

# The History of Hypospadias and Hypospadias Repairs

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Currently, hypospadias is defined as an abnormal ventral urethral meatus, an abnormal ventral penile curvature, and a deficiency of ventral prepuceal skin or a “dorsal hood.” This constellation of anomalies still presents a challenge for the skilled hypospadias surgeon. Questions regarding embryology, androgen deficiency, the urethral plate, and etiology of the penile curvature remain. Indeed, much of the ventral curvature can be attributed to a deficiency of ventral penile shaft skin rather than corporal disproportion. For the purposes of this review, we will use the term *chordee* for consistency. Clinical and basic science researchers continue to investigate many of these contributory factors to achieve a clear understanding of hypospadias and, through this knowledge, improve surgical outcome. To advance the field of “hypospadiology,” one must acknowledge the contributors whose work has helped to evolve older techniques into the modern hypospadias repair. Through 2 centuries, many surgeons have invented operations to repair hypospadias and many others have modified the operations. This review will include many of the repairs described and highlight critical changes in thought regarding the hypospadias repair. Many of the historical sources described in this article were initially compiled by the authors who contributed to *Plastic and Reconstructive Surgery of the Genital Area*, especially Blair O. Rogers.<sup>1</sup>

In the 1st century AD, the Roman Empire retained records from many of the early physicians and surgeons. Early European and North African surgeons likely shared knowledge among medical practitioners from ancient Rome, Greece, and Egypt. Hypospadias, as a congenital anomaly, was first documented in the medical literature in the 1st and 2nd centuries AD by Heliodorus, Antyllus, and Galen.<sup>2,3</sup> References to hypospadias and hypospadias repairs from this time period have been found in Greek, Roman, and Egyptian texts. Early writers documented the abnormal urethral meatus below the corona of the glans, and concern was exhibited regarding proper voiding and the ability to procreate. Galen (AD 130-199), physician to

the gladiators in Rome, specifically emphasized the impact of *chordee* by stating that “the curvature of the penis prevents its normal overflow from being conveyed forwards. The theory is confirmed by the ability to beget children if the frenum is divided.”<sup>4</sup> The repair described was a partial penectomy to the level of the urethral meatus with a conical incision to preserve a glanular shape. Hemostasis was achieved by compressive dressings, vinegar water, and cauterization if necessary.<sup>5</sup>

The fall of the Roman Empire heralded the arrival of the Middle Ages, in which the majority of medical texts originated from Islamic physicians. Albucasis of Cordoba (936-1013) described a similar surgical repair of hypospadias, stating a surgeon must “cut the head of the glans, at the eminence [or the presenting aperture of the meatus] with a knife or a bistoury, as if you were cutting a quill or as if you want to carve a piece of wood, in the manner of reestablishing a natural shape of the glans, and in which the meatus falls into a median position where it should be.”<sup>6</sup> Serefeddin Sabuncuoglu, one of the most influential surgeons of the early Ottoman Empire, was influenced by Albucasis and practiced surgery in what is now northern Turkey.<sup>7</sup> Sabuncuoglu wrote an extensive textbook that incorporated aspects of neurosurgery, thoracic surgery, orthopedics, and urological surgery. He describes a hypospadias repair similar to that of Albucasis but carefully included the necessity of placement of a thin catheter into the urethra to maintain patency. In addition, he cautions his students regarding hemostasis by stating “as there would be much bleeding during this surgery, it needs to be done very carefully and the bleeding has to be controlled by a cautery.”<sup>8</sup> Sabuncuoglu’s surgical texts included many paintings and calligraphy detailing surgical instruments and techniques. Urological depictions included meatal dilation and circumcision techniques (Figures 1 and 2).<sup>9</sup>

As the middle ages ended, the Renaissance period truly represented a rebirth in medical thought and technique. The first advances from the partial penectomy technique described previously were documented in the early part of the Renaissance period. In Portugal, Amatus *lusitanus* described a canalization technique to repair a penoscrotal hypospadias in a 2-year-old boy. Using a silver cannula, he created a channel distally until reaching the glans.<sup>10</sup> A. Pare (1510-1590), surgeon to the French royalty,

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Figure 1. Treatment of meatal stenosis.



Figure 2. Circumcision technique.

described the accompanying “chordee” as “the bridle or ligament of the yarde [penis] too short, so that the yarde cannot stand straight, but crooked, and as it were turned downwards; in these also the generation of children is hindred, because the seed cannot be cast directly and plentifully into the wombe. Therefore this ligament must be cut with much dexterity.”<sup>11</sup> In Malta, 2 physicians testified in an annulment hearing. Mathia, the wife, alleged her husband’s hypospadias and chordee hindered the ability to perform the conjugal and procreative act. According to court proceedings, the annulment was granted after both physicians examined her husband in court.<sup>12</sup>

The beginning of the 18th century saw an expansion of surgical texts and training. In 1707, P. Dionis (1643-1718), a French surgeon and anatomist, wrote an exhaustive text titled *Cours d’operations de Chirurgie demonstrees au Jardin royal à Paris* (A course of Surgical Operation Demonstrated in the Royal Garden in Paris). This text was the first comprehensive surgical text since the Renaissance.<sup>13</sup> This book contained a detailed description of the hypospadias repair. This text included details regarding the placement of a tunneled leaden pipe to bridge the distance between the hypospadiac meatus and the newly created glanular meatus. In much greater detail, Dionis also relates how to close the hypospadiac meatus and how to secure the leaden pipe to the penis. He also describes the repair of the associated chordee: “by a light scissure of the incision knife or scissars X, we cut the bridle across, in the same manner that we do the string

under the tongue, and so by a very light operation remedy two inconveniences which it causes.”<sup>14</sup> The 2 inconveniences to which Dionis alludes are the inability to void while standing and the inability to direct semen into the vaginal canal properly.

The 19th century was characterized by a diversification of surgical technique. Some of these new surgical explorations were especially inhumane and failed from a clinical standpoint. Other novel operations truly advanced the field of surgery. Dupuytren described a modification of the canalization canal. The canal thus formed was then cauterized with a red-hot wire, and after the severe inflammation had passed, he kept the new canal open with elastic catheters.<sup>15,16</sup> In stark contrast, Liston, a London surgeon, reported in 1838 the successful hypospadias repair that used a preputial flap.<sup>17</sup> Mettauer, an American surgeon, described a similar repair in 1842. Mettauer continued to study urethral anomalies, such as hypospadias and epispadias and detailed his findings with extensive texts on the subject. He described a technique for proximal hypospadias repair that included denuding the hypospadiac meatus and reapproximating the denuded edges. In addition, Mettauer recognized the skin tethering responsible for much of the ventral chordee. He recommended a “succession of subcutaneous incisions until the organ is liberated.”<sup>18</sup>

The next significant advancement in hypospadias surgery occurred in 1874 when Theophile Anger, a French surgeon, reported his novel operation at a surgical meeting in Paris. Anger used a method for epispadias repair that Karl Thiersch had previously described in 1869.<sup>19</sup> Anger created 2 ventral penile skin flaps, with the first flap creating a tubularized neourethra and the second flap used for ventral skin coverage.<sup>20</sup> In 1874, other surgeons began refining these techniques, including Duplay, Nove-Josserand, and Ombredanne. Duplay clearly delineated 3 separate steps in the hypospadias repair: correction of the ventral chordee with a horizontal ventral penile incision that was closed vertically, use of penile skin flaps to create a neourethra, and connection of the neourethra to the proximal meatus.<sup>21</sup> Nove-Josserand used a free graft from the thigh that was placed via trocar and canalization to create a neourethra.<sup>22</sup> Ombredanne was the first surgeon to create a penile pedicle flap to create a neourethra creating a rudimentary flip-flap.<sup>23</sup> Rosenberger used a two-stage repair with scrotal flaps that involved suturing the penis to the scrotum in the first stage.<sup>24</sup> Weller Van Hook, a prominent Chicago physician and surgeon, first described the use of pedicle flap of preputial skin as a tubularized neourethra. He was careful to explain the operation to the mother of a boy with severe penoscrotal hypospadias. Van Hook notes in his description: “It was explained to the mother that the operation must proceed by a series of steps, the number of which could not then be told; and it was definitely agreed that, unless the child’s health failed, the operations should be continued until the penis was brought to a satisfactory state.”<sup>25</sup>

These surgeons and surgical techniques represent nearly 2 centuries of innovation and operative repairs for hypospadias, most often without the aid of anesthesia. These early techniques constructed the foundation of the hypospadias repairs used today.

The repairs of the early to mid-20th century were usually performed in 2 stages. Edmunds advocated a two-stage repair with the chordee release and transfer of preputial skin to the ventrum that would later be tubularized.<sup>26</sup> By separating the chordee repair and the urethroplasty, the operation was simplified and could be performed reproducibly by many surgeons. Unfortunately these early repairs often resulted in a hair-bearing urethra and a ventrally displaced meatus.<sup>27</sup>

Beginning in the late 1950s and 1960s, there was renewed interest in one-stage hypospadias repairs. Before this period, one stage operations were performed using split-thickness free grafts from the thigh or arm that were fraught with complications, including marked contracture. Devine and colleagues published data using free full-thickness skin graft tubes. Devine's group excised the urethral plate and created the graft from preputial skin. The proximal anastomosis was oblique and the glans was incised to create the distal anastomosis. Tunneling through the glans was abandoned because of the high rate of stricture. The ventral defect was covered using a penile or scrotal skin flap. In addition, urine was diverted by placement of a perineal urethrostomy catheter. The use of preputial skin graft was a significant advantage to the previously used graft sites in that the skin is thin, pliable, and hairless.<sup>28</sup> In 1970, Hodgson advanced the technique of using preputial skin by maintaining a pedicle blood supply. He describes transillumination of the dorsal vessels and transfer of the preputial flap from dorsum to ventrum using a buttonhole technique.<sup>29</sup> Asopa and colleagues described a transverse preputial flap urethroplasty also based on the superficial dorsal blood supply of the penis that also maintained the continuity of the urethral plate.<sup>30</sup> Techniques for urethroplasty have been augmented by advances in other aspects of the repair. Skin coverage was advanced by Byars' technique for 2 dorsal preputial flaps. The flaps are rotated to the ventral shaft and are approximated in the midline compensating for the ventral skin deficiency.<sup>31</sup> Many methods of coverage for the neourethra have been developed to decrease the risk of urethrocutaneous fistula formation. The use of tunica vaginalis, dartos flaps, de-epithelialized skin flaps, and local subcutaneous flaps have all been described. In addition, Yerkes and colleagues described the Y to I spongioplasty that uses the distal spongiosum to wrap around the neourethra (Figure 3).<sup>32</sup> As the modern hypospadias repair evolved, surgeons modified the chordee repair to address potential corporal disproportion. In 1965, Nesbit described the technique of removing elliptical portions of the dorsal tunica albuginea to correct the ventral curvature.<sup>33</sup> As an alternative, transverse incisions in the ventral tunica albuginea can be closed ver-

**Figure 3.** Y to I urethroplasty. Flip-flap is in place to create a neourethra. The lateral distal spongiosum is then mobilized medially to cover the neourethra.

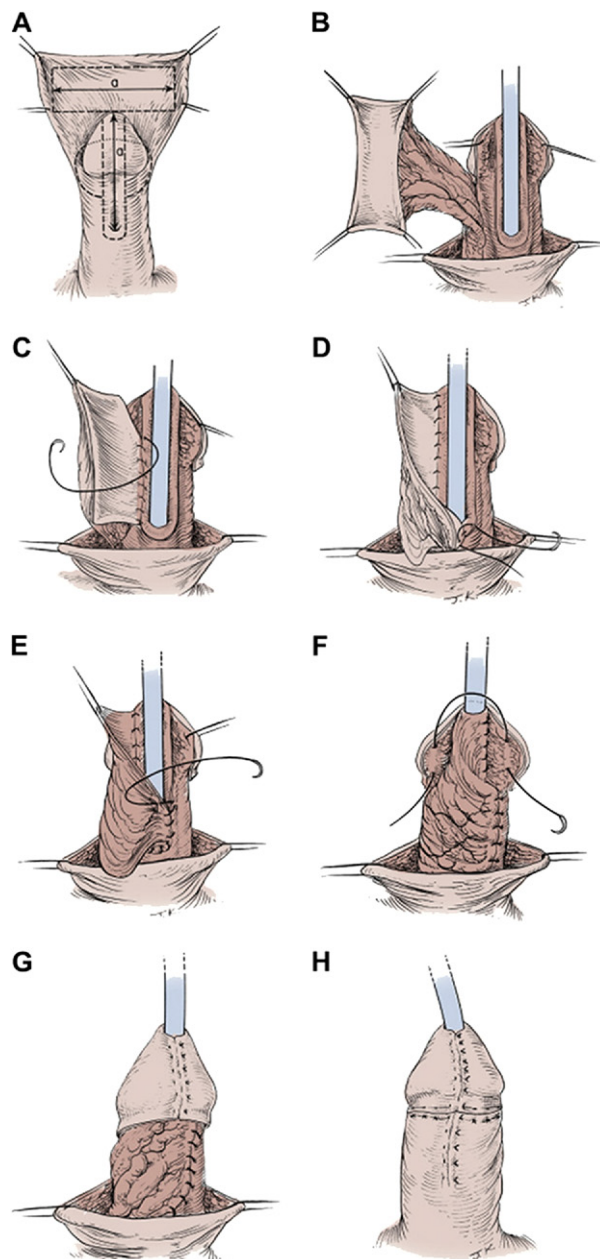
tically to lengthen the ventrum of the penis.<sup>34</sup> In 1994, Duckett and Baskin proposed plication of the dorsal penile shaft by creating 2 transverse incisions and closing the outer edges together, thus shortening the dorsal penile shaft without removing tunical tissue.<sup>35</sup> This dorsal repair was modified by Yachia, who described the creation of longitudinal incisions on the dorsal tunica albuginea that are closed horizontally to shorten the dorsal penile shaft.<sup>36</sup> The use of vertical incisions decreases the potential risk for damage to the neurovascular bundles that traverse the dorsum of the penis. Baskin and colleagues subsequently documented a paucity of neurovascular tissue in the dorsal midline of the penile shaft.<sup>37</sup> As a result, most dorsal plications are now performed at the 12 o'clock position on the dorsum of the penis. The concern regarding penile shortening in patients with a small phallus and severe curvature has led many surgeons to use free grafts to lengthen the ventrum of the penis. Dermal free grafts from nonhair-bearing skin have been used since the 1970s by many surgeons and more recently the use of other materials, such as tunica vaginalis has also been described.<sup>38,39</sup> These hypospadias repair techniques allow for improved appearance and decreased complications.

As results improved, given the increased prevalence of distal and midshaft hypospadias compared with the more proximal defects, interest was also growing in new repairs for mild hypospadias. In the 1980s and 1990s, new less invasive techniques were described. These newer repairs attempted to decrease the operative time, need for catheterization, and required hospitalization. In addition, the limited nature of these repairs minimized the risk of major complications. Duckett described the meatal advancement and glanuloplasty for subcoronal hypospadias. The tenet of this procedure is a modification of the Heineke-Mikulicz principle. In brief, a longitudinal incision is created from the dorsal edge of the meatus to the distal glans groove. This vertical incision is then closed horizontally followed by the ventral glansplasty. This procedure minimized the risk of urethral fistula and was performed as an outpatient procedure without a urethral catheter.<sup>40</sup> This drive for minimally invasive techniques was accompanied by a desire for improved cosmesis. Al-

though many of the previously described repairs were technically successful, the neomeatus fell short of the desired vertical slit-like meatus. This impetus led to further modifications of the hypospadias repair. Zaontz described the glans approximation procedure (GAP) that capitalized on the glanular groove. This groove is incised and then the urethral plate is tubularized.<sup>41</sup> These techniques added to the armamentarium available to the hypospadias surgeon.

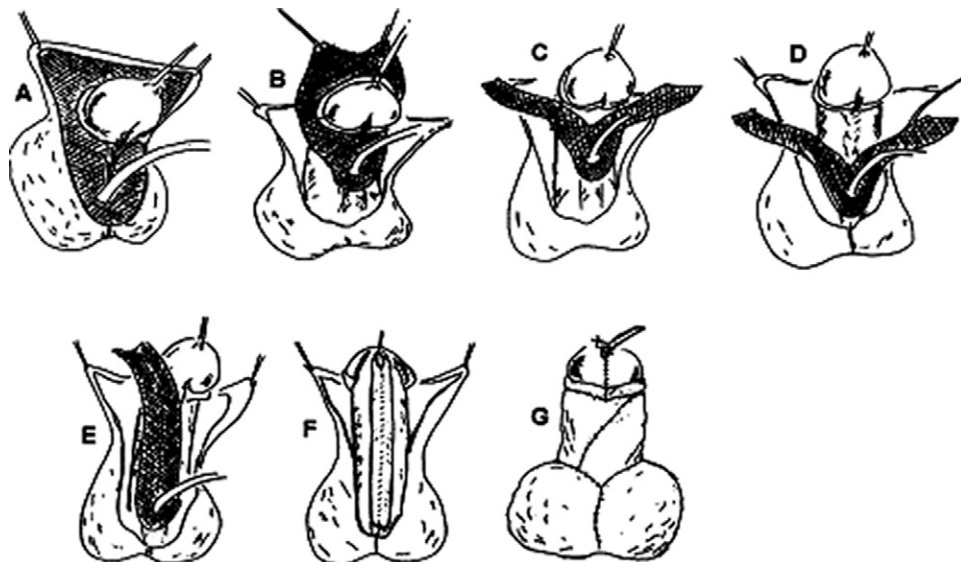
Other surgeons sought to extend the use of primary repairs to include proximal hypospadias. In 1980, Duckett advanced the use of preputial tissue in the description of the transverse preputial island flap technique.<sup>42</sup> In this technique, a transverse flap of dorsal preputial skin is used as a flap or tube to create the neourethra. The transverse preputial island flap was initially described in conjunction with creation of a glans channel, but as glansplasty techniques advanced, the repair used the improved glansplasty technique (Figure 4).<sup>43</sup> Koyanagi described a one-stage repair for proximal hypospadias using lateral penile shaft flaps that are then tubularized ventrally to create a neourethra (Figure 5).<sup>44</sup> As with other repairs, the initial description was advanced by other investigators; Koff and colleagues modified this technique to improve flap vascularity.<sup>45</sup> The one-stage hypospadias repair was redefined by Snodgrass with the description of the tubularized, incised plate urethroplasty (TIP). This technique was initially described for distal hypospadias without significant chordee and diverged from the preputial flaps most commonly used. This technique draws upon similar principles to the GAP procedure previously described and a technique for hinging the urethral plate. In 1989, Rich and colleagues described a midline longitudinal incision in the urethral plate that allowed for hinging of the plate during glansplasty.<sup>46</sup> This technique resulted in the creation of a vertical slit-like meatus. The advantages of the TIP repair include creation of a vertical meatus and the avoidance of vascular pedicle flaps (Figure 6).<sup>47,48</sup> As this technique developed, the indications for use and the modifications also developed. As experience with the TIP repair increased, other investigators expanded its use to include more proximal defects and reoperative repairs.<sup>49,50</sup>

In addition, one of the most complex challenges facing the hypospadias surgeon is the hypospadias “cripple.” These patients have typically undergone multiple failed hypospadias repairs and have little healthy tissue available. Many of the previously described free graft techniques have been attempted for reconstruction after failed repairs. Initially, full-thickness skin grafts with nonhair-bearing skin were used but significant complications arose, including stricture, scarring, and graft shrinkage.<sup>51</sup> Subsequently, the use of bladder mucosa was introduced. Although the stricture and scarring decreased, the need for a retroperitoneal dissection and the high risk of mucosal prolapse of the neomeatus led to the abandonment of this repair.<sup>52</sup> The initial concept for the use

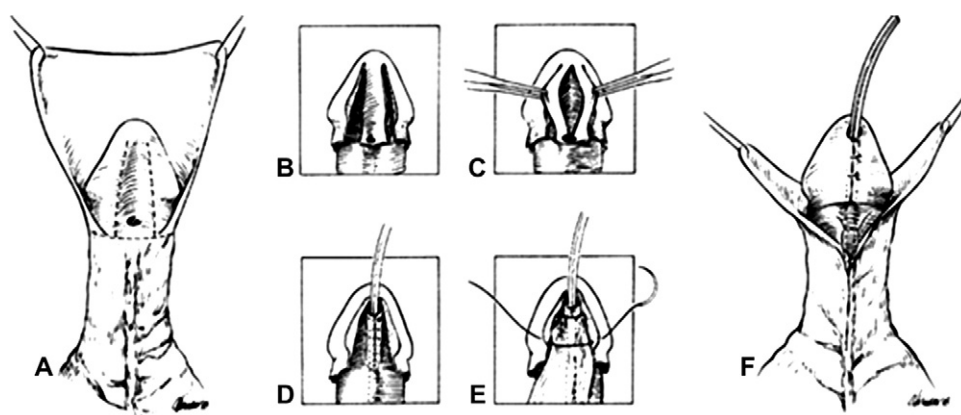


**Figure 4.** Island onlay flap technique. (A) Planned incisions for transverse preputial flap and urethral plate. (B) Isolation of preputial flap on pedicle. (C) Initiation of anastomosis of onlay flap and urethral plate. (D) Proximal approximation. (E) Completion of neourethral channel. (F) Advancement of a second layer of urethral coverage. (G) Glansplasty. (H) Skin coverage and completed repair.

of buccal mucosa for hypospadias repair was suggested in 1941, but the repair did not gain widespread use until the 1980s to 1990s.<sup>53</sup> With long-term follow-up, buccal mucosa has become the most successful of the free graft tissues. Buccal grafts have proven successful in both young patients and older patients with recurrent urethral strictures. Initial reports of buccal graft described a single-stage repair with a tubularized or onlay technique. Meatal stenosis appeared to be the most common complication observed, although scarring was also noted.<sup>54</sup> In the mid-



**Figure 5.** Koyanagi hypospadias repair. **(A)** A circumferential incision is made around the subcoronal margin. **(B)** A second incision begins distal to the meatus and extends laterally on either side of the glans to the dorsum approximately 8 mm below the first incision. **(C, B)** The new flap is split dorsally into 2 strips that are next brought ventrally. **(D)** The urethral plate is excised. **(E)** The 2 skin flaps are anastomosed in the midline. **(F)** The neourethra is tubularized. **(G)** Glansplasty and skin closure re-performed.



**Figure 6.** Tubularized, incised plate urethroplasty. **(A)** Planned circumscribing incision and identification of urethral plate. **(B)** Separation of the urethral plate and glans wings. **(C)** Deep, midline incision into the urethral plate from the meatus to the distal margin. **(D)** Tubularization of the urethral plate. **(E)** Dartos coverage and glansplasty. **(F)** Skin closure and completed repair.

1990s, Bracka presented a personal series of 600 cases and advocated for a two-stage repair for using full-thickness skin grafts.<sup>55</sup> Now, buccal mucosal grafts are performed with a two-stage repair to increase graft take and decrease complications, such as fistula and meatal stenosis.<sup>56</sup>

A greater understanding of the embryology and anatomy of hypospadias provides the modern hypospadias surgeon with a guide for reconstruction of normal anatomy while preserving penile function. By examining human fetuses with and without hypospadias, Baskin and colleagues were able to demonstrate the location of the neurovascular bundles on the dorsum of the penile shaft. These nerves begin as 2 bundles that spread laterally along the corpora but are absent at the dorsal midline, providing a guide for dorsal plication.<sup>57</sup> These authors

also determined that the major difference in anatomy between a normal penis and a hypospadiac penis is an abnormal spongiosum and glans. In addition, the hypospadiac glans has less extensive innervation that may predispose the surgeon to less aggressive glanular dissection.<sup>58</sup> The blood supply of the prepuce has also been delineated with 70% of patients having 1 or 2 predominant vessels that allow for well vascularized preputial flaps. Conversely, 30% of patients had a capillary network present without dominant vessels, thus creating a challenge for the development of preputial flaps.<sup>59</sup> As hypospadias surgery continues to evolve, multiple operative techniques are common for the treatment of hypospadias, from distal to perineal. These techniques progress in large part because of the advances in knowledge re-

garding penile anatomy. Indeed, the technique used must be tailored to the child based upon glanular size, degree of chordee, and location of the meatus. The surgeon must be familiar with the chosen technique as well. Some authors are proponents of one-stage repairs but propose the use of a two-stage repair in extreme cases of perineal hypospadias associated with severe chordee or very small glans size.<sup>60</sup>

The future of hypospadias surgery depends on surgical outcomes and patient satisfaction. Clearly, many hypospadias repairs exist and are in use by thousands of practitioners. Yet, long-term voiding function, sexual performance, and overall patient satisfaction are under-reported. Typically, surgeons perform hypospadias repairs in the first year of life, and follow-up is routine for 6 to 12 months. Given the mobility inherent in today's society, follow-up is difficult even for practitioners who attempt to contact their patients. Even so, a few investigators have detailed long-term outcomes in patients with hypospadias. Reported complication rates vary by severity, type of repair, and institution.<sup>61,62</sup> Surgeon and patient assessments of outcome are often discordant.<sup>63</sup> Patient satisfaction also varies between studies.<sup>64,65</sup> Contacting adult patients who underwent hypospadias repairs in childhood provides essential surgical follow-up. Combining medical data from chart reviews and a patient questionnaire, Hoag and colleagues found that 11% of patients have an ongoing fistula and 10% had a subsequent urethral stricture.<sup>66</sup> These same investigators found that persistent chordee was the most common postsurgical complaint among their patients. A similar study documented that 20% of patients reported persistent chordee but only 30% of those patients found the residual chordee to be a major handicap.<sup>67</sup> Without these data, valid evaluation of various hypospadias repair techniques is limited at best. Although these retrospective studies begin to define the questions for future analysis, initiation of long-term, prospective data collection will present the surgeon with necessary information to advance operative techniques.

Pioneering surgeons have revolutionized techniques for hypospadias repair since the first surgical interventions were undertaken in the 1st century AD. The transition from an extirpative partial penectomy to a reconstructive urethroplasty represented a radical advancement in the treatment of hypospadias. This reconstructive technique was dramatically improved with recognition that the urethral plate did not routinely require transection. In addition, the understanding that the incised urethral plate heals by secondary intention provided the innovation for the TIP repair. The advancement of hypospadias repairs will continue with further investigation of wound-healing properties, a greater understanding of biological susceptibilities, and critical evaluation of the long-term cosmetic and functional results of the current hypospadias techniques.

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