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MAIN TOPIC

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The history of hypospadias

Abstract The dictum, "There is nothing new in surgery not previously described", is particularly true of hypospadias. The major significance of chordee was fully appreciated by Galen in the second century A.D. and then almost forgotten until Mettauer in 1842, all previous surgeons overstressing the position of the orifice. Mettauer recognised skin shortening as a cause of chordee, a fact not rediscovered until 1967. Urethroplasty from penile skin in situ was well described by Thiersch in 1869 and Duplay in 1874; additional covering skin flaps were developed in 1892 by Lauenstein. The modern enthusiasm for pedicle tubes from prepuce was first employed by Van Hook in 1896, Rochet in 1899, Hamilton Russell in 1900, and Mayo in 1901; the "buried skin" technique of Denis Browne was described by Duplay in 1880, although attributed by Browne to Hamilton Russell in 1915. Even scrotal tissues were incorporated in repairs in 1860 (Bouisson). Beck, in 1898, practised a repair for balanitic hypospadias very similar to the modern MAGPI repair, and free grafts, so popularised in the last 20 years, were performed by Nové-Josserand in 1897. We have certainly advanced from the era of the first millenium A.D., in which the treatment was amputation beyond the orifice, but almost all present-day techniques are well-founded in ideas developed by enterprising surgeons of the last century.

Key words History · Chordee · Urethroplasty · Hypospadias

Introduction

Over 300 repairs of hypospadias have been described in the literature, most within the last 60 years. Many authors claim originality of ideas, but a study of historical papers indicates that almost all present-day techniques were well-founded in ideas developed by enterprising surgeons of the last century. This paper researches the work of these early surgeons, who recognised the two main features of hypospadias – chordee and the position of the orifice – and each of these phenomena will be described separately.

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Early texts

Galen (130–199 A.D.), born in Pergaman and physician to the gladiators in Rome, dominated European medicine for 1,500 years until Vesalius (1543) corrected his anatomical mistakes and Harvey (1628) discovered the circulation of the blood. However, to be fair, autopsies were forbidden in Rome, and Galen never saw the inside of a body except through some of the injuries suffered by the gladiators. Amongst his many writings, he was the first to use the term "hypospadias", and more particularly emphasised the major significance of chordee:

> "Thus men afflicted with hypospadias find it impossible to beget children, the meatus being turned away from the extremity of the penis by the frenum, not because they lack fertile sperm, but because the curvature of the penis prevents its normal overflow from being conveyed forwards. This theory is confirmed by the ability to beget children if the frenum is divided". (De usu partium, XV, III) [1].

Apart from occasional references, the next major medical text referring to hypospadias was written by Ambrose Paré (1510–1590) [2]. Despite having no formal education and being apprenticed as a "barber-surgeon", he became surgeon to five French kings and came to fame as a military surgeon. He wrote his magnum opus at the age of 75 years – in French, to the displeasure of the Faculty in Paris, for which protocol dictated that major works should be in Latin. He also described chordee and gave extensive descriptions of hermaphroditic conditions and their overlap with hypospadias.

The third great surgical writer was Pierre Dionis (d. 1718) [3], the founder of modern French surgical education; sixteenth and seventeenth century French surgery still adhered to Galen anatomy. A stimulus to more rational surgery was actually led by Louis XIV, who offered courses

in the royal grounds on anatomy and operations. His director was Pierre Dionis, who wrote a major textbook on surgery, including a reference to hypospadias.

Chordee deformity

After Galen and a brief reference by Oribasus (325–403) that chordee interferes with intercourse, there was almost no reference to chordee for 1,500 years. Surgeons seemed to concentrate their efforts on the position of the orifice. However, two interesting historical vignettes are reported. In 1547 the Duke of Orleans became King Henry II of France. At age 14 years he was married to Catherine de Medici, also 14, to strengthen ties with Italy; she was the daughter of Lorenzo II and her uncle was Pope Clement VII. The king had a mistress, Diane de Poitiers, but despite this alliance and 10 years of trying with Catherine there were no children. He had a severe chordee deformity. He was said to be very robust and loved violent exercise, a degree of athletic prowess he probably needed to perform at all with chordee! However, his surgeon Jean Fernel corrected his chordee, after which he fathered ten children by Catherine, surely one of the most successful outcomes of surgery.

The other reference is to the attitude of the Roman Catholic Church to the deformity. A manuscript has been discovered in the University of Rome [4] of an action by a Maltese woman, Mathia, in 1542 for annulment of marriage on account of a deformity (hypospadias) of her husband. "She alleged that he suffered from a defect in the configuration of his "virile member" on account of which he did not urinate in an natural way like other men". The matter came before an ecclesiastical court – the Bishop's Court – that called two medical witnesses, Dr. Callus and Dr. de Bonellis, to examine the man in the presence of the Court. Their report on the husband is both accurate and colourful: "John's male member was inept or incapable and also useless for deflorating or perforating because it was short and curved, this curvation tending, in the judgement of Dr. Callus, to become more pronounced with rigidity of the penis ···". The Court annulled the marriage based upon Common Law established by ecclesiastical authorities regulating the discipline of priests and lay subjects; marriage is one of the laws and impotence as a "bodily defect" justifies annulment. We may criticise the procedure of having the medical examination carried out in the presence of Court officials, but this was normal practice in the 16th century. In fact, a more objectionable procedure for proof of impotence was practised by some European law courts in which actual sexual intercourse was required to be demonstrated in front of a matrone savante et expérimentée as late as the 18th century.

Not only did surgeons largely ignore the technical challenge of chordee for 1,800 years, they were also slow to recognise its causes. But in 1842, Mettauer [5] in the United States was the first to study the latter, and significantly recognised that skin tethering was the principle cause. He advocated a "succession of subcutaneous incisions until the organ is liberated", a very modern concept. Despite this, the advice was ignored, and indeed, the ignorance was formalised by Étienne Bouisson in 1860 [6], who first emphasised a "central fibrous band" on the corpora as the cause. That misconception continued for more than 100 years, even in Denis Browne's concept, and in my early years we laboriously searched for this central white band distal to the urethral orifice but could rarely find it. The truth of Mettauer's concept of 1842 was not rediscovered until 1967, when D. R. Smith of the United States [7] restressed the significance of skin and subcutaneous shortening and tethering. This concept was then used in the operative procedures of Allen and Spence in 1968 [8] and Lowell King in 1970 [9]. The fact that these are the principal structures of chordee and they are proximal to the orifice was realised by Mettauer 150 years ago!

This late recognition of the essential factors of chordee also ignored the pioneer work of Duplay in 1874 [10] and 1880 [11, 12], who stressed complete chordee release before urethroplasty, of Lauenstein in 1892 [13], who added skin from the pubis to the penis to make up for the deficiency, and of Edmunds in 1913 [14], who moved preputial skin to the shaft. Better known are the procedures of Blair et al. (1933) [15], Blair and Byers (1938) [16], and Byers (1955) [17], who extended the use of preputial flaps to make up for shaft deficiency, now a standard technique in many repairs. Other surgeons had tried other means: Beck, in 1917 [18], had passed the glans through a buttonhole of prepuce to get skin to the ventral surface (a procedure developed later by Nesbitt in 1941) [19]. Physick and also Pancoast in 1844 [20] resected part of the dorsal corpora (another procedure advocated by Nesbitt 100 years later). In 1936 Young [21] popularised the concept of a "congenital short urethra", another erroneous concept, exept in rare circumstances.

Urethral reconstruction

The first "repair" was recorded by Antyl in the first century A.D. [22] as *amputation beyond the orifice*, also advocated by Paul of Aegina (625–690) [23], and this was the only method offered for 1,000 years except for two brief references to skin stretching. Galen stretched the skin over the orifice up to the glans, anchored it with tape, and cut off the excess, also advocated in the Talmud of Jerusalum, Jebamot VIII.

The next period, for almost another 1,000 years until the nineteenth century, might be described as the era of *tunnelling and cannulation*. A tunnel was made by trocar through the penis and the channel kept open by a cannula or bougies until it epithelialised. Abulcasis (936–1013) [24], an Arab physician, used lead bougies, Guy de Chauliac (1363) [25] used a silver cannula, others being Lusitanus (1511–1568) [26], a Portugese, Dionis (1707) [3], who used a lead cannula, Morgagni (1761) [27], Sir Astley Cooper (1815) [28], using a gum elastic catheter as did

Dupuytren (1777–1835) [29] and Mettauer in 1842 [5]. In 1836 Dieffenbach [30] denuded the edges of the urethral gutter and sutured them over a cannula, but it broke down.

Repairs blossomed from the middle of the nineteenth century and ushered in the modern era. All the current methods derive from innovative surgeons of this period. Over 300 repairs have been described and all are based upon the use of three types of tissue: (1) penile and preputial skin; (2) scrotal skin; and (3) free grafts. It will be convenient to take each of these modalities in turn.

Penile and preputial skin

These tissues have been utilised in a number of ways, and at least six modern techniques have evolved: (1) tube in situ, with or without additional flaps; (2) buried skin; (3) pedicle tube grafts; (4) flip-flap grafts from proximal to the orifice; (5) urethral advancement; and (6) meatotomy, hemi-circumcision, meatal advancement and glanuloplasty (MAGPI).

Tube-in-situ, distal to the orifice

Anger, in 1874 [31], first used longitudinal flaps on either side of the urethral groove by overlapping them as a double-breasted suit, but did not denude them. Not surprisingly, they broke down. In 1875 John Wood in London [32] also covered the urethral groove with skin by button-holing the prepuce and joining it to peno-scrotal skin. Again, no actual "tube" was constructed.

Simon Duplay, in France in 1874 [10], first actually tubularised the skin, although Thiersch had performed this procedure for epispadias in 1869. Duplay incised the skin on either side of the urethral groove, bringing the edges together as a tube (although not the orifice itself, which was closed later over a catheter). The outer skin was closed edge-to-edge over the tube. There were many failures, but his first success came after a five-stage repeat. This basic tubularisation of skin distal to the orifice is still used by many surgeons and by the author.

In 1880 [11] Duplay described a second procedure that is remarkable in two respects. Firstly, he made the urethral strip much narrower and sutured it over a catheter, but not as a completely secure tube. In other words, it was a "buried strip", the forerunner of the Denis Browne technique 69 years later. Indeed, Duplay specifically commented on the capacity of the tissues to tubularise themselves: "Although the catheter is not actually covered entirely by skin, I am convinced that this has no ill effect on the formation of the new urethra, stricture formation does not occur so long as half the urethral wall is supplied by skin". The second feature of his repair was that the outer skin was not sutured edge-to-edge but by everted flap-to-flap, the forerunner of the overlapping denuded flaps developed by the author in 1970 [33, 34]. The next stage was to add extra skin to the area by transference of preputial skin, as an initial phase or at the same time. In 1892 Lauenstein [13] added skin from the pubis; in 1899 Beck [35] added a peno-scrotal flap over the tube; in 1913 Edmunds [14] moved prepuce before tubularisation; in 1917 Beck [18] button-holed the glans through the prepuce to obtain extra skin; and in 1932 Ombrédanne [36] did the same procedure and grafted the prepuce onto denuded areas lateral to the tube.

No surgeon of the last 60 years can claim originality for any of these basic concepts. In the author's two-stage repair he grafts preputial skin onto the shaft and glans as a first stage, to obtain thick viable skin for tubularisation in the second stage down to the tip, with a second stage of overlapping double-breasted, denuded flaps over the new urethral tube [33, 34]. But Duplay, Edmunds, Ombrédanne, and Byars laid the foundation.

Buried skin

The concept of a buried strip of skin that would subsequently complete its own tubularisation was popularised by Denis Browne in 1949 [37]. Browne always acknowledged the idea to the Australian surgeon, Hamilton Russell, who in 1915 [38], after excising a urethral stricture, found that if the urethral ends were joined as a flat strip of mucosa and buried, the urethra re-formed. Browne was apparently unaware of the second operation of Duplay in 1880, in which the concept was developed for hypospadias, described previously.

Pedicle skin grafts

In the modern era there has been much enthusiasm for the use of pedicle tubes, generally derived from the prepuce, retaining their principal blood supply on the pedicle and swung ventrally to be anastomosed to the existing urethra. One-stage repairs have been achieved, commencing with the work of Broadbent, Woolf, and Toksu (1961) [39], Des Prez, Persky, and Kiehn (1961) [40] (who first used the words "island flap"), and developed so expertly by Duckett [41]. But like other aspects of hypospadias, surgeons of the last century and early in this century had attempted similar repairs. In 1838 Liston [42] had closed a fistula from a preputial flap – not quite a pedicle tube, but a forerunner. In 1896 Van Hook [43] used a proximally-based pedicle tube of prepuce; in 1899 Rochet [44] swung a pedicle from scrotal skin based on blood supply just proximal to the meatus and tunnelled through the glans and penis; in 1900 Hamilton Russell [45] described a "stole" operation, a pedicle tube of penile and preputial skin; in 1901 [46] C. H. Mayo [46] made a distally based pedicle of prepuce; in 1917 Bevan [47] tunnelled a penile pedicle through the glans; in 1929 Rosenstein [48] used a pedicle graft of bladder mucosa; and in 1940 Davis [49] employed a pedicle from dorsal pedicle skin.

Flipp-flap tube based proximal to the meatus and swung distally

Bouisson had used a proximal peno-scrotal flap swung distally in 1860 [6], but strangely, no one else employed a flip-flap technique until 1932 (Mathieu) [50]. It was one of the few procedures not followed up in the nineteenth century. After Mathieu, it was later developed by Ross et al. (1959) [51] and Mustardé (1965) [52].

Urethral advancement

Beck, in 1898 [35], was probably the first to mobilise the urethra and draw it distally along a trocar track. But like the flip-flap repair, urethral advancement was a late development, first by the British urologist Badenoch (1950) [53] for by-passing a stricture, and later by McGowan and Waterhouse (1964) [54].

MAGPI (meatal advancement and glanuloplasty) [55]

In 1874 Duplay [10] deepened the glanular groove and closed it over a catheter, and in 1898 Beck [35] mobilised the urethra for balanitic hypospadias and sutured it distally – procedures not dissimilar to the modern MAGPI repair.

Scrotal repairs

As scrotal repairs are now rarely performed, the discussion will be brief. In 1860 Bouisson [6] utilised a peno-scrotal flap swung distally, but not tubularised. In 1870 Moutet [56] swung a scrotal flap over the urethral groove and covered its new surface with skin from the pubis, which sloughed. In London in 1875 Wood [32], being unaware of Duplay's work, button-holed the glans through the prepuce and then sutured the prepuce to a peno-scrotal flap. In 1891 Landerer [57] denuded long skin strips on either side of the urethral groove both proximal and distal to the meatus, then buried the penis back into the scrotum so that the denuded strips came together; as a second stage, the penis was released from the scrotum. The modern era was popularised by Cecil (1936) [58] at first by a three-stage technique.

Free grafts

Free grafts have been popular in some countries over the last 30 years, but their use also dates from the last century. Nové Josserand, the third of the great French surgeons of the last century (with Duplay and Ombrédanne), in 1897 [59] utilised an autograft of penile and preputial skin wrapped around a catheter and tunnelled through the penis and glans along a trocar track introduced through a perineal urethrostomy. It strictured, as is the fate of many free grafts. In 1937 McIndoe [60] designed a hollow trocar to introduce a Thiersch graft of penile and preputial skin around a gum electric catheter. In 1941 Humby [51] employed a one-stage technique using a full-thickness prepuce graft, later developed by Devine and Horton in 1955 [62].

Other tissues have also been used. Saphenous vein was used by Tanton, Unger, and Becker in 1909 [63] and by Tuffier in 1910 [64], Cantas in 1911 [65], and Marion 1922 [66]. Fistulae and rejection of grafts resulted. Bladder mucosa was first used by Rosenstein in 1929 [48] as a pedicle graft and as a free graft by Memmelaar in 1947 [67] and Marshall and Spellman in 1955 [68]. Schmieden employed ureter (from operation or autopsy) in 1909 [69] with failure; Lexer used appendix in 1911 [70]; cadaveric urethra was said to be a success by Bourque in 1952 [71], as Pringle claimed for bullock's urethra in 1904 [72]. Even a dog's aorta was used by Legueu [73], but without success.

Conclusion

There is much to be learned from a study of historical papers, especially of surgical developments in the second half of the nineteenth century. Most of the current techniques for hypospadias repair were developed during that time by innovative and enterprising surgeons. The numerous modifications of recent times, often claimed as original or distinctive and bearing the surgeon's name, are little more than minor variations of the pioneer work of the early surgeons. That results are now considerably better is undoubtedly true, but this can be attributed to better anaesthesia, antibiotics, better sterility, practised techniques with large-volume experience, non-irritant catheters and suture material, and better understanding of tissue handling. The credit for the innovative principles of repair is inherited from our surgical colleagues of yester-year.

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