Long-term outcome of feminization surgery: the London experience

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INTRODUCTION

The traditional management of the virilized female infant has centred on restoring 'normality'. Once the diagnosis has been made and the infant assigned to a female sex of rearing, feminizing genital surgery almost inevitably follows. This management is now being criticised from ethical, social and clinical perspectives [1–3]. The current debate is emotive and distressing both for patients who have undergone surgery and for clinicians striving to do their best for their patients in an increasingly litigious world. There are many complex issues to consider before recommending a course of management. Clinicians must have a good knowledge of the current medical literature and should be very clear as to what is supported by a reasonable evidence base, and what is clinical hunch and surmise.

AIMS OF INFANT FEMINIZING GENITOPLASTY

Proponents of feminizing genitoplasty in infancy cite the following as reasons to operate:

- a more stable development of gender identity;
- a better psychosexual and psychosocial outcome;
- relief of parental anxiety;
- provision of a vaginal introitus for psychological relief;
- menstruation and intercourse in adolescence and adulthood.

There is often an unstated assumption in some of the literature promoting infant vaginoplasty that by performing the surgery in infancy the child can be 'cured' and spared the potential psychological trauma of surgery in later childhood or adolescence.

Despite widespread acceptance of these aims of surgery, there are no publications of evidence of the association between genital surgery and an improved psychosocial outcome. There is also no evidence that surgery promotes a stable gender identity development or that gender will develop as assigned. There are increasing data available assessing the effects of surgery on menstruation and sexual function, and here I present past and current data.

THE AVAILABILITY OF LONG-TERM DATA

Objective data on long-term outcomes from representative groups of intersex adults would prove invaluable; unfortunately there is little such published work available. The evidence that there is falls mainly into two groups; psychological follow-up of gender and psychosexual outcomes, and personal surgical series (with operating techniques and variable outcome measures) from clinicians. There are very few studies of long-term outcomes that are not by the original surgeons. Follow-up studies of intersex adults have also been hampered by the widespread policy of nondisclosure, which leaves some adults ignorant of their true diagnosis, and absent from outcome studies.

OUTCOMES OF SURGERY

It is likely that units and individual clinicians will continue to publish small series of patients. To use that outcome data the endpoints should be defined and assessed consistently. Outcome measures should include the following:

- Cosmetic appearance (rated by clinician and patient);
- Anatomical outcome (e.g. size of vagina, ability to inset tampon, have penetrative intercourse);
- Revision rate;
- Complications;
- Psychology and quality of life;
- Sexual function;
- Patient satisfaction.

Such data are patchy or absent from many follow-up studies.

CONFOUNDING FACTORS

Several factors will affect the outcome of surgery; in congenital adrenal hyperplasia (CAH), poor endocrine control leading to ongoing androgenic stimulation can result in to further clitoromegaly and repeated surgical procedures. In all children the cosmetic appearance will change with age. The changes at puberty with pubic hair development and deposition of fat in the labia can result in a dramatic change. An enlarged clitoris in a prepubertal girl will look less prominent after puberty. The patient's opinion of surgery will be altered by her body image, particularly if she has difficulties because of short stature or obesity. Other psychological and psychosocial issues may also affect outcome in intersex women, including an XY karyotype or dissatisfaction with the amount of medical knowledge given to them [4,5].

AVAILABLE DATA TO DATE

There are no standard methods available for cosmetic assessment; attention should be paid to the overall genital proportions and symmetry, distribution of pubic hair, size and shape of the clitoris, labia and introitus, and the genital skin quality, including scarring, rugosity and pigmentation. Whether the cosmetic assessment has been by the clinicians or by the patient is important and should be noted. The patient is likely to be less concerned about the doctor's opinion than her own or that of a potential partner. It is not known whether a partner who is not medically trained would be more or less accepting of genital variation. Where cosmetic appearance has been noted, it is generally poor, with reports varying from 28 to 46% having an unsatisfactory appearance after clitoral surgery [6,7].

Anatomical and cosmetic outcomes are not the same thing; it is possible to have a good cosmetic outcome and yet still have a vagina too narrow for sexual intercourse. It is also possible to have a good anatomical outcome, i.e. a normal calibre vagina, and yet still have

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a poor sexual outcome. Vaginal stenosis is the main anatomical problem and is common, occurring in 36–100% after surgery in infancy [7,8].

Even after a 'one-stage' genitoplasty, revision at adolescence is frequently necessary and the family should be prepared for this. Revision surgery may be on the clitoris, with up to 44% undergoing clitoral revision [9], or the vagina, with estimates of up to 80% revision rate [10].

The complications of genital surgery include urinary infection [11], fistulae [12], clitoral pain and haematocolpos. Incontinence and urethral strictures have also been reported. There are few available data on the long-term urinary function of women after surgery.

The clitoris is an erotically important sensory organ, and its only known function is in contributing to female orgasm. However, sexual response is multifactorial and the exact contribution of the clitoral glans, clitoral hood and clitoral corpora to orgasm is only poorly understood. Recent work on the innervation of the clitoris has shown nerves surrounding the tunica with many perforating branches entering the dorsal aspect of the corporeal body and glans [13]. Any incision to the clitoral glans, corpora or hood may risk damage to the innervation. There are numerous papers on clitoral surgery but only a handful provide follow-up data on psychosexual outcomes. The objectivity of all the follow-up reports to date is poor, and the methods of assessment are either vague or unreported. In only one study were attempts made to objectively evaluate clitoral sensory innervation after surgery [14]. In that study five of six infants were shown to have preservation of a genital electromyographic response at the glans clitoris after stimulating the clitoral dorsal neurovascular bundle, as assessed both before and immediately after clitoral reduction surgery. These patients were all children and so have had no psychosexual evaluation. This study assessed conduction in large myelinated fibres rather than the more appropriate small-diameter myelinated and unmyelinated fibres [15,16].

Two studies assessed in more detail psychosexual function in intersex women after genital surgery. The first compared 34 women with CAH to their sisters without CAH [17]. The CAH group were less likely than their sisters to be sexually active and more likely to have orgasmic dysfunction (33% vs 0%, respectively). The second study of 19 women with CAH compared them to a control group of women with diabetes [18]. Again, those with CAH had significantly less sexual experience, worse orgasmic dysfunction, and were more likely to report problems with penetration. They attributed their difficulties to their surgery.

LONG-TERM SEXUAL FUNCTION IN ADULT WOMEN WITH INTERSEX CONDITIONS

At University College London Hospital (UCLH) we have a multidisciplinary adolescent and adult intersex clinic, accepting referrals from all over the UK. We are also closely linked with the nearby Great Ormond Street Hospital and provide a seamless transition from the paediatric intersex service to the adult service. This means there are many patients for follow-up studies and we have been working for some time on adult outcomes. Our recent work has focused on the following areas:

- Anatomical and cosmetic outcomes in adolescence before sexual activity.
- Sexual function in complete androgen insensitivity syndrome [4].
- Sexual function in adult women with ambiguous genitalia with and without previous feminizing surgery.
- Neurological assessment of clitoral and vaginal sensation after feminizing surgery.

In the study of anatomical and cosmetic outcomes in adolescence [19] we examined 44 adolescents (mean age 15 years) under general anaesthetic to evaluate the cosmetic and anatomical outcome. All had undergone feminizing genital surgery for ambiguous genitalia; 43 (98%) would require further treatment to the vagina before using tampons or undertaking sexual activity. Ten girls would need to use dilators and the other 33 required more genital surgery. There were also many repeated surgical procedures before referral at adolescence for gynaecological assessment; 31% had already had two or more vaginoplasties and 26% had had two or more clitoral procedures. We concluded that repeat procedures were common and could be avoided by deferring the primary vaginal procedure until adolescence.

The study of sexual function in women with ambiguous genitalia with and without previous feminizing surgery [20] included 37 adult women. Of this group 24 had undergone feminizing genital surgery and 13 had not. All were assessed using the Golombok Rust Inventory of Sexual Satisfaction, a UK standardized questionnaire assessing seven areas of female sexual dysfunction and giving an overall score. All patients were also examined gynaecologically. Overall sexual function scores were poorer in both groups than in a standard UK population of women, but there were also other significant differences between the groups. Those who had undergone clitoral surgery were significantly less likely to achieve orgasm than those who had not had surgery (26% anorgasmia vs 0%, respectively). There was no difference in outcome with the type of clitoral procedure, although there were few of each different procedure. The study suggests that cosmetic surgery to the clitoris does not ensure improved adult sexual function and indeed might cause damage.

In the current project on sensitivity testing [21] we measure genital sensation in adult women with CAH who have undergone previous feminizing surgery. Clitoral and vaginal sensation is measured using thermal, vibratory and light-touch sensory thresholds using a GenitoSensory analyser (GSA Medoc Ltd) and Von Frey filaments. Pilot data compared study patients with the normal control values already published in 'normal' women [22]. Preliminary results show markedly impaired sensation [21].

CONCLUSION

Surgery has been regarded as the cornerstone of treatment for virilized female infants and parents. Immediate cosmetic results can be good and this may relieve the concern of both parents and clinicians. However, there is very scanty evidence of a satisfactory postpubertal cosmetic or anatomical outcome. Surgical revision of the vagina to allow tampon use and intercourse is necessary in most patients. If vaginal surgery were deferred it would limit the total number of operations for each individual and may reduce the substantial risk of fibrotic introital stenosis. It is now unacceptable to claim that clitoral surgery does not affect sexual function, although the magnitude of this effect needs further evaluation. In the absence of firm evidence that infant feminizing genital surgery benefits psychological outcome, then the option of no infant genital surgery must be discussed with the family.

The current management of affected patients and their families is difficult and no consensus amongst clinicians has yet been reached. Each case must be considered individually with a clear commitment to transparency of discussion, including possible detrimental effects upon future sexual function. Adequate and informed long-term psychological support should be available to all families whether or not they elect to have surgery.

REFERENCES

2 **Kessler SJ.** *Lessons From The Intersexed.* New Brunswick, New Jersey: Rutgers University Press, 1990.

3 **Phornphutkul C, Fausto-Sterling A, Gruppuso PA.** Gender self-reassignment in an XY adolescent female born with ambiguous genitalia. *Pediatrics* 2000; **106**: 135–7

4 **Minto CL, Liao LM, Conway GS, Creighton SM.** Sexual function in complete androgen insensitivity syndrome. *Fertil Steril* 2003; **80**: 157–64

5 **Migeon CJ, Wisniewski AB, Brown TR *et al.*** 46XY intersex individuals: phenotypic and etiologic classification, knowledge of condition and satisfaction with knowledge in adulthood. *Pediatrics* 2002; **110**: E32

6 **Randolph J, Hung W, Rathlev MC.** Clitoroplasty for females born with ambiguous genitalia. A long-term study of 37 patients. *J Ped Surg* 1981; **16**: 882–7

7 **Alizai NK, Thomas DFM, Lilford RJ, Batchelor AGG, Johnson N.** Feminizing genitoplasty for congenital adrenal hyperplasia. What happens at puberty? *J Urol* 1999; **161**: 1588

8 **Krege S, Walz KH, Hauffa BP, Korner I, Rubben H.** Long-term follow-up of female patients with congenital adrenal hyperplasia from 21-hydroxylase deficiency with special emphasis on the results of vaginoplasty. *BJU Int* 2000; **86**: 253–8

9 **Newman K, Randolph J, Anderson K.** The surgical management of infants and children with ambiguous genitalia. Lessons learned from 25 years. *Ann Surg* 1992; **215**: 644–53

10 **Bailez MM, Gearhart JP, Migeon C, Rock J.** Vaginal reconstruction after initial construction of the external genitalia in girls with salt-wasting adrenal hyperplasia. *J Urol* 1992; **148**: 680–2

11 **Lobe TE, Woodall DL, Richards GE, Cavallo A, Meyer WJ.** The complications of surgery for intersex: changing patterns over two decades. *J Ped Surg* 1987; **22**: 651–2

12 **Rink RC, Adams MC.** Feminizing genitoplasty: state of the art. *World J Urol* 1998; **16**: 212–8

13 **Baskin LS, Erol A, Li YW, Liu WH, Kurzrock E, Cunha GR.** Anatomical studies of the human clitoris. *J Urol* 1999; **162**: 1015–20

14 **Gearhart JP, Burnett A, Owen JH.** Measurement of pudendal evoked potentials during feminizing genitoplasty: technique and applications. *J Urol* 1995; **153**: 486–7

15 **Chase C.** Re: Measurement of pudendal evoked potentials during feminizing genitoplasty: technique and applications. *J Urol* 1996; **156**: 1139–40

16 **Lundberg PO.** Physiology of female sexual function and effect of neurologic disease. In **Fowler CJ**, ed. *Neurology of Bladder, Bowel and Sexual Dysfunction.* Woburn, MA: Butterworth Heinemann, 1999: 33–46

17 **Dittmann RW, Kappes ME, Kappes MH.** Sexual behavior in adolescent and adult females with congenital adrenal hyperplasia. *Psychoneuroendocrinology* 1992; **17**: 153–70

18 **May B, Boyle M, Grant D.** A comparative study of sexual experiences. *J Health Psychol* 1996; **1**: 479–92

19 **Creighton SM, Minto CL, Steele SJ.** Objective cosmetic and anatomical outcomes at adolescence of feminising surgery for ambiguous genitalia done in childhood. *Lancet* 2001; **358**: 124–5

20 **Minto CL, Liao KL, Woodhouse CRJ, Ransley PG, Creighton SM.** The effect of clitoral surgery on sexual outcome in individuals who have intersex conditions with ambiguous genitalia: a cross-sectional study. *Lancet* 2003; **361**: 1252–7

21 **Crouch NS, Minto CL, Liao KLM, Woodhouse CRJ, Creighton SM.** Genital sensation following feminizing genitoplasty for CAH: a pilot study. *BJU Int* 2004; **93**: 135–8

22 **Vardi Y, Gruenwald I, Sprecher E, Gertman I, Yarnitsky D.** Normative values for female genital sensation. *Urology* 2003; **56**: 1035–40

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