History of hypospadias: Lost in translation

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The evolution of hypospadias surgery can be classified under 3 periods that were closely related to advances in surgical instruments, introduction of anesthesia, and newer suture materials. Stretching dominated the early period, tunneling during the Middle Ages, and flaps during the modern period. Suture materials have included at various time silver wires, horsehair, and stainless steel.

Examination and translation of the original manuscripts showed that Galen recommended stretching and suturing of glanular hypospadias and not amputation or partial penectomy as has been currently reported. The term chordee was first introduced in the 18th century in relation to gonorrhea and was defined as “painful imperfect erection of the penis during gonorrhea with downward incurvation.” This was a common complication of gonorrhea before the introduction of antibiotics. Mettauer, Duplay, Mayo, and others used the terms incurvation, ventral deformity, ventral curvature, and others. Smith in 1938 was the first surgeon to use the term congenital chordee in direct relation to hypospadias.

The use of prepuce for urethroplasty, popular now, was first reported by Liston in 1838, Rochet in 1899, Russell in 1900, and Mayo in 1901.

The two stage repair performed in the early 20th century differed from that in the early 21st century in that urethroplasty was performed in the first stage and only anastomosis to the original meatus was performed in the second stage. The two-stage repair, currently known as Bracka’s two-stage repair, was first described in 1962 by Clouter.

The use of the urethral plate in epispadias was first described by Liston in 1838, Thiersch in 1869 and by Anger & Duplay in hypospadias in 1874. Partially epithelialized urethroplasty using the urethral plate was described by Duplay in 1880, Russell 1915, Denis Browne 1940, Reddy 1975, Orkiszewski 1987, Rich 1989, and Snodgrass in 1994.

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Hypospadias is the most common congenital malformation affecting the penis with incidence approaching 1% of newborn boys [1]. The London surgeon, Twistington Higgins in 1941 wrote, “Hypospadias is a grievous deformity, which must ever move us to the highest surgical endeavor. The refashioning of the urethra offers a problem as formidable as any in the wide field of our art. The fruits of success are beyond rubies. The gratitude of a boy has to be experienced to be believed” [2]. To advance, one must acknowledge the work of the pioneers whose works have paved the way to the current state of the art.

Published work on the history of hypospadias [3–6] has suffered somewhat from the reiteration of quotes from earlier papers rather than the actual examination of original sources. This review contains my impressions based on professional translation of the original sources and direct observation rather than simple reiteration.

1. Galen and amputation or partial penectomy

Galen of Pergamon (129–199 AD) was one of the most influential physicians of his era and for many centuries after because of his position as personal physician to several Roman emperors and his many treatises. He spent a year as a scholar in Alexandria, Egypt and is credited as the first to use the term hypospadias to describe the condition where the urethral opening is on the undersurface of the penis [3–6].

Most publications on the history of hypospadias quote Galen as recommending “amputation or partial penectomy to the level of the urethral meatus with a conical incision to preserve a glanular shape” [3–6].

I began to wonder about the accuracy of translation and transfer of knowledge over the centuries as glanular or even coronal hypospadias does not interfere with urination or procreation. In addition, as the glans is the most sensitive part of the penis, there seems to be little logic in making it worse than it is without surgery. I was able to access and translate the original ancient Greek manuscript of Galen (Omnia Opera, volume 10, page 1001–1003) (Fig. 1). In actuality, he did not recommend partial amputation or amputation but refreshing of skin edges and skin stretching to the tip of the penis. This technique he used successfully to treat patients with amputation of the nose and ear "Some
other times removing excess skin with the scalpel from the base of the glans and pushing them downward and then fix them as already said with a gummy substance. This treatment resembles the one we use for amputations and deficiencies of the nose and lips and ears. So we can treat also these diseases by removing partly the skin from both sides and then we re-approximate the skin after removing the surplus of both parts and then we suture them together” [7].

2. Chordee (penile curvature)

The significance of penile curvature associated with hypospadias was appreciated by Galen and Oribasus who noted that penile curvature interferes with intercourse. The French anatomist and surgeon, Ambroise Paré (1510–1590) [8], described the accompanying chordee as to prevent “the seed to be cast directly and plentifully into the womb.” He induced an artificial erection of the penis 400 years before the description by Gittes [9]. Jean Fernel famouly corrected the chordee of Henry II, King of France in 1547, who had been married for 10 years without children, thereafter he fathered 10 children [5].

The term chordee was first introduced into the medical literature in 1708 from the French and was defined as “painful inflammatory downward curving of the penis” [10]. This affection of the penis was most common in the second and third weeks of gonorrhea and occurred chiefly at night [11] (Fig. 2).

Forster in 1860 wrote, “I saw a child 8 months old in 1857. The penis was (without erection) in a state resembling chordee, bent like a siphon. The only explanation that could be obtained from the mother was that whilst pregnant, her husband had gonorrhea, and suffered much from chordee” [12].

A number of other terms have also been in common use to describe the curvature element of hypospadias. Mettauer [13] (1842) and Mayo [14] (1901) used the terms incurvation, ventral deformity or ventral curvature. Nesbit in his first article on hypospadias in 1941 used the term ventral curvature [15].

The first surgeon that I could identify who used the term chordee in direct relation to hypospadias was Clinton Smith (1938) [16]. He wrote: “Recalling the embryological aspects momentarily, all hypospadias are associated with some degree of congential chordee.” This varies from slight downward bending of the glans to complete anchorage of the glans in the perineum in exaggerated instances of the third degree type.” He used the term congenital chorddee to differentiate it from the then – classic chorddee associated with gonorrhea.

3. The urethral plate

Glenister (1954) was the first to introduce the embryological term urethral plate and he defined it as “an outgrowth from the anterior walls of the urogenital sinus” [17]. Paul & Kanagauntheram (1956) defined the urethral plate as “the moist pink gutter of mucus membrane with (a) well-defined muco-cutaneous line and extends from the urethral orifice to the base of the glans” [18]. John Duckett used the term urethral plate to define the skin and spongiosal tissue which lies distal to the urethral meatus in a hypospadias and goes out onto the ventral glans [19].

To date there is no clear definition of the width, depth, type of tissue and extension of the urethral plate in clinical practice. Surgeons who rely totally on the urethral plate to repair hypospadias still use vague definitions such as “the urethral plate is the strip of tissue that extends distal from the hypospadiac meatus to the tip of the glans” [20].

4. Evolution of hypospadias repair

The evolution of the surgical management of hypospadias can be grouped into three distinct periods, the first starts in first 3 centuries AD, the second during the Middle Ages or Medieval period and the third period runs from the 19th century until today. These periods seem to be closely linked to advances in instrument manufacture, anesthesia and suture material.

4.1. The early period

This could alternatively be called the “Period of Stretching and? Amputation?” As instruments were very primitive and there was no anesthesia nor the appropriate suture materials, it is understandable that penile skin stretching over the urethra to the tip of the penis with a pin or gummy materials extracted from plants was the preferred surgical approach.

There is no preserved documentation of the work of the Alexandria surgeons. However, Oribasius, doctor of the Roman emperor Julian the Apostate (4th century) left a medical encyclopedia of 70 volumes (Iatrikai Synagogi, Collecta Medica). Only a third of his writings are preserved and in book 44 of Collecta Medica, he wrote [21,22], “drawn from Heliodorus and Antyllus from Alexandria, Egypt (1st century AD) who were the first to describe urethral opening malformation “Among certain individuals, the glans, because of an inborn defect, is not pierced according to nature, but the hole is found below the brake (frenum as is called in Greek), at the termination of the glans. For this reason, the patient can neither urinate in front, unless the penis is raised sharply towards the pubes, nor procreate children because the sperm cannot be directed in a straight line into the uterus, but forms on the side of the vagina... Sometimes the hole is situated far from the frenum; in the middle of the urethra, these cases are incurable. The simplest, best, and least dangerous operative procedure is what is called operation by resection, the patient is placed on his back and the glans is fully raised with the left hand. This part is then cut with the edge of the scalpel at the level of the crown. However, the incision
should not be slightly oblique but should resemble a delicate carving, leaving a projection representing the form of a glans. If little blood flows, it should be stopped with a bandage and vinegar; if this does not suffice medications or cautery should be used. The resection must be done in the glans rather than in the penis, for because of its compact structure, the glans is less likely to haemorrhage.

As mentioned before, Galen did not recommend partial penectomy or amputation but refreshing of skin edges and skin stretching to the tip of the penis. Oribasius, Celsius, Fallopio and Paul of Aegina (625–690 AD) followed the same approach of Heliodorus, Antyllus and Galen for hypospadias.

4.2. The middle period

The fall of the Roman Empire heralded the arrival of the middle ages, in which the majority of medical texts originated from Islamic physicians. Abu Al Qasim Al Zahrawi (known as Albucasis) of Cordoba, Andalusia, Spain (930–1013) (Fig. 3) wrote a medical encyclopedia called Al Tasreef (influenced by the work of Hippocrates, Galen and Paul of Aegina). In 1150 AD, Gerard of Crimona translated “Al-Tasreef” into Latin and it remained the most important reference book on surgery until the end of eighteenth century [24]. Albucasis described (with illustrations) the tonsil guillotine, the true scissors, the syringe, the vaginal speculum, the lithotrite, the Chamberlin’s obstetric forceps and used animal gut as a suture material. He even suggested the use of animal teeth to replace lost teeth. His surgical instruments are still preserved in the Calxa Hurra Museum in Cordoba (Fig. 3). He was a strong advocate of cautery to treat diseases.

The author accessed a copy of the original book “AL-Tasreef” in its original Arabic language and translated what he actually wrote about hypospadias in chapter fifty five [24].

“Some boys are born and the glans is without a hole or with a narrow meatus or the meatus may be displaced. When the glans is without a hole, you should penetrate the glans with a trocar made of lead and tie it for 3–4 days. It should be taken off when the boy needs to urinate and then put it back. It is possible that the passage of urine may help keep the tract open. Those who have a narrow meatus should be dilated with lead trocar as we mentioned for many days.

Some of those boys with a displaced meatus cannot pass urine in a forward stream and cannot procreate, as the semen cannot reach the uterus. For those children, the child should lie on his back, hold the penis with your left hand and incise the head of the glans with a knife to have the opening in the middle and shave the glans as if you were cutting a quill or as if you want to carve a piece of wood in manner of reestablishing as natural shape of the glans and in which the meatus falls into the median position where it should be. You should be careful and control bleeding using cautery if needed.”

In other words, Albucasis described two techniques for hypospadias repair: a) Tunneling for those with imperforate glans and b) Carving for those with displaced meatus. Albucasis is credited to be the first to describe tunneling for the correction of hypospadias. Serefeddin Sabuncuglu of Anatolia (Turkey) was influenced by Albucasis and left a tin catheter into the urethra to maintain patency.

In strong contrast, Professor Robert Liston (1794–1847), a Scottish surgeon who worked as a professor in University College Hospital, London, reported in 1837 the successful use of prepuce to line the tunnel and avoid its closure. The authors could access his original book “Practical Surgery” in 1837 (Fig. 4) where he wrote [26]:

“This hypospadias, as it is denominated, can occasionally be remedied. Attempts have been made by perforating the glans in the course of the natural canal, and by the insertion of a tube in the perforation, to carry forward the current and elongate the urethra permanently; this method does not answer; there is a want of substance in the part, and sloughing is generally the consequence of the attempt. I have sometimes succeed, and in cases where other means have been tried unsuccessfully, in protecting the exposed and irritable lining membrane of the passage, and continuing the canal open to the apex of the organ, by turning back a portion of the prepuce and uniting it without any twist, the lining membrane presenting outwards; patients have been thus relieved from the frequent calls to make water, the nocturnal emissions, and other unpleasant consequences.”

He even described in his book the correction of epispadias in a 23-year-old man similar to that of Thiersch in 1869 [27].
4.3. The modern period

This was sparked by the introduction of ether anesthesia in 1840s [28] which enabled surgeons to perform sophisticated urethral surgery. Although published by Liston 1838, the modern era in hypospadias surgery started in 1869 when Karl Thiersch [27] from Leipzig, Germany, described his technique of two longitudinal skin incisions for the correction of epispadias in 1869. His student, Theophile Anger [29] from Paris applied his principle to repair penoscrotal hypospadias successfully and reported it in a surgical meeting.

5. Grading of hypospadias

Currently, primary hypospadias may be classified into 4 grades:

5.1. Grade I or glanular hypospadias

Carl Beck & Hacker are probably the pioneers for the modern correction of glanular hypospadias. Carl Beck (1856–1911) studied medicine in Heidelberg and Berlin before he went to New York in 1882 and established the New York post-graduate medical school. He published the principle of urethral mobilization and meatal advancement in 1898 in the New York medical journal [30]. Hacker published the same principle, the same year in Germany [31]. This principle has been modified by many others [32] including: Horton & Devine (triangularized glans) in 1961, Mustarde in 1965 (V-flap), Duckett in 1981 as the meatal advancement and glanuloplasty incorporated (MAGPI), Koff in 1981, Arap (M configuration), Harrison and Grobbelaar (urethral advancement and glanuloplasty procedure or UGPI) (1997) and lately Hadidi in 2010 (double Y glanuloplasty technique).

5.2. Grade II or distal hypospadias

There are two main approaches to the operative repair. Anger [29] and Duplay [33] described the tubularization of the urethral plate and ventral penile skin to reconstruct fully epithelialized urethra in 1874. Duplay [34], 6 years later in 1880, reported his second operation where he wrapped the urethral plate around a catheter but did not suture the edges together and covered it with skin. This principle of forming an incomplete new urethra was further popularized in the 1940s by Denis Browne [35]. Incision of the urethral plate was first described by Reddy [36] in 1975, Orkiszewski [37] in 1987, Rich [38] in 1989 and Snodgrass [39] in 1994. To reduce urethral strictures and functional urethral obstruction associated with incision of the urethral plate [40], Malone [41], Gundeti [42] and Ferro [43] described covering of the raw surface resulting from incision of the urethral plate with grafts.

The use of meatal-based flap is the other popular approach for distal hypospadias and withstood the test of time for more than 80 years. This was pioneered by Mathieu [44] in 1932 although the principle was described earlier by Wood in 1875, Omberedanne in 1911, and Bevan in 1917 [3]. Other modifications of the same principle to produce a slit-
5.3. Grade III or proximal hypospadias without severe deep chordee

Tubularization of the urethral plate as mentioned above is common. Other popular techniques include the preputial island onlay flap described by Elder et al. [49] in 1987 and the lateral based onlay flap (LABO) by Hadidi [50] in 2012.

5.4. Grade IV or perineal hypospadias with severe deep chordee

There are two principles behind the operative repair for these:


b) Koyanagi [61] (1983) described the use of lateral penile skin and foreskin from both sides for urethral reconstruction. This was further modified by Snow [62] (1994), Emir [63] (2000), Hayashi [64] and Hadidi [65] (2014).

5.5. Grade V or cicatricial or crippled hypospadias

One of the most complex challenges facing the hypospadias surgeon is the cicatricial complicated hypospadias. These patients have typically undergone multiple failed hypospadias repairs and have little healthy tissue available. Several authors use the term hypospadias cripple [3,6]. However, parents find this term offensive and insulting and it is probably better to use the term cicatricial or Grade V hypospadias. Such patients require the use of grafts.

Nove-Josserand [66] in 1897 was the first to use a free graft from the thigh that was placed around a trocar and inserted through the glans to create a neourethra. Memmelaar [67] first described the use of bladder mucosa in 1947, which was popularized by Hendren and Reda [68] (1986) and Ransley [69] et al. (1987) but it became less popular because of mucosal prolapse through the meatus and a high incidence of strictures and stenosis.

Devine and Horton reported the use of free preputial graft in 1961 [70] as a single stage repair with a tubularized grafting technique. Meatal stenosis, stricture and scarring were the common complications observed [71], Cloutier [72] in 1962 recommended the use of preputial skin graft as a first stage and reconstruct the new urethra 6 months later as the second stage. The Cloutier technique became popular in 1990s after Bracka presented a personal series of 600 cases using preputial skin graft or buccal mucosa for correction of all forms of hypospadias [73].

Humby [74] first reported the use of buccal mucosa for hypospadias repair (1941). It became popular after the publications of Duckett [75] (1986) and Dessanti [76] et al. (1992). Now, preputial skin, non-hair bearing skin or buccal mucosal grafts are used as a two-stage repair to increase graft take and decrease complications, such as urethral stricture and meatal stenosis.

6. Stages of repair

The repairs of the late 19th century were 3 stages repair. Duplay [34] recommended 3 steps or stages of repair: 1) excision of chordee, 2) neo-urethra tube reconstruction, 3) Anastomosis of the neo-urethra to the proximal native urethra.

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 [77]. 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 [78].

In the late 1950s and 1960s, there was renewed interest in one-stage hypospadias repairs [54–56]. 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 et al. published data using free full-thickness skin graft tubes as one-stage repair. They 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 [78].

In the early 21st century, the neo-urethra in hypospadias Grades I, II and III is commonly reconstructed in a single stage. Perineal hypospadias (Grade IV) and cicatricial complicated hypospadias (Grade V) are preferably reconstructed in a 2-stage repair with excision of the deep chordae or cicatricial scar and laying of healthy epithelium or graft (preputial if available or buccal mucosa) in the first operation and urethroplasty is delayed 3–6 months later.

7. Suture materials

Advances in suture material played an important role in improving the results of hypospadias surgery and reducing complications. Suture materials used included animal intestine, silver wire, horse hair, stainless steel and synthetic suture materials (Fig. 5).

Albucasis used suture material made of animal intestine [24], Duplay preferred to use silver wire [34]. In 1917, Beck wrote “Intraurethral sutures, no matter how fine is bad. The catgut swells and loosens and favors leakage. The borders and stitch holes are infiltrated with urine and becomes easily infected, thus the suture is spoiled. Fine horse-hair is the ideal material and it need not be removed from inside of the urethra; it drops out when it is not needed, often months after operation” [79]. Mathieu [44] used silk sutures that were coming out through the urethra (Fig. 5).

Nesbit [15] in his article in 1941 wrote: “we prefer the Duplay urethroplasty using stainless steel subcuticular sutures closure as suggested to us by Lloyd Reynolds... these stainless steel sutures are accompanied by a minimum of reaction and infection and are easily removed on the fourteenth postoperative day.” Denis Browne [35] used glass beads and metal pears to secure his sutures. Most surgeons nowadays tend to use synthetic absorbable sutures like polyglaclin (vicryl, Ethicon) or polydioxanone (PDS©, Ethicon), polyglyconate (Maxon©, Davis & Geck) or poliglecaprone 25 (Monocryl©, Ethicon) as fine as 6/0 and 7/0.

In spite of more than 400 operations described for hypospadias repair and thousands of publications, there is still a lot to learn about the normal development of the urethra [80] and the pathogenesis of hypospadias. We need to use a standardized international classification of the different forms and degrees of hypospadias. We need to be more objective and rely on standard long-term assessment of hypospadias surgery.

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