Nerve Sparing Ventral Clitoroplasty: Analysis of Clitoral Sensitivity and Viability

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Purpose: Enlargement of the clitoris is often a prominent manifestation of virilizing congenital adrenal hyperplasia and other disorders of sexual development. Controversy persists regarding the viability and sensitivity of the clitoris following clitoroplasty. We present 51 consecutive cases of nerve sparing ventral clitoroplasty performed by a single surgeon.

Materials and Methods: Nerve sparing ventral clitoroplasty was performed in all patients. Postoperative evaluation for clitoral viability included gross examination and capillary perfusion testing. Patients older than 5 years were evaluated for clitoral sensory testing and vibratory sensory testing.

Results: A total of 51 patients 4 months to 24 years old (mean age \pm SD 4.6 \pm 6.8 years) with clitorimegaly underwent nerve sparing ventral clitoroplasty. Of the patients 41 had capillary perfusion testing of the clitoris, of whom all had a viable clitoris. Ten of the 41 patients underwent clitoral sensory testing. Patients reported an average degree of sensation of 3.6 \pm 0.9 at the labia minora and 4.8 \pm 0.4 at the clitoris. Nine of the 10 patients also underwent vibratory sensory testing. Average values for the introitus, clitoris, labia and thigh were 3.56, 1.61, 5.08, and 5.83, respectively. Mean time after surgery for the patients who underwent clitoral sensory testing/vibratory sensory testing was 2.0 \pm 0.8 years. No variations in the sensitivity results were reported at followup in 2 patients.

Conclusions: To our knowledge this is the largest report of followup testing of clitoral viability and sensation after clitoroplasty. Continued long-term followup is ongoing to document long-term sexual function using this nerve sparing ventral approach for clitoroplasty.

Key Words: abnormalities; clitoris; adrenal hyperplasia, congenital; surgery, plastic

pproximately 1/2,000 infants is born each year worldwide with genital ambiguity.1 Management of the physical and potential psychological effects faced by these children and their families requires a compassionate and multidisciplinary approach. The surgical treatment of patients with ambiguous genitalia has been the focus of debate for many years and it remains in large part unresolved. This is especially true with regard to the surgical reconstruction of the masculinized clitoris. Several concerns regarding the current body of knowledge exist and relate to a lack of early and long-term followup of patients undergoing clitoral surgery. Additionally, long-term outcome studies that are available examine clitoral sensation and cosmesis in women after surgical techniques that are no longer used or advised.² Furthermore, it is likely that many of these women were treated at centers where there was limited experience.

Surgical correction of the enlarged clitoris has undergone many changes in the past 3 decades. Initially, clitoridectomy was performed. Recession clitoroplasty was later devised as an alternative. Today, reduction clitoroplasty, where the glans is preserved and part of the erectile bodies are excised,

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is the most widely accepted and used technique.³ Despite advancements in surgical technique controversy persists regarding the viability and sensitivity of the clitoris following reduction clitoroplasty. Additionally, the long-term outcome of sexual function using this technique remains unclear. At a minimum, optimal sexual function in patients undergoing reduction clitoroplasty requires preservation of the innervation and vascular supply to the glans clitoris.⁴ We report our results using an NSVC technique for reduction clitoroplasty based on the current understanding of female clitoral anatomy.

MATERIALS AND METHODS

Under institution review board approval from our medical institution, we retrospectively reviewed the charts of patients that underwent NSVC, as described by Poppas et al, 5 as performed by a single surgeon at 1 institution between 1996 and 2005 for clitorimegaly associated with ambiguous genitalia. Records were also reviewed with respect to diagnosis, patient age at presentation, age at surgery, diagnostic procedures and gender assignments. All patients were assessed and treated by a multidisciplinary team consisting of a pediatric urologist, pediatric endo-psychiatrist and geneticist.

All patients followed routine preoperative protocol, including a liquid diet 48 hours before surgery. Patients were admitted to the hospital the day before surgery for final

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assessment by the multidisciplinary team. Medical treatment was initiated by the endocrinologist in patients who required medical therapy. Patients also received preoperative antibiotics on call to the operating room.

A total of 51 patients 4 months to 24 years old (mean \pm age SD 4.6 \pm 6.8 years) with clitorimegaly associated with ambiguous genitalia underwent NSVC. Of the patients 32 were between 0 and 2 years old, 5 were 2 to 5 years old and 11 were older than 5 years. Of these patients 46 (90%) were genetic females with congenital adrenal hyperplasia, 3 (6%) were 46 XY who had undergone sex reassignment surgeries and 2 (4%) were 46 XX disorder of sexual development. On initial examination, 4 (8%), 22 (43%), 11 (21%) and 6 patients (12%) were rated Prader score II to V, respectively. The remaining 16% of the patients were undocumented. Phallus length was 1.0 to 4.5 cm (average 2.4).

Clitoral Testing

Clitoral viability was evaluated postoperatively by gross examination and CPT. Capillary perfusion of the clitoris was performed by applying pressure to the clitoris until it blanched, when pressure was removed. Capillary perfusion time was determined by the number of seconds until complete reperfusion of the glans. This was compared to the nail bed capillary perfusion test.

Patients older than 5 years were considered candidates for CST. CST was performed using a cotton tip applicator. Using a scale of 0—no sensation to 5—maximum sensation, the patient was asked to report the degree of sensation at various points of the inner thigh and genitalia (labia majora, labia minora, vaginal introitus and clitoris). Inner thigh stimulation was set at level 3 for each patient and used as a baseline to compare other areas tested. In addition, these patients also had vibratory sensory testing performed using a biothesiometer designed to quantify the ability of patients to detect vibratory stimuli. A biothesiometer is most commonly used in testing for neurological diseases, such as peripheral neuropathy.6 The device generates a vibratory stimulus of varying amplitudes that can be gradually increased until the sensation is perceived by patients. A quantitative measure was established by recording the amplitude of vibration (on a scale from 1 to 10), which correlated with the threshold. The amplitude of vibration was inversely proportional to vibratory sensing ability. Two patients underwent sensory evaluation before and after surgery.

RESULTS

Evaluation of postoperative results included assessment of overall cosmesis, CPT and in select patients CST of the clitoris. Followup was 1 week to 8 years (mean 24.4 months). Two patients were lost to followup. No infections, ischemic tissue loss or other postoperative complications were identified. None of the patients in the study group were identified as requiring further revision of the clitoris.

A total of 49 patients (96%) had CPT of the clitoris. All patients had a viable clitoris with normal CPT compared to nail bed testing (less than 5 seconds). Average clitoral reperfusion was 2.8 \pm 0.4 seconds. Ten of the 49 patients (20%) were older than 5 years and were considered candidates for CST. Patients reported an average degree of sensation of 3.6 \pm 0.9 at the labia minora and 4.8 \pm 0.4 at the clitoris. Nine of the 10 patients also underwent clitoral vibratory testing.

Area Tested	No. Pts	Mean ± SD
Capillary refill	49	2.8 ± 0.4
Clitoral sensitivity:	10	
Inner thigh		Referent
Labia minora		3.6 ± 0.9
Clitoris		4.8 ± 0.4
Vibratory sensory testing:	9	
Inner thigh		5.83 ± 1.2
Labia minora		5.08 ± 1.7
Vaginal introitus		3.56 ± 0.5
Clitoris		1.61 ± 0.6

Average values for the introitus, clitoris, labia and thigh were 3.56, 1.61, 5.08, and 5,.83 respectively (see table). Mean time after surgery for the patients who underwent clitoral testing was 2.0 ± 0.8 years. Two patients showed no change in sensation when tested before and after clitoroplasty.

DISCUSSION

Various techniques have been described to correct clitorimegaly. Until the 1960s, the clitoris was typically amputated, resulting in a female appearing perineum but poor sexual function. Alternatives to clitoridectomy were later described, including recession and relocation of the enlarged clitoris. The clitoris was preserved by burying and imbricating the corporeal shaft and excess glans clitoris beneath the pubis. While these procedures maintained clitoral innervation and function with good cosmetic result, there were many disadvantages, including pain during stimulation due to the trapping of erectile tissue beneath scar. In 1970, Randolph and Hung devised a new technique for the correction of enlarged clitoris, that is reduction clitoroplasty.9 A reduction clitoroplasty involves excision of a portion or all of the shaft with preservation of the glans. Spence and Allen later described subtotal resection of the clitoral shaft with preservation of the glans. 10 The major disadvantage to these procedures is that the primary vascular and nervous supply of the glans may be interrupted and the viability of the glans clitoris relies on collateral supply from surrounding soft tissue.

These alternative surgical techniques were followed by descriptions of methods to preserve the neurovascular bundle through a ventral dissection. 11,12 Wedge resection of the glans clitoris has been described for the management of marked clitorimegaly. The glans and corporeal bodies are bivalved in a coronal plane, removing approximately 50% of the girth of the ventral corporeal tissue down to the level of the base of the shaft. No bulging has been reported with stimulation and patients have good sensation and clitoral function. Despite these advances in operative technique, the published literature describing surgical outcomes and functional results reflect complications of older procedures that are no longer used.

Early studies by Money et al showed satisfactory sexual response in females following clitorectomy. ¹⁴ In 1966 Gross et al reported a series of 47 patients who underwent clitoridectomy with good cosmetic results. ¹⁵ Clitoral recession and relocation of the enlarged clitoris has been reported with disadvantages, including painful erections, abnormalities in sexual function, compromised appearance and progression of clitoral enlargement in poorly controlled patients on steroids. Reduction clitoroplasty, as described by Randolph and Hung, was performed on 14 patients with good results in

terms of erection and cosmetic appearance. However, no followup information on the sexual function or clitoral sensation of these patients was reported.

Reduction clitoroplasty in which part or the entire shaft is excised with glans preservation is the most accepted and widely used technique today. Various problems have been associated with reduction clitoroplasty, ranging from loss of sensation and sexual function to sloughing of the glans. 16,17 There are a number of published studies of clitoral sensitivity after clitoroplasty. Minto et al examined 39 adults with a history of ambiguous genitalia who underwent feminizing genitoplasty using a validated sexual function questionnaire and genital examination. 18 They found that patients who had undergone clitoral surgery had significantly higher difficulties with sensuality compared to the no surgery group. The 2 groups had overall difficulties with orgasm. Krege et al assessed long-term followup in 27 patients who underwent clitoroplasty and vaginoplasty. They reported the presence of clitoral sensation in 6 patients who underwent clitoroplasty, but only through qualitative description from a nonvalidated questionnaire. However, none of these reports provided an objective assessment of clitoral viability or followup testing of clitoral sensitivity.

Crouch et al described the initial results from a pilot study involving 6 women who had undergone varying degrees of clitoral surgery. 19 They obtained post-coital questionnaire results as well as specialized sexual function assessment. Light touch, thermal and vibratory sensation thresholds were evaluated for the clitoris and vagina using a genitosensory analyzer and Von Frey filaments. All 6 women were shown to have highly abnormal results for sensation of the clitoris, but these women underwent clitoral surgical procedures that are no longer used. Frost-Arner et al described a similar group of postpubertal women who underwent reduction clitoroplasty at a mean age of 20 years.²⁰ They compared vibration and light touch/deep pressure sensation in this cohort compared to those in a group of normal women. In contrast to the previous studies, they found no difference between women who had single clitoral reduction and the normal cohort. The heterogeneous nature of these data regarding clitoral sensitivity following clitoroplasty stems from the wide variety of surgical techniques used historically in this specialized patient population.

In 2002, a consensus statement recommended that it is crucial to preserve the neurovascular bundle, the glans and the preputial skin of the glans if clitoral reduction is to be performed.⁴ Preservation of the neurovascular bundle relies on an intimate understanding of anatomy. Baskin et al reported a computerized 3-dimensional picture analysis of 14 normal human fetal clitoral specimens.²¹ This permitted a detailed description of the normal fetal clitoral nerve bundles and corporeal bodies. Gearhart et al performed dorsal neurovascular bundle nerve conduction studies at the time of feminizing genitoplasty in 6 patients with an average age of 13 months. They concluded that modern, nerve sparing techniques allow preservation of the neurovascular bundle, which may permit normal sexual function in adulthood.²² The Intersex Society of North America responded by citing the experience of adults who had undergone feminizing genitoplasty as infants who have normal nerve conduction studies and yet have an absence of clitoral sensation and orgasmic experience. Therefore, despite precise anatomical models and careful surgical technique

during clitoroplasty, short-term and long-term followup are needed.

There is an obvious concern relating to attempting to quantify outcomes in clitoral surgery. There are no substantiated tests for assessment of the viability and function of the clitoris in unaffected women. Therefore, we have chosen the capillary perfusion test as a potential model for viability related to clitoral perfusion. Capillary refill testing of the nail bed has long since been used as an overall measure of perfusion, in the setting of dehydration or hypovolemia. The recent meta-analysis by Steiner et al cited capillary refill time, in combination with other clinical parameters such as skin turgor and respiratory pattern, as a useful initial assessment of dehydration.²³ One study used capillary nail bed refill time as an indication of low upper body blood flow in premature infants.²⁴ Similarly, we used this simple technique as estimation of the perfusion of the clitoris, as compared to normal values for capillary refill time of the nail bed. 25

Based on a clear understanding of the female clitoral anatomy we used a modified NSVC technique.⁵ This technique resulted in excellent cosmetic outcome and no postoperative ischemic complications. In addition, the vascular supply to the clitoris was well maintained, as evidenced by a mean CPT of 2.96 seconds (normal less than 5) in all patients tested. CST and vibratory testing results revealed heightened clitoral sensitivity relative to surrounding genitalia. Unlike other studies in the literature, all patients in this study underwent NSVC, which was performed by a single surgeon. To our knowledge this is the largest cohort with followup clitoral viability and sensitivity ever reported. A limitation of this study was the lack of a normal cohort for comparison. Although the results presented suggest relative normal clitoral sensitivity in these patients, continued followup is indicated as well as the use of validated instruments to determine sexual function after the cohort becomes sexually active. At that time, it may be feasible to obtain similar test results from an age matched normal cohort for comparison.

CONCLUSIONS

To our knowledge this is the first report of followup testing of clitoral viability and sensation after reduction clitoroplasty. The nerve sparing reduction clitoroplasty described in this report leaves the dorsal neurovascular bundles of the corporeal bodies and the glans clitoris intact. This is a safe and reliable approach to correct the enlarged clitoris. Sexual and social function of our patient cohort is difficult to assess until all patients reach sexual maturity and adolescence. Continued, long-term followup is ongoing to document long-term sexual function using this nerve sparing approach for clitoroplasty.

Abbreviations and Acronyms

CPT = capillary perfusion testing CST = clitoral sensitivity testing

NSVC = nerve sparing ventral clitoroplasty

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