

# Material and methods

We retrospectively reviewed medical files of patients who underwent FG at our center from 1988 to 2000. In addition, patients were asked to complete the following standardized questionnaires: Female Genital Self-Image Scale (FGSIS), dysfunctional voiding and incontinence symptoms score (DVISS), and Female Sexual Function Questionnaire (FSFI).

#### Results

patients underwent one-stage FG; of those, 36 with median age of 21 years (range 18—30 years) who completed the puberty period were included into this study. Of the participants, 15 (41.6%) lived in a committed partnership, and 12 (80%) had sexual intercourse on a regular basis. Total

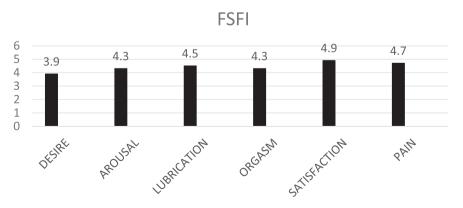
FSFI was 22.2 + 3.1 (mean + SE). Domain scores were 3.9, 4.3, 4.5, 4.3, 4.9, and 4.7 for desire, arousal, lubrication, orgasm, satisfaction, and pain, respectively (Summary Figure). None of the patients reported on daytime incontinence or enuresis. Female genital self-image was found to be positive in all patients and related positively to women's sexual function.

#### Discussion

The general rate of the sexual activity of our studied population was similar to that published in the literature 80% in our study vs 60–87% published in the literature. The overall FSFI was 22, showing some evidence of the female sexual dysfunction. However, only one patient regarding pain during vaginal penetration demonstrated mild dyspareunia. At the same time, we have not found any desire, arousal, lubrication, orgasm, satisfaction problems. Furthermore none of our patients complained on lower urinary tract symptoms during questioning, while three girls with congenital adrenal hyperplasia presented during follow-up with dribbling incontinence, which resolved with introitoplasty and creation of the wide vaginal opening.

# Conclusions

Our data demonstrate that one -tage FG in childhood is a long-term effective surgical procedure that does not cause voiding dysfunction. If a secondary intervention is required prior to the sexual life, simple introitoplasty usually solves the problem. The FSFI score showed overall acceptable sexual function, whereas mild dyspareunia was a major complain.



Summary Figure Female Sexual Function Index (FSFI).

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# Introduction

The evaluation and initial management of the neonate with ambiguous genitalia must be regarded as a medical and psychosocial emergency and be handled with great sensitivity toward the family [1-5]. Clinical management usually includes surgery to render the genital appearance more feminine in infancy and childhood [6,7]. There are a several aims for feminizing genitoplasty (FG), which include: an adequate opening for the vagina into perineum with normal looking wet introitus, fully separated urethral orifice from vagina, and removing phallic erectile tissue while preserving glandular innervation and blood supply [8]. The data on long-term outcome of FG with regard to the sexual function and voiding status are limited and nearly always retrospective, reviewing only small numbers of patients [9.10]. Furthermore, most articles only looked at the success as an adequately open vagina with a feminized appearance. Success should include normal erotic sensation, lubrication, sexual satisfaction, orgasm, and intercourse without discomfort. These latter factors are just beginning to be studied. Cosmetic and early functional results have uniformly been reported to be good. Long-term outcomes are of more concern. Therefore, we have designed this study aiming to investigate the need for reoperation, sexual function, urinary symptoms status, and self-image outcomes experienced by post-pubertal females with disorders of sexual development (DSD) following FG.

## Patients and methods

Following an ethical committee approval, we have reviewed the medical charts of post-pubertal DSD patients who underwent FG at our center from 1988 to 2000.

Our regimen for follow-up, surgical technique, and indications for surgical intervention in DSD children who undergo FG has been previously described [8,11]. In brief, all patients were referred following complete evaluation of gender and chromosomal and biochemical data by pediatric endocrinologists. All patients underwent previously described one-stage FG with slight modifications, which have been adapted over the last decades because of new acquired data on surgical anatomy of urogenital sinus and its neurovascular supply [12,13].

The patients were placed in an exaggerated lithotomy position, and the surgery was performed via the perineal approach. The operation begins with cystoscopy and the placement of Foley or Fogarty balloon catheter into vagina and urinary bladder via common channel. Vertical incisions of the phallic skin on ventral and dorsal surfaces and degloving of the phallus circumferentially are made. The ventral incision is extended to the bottom of the labioscrotal folds in a Y shape and then terminates in an inverted U shaped perineal flap to provide good exposure of the urogenital sinus. The urogenital sinus, including the vagina and urethra, is then completely mobilized en bloc from the corporeal bodies. The dissection is done between the two crura of corpora cavernosa and mobilization continues below the lower rami of the pubis. Thereafter, the dissection between the lateral and posterior walls of the sinus, including the vagina and the anterior rectal wall, is

completed circumferentially so that the posterior wall of the vagina can be brought to the perineum without tension. At this point, subtotal Kogan clitoroplasty is performed. The plane of cleavage is developed between the corpora and dorsal neurovascular bundle via Buck's fascia, taking care to preserve the tunica of the corporeal body. Subsequently, resection of the corpora is performed from the glans to the proximal part passing the bifurcation. The stumps of the corpora are placed and sutured by running five-zero polyglactin sutures below the pubic bone. The proximal phallic skin sutured to the preputial skin and left around the glans clitoris corona creates a preputial hood.

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The posterior vaginal wall is opened over the balloon of the catheter, which previously is placed into vagina between the traction sutures, and the connection between the vagina and urogenital sinus is closed from the internal surface of the vagina. The vagina is opened into an adequate caliber to prevent future stenosis. The previously dissected posterior inverted U is sutured to the dorsal vaginal wall. The redundant distal part of the urogenital sinus is partially opened on the dorsal surface, leaving a long-enough urethra. The inverted open strip of epithelium is sutured to the anterior and lateral vaginal walls, enabling creation of a wet and wide introitus. The preputial and phallic skin is completely split down the midline and sutured around the clitoris and to the lateral epithelial edges of the aforementioned urogenital strips to create the labia minora. The lateral labioscrotal skin can be brought down and sutured to the corners between the poste U flap and lateral skin to form the labia majora.

We have followed our patients at three months, six months, and one year after surgery. The anatomical assessment was focused on vaginal and clitoral dimensions utilizing previously published recommendations and our own experience [12-14]. Vaginal length is measured in centimeters from the posterior fourchette to the posterior fornix or vaginal vault if no cervix is present, and capacity and suppleness are recorded. Capacity is measured by insertion of vaginal dilators of graduating size (Cooper Surgical, USA). Suppleness is the ease with which the vagina stretches and to what extent this is affected by scar tissue. It is very difficult to assess objectively the vaginal suppleness; however, we have utilized the following assessment; if no pressure is needed to introduce a dilator, a score of 1 is given; a score of 2 after moderate pressure, and score 3 when firm pressure is required. The clitoris is measured using a sterile ruler from the base to the tip of the glans clitoris on the dorsal surface and deemed normal if less than 3.5 cm. It is deemed absent when not visible and no corporal tissue is palpable. Cosmetic criteria include the appearance of the glans and clitoral corpora, the presence of a clitoral hood, the appearance of labia minora and majora (rugosity, scarring, and pigmentation), vaginal introital position, and appearance along with overall genital proportions. The overall cosmesis is graded by the surgical team as: good (external genitalia more or less typical for women; unusual features not identified), satisfactory (up to two mildly unusual features identified), or poor (highly atypical, three or more unusual features identified). Examples of unusual features would include an absent clitoral hood or absent or markedly asymmetrical labia minora.

including desire, arousal, lubrication, orgasm, satisfaction, and pain.

The individual domain scores and full-scale (overall) score of the FSFI can be derived from the computational formula outlined in the table. For individual domain scores, add the scores of the individual items that comprise the domain and multiply the sum by the domain factor.

Add the six domain scores to obtain the full scale score. It should be noted that within the individual domains, a domain score of zero indicates that the subject reported having no sexual activity during the past month. Subject scores can be entered in the right-hand column. Full-scale score range is 2.0–36.0. The higher the total score (maximum 36.0), the better is the sexual function in female patients; a domain score of zero indicates that the subject reported having no sexual activity during the past month, a value of 26.55 was classified as having a risk of sexual dysfunction.

Commercially available software Graph Pad Prism 6.01 (Graph Pad prism, Prism 6 for Windows, version 6) utilizing Fisher exact test and unpaired t-test were used for statistical evaluation, with P-value of less than 0.05 considered as significant.

# **Results**

Patient demographics, intraoperative details, and surgical outcome were obtained from a continuously updated database (Table 1.).

In total, 56 patients underwent one-stage FG. Of those, 36 (64.3%) who have completed the puberty and fulfilled all requirements of aforementioned follow-up were included

In those cases where repeat examinations showed a wet and wide introitus and no evidence of fibrosis or scarring of the perineum, we have continued with annual follow-up till puberty. If the patients have had normal menstruations and their physical examination was normal in terms of the introitus appearance, we have continued with annual follow-up till the age of 16-17 years. At this stage, all girls underwent vaginal calibration if we were able to calibrate the vaginal opening easily with a 22 to 24Fr bougie (Sklar Surgical Instruments, USA); we have recommended annual examination till sexual activities commencement. If one of the patients showed symptoms of the introital stenosis, vaginal calibration under general anesthesia was performed. In the majority of the cases, an episiotomy-like operation consisted of longitudinal incision of the perineal skin with extension into vaginal mucosa at 5- and 7-o'clock position and suturing between vaginal mucosa and perineal skin utilizing the Heineke Mikulich principle with introitus widening is performed followed by vaginal calibration done at home by the patients under the supervision of the nurse practitioner. If the episiotomy was insufficient to achieve a desired vaginal caliber, redo FG was performed followed by the self-performed vaginal calibration utilizing above mentioned regimen. Clitoral surgery is offered to girls and women whose clitoris is enlarged and in the presence of distressing symptoms. Psychological assessment and assistance were available to all participants.

To conduct this study, only post-pubertal patients who have completed aforementioned follow-up were included. An initial invitation letter and information leaflet were sent using a clinical database. The study consisted of completion of a telephoned questionnaire. Time to complete the survey ranged from approximately 10 to 20 min. All participants, moreover, were asked to provide a written permission to obtain existing hospital notes to ascertain which operations have been performed.

# Female Genital Self-Image Scale (Appendix A)

The questionnaire included items related to how the woman feels about her own genitals (the vulva and the vagina). The woman needed to indicate how strongly she agrees or disagrees with each statement.

# Dysfunctional voiding and incontinence symptoms score (DVISS) questionnaire (Appendix B)

We have previously published our experience utilizing DVISS in our routine practice [15]. The questionnaire consisted of 13 questions that discussed daytime symptoms, nighttime symptoms, voiding habits, bowel habits, in addition to one quality of life question at the end of the study.

# Female Sexual Function Index (FSFI) questionnaire (Appendix C)

The questionnaire consists of 19 questions with five response options, ranging from never to always in five increments. A global sexual function score is given, indicating general difficulties: six subscale scores are calculated,

**Table 1** Patient demographics, intra-operative details, and surgical outcome.

and sargical outcome.	
Age at primary surgery	Mean years ( $\pm$ SD)
CAH	$0.9 \pm 0.3$
Others	$7\pm3.5$
Diagnosis	N (%)
CAH	33 (91.7%)
Others	3 (8.3%)
Primary surgery	N (%)
Total mobilization of	7 (19.4%)
urogenital sinus (TMUS)	
Partial mobilization of	23 (64%)
urogenital sinus (PMUS)	
Other vaginal surgery	6 (16.6%)
Clitoreduction	30 (83.3%)
Secondary surgery	N (%)
Introitoplasty	7 (19.4%)
Redo clitororeduction	2 (5.6%)
Relationship status	N (%)
Currently in a relationship	15 (41.6%)
Sexually active on the	12 (80%)
permanent basis	

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into this study. Of the 36 patients, 33 (91.7%) had congenital adrenal hyperplasia (CAH), one (2.8%) had mixed gonadal dysgenesis, and two (5.6%) were 46 XX ovotestis DSD. Seven (19.4%) children required total, 23 (64%) partial mobilization of urogenital sinus, four (11.1%) underwent Fortunoff flap only, and two (5.5%) required simple vaginal cut back. Of those, 30 (83.3%) required subtotal clitororeduction during FG. The mean age was 0.9  $\pm$  0.3 years (mean  $\pm$  SD) of the patients with CAH and 7  $\pm$  3.5 of the remainders.

During follow-up, seven (19.4%) required secondary surgery consisting of redo-clitoreduction with introitoplasty in two (5.5%) both due to insufficient androgen suppression and introitoplasty only in the remaining five (13.9%).

The median age of the reviewed in this study patients was 21 years (range 18–30 years). Fifteen (41.6%) of the 36 patients lived in a committed partnership, and 12 (33.3%) had sexual intercourse on a regular basis and fully responded on the questionnaires. These 21 (58.3%) of the 36 patients who have not completed the questionnaire clarified they have not been sexually active, and so, do not feel they are able to give accurate answers and they have been excluded from the study group. However, on the question, "Do you think that the previous history of the vaginal surgery is the cause of absence of the sexual life?," majority responded that they do not see any connection, while eight (39.1%) could not rule out this cause completely.

Total FSFI was 22.2  $\pm$  3.1 (mean  $\pm$  SE). Domain scores were 3.9, 4.3, 4.5, 4.3, 4.9, and 4.7 for desire, arousal, lubrication, orgasm, satisfaction, and pain, respectively (Fig. 1).

All patients have achieved continence without further surgical intervention. None presented with any urinary complaints at the time of study. However, three girls reported daytime and nighttime incontinence after toilettraining. These three patients complained on occasional dribbling incontinence during the adolescence period. On their examination, introital stenosis was diagnosed, and they underwent subsequent introitoplasty. Following surgery, all urinary symptoms disappeared. We have attributed these symptoms to the presence of the urine inside the vagina as a result of urethra vaginal reflux and inability to drainage spontaneously out of vagina due to stenotic introitus.

Finally, female genital self-image was found to be positive in all patients (all the patients have answered with AGREE) and was significantly correlated with female sexual function.

# Discussion

Sexual health is a state of physical, emotional, mental, and social well-being in relation to sexuality; it is not merely the absence of disease, dysfunction, or infirmity [16,17]. Another aspect of the general health is a state of the urinary bladder functioning and presence of the lower urinary tract symptoms (LUTS). The majority of the reports on postpubertal women with history of feminizing genital surgery with regard to the sexual status and voiding function addressed outdated surgical techniques and did not have a long-term fully completed follow-up [9,10]. Crouch et al. published their results concerning the effects of childhood surgery on genital sensitivity and sexual function in adulthood [9]. In their study, sexual function difficulties were more severe in women who underwent surgery, especially vaginal penetration difficulties and intercourse frequency. Linear relationships were observed for impairment to sensitivity and severity of sexual difficulties.

However, the authors have emphasized that all studied patents underwent outdated surgical techniques, often without neurovascular bundle preservation. They suggested that it will be interesting to evaluate the outcome data on surgery performed in the 1990s when these individuals become sexually active.

All our patients underwent one-stage FG utilizing modern technique of mobilization of the urogenital sinus; furthermore, all patients who have had enlarged clitoris underwent subtotal clitororeduction with neurovascular preservation. Our technique has been evolved over the study period adapting a new knowledge regarding neurovascular anatomy of the urogenital sinus and clitoris. Of 36 studied patients, 41.6% lived in a committed partnership, and of those, 80% had sexual intercourse on a regular basis and fully responded on the questionnaires. The remaining 58% of the included into this study patient were not willing to take part in the project. They have cited that they are not living in a committed partnership and have no sexual activity on a regular basis and therefore do not feel that

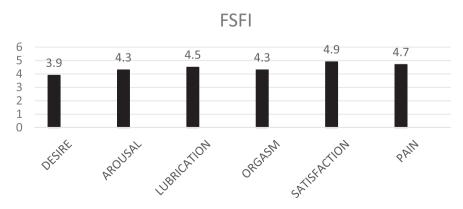


Fig. 1 Female Sexual Function Index (FSFI).

they could make substantial contribution to the study. One has to keep in mind that our study population is very heterogeneous and consisted of different cultural groups, including orthodox Jews and Muslim population, who are not ready very often to discuss their sexual life even with their physician.

However, the general rate of the sexual activity of our population was similar to that published in the literature 80% in our study vs 60-87% published in the literature. Moreover, it has been shown that up to 80% of those who are not engaged in regular sexual activity do not have a partner. The overall FSFI was 22, showing some evidence of the female sexual dysfunction. However, only one patient regarding pain during vaginal penetration demonstrated mild dyspareunia. At the same time, we have not found any desire, arousal, lubrication, orgasm, satisfaction problems. Female sexual dysfunction is not rare finding in the general population [17-20]. The often-cited Laumann study reports 32% desire disorder, 28% orgasmic difficulty, 21% dyspareunia, and 27% stating sex was not pleasurable in the North American population [18]. The Women's International Survey on Health and Sexuality of European Women used the current DSM-IV categories and found 29% desire disorder, 22% arousal disorder, 19% orgasmic disorder, and 14% dyspareunia in over 1300 women between the ages of 20 and 70 years [20]. No doubt that vaginal surgery causes mild dyspareunia in our patients, but it is not associated with significant decrease in the overall sexual satisfaction. Moreover, all our patients were satisfied with appearance of their genitalia. These findings were significantly correlated with female sexual function. Another intriguing issue in this group of patients is LUTS appearance following FG [21,22]. In our clinical practice, we have not noted severe LUTS in female DSD patients. Because of previous contradictory results, we aimed to evaluate the prevalence of LUTS in adult females who had undergone surgery. In a study where the Bristol female incontinence questionnaire was used, 68% of the patients with CAH had urge urinary incontinence and 47% had stress incontinence compared to controls with 16% of urge incontinence and 26% of stress incontinence [21]. In contradiction, a recent study by Fagerholm et al. showed that although LUTS are as common in female DSD patients with FG, the occurrence of LUTS was the same as are in controls [22]. Moreover, some degree of distressing incontinence occurred in 13-25% of the young female patients and the controls. None of our patients complained of LUTS during questioning; however, three girls with CAH presented during follow-up with dribbling incontinence, which resolved with introitoplasty and creation of the wide vaginal opening. Moreover, four patients suffered of low UTI, which also resolved following introitoplasty and self-administrated calibration. We tend to speculate that urethra vaginal reflux into stenotic vagina and difficulties in the urinary drainage due to stenotic opening in such situation may be responsible for the patients complains and could be sorted out with surgical creation and maintenance of the wide introitus. Genital surgery in infancy remains common, while majority of the children undergo one-stage FG with clitororeduction as an integral part of the surgery [23]. There were some reports showing increased number of surgical complications and dissatisfaction among some who had experienced early

genitoplasty, intersex advocacy, and human rights organizations condemn early DSD-related surgery unless medically urgent. A counterpoint is reflected in parents who recall strong wishes to surgically "normalize" their child's sexual anatomy in infancy and early childhood; they viewed genital surgery as "obvious" and "necessary" to assure their child's positive psychosocial and psychosexual adaptation. This perspective is bolstered by follow-up studies of patients who had received early surgery, largely focused on women who had received early FG, suggesting predominantly favorable attitudes toward early feminizing procedures. Cultural attitude of the society of our patients tends toward early "surgical rehabilitation." In the reviewed-here group of patients, 19% required introtoplasty in the long-term follow-up, while 5.5% required reclitororeduction due to clitoris regrowth. Clitoral regrowth after clitoral reduction surgery is not well understood, but has been attributed to increased androgen production at puberty or to poor control in CAH. Surgery to the clitoris risks nerve damage; the risk is likely to be cumulative with multiple operations. It is possible that clitoral surgery performed in the 1970s and 1980s was more radical, leaving little or no clitoral tissue, whereas, later, surgery was less aggressive, leaving a greater potential for clitoral regrowth at puberty. Both our patients showed clitoris regrowth due to insufficient androgen suppression. We have managed vaginal opening stenosis with simple episiotomy and self-administrated calibration then after. Although no objective information was available regarding sensitivity in genital areas in this cohort of patients, all reviewed-here patients were satisfied with the appearance of their genitalia and their positive female genital selfimage; furthermore, we have found a significant correlation between positive genital self-image and female sexual function in our patients.

This manuscript is not without limitations, which are worth to be mentioned. We have performed analysis of our patients in the retrospective fashion and subject to all the flaws inherent to any such study. However, all reviewedhere patients have completed post-pubertal evaluation, and all were available for the telephone interview. Those patients who declined to take part in the study have cited absence of the committed partnership as a reason for unwillingness to take part in this project. Moreover, the majority of these patients did not think that the history of vaginal surgery causes avoidance of sexual activity. We have presented here the results of very heterogeneous population, which address sexual issues in rather different ways. However, our patients reflect a multicultural face of our society, which we have to take in consideration upon surgical decision-making with regard to the timing of genitoplasty and clitoreduction. Furthermore, we have performed a retrospective analysis of patients who underwent different types of FG from total mobilization of UGS to simple cut back. However, we did not think that it will be wise to omit some patients from this study, as it reflects our daily practice, and we all could learn from these data and use it upon surgery counseling. Finally, we have utilized in this study DVISS questionnaire to assess LUTS in our patients, although this questionnaire was suggested for children with dysfunctional voiding. However, we have extensively published the results with the use of + MODEL

this in the children and adults alike in our previous publications [16].

## Conclusions

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Our data demonstrate that one-stage FG in childhood is a long-term effective surgical procedure that does not cause voiding dysfunction. If a secondary intervention is required prior to the sexual life, simple introitoplasty usually solves the problem. The sexual activity rate in the present study was similar to that reported in the literature, with total FSFI score showing overall acceptable sexual function, whereas mild dyspareunia was a major complain.

## **Author statements**

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None.

# Conflict of interest

None.

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# Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jpurol.2019.11.017.