Sexual Function and Attitudes Toward Surgery After Feminizing Genitoplasty

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Abbreviations and Acronyms

AIS = androgen insensitivity syndrome

CAH = congenital adrenal hyperplasia

CAIS = complete androgen insensitivity syndrome

FSFI = Female Sexual Function Index

PAIS = partial androgen insensitivity syndrome

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Purpose: Sexual function and attitudes toward surgery were evaluated in females who had undergone feminizing genitoplasty in childhood.

Materials and Methods: Sexual function and attitudes toward surgery were assessed by a questionnaire in 24 females who had undergone genitoplasty in childhood. Of 16 females who were prenatally exposed to androgens 15 had congenital adrenal hyperplasia and 8 had androgen insensitivity. A total of 18 patients who had reached adulthood were compared with 900 age matched normal controls by using the Female Sexual Function Index questionnaire.

Results: Of the 24 patients 19 had undergone clitoral reduction and 21 had undergone reconstruction of the vaginal introitus. Sigmoid bowel had been used in vaginal reconstruction in 5 patients. There were 17 patients who believed that the genital operation was performed at a proper age, 3 who thought it was done too late while none thought it was performed at too young an age. Two patients regretted the operation, 1 of whom had undergone clitoral resection without nerve preservation and the other had a sigmoid vagina. The control group had more often and earlier (median age 17 vs 19 years) experiences with sexual intercourse. Overall sexual function was similar in the sexually active controls and patients. Decreased sexual desire and problems in achieving orgasm were common but severe pain experiences during penetrative sex were rare in both groups.

Conclusions: Sexual intercoital relationships started later in females who underwent genital reconstruction in childhood. Early surgery is preferred by the patients and satisfactory sex life is possible in adulthood.

Key Words: disorders of sex development; adrenal hyperplasia, congenital; androgen-insensitivity syndrome; sexual dysfunction, physiological

ACCORDING to a recent international consensus statement, genital reconstructive surgery in females with disorders of sexual differentiation should be performed only if the patient is severely virilized.[1] No strict guidelines were given for the timing of surgery in female patients, although it was stated that reconstruction in infancy usually requires revision at puberty. There has been criticism against feminizing genitoplasty, especially if it is performed in early childhood, because it can lead to impaired sexual function.[2–4] However, satisfactory results with early surgery have been reported.[5,6]

We performed a questionnaire study to analyze the sexual function of adult females who had undergone genital

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operations mainly with nerve preservation methods in childhood. The sexual function of the patients was compared to that of a large number of age matched controls.[7] In addition to the patient attitudes the results of surgery were evaluated.

MATERIALS AND METHODS

The operative database of the Hospital for Children and Adolescents, University of Helsinki was retrospectively reviewed for feminizing genitoplasty between 1980 and 2008. A total of 45 patients older than 15 years old were identified and were mailed a questionnaire up to 3 times. Of these patients 24 (53%) returned the questionnaire. Of the 24 patients 16 had prenatal androgen exposure and 46,XX karyotype (CAH group), and 8 had 46,XY karyotype (AIS group). In the CAH group 15 patients had congenital adrenal hyperplasia (3 had a simple virilizing form and 12 had a salt losing form) and 1 had virilization due to a maternal virilizing tumor. In the AIS group 3 patients had complete and 5 had partial androgen insensitivity. None of the patients had a molecular etiology defined.

Patient attitudes were evaluated with several questions and response categories. Was the genital surgery done at the proper age? (too young, proper age, too old, should not have been done), what kind of memories are associated with the operation? (none, positive, reasonably positive, distressing, very distressing), how have you experienced vaginal self-dilations? (easy or distressing, and were they painful), how do you find the sensitivity of your clitoris? (good or poor, and is it painful), is your vagina too tight? (no, yes), are you satisfied with the function of your genitals? (yes, no, why not) and are you satisfied with your sex life? (yes, no, why not). The 18 patients who were older than 18 years old were asked to fill in a Finnish version (translated from English to Finnish by the authors) of a 19-item validated FSFI questionnaire for the assessment of 6 domains of female sexual function (desire, arousal, lubrication, orgasm, satisfaction, and pain) during the last 4 weeks.[8] In the FSFI questionnaire the range between the minimum and maximum total score was 1.2 to 36 and a total FSFI score less than 26.55 was considered abnormal.[9] Patients and controls with no sexual activity during the last 4 weeks were excluded from the FSFI analysis.[10] The FSFI of the patients was compared with that of females of a normal population using a cohort of 900 age matched controls from a Finnish population based sample of twins and their siblings.[7] The Finnish FSFI versions used for the patients and the controls were basically identical. Controls were chosen so that each patient got 50 controls of same age. An exception was the oldest patient (36 years old) who got 6 controls who were 35 years old, 37 controls who were 36 years old and 7 controls who were 37 years old. The prevalence of problems in sexual function was evaluated from the FSFI questionnaire. The subjects were also asked if they have had sex with a partner (yes, no), at what age they had sex for the first time, have they had sex during the last 4 weeks and has the partner(s) been male(s) and/or female(s). The prevalence of sexual dysfunction was calculated using FSFI questions 1, 3, 7, 11 and 17, and options 1 and 2 for those questions.[8] The Mann-Whitney U test was used to compare continuous variables and Fisher's exact test was used to compare categorical variables (Statview® 5.0.1) with $p < 0.05$ considered significant.

RESULTS

Patient age ranged from 15.5 to 36.7 years (median 25). Of the 24 patients 19 had undergone clitoral reduction plasty at a median age of 3.8 years (range 0.4 to 14.3, table 1). Clitoroplasty was performed by resecting the distal part of the corpora cavernosa, and preserving the corpus spongiosum and urethral plate together with the glans. The dorsal neurovascular bundle was macroscopically preserved in all but 1 of the patients. Of 19 patients (37%) 7 had undergone minor revision procedures at an older age (3 patients had 1 reoperation, 2 had repeat surgery 2 times and 2 had repeat surgery 3 times) because of continuous growth of the glans.

All 16 patients in the CAH group and 5 of the 8 in the AIS group had undergone reconstruction of the vaginal introitus or merely the common urogenital sinus (inverted U-flap usually to the base of hymen in 14 and longitudinal incision(s) in 7 (table 1). Median patient age at the first introitoplasty in these 21 patients was 4.5 years (range 0.4 to 19.2). Nine patients (43%) later needed surgery for a narrowed

Table 1. *Type of genital surgery by diagnostic group*

	No. CAH × Group			No. AIS × Group	
	Simple Virilizing	Salt Losing	Virilizing Tumor	PAIS	CAIS
No. Pts	3	12	1	5	3
Clitoroplasty	3	10	1	5	0
Repeat clitoroplasty	1	5	0	1	0
Introitoplasty	3	12	1	4	1
Repeat introitoplasty	0	6	1	2	0
Sigmoid vagina	0	1	0	2	2
Vaginal dilation in pt under anesthesia	1	6	0	4	2
Vaginal self-dilation	1	5	1	4	2
Dilation without vaginal operation	0	0	0	0	1
No need for vaginal dilation	2	5	0	1	0

distal vagina (1 patient had 1 reoperation, 5 had 2 reoperations, 3 had 3 reoperations). In addition, 13 of the 24 patients (54%) required vaginal dilations under general anesthesia starting at a median age of 13 years (range 2 to 19). Fourteen patients (58%) had performed vaginal self-dilations starting at a median age of 17 years (range 14 to 23). Sigmoid vagina had been reconstructed in 5 patients with severe vaginal hypoplasia at age 7, 14, 15, 19 and 21 years. One patient with CAH had a pull-through vaginoplasty.

Surgical Outcomes

Five patients had no memories concerning the genital operations, 11 had positive or reasonably positive memories, and 3 had distressing or very distressing memories. Of the patients with distressing memories 1 had clitoral reduction and introitoplasty at age 5 years, the second had vaginal introitoplasty at the age of 9 years, and the third patient had sigmoid vagina replacement with a temporary protecting colostoma at age 15 years and introitoplasty at age 19. Of the 14 patients with vaginal self-dilations 6 experienced the dilations as distressing.

None of the patients believed that the surgery was performed too early while 17 thought that it was performed at a proper age. Median patient age at the first operation was 2.1 years (range 0.4 to 14.8). Three patients (all with CAH) believed that the surgery was performed too late. Of these patients 2 had clitoral reduction and vaginal introitoplasty at age 14 and 17 years, respectively, and the third patient had vaginal introitoplasty at age 9 years. Two patients (both in the AIS group) believed that surgery should not have been done at all. Of these patients 1 with CAIS had undergone vaginal reconstruction with sigmoid bowel and the other with PAIS had clitoral reduction without preservation of the dorsal neurovascular bundle in the early series.

None of the patients reported pain at the clitoral area, 21 of the 24 considered clitoral sensation to be good, and 3 thought that clitoral sensation was poor or very poor. The patient with very poor sensation had had clitoroplasty without preserving the neurovascular bundle and the other 2 patients with poor sensation had undergone repeat clitoral surgery.

There were 17 patients (77%) who were satisfied and 5 (23%) who were dissatisfied with genital function. Of the 5 dissatisfied patients 3 experienced their vagina as too tight or tender. Two patients were dissatisfied because of poor clitoral sensation. Despite repeat surgery and an intense dilation program overall 8 of 21 (38%) patients (6 of 14 [43%] in the CAH group and 2 of 7 [29%] in the AIS group) considered their vaginas were somewhat too tight.

Sexual Function

Of the 18 adult patients 14 (77.8%, 7 [64%] with CAH, 3 [100%] with CAIS and 4 [100%] with PAIS) and of 867 controls 816 (94.1%, $p = 0.080$) had started having sex. The patients had started sex life with a partner at a later age than the controls (19.2 ± 3.6 vs 17.1 ± 2.4 years, $p = 0.002$). Also, sexual activity with a partner was more frequent in the controls than in the patients (tables 1 and 2). The sexual relationship had been heterosexual in 6 patients (86%) and both with male and female partner(s) in 1 patient 14% in the CAH group. In the AIS group the relationship had been heterosexual in 4 patients (57%). Of the 3 patients with CAIS 1 had a relationship with male and female partners, and 2 of the 4 adult patients with PAIS had homosexual relationships. In the sexually active patients the median number of lifetime partners was 4.2 (range 1 to 10) and 3 of the patients with CAH had children.

Sexually active patients had sexual function similar to that of the controls (tables 2 and 3). Low sexual desire and orgasmic problems were common in the patients and the controls. Of the 5 patients with infrequent orgasms 3 had undergone clitoral reduction and 1 of them reported poor sensation in the clitoris. Experiences of frequent pain were rare in the patients and the controls. However, the pain index was worse in the patients (especially in the CAH group) than in the controls. Total FSFI score was similar in the sexually active patients and controls. Furthermore, the 3 adult patients who were not currently sexually active reported dissatisfaction with their sexual life. Of these dissatisfied patients 1 was a woman with CAH who was currently pregnant, the second was a patient with PAIS with an insensitive clitoris, and the third was the afore-

Table 2. *Sexual activity and prevalence of sexual function problems*

	No. Pos Answers/Total No. Answers (%)			
	CAH	AIS	CAH + AIS	Controls
Sexually active with a partner	7/11 (64)*	5/7 (71)	12/18 (67)*	854/985 (86.7)
Sexual desire only seldomly	3/10 (30)	1/7 (14)	4/17 (24)	591/897 (65.9)
Sexual arousal only seldomly	1/8 (13)	1/7 (14)	2/15 (13.3)	41/831 (4.9)
Lubrication only seldomly	0/7 (0)	2/7 (29)	2/14 (14.3)	26/816 (3.2)
Orgasm only seldomly	3/7 (43)	2/7 (29)	5/14 (35.7)	192/821 (23.4)
Frequent pain during penetration	0/6 (0)	0/5 (0)	0/11 (0)	35/741 (4.7)

* Difference was significant ($p < 0.05$) compared to controls.

Table 3. *FSFI domain scores in sexually active patients and age matched sexually active controls*

	No. (mean ± SD score)							
	CAH		AIS		CAH + AIS		Controls	
Desire	10	(4.0 ± 1.5)	7	(4.5 ± 1.6)*	17	(4.2 ± 1.5)*	895	(3.2 ± 0.9)
Arousal	7	(5.1 ± 0.6)	7	(5.0 ± 1.7)	14	(5.0 ± 1.2)	810	(5.0 ± 0.9)
Lubrication	7	(5.6 ± 0.5)	7	(4.5 ± 2.1)	14	(5.0 ± 1.6)	802	(5.6 ± 0.7)
Orgasm	7	(4.2 ± 2.0)	6	(4.2 ± 1.6)	14	(4.2 ± 1.7)	807	(4.5 ± 1.4)
Satisfaction	7	(4.9 ± 0.8)	5	(5.6 ± 0.4)	12	(5.2 ± 0.7)	748	(5.0 ± 1.0)
Pain	7	(4.7 ± 0.7)*	4	(5.1 ± 0.9)	10	(4.9 ± 0.7)*	726	(5.4 ± 0.8)
Total score	7	(28.7 ± 3.6)	4	(32.3 ± 1.9)†	11	(30.0 ± 3.5)	697	(28.8 ± 3.8)
No./total (%) abnormal total score (less than 26.55)	2/7	(29)	0/4	(0)	2/11	(18)	158/697	(22.7)

* Difference was very significant ($p < 0.01$) compared to controls.
† Difference was significant ($p < 0.05$) compared to controls.

mentioned patient with CAIS with a sigmoid vagina who regretted the operation and who had distressing hospital memories.

DISCUSSION

The patients of the present study started their sex life with a partner an average of 2 years later than the controls. The results are not surprising because in some patients the vaginal dilations were still occurring in early adulthood. Also the percentage of sexually active patients was slightly smaller than in the controls. Previously decreased sexual activity has been reported in patients with CAH,[4,11,12] especially in those with the severe salt losing form.[3,12]

In previous studies of patients with CAH homosexual or bisexual orientation was seen in 10% to 40%.[12,13] In our CAH group the sexual relationship had been heterosexual in 86% and bisexual in 14% of the interviewed patients. The incidence of homosexual or bisexual activity was higher than in a previous Finnish study where homosexual activity was detected in 1.2% of normal twin population.[14]

Sexual behavior has not been studied in patients with AIS as much as in those with CAH. However, it has been considered that sexual orientation in patients with CAIS is similar to normal females.[15] In our study nonheterosexual orientation was more common. Of the 3 patients with CAIS 1 had bisexual behavior and 2 of the 4 patients with PAIS had homosexual behavior.

In this study 23% of the controls and 18% of the sexually active patients had abnormal total FSFI scores. The prevalence of sexual dysfunction in our sexually active controls, and in patients in the CAH and AIS groups was similar to that reported in some previous studies.[16,17] However, our results clearly differ from those of a French study in which the patients with CAH reported largely reduced sexual function despite preservation of dorsal nerves.[12] It has been shown that clitoral amputation leads to impaired sexual function.[4,18] In addition, in our study the importance of nerve preservation in clitoris surgery became evident.

The need for surgical revision has been reported to be 25% to 37% after vaginoplasty.[18,19] In addition, many patients need vaginal dilation especially if surgery is performed before puberty. In our study usually only the urogenital sinus up to the hymen was operated initially. Later 43% of the patients needed operations for a tight vagina, 52% had undergone vaginal dilation under general anesthesia and 62% had performed vaginal self-dilation. Despite active treatment 38% of the patients experienced that the vagina was too narrow. Also in other studies approximately half of the patients with AIS and CAH experienced a narrow vagina.[2,18]

Operative enlargement of the vaginal introitus was not experienced as especially distressful by most of the patients in our study. However, 46% of the patients considered the vaginal self-dilations unpleasant. Similarly in a previous study of patients with AIS only 30% were satisfied with the dilation treatment.[2] In another study only 50% of patients with CAH believed that dilations were beneficial.[3] However, dilations are proven to be an efficient treatment modality in patients with AIS when there is a reasonable starting length of the vagina and good patient motivation.[20] Currently in our unit we start vaginal dilations after puberty when the patient herself is motivated for the treatment. In our clinical experience successful self-calibrations or dilations may reduce the anxiety and fear of first intercourse.

The opinions regarding the timing of the genitoplasty have been controversial. Some have advocated early genitoplasty because the operative circumstances may be optimal and the results are good.[21,22] In 1 study most patients with an early intervention of disorders of sexual differentiation had positive psychosocial and psychosexual outcomes.[6] Others have favored delaying the surgery because of better cosmetic results or fewer compli-

cations.[19] We found only 3 studies in which the patients had been asked about timing of the genitoplasty.[3,18,23] According to these studies most patients with CAH preferred early genitoplasty. Also, most 46,XY patients preferred to undergo the operation before adulthood. In the present study we had similar findings. None of our patients believed that the operation was performed too early but 3 thought it was performed too late. With early introitoplasty patients avoided an unpleasant primary operation for the urogenital sinus at the difficult age when starting menstruation.

Based on these results we prefer early genitoplasty in infancy for prominent clitoris and common urogenital sinus. However, the family must be informed that clitoroplasty carries potential risks of impaired sensitivity and procedures for a narrow vagina are often required before intercourse. In the present series we did not find early introitoplasty to be harmful. Furthermore, it seems that later operations for a narrow vaginal introitus are not stressful for most of the patients. On the other hand, vaginal reconstruction (eg with intestine) because of vaginal hypoplasia should be delayed until young adulthood as recommended recently if it turns out to be necessary at all.[24] We did not find differences in sexual function between patients with CAH and those with AIS, but this may result from the small sample size.

According to our study many females with CAH or AIS start sex life with a partner later than normal females. However, most of the sexually active females have a satisfactory sexual life, which is reassuring for the surgeon and the families.

REFERENCES

1. Lee PA, Houk CP, Ahmed F et al: Consensus statement on management of intersex disorders. Pediatrics 2006; **118:** 488.
2. Minto C, Liao LM, Woodhouse CR et al: The effect of clitoral surgery on sexual outcome in individuals who have intersex conditions with ambiguous genitalia: a cross-sectional study. Lancet 2003; **361:** 1252.
3. Wisniewski AB, Migeon CJ, Malouf MA et al: Psychosexual outcome in women affected by congenital adrenal hyperplasia due to 21-hydroxylase deficiency. J Urol 2004; **171:** 2497.
4. Crouch NS, Liao LM, Woodhouse CR et al: Sexual function and genital sensitivity following feminizing genitoplasty for congenital adrenal hyperplasia. J Urol 2008; **179:** 634.
5. Lee PA and Witchel SF: Genital surgery among females with congenital adrenal hyperplasias: changes over the past five decades. J Pediatr Endocrinol Metab 2002; **15:** 1473.
6. Warne G, Grover S, Hutson J et al: A long-term outcome study of intersex conditions. J Pediatr Endocrinol Metab 2005; **18:** 555.
7. Witting K, Santtila P, Jern P et al: Evaluation of the female sexual function index in a population based sample from Finland. Arch Sex Behav 2008; **37:** 912.
8. Rosen R, Brown C, Heiman J et al: The Female Sexual Function Index (FSFI): a multidimensional self-report instrument for the assessment of female sexual function. J Sex Marital Ther 2000; **26:** 191.
9. Wiegel M, Meston C and Rosen R: The female sexual function index (FSFI): cross-validation and development of clinical cut off scores. J Sex Marital Ther 2005; **31:** 1.
10. Meyer-Bahlburg HF and Dolezal C: The female sexual function index: a methological critique and suggestions for improvement. J Sex Marital Ther 2007; **33:** 217.
11. Kuhnle U and Bullinger M: Outcome of congenital adrenal hyperplasia. Pediatr Surg Int 1997; **12:** 511.
12. Gastaud F, Bouvattier C, Duranteau L et al: Impaired sexual and reproductive outcomes in women with classical forms of congenital adrenal hyperplasia. J Clin Endocrinol Metab 2007; **92:** 1391.
13. Meyer-Bahlburg HF, Dolezal C, Baker SW et al: Sexual orientation in women with classical or non-classical congenital adrenal hyperplasia as a function of degree of prenatal androgen excess. Arch Sex Behav 2008; **37:** 85.
14. Santtila P, Sandnabba NK, Harlaar N et al: Potential for homosexual response is prevalent and genetic. Biol Psychol 2008; **77:** 102.
15. Hines M, Ahmed SF and Hughes IA: Psychological outcomes and gender-related development in complete androgen insensitivity syndrome. Arch Sex Behav 2003; **32:** 93.
16. Laumann EO, Paik A and Rosen RC: Sexual dysfunction in the United States: prevalence and predictors. JAMA 1999; **281:** 537.
17. King M, Holt V and Nazareth I: Women's views of their sexual difficulties: agreement and disagreement with clinical diagnoses. Arch Sex Behav 2007; **36:** 281.
18. Nordenskjöld A, Holmdahl G, Frisen L et al: Type of mutation and surgical procedure affect long-term quality of life for women with congenital adrenal hyperplasia. J Clin Endocrinol Metab 2008; **93:** 380.
19. Burgu B, Duffy PG, Cuckow P et al: Long-term outcome of vaginal reconstruction: comparing techniques and timing. J Pediatr Urol 2007; **3:** 316.
20. Ismail-Pratt IS, Bikoo M, Liao LM et al: Normalization of the vagina by dilator treatment alone in complete androgen insensitivity syndrome and Mayer-Rokitansky-Kuster-Hauser syndrome. Hum Reprod 2007; **22:** 2020.
21. Rink RC and Adams MC: Feminizing genitoplasty: state of the art. World J Urol 1998; **16:** 212.
22. Crawford JM, Warne G, Grover S et al: Results from a pediatric surgical centre justify early intervention in disorders of sex development. J Pediatr Surg 2009; **44:** 413.
23. Meyer-Bahlburg HF, Migeon CJ, Berkovitz GD et al: Attitudes of adult 46, XY intersex persons to clinical management policies. J Urol 2004; **171:** 1615.
24. Purves JT, Miles-Thomas J, Migeon C et al: Complete androgen insensitivity: the role of the surgeon. J Urol 2008; **180:** 1716.