Phalloplasty in Complete Aphallia and Ambiguous Genitalia

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ABSTRACT

The most common indications for phalloplasty in children include aphallia, micropenis/severe penile inadequacy, ambiguous genitalia, phallic inadequacy associated with epispadias/bladder exstrophy and female to male gender reassignment in adolescents. There are many surgical options for phalloplasty; both local pedicled tissue as well as free tissue transfer. The advantages of local tissue include a more concealed donor site, less complex operation and potentially faster recovery. However, pedicled options are generally less sensate, making placement of a penile prosthesis more risky and many children with bladder exstrophy have been previously operated upon making the blood supply for local pedicled flaps less reliable. Here the authors discuss free tissue transfer, including the radial forearm, the anterolateral thigh, the scapula and latissimus, and the fibula free flaps, as well as local rotational flaps from the abdomen, groin, and thigh. The goal of reconstruction should be an aesthetic and functional (ability to penetrate) phallus, which provides tactile and erogenous sensation, and the ability to urinate standing. Ideally, the operation should be completed in one to two operations with minimal donor site morbidity. There are advantages and disadvantages of each of flap and thus the choice of donor site should be a combination of the patient's preference and surgeon's ability to produce a consistent result.

KEYWORDS: Phalloplasty, children bladder exstrophy, aphallia

The most common indications for phalloplasty in children include aphallia, micropenis/severe penile inadequacy, ambiguous genitalia, phallic inadequacy associated with epispadias/bladder exstrophy, and female to male gender reassignment in adolescents. The reported incidence of bladder exstrophy ranges from 1:10,000 to 1:50,000 live births. Children are born with separated pelvic bones, exposed and everted bladder, severe penile deformity, and the urethral opening proximally on the dorsum of the penis. The anterior corporal length is at least 50% shorter (see Fig. 1). The incidence of cloacal exstrophy is esti-

mated to be 1:200,000 to 1:400,000 live births. Children are born with significant diastasis of the pubic symphysis: The hindgut and cecum are exstrophied between two halves of the bladder, and the phallus and scrotum are also separated into two halves (see Fig. 2).¹ Males with bladder or cloacal exstrophy can have severely ambiguous genitalia and make up the bulk of complex pediatric genital reconstruction. Historically, some of these children have been gender-converted from male to female during infancy and later elect to have a phalloplasty as an adolescent or adult.

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Figure 1 (A) A 6-month-old boy with epispadias. (B) An 8-month-old boy with bladder exstrophy.

SURGICAL OPTIONS FOR PHALLOPLASTY

There are many surgical options for phalloplasty—both local pedicled tissue as well as free tissue transfer. The advantages of local tissue include a more concealed donor site, a less complex operation, and a potentially faster recovery. However, pedicled options are generally

Figure 2 A 12-month-old boy with cloacal exstrophy.

less sensate, making placement of a penile prosthesis more risky.² We will discuss free tissue transfer from the radial forearm, the anterolateral thigh, the scapula and latissimus, and the fibula, as well as local rotational flaps from the abdomen, groin, and thigh. The goal of reconstruction should be an aesthetic and functional (ability to penetrate) phallus, which provides tactile and erogenous sensation, the ability to urinate standing and ideally be completed in one to two operations with minimal donor site morbidity.³ There are advantages and disadvantages of each of flap and thus the choice of donor site should be a combination of the patient's preference and surgeon's ability to produce a consistent result.

Pedicled Groin Flaps

The first reported phalloplasty was a tubularized pedicled flap from the abdomen performed by Bogoras in 1936.⁴ As the field of microsurgery advances, pedicled flaps for penile reconstruction have become less popular. Generally, pedicled flaps are aesthetically inferior and less sensate, precluding insertion of a penile stiffener. Despite a more limited use, pedicled flaps have lower failure rates, donor sites that are easy to conceal, and decreased operative time. Although total free flap failure rates are generally less than 1% partial flap loss, compromising patency of the urethra and final penis length can be as high as 12%.³ Partial flap loss does appear to be associated with tobacco use.

Radial Forearm Free Flap

The radial forearm free flap is one of the most common donor sites for penile reconstruction. Its use for phalloplasty was first described by Song in 1982⁵ and modified by Biemer in 1988 to include a portion of the radius for rigidity.⁶ The donor site is relatively hairless, thin, and supple allowing the flap to be easily tubed and shaped into a penis. The flap has a long pedicle and can be innervated. The lateral and medial antebrachial cutaneous nerves can be coapted to the ilioinguinal and dorsal penile nerves providing both tactile protective and erogenous sensation. The radial artery of the flap can be anastomosed to the profunda femoris, lateral circumflex femoral, circumflex iliac, or the inferior epigastric artery and the venae comitantes; or the cephalic vein of the flap can be anastomosed to branches of the greater saphenous vein (Fig. 3).

Many children with exstrophy will have a continent umbilical bladder stoma and will have no need for urethral reconstruction as this frequently results in incontinence. For these children, the glans of the native penis is de-epithelialized and brought out of the proximal, ventral surface of the neophallus (see Fig. 4). This allows for ejaculation from the native urethra. Without

Figure 3 (A) the outline of the radial forearm phalloplasty flap on the arm. The lateral and medial antebrachial cutaneous nerves can be coapted to the ilioinguinal and dorsal penile nerves. The radial artery of the flap can be anastomosed to either the profunda femoris, lateral circumflex femoral, circumflex iliac, or the inferior epigastric artery. The venae comitantes and the cephalic vein of the flap can be anastomosed to branches of the greater saphenous vein. (B) Illustration of the flap following inset, anastomosis, and coaptation.

Figure 4 Incorporation of the native glans and urethra into the base of the ventral surface of the phalloplasty. These patients have a continent umbilical stoma and therefore do not void through the penis.

the normal peristalsis provided by the corpora bodies, the ejaculate will not reach the tip of the tip of the neophallus if complete urethral reconstruction is undertaken.

For those children who require urethral reconstruction, the flap is designed on the distal portion of the forearm and is divided into three parts. The outer skin envelope of the penis, the deepithelialized portion is used to separate the outer portion of the reconstructed penis from the urethra and the ulnar-sided skin paddle is used to create the urethra (see Fig. 5). The aesthetics of the phallus can be refined by the creation of a corona using a local flap and full thickness skin graft as described by Monstrey et al³ (see Fig. 6). The radial forearm donor site can be closed using a split thickness unmeshed skin graft harvested from the thigh. The donor site is usually too large to cover with a full thickness skin graft. Erectile function can be achieved using either a portion of vascularized radius harvested with the flap or a penile prosthesis inserted at a second procedure 10 to 12 months later after tactile sensation has been restored. A transurethral 16F Foley catheter is left in place for 2 weeks to reduce the risk of stricture and fistula formation while the neo-urethra is healing. Tattooing of the corona to match the color of the areola is performed at 3 months before sensation returns (see Figs. 7, 8, and 9).

Figure 5 (A) The flap is divided into three sections, the outer skin envelope of the penis, the deepithelialized portion, which is used to separate the outer portion of the reconstructed penis from the urethra, and the ulnar-sided skin paddle used to create the urethra. (B) The entire flap is tubed around a 16-French Foley catheter.

Figure 6 To create a glans, the distal portion of the flap is deepithelialized and then curled under. A full thickness skin graft, harvested from the groin is then placed just below.

Monstrey et al have one of the largest series of radial forearm phalloplasties with 287 patients; 100% of patients had tactile sensation at one year, making insertion of a penile prosthesis safe, and 80% had erogenous sensation.³ Complications with the penile implant were most common, with 44% of patients requiring removal due to infection, erosion, or leakage. Urologic complications were second most common, 41% overall, with fistulas being more common than strictures.³ The advantage of the radial forearm free flap is that it is a thin, pliable, relatively hairless skin paddle that provides erogenous sensation, and allows urethral reconstruction in a single stage. The disadvantage is that the donor site can be difficult to conceal and a rigid prosthesis is required for penetration. Given the forearm donor site for a phalloplasty is much larger than that used for head and neck reconstruction, one might speculate that the associated morbidity is higher. Selvaggi retrospectively reviewed the donor site morbidity in 129 patients who had undergone radial forearm phalloplasty.⁷ The average flap size was 228 cm², often including the Palmaris tendon and with an incision extending to the antebrachial fossa. The complications and rates were comparable to that of small radial forearm flaps and included partial skin graft loss, decreased sensitivity, swelling, decreased range of motion (resolved with hand therapy).

Figure 7 (A) Partial amputation of the penis following an explosion of an improvised explosive device. (B) After penile and urethral reconstruction and tattooing.

There was no difference in patient reports of aesthetic satisfaction between the full thickness and split thickness skin grafts.

Anterolateral Thigh Flap

The anterolateral thigh (ALT) flap can be used for both pedicled and free flap phalloplasty. Descamps et al report on a small series of children with aphallia reconstructed with a pedicled sensate ALT flap.⁸ Felici et al were the first to describe the use of the free ALT for phalloplasty.⁹ The flap is based on the descending branch of the lateral femoral circumflex vessels with innervation provided by the lateral femoral cutaneous nerve. Rubino et al describe harvesting the flap with a 3 cm cuff of fascia, which can be tubed to provide a neotunica to cover the penile prosthesis.¹⁰ To create a neo-urethra, a separate skin graft must be harvested, wrapped around a catheter, and sewn to the native urethra. Mutaft et al describe a pedicled chimeric flap in which the sartorius is harvested and tubed to create the neo-urethra within the tubed ALT.^{11,12} The advantage of the sartorius is that it is well vascularized and hairless. The pedicled ALT is particularly useful when the pelvic vascular anatomy is uncertain such as in children who have undergone multiple procedures for bladder exstrophy. Lumen et al compared the radial forearm free flap to the pedicled ALT flap and found that although some patients felt the ALT donor site was easier to conceal and the operative time was decreased, the sensitivity of the ALT compared with the radial forearm is markedly decreased.^{13,14}

Figure 8 (A) A 16-year-old boy with severe micropenis and bladder exstrophy (B) who underwent radial forearm phalloplasty. Urethral reconstruction was not undertaken as the patient has a continent umbilical stoma.

Figure 9 (A) This is a 17-year-old patient with micropenis and bladder exstrophy who underwent a radial forearm phalloplasty. (B) Flap after tubularization and creation of the neoglans. Urethral reconstruction was not performed as the patient has a continent umbilical stoma. (C) Immediately following anastomosis. The corona is deliberately peaked on the ventral surface of the neophallus to simulate the natural shape of the glans-shaft junction. (D) Two months postop prior to tattooing.

Myocutaneous Latissimus Dorsi Flap

Perovic et al have reported extensively on the use of the myocutaneous latissimus dorsi flap for penile reconstruction.^{15,16} The advantages of this flap include minimal donor site morbidity with a scar that can be concealed, a

relatively hairless donor site, and good aesthetic outcomes. However, given that the flap is taken from a less sensitive area of the body, tactile sensation may be limited and erogenous sensation can be difficult to achieve. The latissimus dorsi myocutaneous flap is based

on the thoracodorsal vessels and nerve; it is anastomosed to the femoral artery and saphenous vein or the deep inferior epigastric artery and vein. The nerve coaptation is performed to the dorsal nerve of the penis or the ilioinguinal nerve. Perovic et al describe a 17×15 cm rectangular skin paddle used for the shaft, an intervening 1-cm deepithelialized strip with a distal circular component for glans reconstruction centered over the pedicle (Fig. 10).¹⁶ Only a thin strip of muscle around the pedicle is harvested, allowing the flap to be easily tabularized. In most cases, the donor site can be closed primarily with the incision closely approximating the mid axillary line, but occasionally a split thickness skin graft is required. This technique yields a phallus that is 13- to 16-cm long and 10 to 12 cm in circumference.^{15,16} Urethroplasty is done in two stages. A buccal mucosa graft is placed and the corporal remnants are incorporated into the neophallus at 3 months. At 6 months, the graft is tabularized and connected to the native urethra. Placement of a penile prosthesis is delayed until late adolescence, but the muscle provides a well-vascularized bed to minimize the risk of extrusion. The large volume of vascularized tissue associated with the latissimus myocutaneous flap allows successful urethroplasty, insertion of a penile prosthesis, and average to above average phallus size. Vesely et al describe the innervation of the latissimus for erectile function. When the patient

Figure 10 This illustrates the design for the myocutaneous latissimus flap described by Perovic. The flap is centered over the pedicle and measures 17×15 cm, with a rectangular skin paddle for the shaft, an intervening 1-cm deepithelialized strip, and a distal circular component for glans reconstruction. Only a thin strip of muscle around the pedicle is harvested, allowing the flap to be easily tabularized.

contracts the adductors and the gracilis, the latissimus contracts shortening, widening, and stiffening the neophallus. However, during walking and when not sexually active the neophallus is flaccid.¹⁷ The flap is composed of somatic tissue, which grows linearly with the child unlike androgen sensitive tissue that grows exponentially at puberty. When performing phalloplasty in children one should aim for a near adult size penis as growth of the somatic tissue will be linear.¹⁶

Fibula Osteocutaneous Flap

Sadove et al were the first to introduce the use of the fibula flap for phalloplasty in 1992.¹⁸ This flap can be harvested in a single stage or prelaminated to create a neo-urethra (Fig. 11).^{19,20} Sadove et al found that prelamination of the neo-urethra reduced the incidence of urethral fistulas²¹ and better controlled girth because the flap is wrapped around the urethra rather than tubed. For prelamination of the neo-urethra a full thickness skin graft wrapped around a catheter is inserted into the leg. At a second stage, the fibula osteocutaneous flap is harvested based on the peroneal vessels along the posterior interosseous septum, leaving 7 cm of fibula bone proximally and distally. The skin paddle can be oriented horizontally and then tubed, or vertically and then folded on itself. For the vertical skin pattern, the distal skin

Figure 11 Illustration of the prelamination of the neo-urethra in the fibula free flap using a full thickness skin graft tubed over a Foley catheter.

paddle contains the fibula and the prelaminated urethra and is then folded to form the ventral side of the penis. The proximal skin paddle is more sensate and forms the dorsal side of the penis (Fig. 12). The sensation of the flap is supplied by the lateral sural cutaneous nerve (LSCN), which courses posterior to the septum in 74% of cases with an anterior branch in 26% of cases.²² Woerdeman et al found that in 54% of cases the LSCN was within 3 cm of the septum, but in 86% of cases within 4 cm of the septum.²² Thus, the skin paddle should be placed posterior to the septum and in the proximal two thirds of the lower leg. Hage et al recommend preoperative sensory marking by injecting lidocaine over the biceps femoris tendon and marking the resulting numb area on the lateral aspect of the lower leg.²⁰ The bone is then fixed to the penile corpora cavernosa or the pubic symphysis. Hage et al suggest making the bone ~2 cm longer than the skin paddle so that the phallus is not floppy. The fibula provides rigidity and in many cases obviates the need for a penile prosthesis, which can be fraught with complications. However, the bone is subject to resorption, warping, and fracture over time and some find it difficult to conceal in pants. Dabernig et al have described a peroneal fasciocutaneous flap of the lower leg that does not include the fibula.²³ This technique addresses the criticism that the permanent rigidity of the osteocutaneous fibula flap can be difficult to conceal. The primary advantage of the fibula site is the ease of concealing the

Figure 12 This drawing illustrates the vertically oriented flap. The distal skin paddle contains the fibula and the prelaminated urethra and is then folded to form the ventral side of the penis. The proximal skin paddle is more sensate and forms the dorsal side of the penis.

donor site.²⁴ In early experience with the flap, the rate of urethral fistulas was very high. Prelamination of the urethra has reduced the incidence, but does add an additional step in the reconstructive process.

Urinary complications including strictures and fistulas are the most common and frustrating complications of phalloplasty and have been reported to be as high as 80% in some series.³ Monstrey et al report on the largest series of radial forearm phalloplasties ($n = 287$) in which the urethroplasty is completed at the time of the flap. Forty one percent of patients in their series had urethral complications, but only 18% required reoperation. Perovic describes a two-stage urethroplasty that is begun 3–6 months following the musculocutaneous latissimus dorsi flap and reports an overall urethral complication rate of 12.5% (2 patients) with only 1 of 16 patients requiring reoperation for urethral fistula.¹⁶ Many who use the free fibula as a donor site for phalloplasty feel that prelamination of the urethra as well as a two-stage urethroplasty decreases the incidence of stricture and fistulas.^{19,20} Schaff et al prelaminate both osteocutaneous radial and fibula flaps in an effort to reduce urethral complications. In their series, 32.6% of patients developed a stricture and 16% developed a fistula (4 of the 6 patients with fistulas required reoperation).²⁵

Sensation and Sexual Function

The dorsal penile nerve provides erogenous sensation and the ilioinguinal nerve provides tactile and protective sensation. Studies that have compared the sensitivity of different flaps, both erogenous and protective, have found the radial forearm free flap to be the most sensitive. Lumen et al found sensation of the radial forearm phalloplasty to be superior to the sensation of the pedicled ALT flap in transgender patients.¹⁴ Papadopoulos et al compared the free sensate osteofasciocutaneous forearm flap to the free sensate osteocutaneous fibula flap in phalloplasties for transgender individuals and found the sensitivity of the forearm flap to be subjectively better than fibula flap.²⁶ Much of the data on erogenous sensation is self-reported data from the transgender literature. Both DeCuypere et al and Selvaggi et al report 100% ability to achieve orgasm in their respective study populations of transgender patients who underwent radial forearm phalloplasty with coaptation of the medial and lateral antebrachial nerves to the ilioinguinal nerve and dorsal sensory nerve.^{27,28}

Penile Prosthesis

Rigidity required for penetration is a key component of the ideal phalloplasty, but is the portion of the reconstruction with the highest rate of complication. Complications include erosion, exposure, infection, mechanical failure, and penile fibrosis. Autologous tissues including

bone and cartilage have been used; however, these materials can resorb over time or fracture and leave the patient with a permanently rigid penis that can be embarrassing and difficult to conceal. Fasciocutaneous phalloplasties require the insertion of a penile prosthesis for penetration. It is generally felt that insertion of a penile prosthesis should be done at a second stage when the neophallus is completely healed and protective sensation has returned at around a year after the initial surgery.²⁹ Hoebeke et al report on their experience with hydraulic erectile implants in 129 female to male transsexual radial forearm phalloplasties. The total complication rate resulting in removal or significant revision was 41%, 11.9% for infection, 8.1% for extrusion, 9.2% for leak, 14.6% for malposition, and 13% for total dysfunction.³⁰ The explant rate in Monstrey et al's series of 130 penile prosthesis in radial forearm phalloplasties is comparable at 44.6%.³

Timing of Reconstruction

Many feel that phalloplasty should be undertaken prior to or just around the time of puberty (10–14 years) in an effort to minimize the psychological trauma associated with genital inadequacy.^{15,31} All phalloplasty techniques use somatic tissue that is not hormone responsive and will grow commensurate with the rest of the body. A native penis, however, grows exponentially during puberty in response to hormonal surges. Thus, when reconstructing a pediatric patient a near adult size phallus should be planned as growth of the somatic tissue will be linear.^{16,31}

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