

Psychosexual and psychosocial adjustment of hypospadias patients

De psychoseksuele en psychosociale ontwikkeling van
hypospadie patiënten

This research was financially supported by the Sophia Foundation for Medical Research, the Gerrit Jan Mulder Foundation, the Esser Foundation, and the Department of Plastic and Reconstructive Surgery, University Hospital "Dijkzigt" Rotterdam.

Cover photo by Wil Hofman ©

Cover design by Maarten van den Bosch and Marc Mureau

Printed by Optima Druk, Molenaarsgraaf, The Netherlands

Psychosexual and psychosocial adjustment of hypospadias patients

De psychoseksuele en psychosociale ontwikkeling van hypospadie patiënten

Proefschrift

ter verkrijging van de graad van doctor
aan de Erasmus Universiteit Rotterdam
op gezag van de Rector Magnificus
Prof. Dr. P.W.C. Akkermans M.A.
en volgens besluit van het College voor Promoties.

De openbare verdediging zal plaatsvinden op
woensdag 6 september 1995 om 11.45 uur

door

Marcus Antonius Maria Mureau

geboren te Prinsenbeek.

Promotiecommissie

Promotores: Prof. Dr. F.C. Verhulst
Prof. Dr. A.K. Slob

Co-promotor: Mw. Dr. F.M.E. Slijper

Overige leden: Prof. Dr. J.C. van der Meulen
Prof. Dr. R.J. Scholtmeijer
Prof. Dr. J. Passchier

Contents

List of contributing authors		iii
Chapter 1	General Introduction	1
Chapter 2	Literature review and study aims	21
Chapter 3	Method	37
Chapter 4	Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study <i>Accepted for publication in the Journal of Urology, 1995</i>	43
Chapter 5	Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study <i>Accepted for publication in the Journal of Urology, 1995</i>	61
Chapter 6	Genital perception of children, adolescents, and adults operated on for hypospadias: A comparative study <i>Accepted for publication in the Journal of Sex Research, 1995</i>	81
Chapter 7	Satisfaction with penile appearance following different types of hypospadias surgery: The patients' and the surgeon's view <i>Accepted for publication in the Journal of Urology, 1995</i>	105
Chapter 8	Psychosocial functioning of children, adolescents, and adults following hypospadias surgery: A comparative study <i>Submitted for publication, 1995</i>	117
Chapter 9	Evaluation and recommendations	141
Appendix A ₁	Interview for Psychosexual Development and Functioning of Hypospadiac Adults (IPDF-HA)	153

Appendix A ₂	Genital Perception Scale (GPS)	159
Appendix B ₁	Interview for Psychosexual Development and Functioning of Hypospadiac Boys (IPDF-HB)	161
Appendix B ₂	Junior Genital Perception Scale (GPS-J)	175
Summary		181
Samenvatting		189
Dankwoord		197
Curriculum vitae		201

Contributing authors

Ms. Dr. Froukje M. E. Slijper, child psychologist,
Department of Child and Adolescent Psychiatry,
Sophia Children's Hospital Rotterdam,
Dr. Molewaterplein 60,
3015 GJ Rotterdam.

Prof. Dr. A. Koos Slob, sexologist,
Department of Endocrinology and Reproduction,
Faculty of Medicine and Health Sciences,
Erasmus University Rotterdam,
Dr. Molewaterplein 50,
3015 GE Rotterdam.

Prof. Dr. Frank C. Verhulst, child psychiatrist,
Department of Child and Adolescent Psychiatry,
Sophia Children's Hospital Rotterdam,
Dr. Molewaterplein 60,
3015 GJ Rotterdam.

Dr. Rien J. M. Nijman, pediatric urologist,
Department of Pediatric Urology,
Sophia Children's Hospital Rotterdam,
Dr. Molewaterplein 60,
3015 GJ Rotterdam.

Prof. Dr. Jacques C. van der Meulen, plastic surgeon,
Department of Plastic and Reconstructive Surgery,
Academic Hospital "Dijkzigt" Rotterdam,
Dr. Molewaterplein 40,
3015 GD Rotterdam.

CHAPTER 1

GENERAL INTRODUCTION

1.1 INTRODUCTION

Hypospadias is a common congenital urogenital anomaly, occurring predominantly in boys and requiring surgical correction in the majority of cases. Before the psychosexual and psychosocial consequences of hypospadias are described in the present thesis, the embryology, etiology, morphology, and surgical treatment of hypospadias will be explained in the next four paragraphs.

1.2 EMBRYOLOGY

1.2.1 NORMAL SEXUAL DIFFERENTIATION

(Paragraph 1.2.1 is for the major part derived from two review papers by Blyth & Duckett,¹ and Wilson, Griffin, & Russell.²) Normal sexual differentiation during embryogenesis consists of three sequential, ordered, and inter-related processes (figure 1.1). The first process involves the establishment of *chromosomal sex* at the time of fertilization. In mammals, the heterogametic sex (XY) is male and the homogametic sex (XX) is female. Before 7 weeks of life, the human fetus is phenotypically indifferent with female and male embryos sharing common primordia (two gonadal ridges, two Wolffian or mesonephric ducts, and two Müllerian or paramesonephric ducts). Recently, DNA sequences on the short arm of the Y gene (SRY = Sex determining Region of the Y gene, or TDF gene = Testis Determining Factor gene) have been identified, which are considered to be responsible for the differentiation of the gonadal ridges into

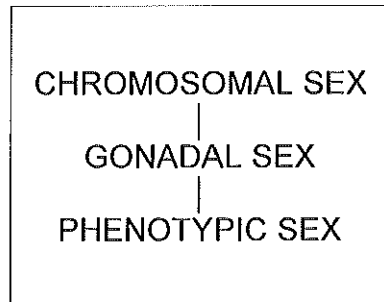


Figure 1.1. Sequence of events in sexual differentiation

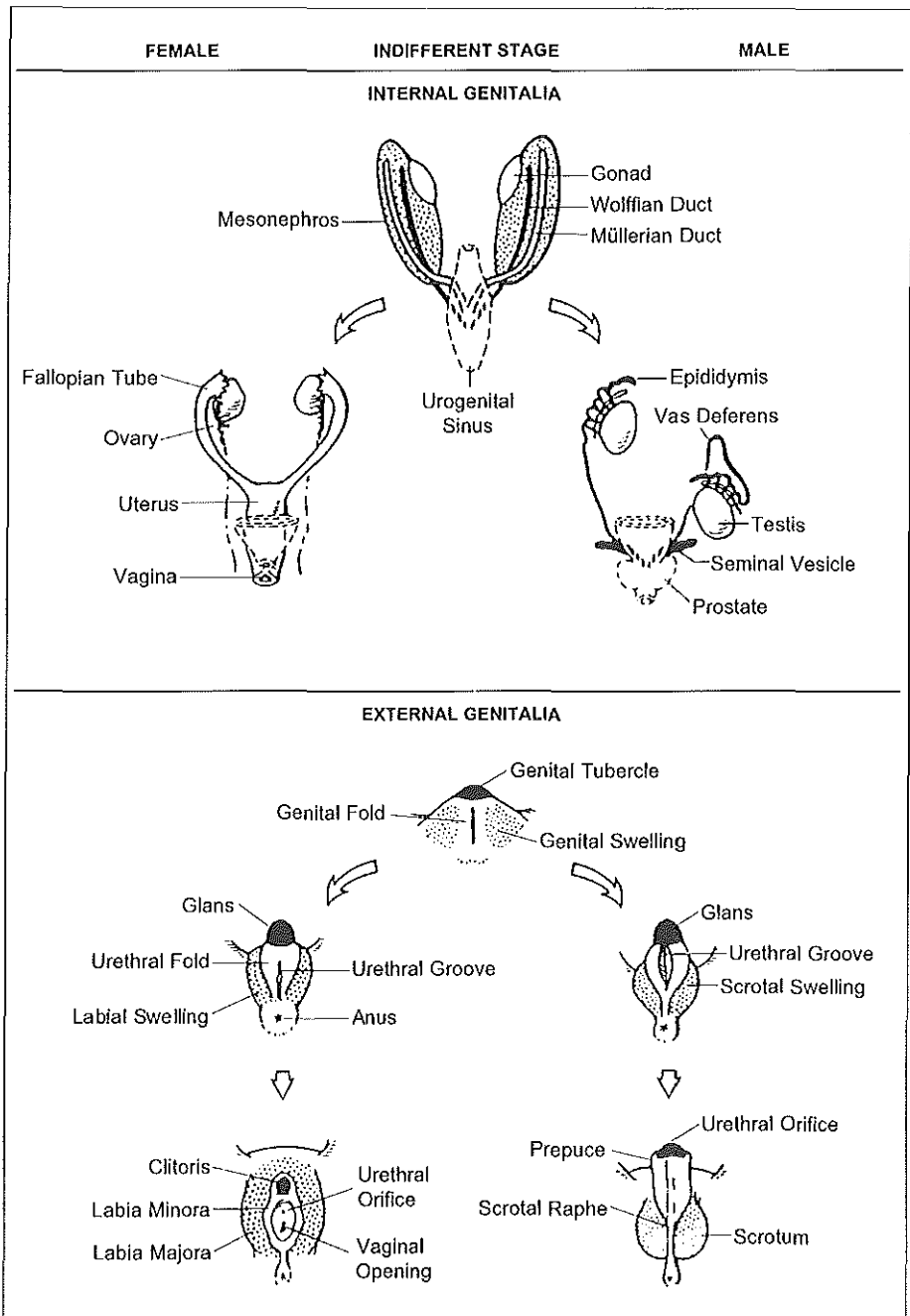


Figure 1.2. Phenotypic differentiation of internal ducts and external genitalia in male and female embryos

normal testes. This latter differentiation is the second process of sexual differentiation, which is known as the translation of the chromosomal sex into the *gonadal sex*. The testicular gonads differentiate further into primordial germ cells which become the spermatogonia, Sertoli cells, and Leydig cells. The third phase of sexual differentiation, the translation of gonadal sex into *phenotypic sex*, takes place between the 7th and 16th week of gestation and is a direct consequence of the type of gonad formed and the endocrine secretions of the fetal testes. In the formation of the phenotypic sex, the indifferent internal (Wolffian and Müllerian ducts) and external primordia (genital tubercle, genital folds, and genital swellings) are converted to male or female forms. In the male, the Wolffian ducts give rise to the epididymides, vasa deferentia, and seminal vesicles, and the Müllerian ducts either disappear or persist as testicular appendage. In the female, the Müllerian ducts give rise to the fallopian tubes, uterus, and upper segment of the vagina, while the Wolffian ducts either disappear or persist as Gartner's ducts. The internal genital tracts in males and females arise from *different* primordia, contrary to the external genitalia and lower urogenital tracts, which develop from *common* primordia in both sexes (genital tubercle, genital folds, and genital swellings; see figure 1.2). In the female, the genital tubercle becomes the clitoris, the genital swellings form the labia majora, and the genital folds are converted into the labia minora. In the male, fusion and elongation of the urethral folds cause formation of the urethra and the shaft of the penis and bring the urethral orifice to the genital tubercle (glans penis). The fused genital swellings become the scrotum (figure 1.2).

The previously described formation of the phenotypic sex develops in the female direction, unless the gonads produce three testicular hormones: testosterone (a steroid secreted by the Leydig cells), anti-Müllerian hormone (AMH, a glycoprotein secreted by the Sertoli cells), and dihydrotestosterone (DHT, formed by reduction of testosterone through the enzyme steroid 5 α -reductase). Masculinization of the fetus is a direct result of the testicular hormones. Stimulated and regulated by human chorionic gonadotropins (secreted by the placenta), the testicular Leydig cells begin to produce and secrete testosterone which acts directly upon the Wolffian ducts and induces

the development of the epididymides, vasa deferentia, and seminal vesicles. AMH causes regression of the Müllerian ducts and prevents the development of fallopian tubes and the uterus in males. These processes are completed at about 13 weeks of gestation. DHT acts in the urogenital sinus to induce development of the male urethra and prostate, and in the urogenital tubercle, swellings, and folds to cause the midline fusion, elongation, and enlargement that forms the penis and scrotum. These latter processes are completed at 16 weeks of gestation.

1.2.2 DEVELOPMENT OF THE ANTERIOR URETHRA

(Paragraph 1.2.2 is for the major part derived from a recent study by Altemus & Hutchins.³) The development of the anterior urethra can be divided into two processes: development of the *bulbar* and *spongy* (penile) urethra, and development of the *glanular* urethra. The formation of the bulbar and spongy urethra occurs by mesenchyme proliferation (the urethral plate), underlying the epithelium of the urethral folds. This causes the creation of a tubular urethra through epithelial fusion in the ventral midline of the penis (figure 1.3). The tubular urethra becomes invested by mesenchyme, which forms the future corpus spongiosum. This latter process of fusion of the urethral folds begins proximally and extends distally to the glans penis. The successful completion of the anterior urethra involves three processes

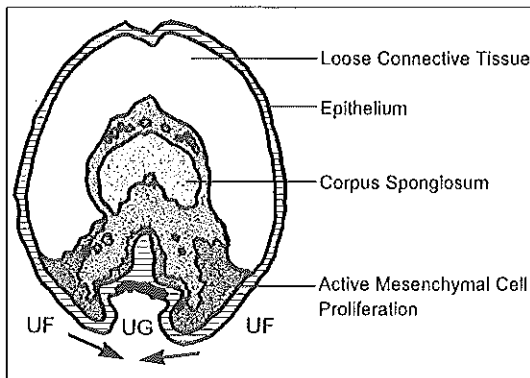


Figure 1.3. Transverse plane of the penis; UG = urethral groove; UF = urethral fold

which must act in synchrony.

First, growth of the skin over the glans penis forms the prepuce, and covers and closes the ventral aspect of the urethral plate by the frenulum. Second, the mesenchyme which forms the corpus spongiosum becomes invested by and fuses with the mesenchyme of the glans penis. Finally, the solid ure-

thral plate within the glans becomes canalized and acquires continuity with the spongy urethra. The formation of the urethra distal to the coronal sulcus has been a subject of controversy, but it is now believed that its development involves two separate processes. The *distal* part of the glanular urethra, the future navicular fossa, is the result of epithelial ingrowth from a terminal epithelial tag at the tip of the glans penis, whereas the *proximal* part of the glanular urethra is formed by fusion of the urethral folds and subsequent canalization of the urethral plate.

1.3 INCIDENCE AND ETIOLOGY OF HYPOSPADIAS

Hypospadias is a common urogenital anomaly, with a reported incidence ranging from 0.8 to 8.2 per 1,000 live male births.⁴ Studies which investigated the etiology of hypospadias showed equivocal results (see table 1.1).⁵⁻¹⁵ Some investigators proposed autosomal recessive inheritance with incomplete expression or autosomal dominant inheritance with low penetrance, but most authors favor a multifactorial model, in which multiple genetic factors combine with environmental factors during early pregnancy.^{5, 6} However, some studies were able to detect environmental factors, whereas other investigations were not.^{5, 6} In some cases of hypospadias chromosomal aberrations,⁴ or sex chromosomal aberrations in combination with endocrinopathies such as poor testosterone response to human chorionic gonadotropin stimulations^{10, 11} or androgen receptor defects were found,⁷⁻⁹ whereas other investigators could not find androgen receptor defects.¹²⁻¹⁵ Until now, however, the exact cause of isolated hypospadias remains unknown in most cases.¹³

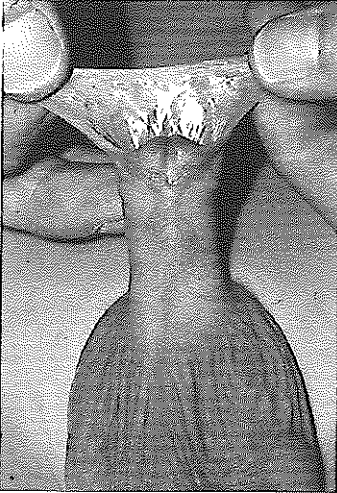
1.4 DEVELOPMENT AND MORPHOLOGY OF HYPOSPADIAS

The term hypospadias derives its origin from the Greek word "hypo" meaning under and "spadon" which denotes a rent or fissure.⁴ Hypospadias is caused by a disturbance in the fusion of the urethral folds, a disturbance in the canalization of the urethral plate or a combination of both,⁶ resulting in an abnormal position of the urethral orifice (see pictures 1.1 to 1.4). It is believed that the synergistic activity of the urethral epithelium and penile mesenchymal tissue is arrested at some stage of normal development which

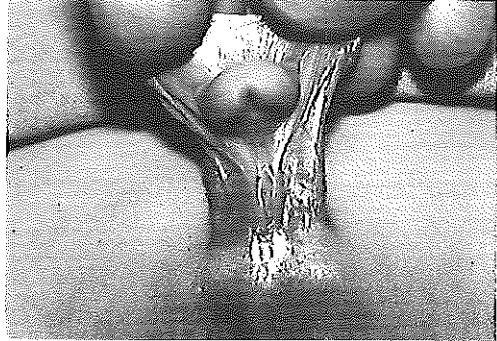
Chapter 1

Table 1.1. Factors which have been suggested to play a role in the etiology of hypospadias

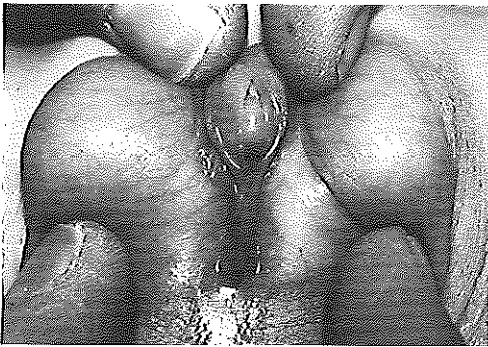
Genetic factors	Endocrinological factors	Environmental factors
Autosomal recessive inheritance ^{5,6} (e.g., steroid 5 α -reductase 2 deficiency; ² syndromes as the Opitz syndrome ⁴).	Testosterone synthesis enzyme deficiencies or defects (e.g., steroid 5 α -reductase 2 deficiency, ² 17-ketosteroid reductase deficiency, ¹¹ 17, 20-desmolase deficiency ¹¹).	Hormonal compounds in the first trimester of pregnancy (e.g., progestin, progestagen compounds). ^{5,6}
Autosomal dominant inheritance, with low penetrance. ^{5,6}	Androgen receptor synthesis disorders (e.g., {partial} androgen insensitivity ^{8,9}).	Hormonal compounds before pregnancy (e.g., oral contraceptives). ^{5,6}
Multifactorial inheritance, with multiple genetic factors in combination with environmental factors. ^{5,6}	Delay in maturation/poor function of the hypothalamic-pituitary-testicular axis (e.g., poor testosterone response following human chorionic gonadotropin stimulation ^{10,11}).	Paternal factors (e.g., testicular anomalies as cryptorchidism, varicocele, atrophic testes, following mumps orchitis; age). ⁴
Y linked dominant inheritance. ⁴		Maternal factors (e.g., parity, threatened abortions, age, early menarche, caesarian section). ^{5,6}
X linked recessive inheritance ({partial} androgen insensitivity syndrome ^{8,9}).		Child factors (e.g., low birth weight, prematurity, small birth length, birth order). ^{5,6}
Autosomal chromosome aberrations (e.g., partial deletion of the short arm of chromosome 4 ⁴).		Exposure to toxins (e.g., cigarettes, alcohol, drugs) or radiation during pregnancy. ^{5,6}
Sex chromosome aberrations (e.g., 46XY/45XO, 47XXY/46XY ⁷).		Low placental weight. ⁶
		Seasonal influences. ^{5,6}



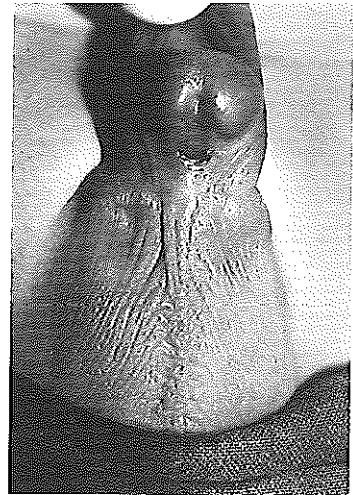
Picture 1.1. Coronal hypospadias



Picture 1.2. Penoscrotal hypospadias



Picture 1.3. Perineoscrotal hypospadias



Picture 1.4. Penile hypospadias

results in the hypospadiac penis.³ In most cases there exists a typical triad of penile skin anomalies, namely deficiency of skin on the ventral aspect, an oblique raphe on each lateral aspect, and a surplus of skin on the dorsal aspect of the penis.¹⁶ The cessation of normal preputial development accounts for the above mentioned skin anomalies and lack of frenulum, and is directly related to failure of complete fusion of the urethral folds and failure to canalize the urethral plate completely.³ Preputial growth over the glans penis is necessary to seal the urethral orifice at the coronal sulcus.³ In many cases of hypospadias the corpus spongiosum beyond the urethral orifice is

absent, which is attributed to failure of fusion of the urethral folds and subsequent insufficient condensation of mesenchymal tissue to the arrested development of the epithelium of the urethra.³ In some cases, there also exists *chordee*, a fibrous band or a fan-shaped area of fibrosis distal to the meatus, which represents the hypoplastic urethral plate in which the urethra was to develop.¹⁶

In a dichotomized classification *distal* hypospadias refers to conditions in which the position of the urethral meatus is situated on the glans penis, the coronal sulcus, or just proximal to the coronal sulcus, and *proximal* hypospadias refers to conditions in which the urethra opens at some point along the penile shaft, at the penoscrotal junction, or on the perineum. Penile curvature (as a result of ventral skin deficiency or chordee), penile underdevelopment, and occasionally penoscrotal transposition, cryptorchidism, as well as an enlargement of the prostatic utricle may be observed; these latter concomitant anomalies are found more often in proximal than in distal hypospadias.¹⁶ Distal hypospadias accounts for approximately 70% of all cases.⁴

1.5 SURGICAL TREATMENT

1.5.1 INTRODUCTION

Surgical reconstruction of the urethra and straightening of the penis is necessary to ensure voiding in a standing position and unhampered adult sexual functioning. Another important goal of hypospadias surgery is the achievement of a cosmetic appearance of the penis which is as "normal" as possible. More than 150 different surgical techniques and modifications to correct hypospadias have been described.⁴ Eight surgical techniques which were performed on patients in the present study will be described in detail in paragraphs 1.5.3 and 1.5.4, and a brief historical review of hypospadias surgery is given in the next paragraph.

1.5.2 HISTORICAL REVIEW OF HYPOSPADIAS SURGERY

(Paragraph 1.5.2 contains several parts derived from De Vries¹⁷ and Wood-Smith.¹⁸) Since the first description of the correction of hypospadias in the second century by Galen (131-201 A.D.) who used a canalization

technique, little progress was made in subsequent centuries, until Dieffenbach (1837) unsuccessfully tried to reconstruct the urethra by raising a fold of skin along either side of the proposed urethra by suturing the edges together. In 1875, Anger successfully applied Thiersch's method for correcting epispadias (1869) to hypospadias. Anger as well as Duplay (1874) employed the principle of the "buried skin strip" in the reconstruction of the urethra. Later in 1936 Dennis Browne, whose technique became very popular, re-introduced the same principles for reconstructing the urethra. Because the Denis Browne method was complicated by a high fistula rate and an unequally calibered urethra in many cases, the buried skin strip procedure was further improved by among others Cecil (1952), Van der Meulen (1964), and Farkas (1967). Byars (1951) reconstructed the neo-urethra by first tubing the skin flap before the wound was closed, suturing the penile skin over the neo-urethra. This technique has been used by many different surgeons and has produced reliable, reproducible functional results. Another approach, originally described by Wood (1875) was the development of a meatal based flap. A flap of penile skin is created proximal to the meatus with a perimeatal hinge which is raised and sutured to two incisions on either side of the proposed urethra. This principle with minor variations (Ombredanne, 1911; Mathieu, 1932) has been used until today without the serious problems of fistula formation. A third concept which was launched by Nové-Josserand (1897) and revived by McIndoe in 1937 was the use of a free unvascularized skin graft, which was tubed and pulled through a tunnel in the penis which was made with a cutting troicart. This technique never became very popular because of the high rates of urethral fistulas and strictures. Many types of grafts have been tried to reconstruct the urethra with variable degrees of success: for instance the ureter (Schmieden, 1909), the appendix (Axhausen, 1918; McGuire, 1927), the vena saphena magna (Tanton, 1910; Adlercreutz, 1918), the tunica vaginalis (Rivoir, 1954), and bladder mucosa (Mummelaar, 1947; Marschall & Spellman, 1955). Recently, the use of buccal mucosa has become popular for the treatment of patients who have been treated unsuccessfully many times, and consequently do not have enough penile skin to reconstruct the urethra.¹⁹ Other methods which were developed used scrotal

skin flaps either as part of the urethra (Rosenberger, 1891; Landerer, 1891; Bidder, 1892) or for compensating the shortness of ventral penile skin (Rochet, 1899; Beck, 1917; Cecil, 1932; Blair & Byars, 1938; Culp, 1951). Disadvantages of these procedures are the inclusion of hair bearing skin in the urethral canal and obstruction caused by the accumulation of calcareous deposits on these hairs. A different concept is the urethral advancement procedure by Beck (1900) who made use of the elastic properties of the urethra. This procedure is still used today (Koff, 1981). Another approach, described by Duckett in 1981, is the advancement of the meatus which is known as the Meatal Advancement and Glanuloplasty Incorporated (MAG-PI) technique. The concept of a tubed pedicle flap from the dorsum of the penis to form the new urethra was developed by Davis (1940). This principle was further refined by Broadbent, Woolf, and Toksu (1961). The technique of a tubed vascularized flap was originally described by Hook (1896) and Mayo (1901), who formed such a flap from the prepuce for the first time. In 1961, Desprez described a method using a tubed skin flap of one of the lateral wings of the prepuce. A few years later Asopa (1971) and Standoli (1979) described a technique which splits the prepuce and tubularizes the inner or outer surface of the prepuce to reconstruct the urethra. A more recent description of such a so called Transverse Preputial Island Flap (TPIF) technique was given by Duckett. A variant of this latter technique is the Onlay Preputial Island Flap (OPIF) technique which keeps the urethral plate intact to form the dorsal wall of the neo-urethra (e.g., Perović, 1981).

Some of the previously mentioned procedures are one-stage procedures, whereas other techniques are multiple staged procedures. In the first stage(s) of the latter procedures the penis is straightened by resecting the chordee or by transposing penile or preputial skin from the dorsal side to the ventral side of the penis. The main motive for performing multiple staged procedures is to keep the complication rate at an acceptable level. Because in the majority of cases chordee is absent and surgical techniques and materials have been refined recently, one-stage procedures which straighten the penis and reconstruct the urethra simultaneously have become very popular. Examples of such one-stage procedures which have been used until today are

the Mathieu procedure (1932), the King procedure (1970), the Hodgson I, II, and III procedures (1972), the MAGPI procedure (1981), the TPIF procedure originally described by Asopa (1971) and later by Standoli (1979) and Duckett (1981), and the Horton/Devine flip flap procedure (1973). Because plastic surgeons and pediatric urologists continuously try to improve the cosmetic surgical results to make the penis look as "natural" as possible, another change in the concept of correcting hypospadias has been the development of *terminalizing* surgical techniques which split, core, or tunnel through the glans to create a true terminal glanular meatus, replacing earlier *ventralizing* surgical techniques, which bring the meatus onto the underside of the glans or coronal sulcus.²⁰ A third change in the treatment of hypospadias has been the earlier timing of surgery. The age at which surgery is performed has been reduced from school age (6 to 12 years) to between the ages of 6 and 18 months. Surgeons have performed hypospadias surgery at increasing earlier ages, since improved microsurgery and anesthesia have made it feasible to operate very young children.^{21, 22} Furthermore, studies on the timing of hypospadias surgery seem to indicate it is psychologically less harmful for the child to operate between the ages of 6 and 18 months,^{22, 23} although there still is a dearth of knowledge about the timing of genital surgery that is based on empirical studies.

1.5.3 VENTRALIZING REPAIRS

*Van der Meulen repair.*²⁴ Van der Meulen developed a one-stage urethroplasty for patients with distal hypospadias and a two-stage ortho-urethroplasty, which straightens the penis in the first stage and reconstructs the urethra in the second stage for patients with proximal hypospadias. The basic principle of the Van der Meulen technique is the ventral rotation of penile skin which corrects the shortage of ventral skin and the surplus of dorsal skin. The ventrally rotated skin flap is used for the reconstruction of the distal urethra and closure of the wound. The reconstructed neo-meatus opens at the undersurface of the glans penis, and penile skin covers the ventral side of the glans (see figure 1.4).

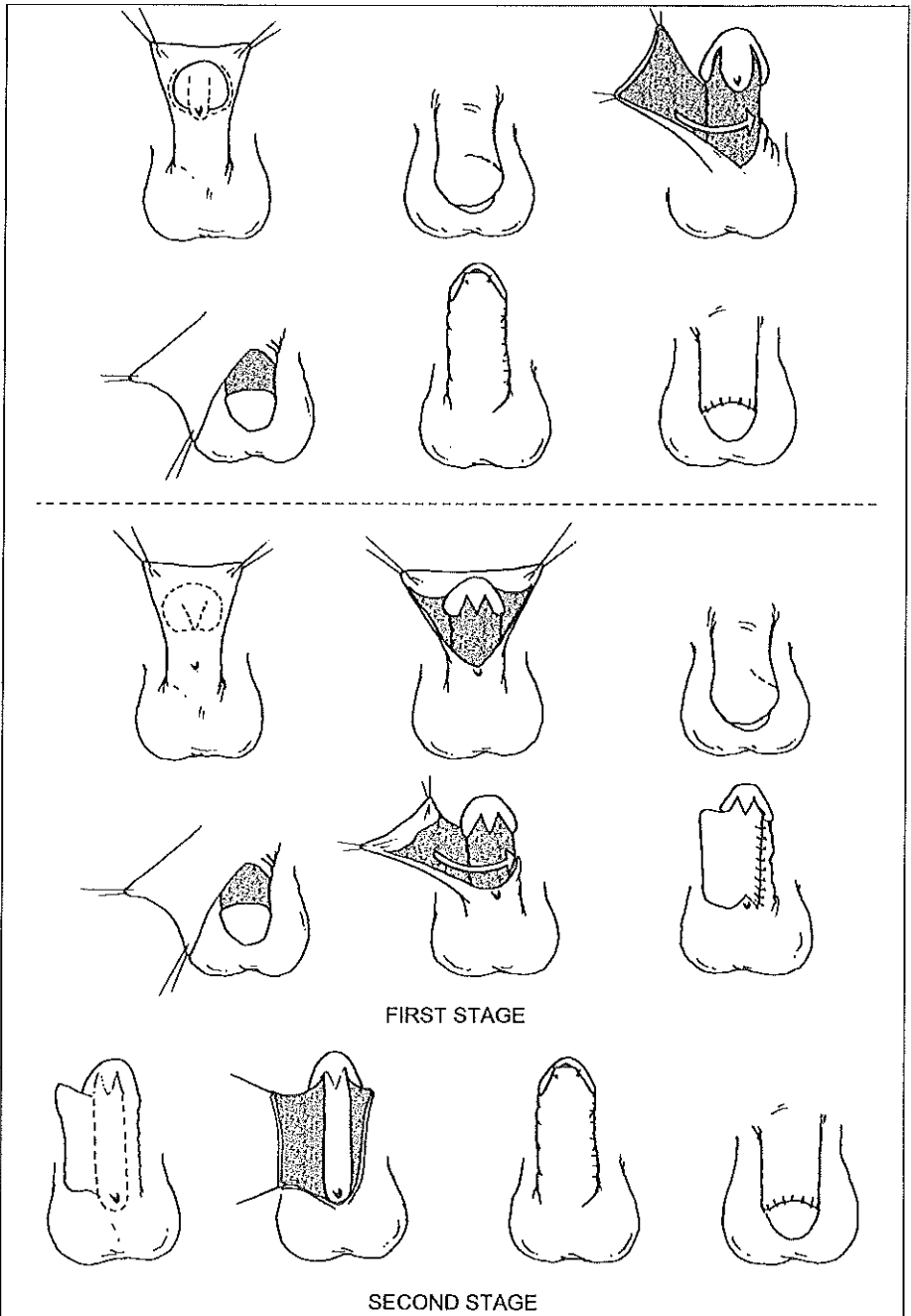


Figure 1.4. Schematic drawing of the Van der Meulen repair. Above dotted line: One-stage urethroplasty for distal hypospadias. Below dotted line: Two-stage ortho-urethroplasty for proximal hypospadias

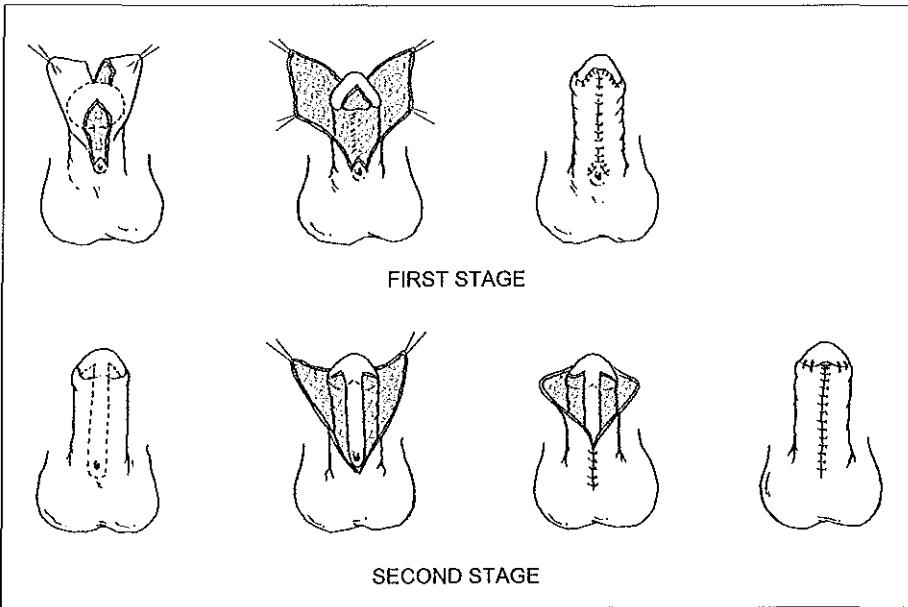


Figure 1.5. Schematic drawing of the Byars/Browne repair for both distal and proximal hypospadias

*Byars/Browne repair.*²⁵ The Byars/Brown technique is a two-stage hypospadias repair, irrespective of the severity of hypospadias. In the first stage the penis is straightened, the glans is split ventrally and the dorsal surplus of prepuce skin is used to close the defect on the ventral side of the penis. In the second stage the urethra is reconstructed. A skin strip is created by making longitudinal incisions on either side of the proposed urethra. The urethra is reconstructed by approximating the edges of the ventral penile skin which are sutured together over the buried skin strip closing the wound. As is the case in the Van der Meulen procedure, the Byars/Browne repair leaves a meatus at the undersurface of the glans penis with penile skin on the ventral side of the glans (see figure 1.5).

1.5.4 TERMINALIZING REPAIRS

*MAGPI (Meatal Advancement and Glanuloplasty Incorporated).*²⁶ The MAGPI procedure is a one-stage repair for correcting coronal and glanular hypospadias. First, the dorsal wall of the meatus is advanced by transversally closing a longitudinal incision which is made in the dorsal wall of the meatus

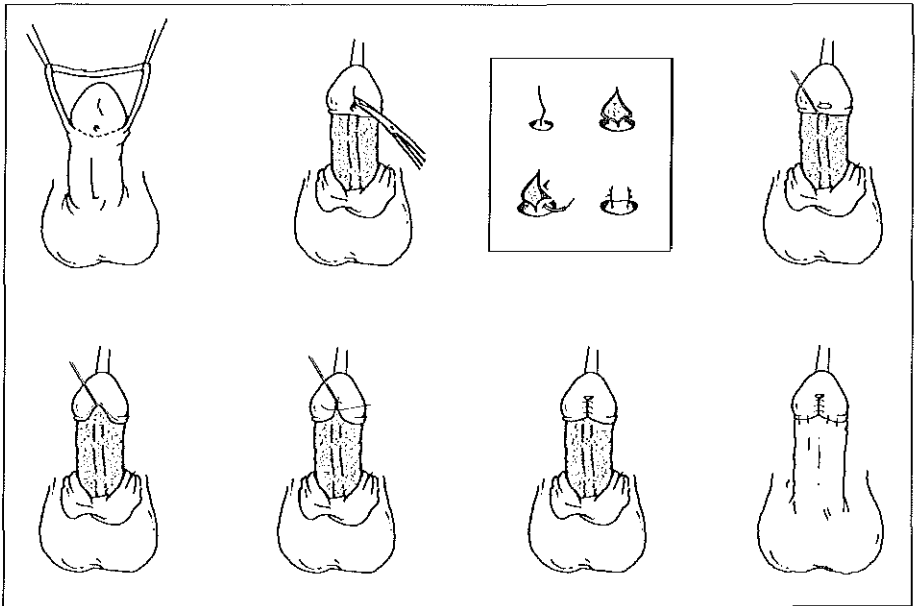


Figure 1.6. Schematic drawing of the MAGPI repair for glanular and coronal hypospadias

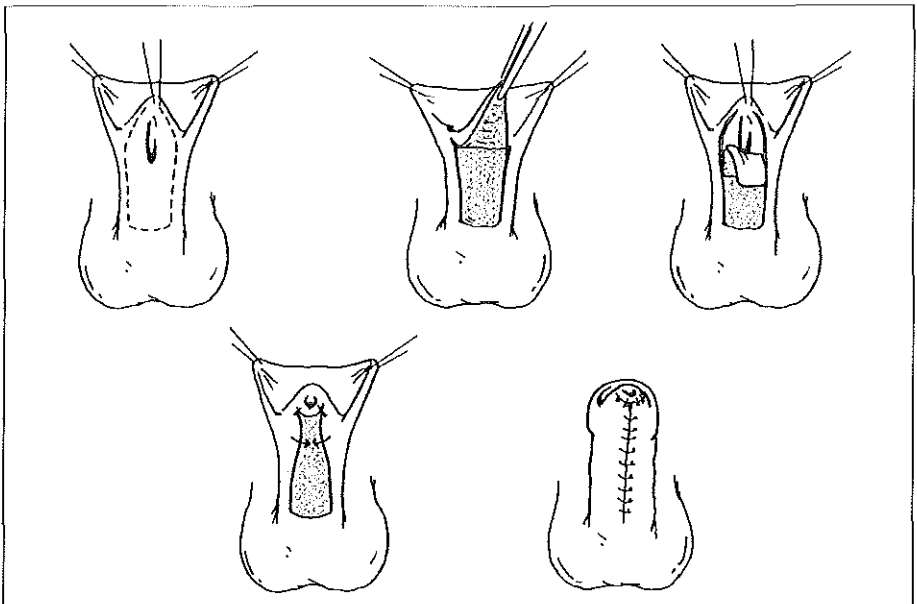


Figure 1.7. Schematic drawing of the Mathieu repair for glanular and coronal hypospadias

(Heineke-Mikulicz closure). Next, the ventral wall of the meatus is advanced and the glanular shape is improved by pulling the glanular edge distally (creating an inverted V-shape) and suturing the lateral edges together in the midline. Consequently, a terminal intraglanular meatus is created (see figure 1.6).

*Mathieu procedure.*²⁷ The Mathieu technique is a one-stage procedure to correct subcoronal or more distal forms of hypospadias. A flap of penile skin is created proximal to the meatus with a perimeatal hinge which is raised and sutured to two paramedian glanular incisions. Next, the closure is completed by suturing the external edges of the glans over the wound leaving a terminal intraglanular meatus (see figure 1.7).

*GAP (Glans Approximation Procedure).*²⁸ This one-stage repair for coronal and glanular hypospadias is performed specifically on patients with an excessively deep glanular groove. A U-shaped de-epithelialized area of the glans is created on each lateral side of and proximal to the meatus. Subsequently, the edges of the wound are approximated in the ventral midline of the glans creating a terminal intraglanular meatus. A subcutaneous layer is

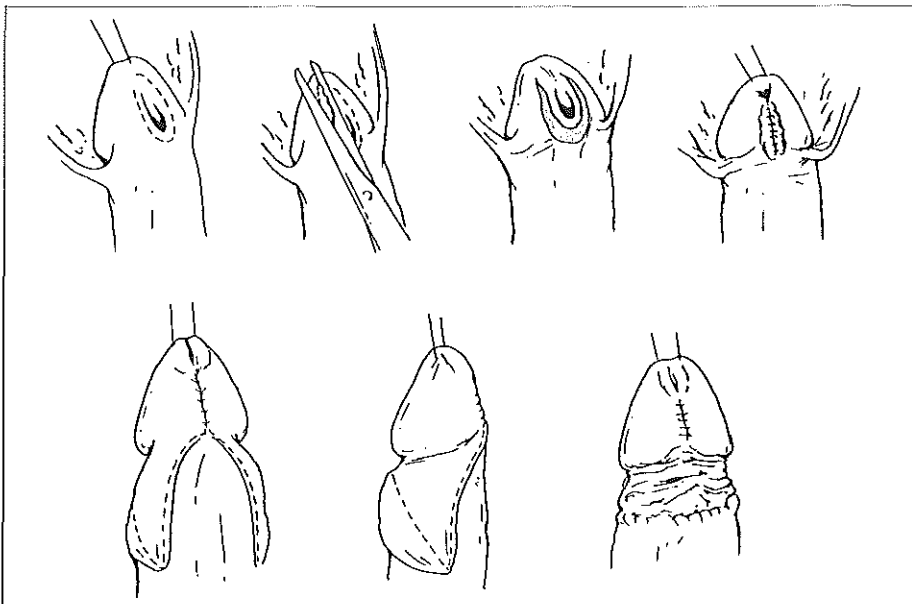


Figure 1.8. Schematic drawing of the GAP for glanular and coronal hypospadias

used to cover the anastomosis to further reduce the likelihood of fistula formation (see figure 1.8).

*Urethral advancement.*²⁹ Urethral advancement is a one-stage procedure suitable for patients with distal hypospadias and a urethra of sufficient length. The urethra with the corpus spongiosum are separated from the corpora cavernosa to the base of the penis. The urethra is pulled through a created subepithelial tunnel of the glans leaving a terminal intraglanular meatus (see figure 1.9).

*TPIF (Transverse Preputial Island Flap) repair.*³⁰ The TPIF technique is a one-stage repair for severe proximal hypospadias with a penile curvature. After the chordee is released and the dorsal and ventral surfaces of the prepuce are separated, the neo-urethra is created through tubularizing the ventral surface of the prepuce. Next, the urethra is pulled through the tunneled glans creating a terminal intraglanular meatus and the proximal part of the neo-urethra is anastomosed with the dystopic urethral orifice. Rather than a tunneling procedure, the glans may be split in the midline, leaving a triangular strip of mucosa to which the neo-urethra is sutured. All suture lines are

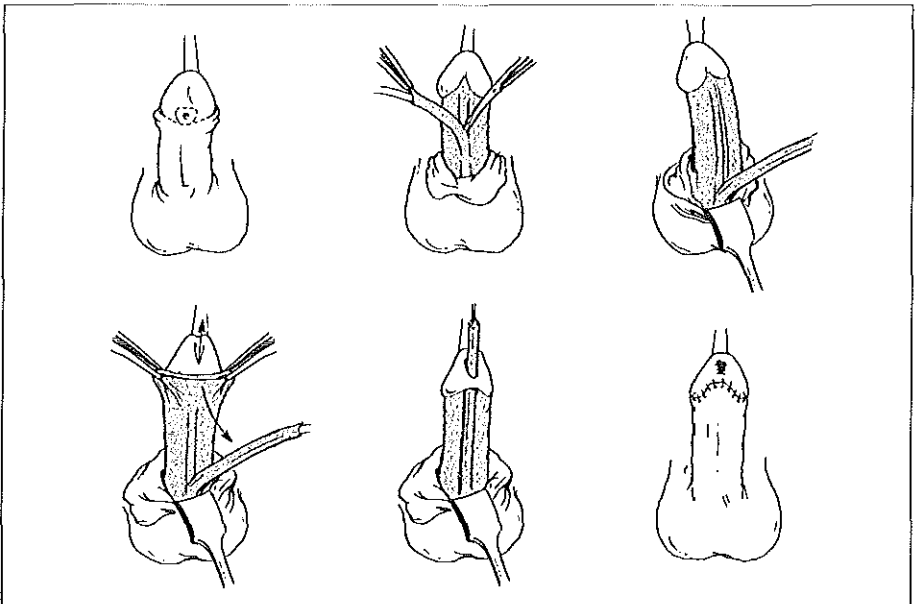


Figure 1.9. Schematic drawing of the urethral advancement repair for distal hypospadias

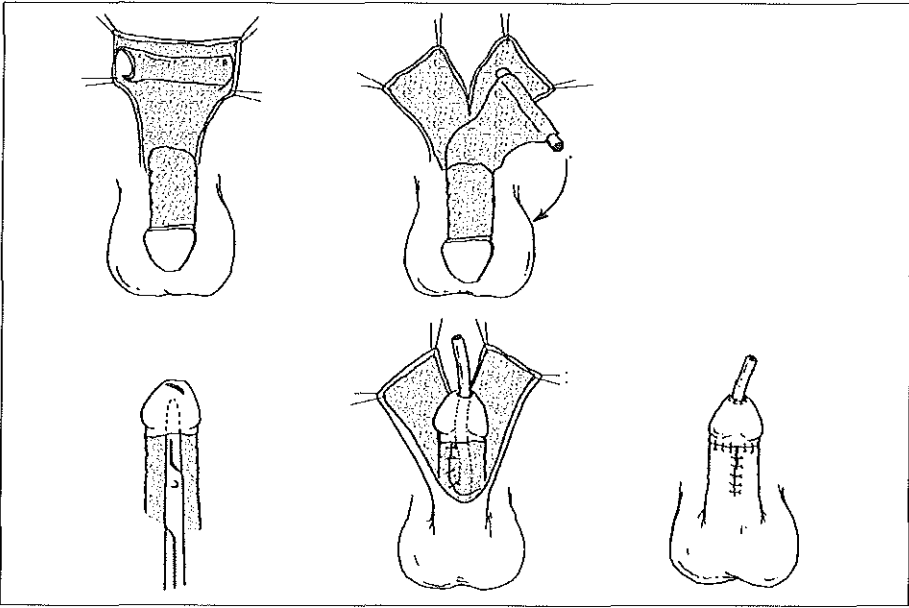


Figure 1.10. Schematic drawing of the TPIF for proximal hypospadias

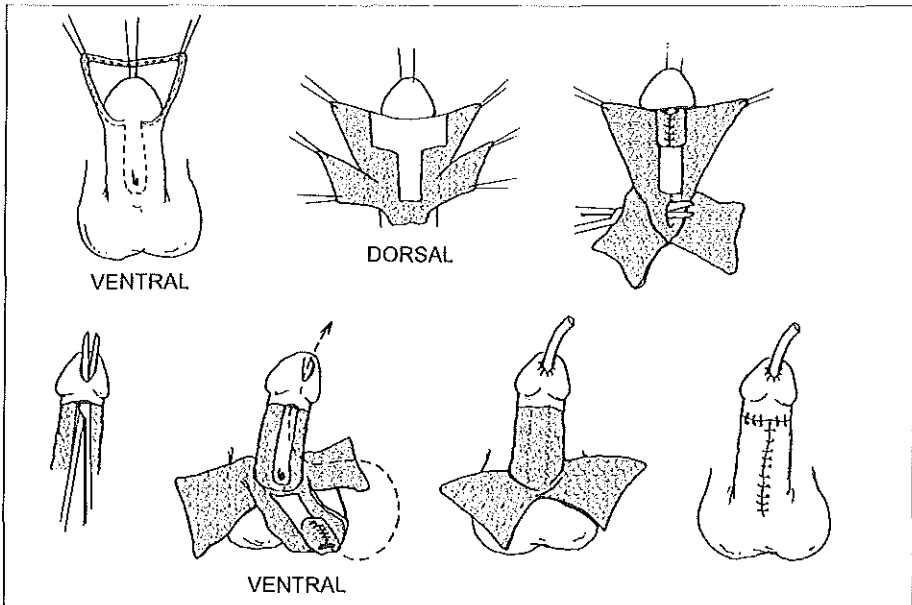


Figure 1.11. Schematic drawing of the OPIF repair for proximal hypospadias

covered with a second layer. Next, the wound is closed with the dorsal surface of the prepuce (see figure 1.10). Since the blood supply to the prepuce is very delicate, some surgeons prefer to use a so-called doublefaced island flap, leaving both outer and inner surface of the prepuce attached. However, cosmetically this latter technique is inferior.

*OPIF (Onlay Preputial Island Flap) repair.*³¹ The OPIF procedure is a one-stage repair for proximal hypospadias in which the urethral plate is mobilized without dividing it, contrary to the TPIF. A skin flap on the *dorsal side* of the prepuce (rather than the ventral side) is created and is put to the ventral side of the penis by making a buttonhole at the base of the flap. Next, the skin flap is layed on the urethral plate and sutured on both sides. The distal part of the urethra, which is tubularized, is pulled through the tunneled glans creating a terminal intraglanular meatus. The wound is closed with penile skin and prepuce (see figure 1.11).

1.6 REFERENCES

1. Blyth, B., & Duckett, J. W. Jr. (1991). Gonadal differentiation: A review of the physiological process and influencing factors based on recent experimental evidence. *Journal of Urology*, 145, 689-694.
2. Wilson, J. D., Griffin, J. E., & Russell, D. W. (1993). Steroid 5 α -reductase 2 deficiency. *Endocrine Reviews*, 14 (5), 577-593.
3. Altemus, A. R., & Hutchins, G. M. (1991). Development of the human anterior urethra. *Journal of Urology*, 146, 1085-1093.
4. Levitt, S. B., & Reda, E. F. (1988). Hypospadias. *Pediatric Annals*, 17 (1), 48-57.
5. Stoll, C., Alembik, Y., Roth, M. P., & Dott, B. (1990). Genetic and environmental factors in hypospadias. *Journal of Medical Genetics*, 27, 559-563.
6. Calzolari, E., Contiero, M. R., Roncarati, E., Mattiuz, P. L., & Volpato, S. (1986). Aetiological factors in hypospadias. *Journal of Medical Genetics*, 23, 333-337.
7. Rohatgi, M., Menon, P. S. N., Verma, I. C., & Iyengar, J. K. (1987). The presence of intersexuality in patients with advanced hypospadias and undescended gonads. *Journal of Urology*, 137, 263-267.
8. Batch, J. A., Evans, B. A., Hughes, I. A., & Patterson, M. N. (1993). Mutations of the androgen receptor gene identified in perineal hypospadias. *Journal of Medical Genetics*, 30 (3), 198-201.

9. Batch, J. A., Williams, D. M., Davies, H. R., Brown, B. D., Evans, B. A., Hughes, I. A., & Patterson, M. N. (1992). Role of the androgen receptor in male sexual differentiation. *Hormone Research*, 38 (5-6), 226-229.
10. Gearhart, J. P., Donohoue, P. A., Brown, T. R., Walsh, P. C., & Berkovitz, G. D. (1990). Endocrine evaluation of adults with mild hypospadias. *Journal of Urology*, 144, 274-277.
11. Shima, H., Ikoma, F., Yabumoto, H., Mori, M., Satoh, Y., Terakawa, T., & Fukuchi, M. (1986). Gonadotropin and testosterone response in prepubertal boys with hypospadias. *Journal of Urology*, 135, 539-542.
12. Terakawa, T., Shima, H., Yabumoto, H., Koyama, K., & Ikoma, F. (1990). Androgen receptor levels in patients with isolated hypospadias. *Acta Endocrinologica (Copenh)*, 123, 24-29.
13. Gearhart, J. P., Linhard, H. R., Berkovitz, G. D., Jeffs, R. D., & Brown, T. R. (1988). Androgen receptor levels and 5 α -reductase activities in preputial skin and chordee tissue of boys with isolated hypospadias. *Journal of Urology*, 140, 1243-1246.
14. Eil, C., Crawford, J. D., Donahoe, P. K., Johnsonbaugh, R. E., & Loriaux, D. L. (1984). Fibroblast androgen receptors in patients with genitourinary anomalies. *Journal of Andrology*, 5, 313-320.
15. Evans, B. A., William, D. M., & Hughes, I. A. (1991). Normal postnatal androgen production and action in isolated micropenis and isolated hypospadias. *Archives of Disease in Childhood*, 66 (9), 1033-1036.
16. Van der Meulen, J. C. (1985). Hypospadias. In F. K. Muir (Ed.), *Current operative surgery: Plastic and reconstructive* (pp. 119-146). London: Ballière and Tindall.
17. De Vries, J. D. M. (1986). *Hypospadias repair with the transverse inner preputial island flap technique*. University thesis. Nijmegen, The Netherlands: Catholic University of Nijmegen.
18. Wood-Smith, D. (1964). Hypospadias: Some historical aspects and the evolution of methods of treatment. In J. M. Converse (Ed.), *Reconstructive plastic surgery. Principles and procedures in correction, reconstruction and transplantation* (pp. 2010-2020). London: Saunders Company.
19. Brock, J. W. 3rd (1994). Autologous buccal mucosa graft for urethral reconstruction. *Urology*, 44 (5), 753-755.
20. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
21. Mackay, A. (1983). Hypospadias repair under the age of 1 year. *Australian & New Zealand Journal of Surgery*, 53, 449-452.
22. Manley, C. B. (1982). Elective genital surgery at one year of age: Psychological and surgical considerations. *Surgical Clinics of North America*, 62 (6), 941-953.

Chapter 1

23. Schultz, J. R., Klykylo, W. M., & Wacksman, J. (1983). Timing of elective hypospadias repair in children. *Pediatrics*, 71 (3), 342-351.
24. Van der Meulen, J. C. (1977). The correction of hypospadias. *Plastic & Reconstructive Surgery*, 59 (2), 206-215.
25. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
26. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
27. Mathieu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabalanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.
28. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.
29. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
30. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
31. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadiac [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.

CHAPTER 2

LITERATURE REVIEW AND STUDY AIMS

2.1 INTRODUCTION

Follow-up studies on psychosexual and psychosocial adjustment and satisfaction with post-operative penile appearance of hypospadias patients are scarce. In the next four paragraphs a literature review is presented of previously published studies which have investigated the long-term consequences of hypospadias surgery.

2.2 SEXUAL ADJUSTMENT AND FUNCTIONING OF HYPOSPADIAS PATIENTS

In 1966 Hynie published a descriptive paper on the sexological consequences of hypospadias, indicating the need for proper surgical correction in order to avoid adult sexual dysfunctioning.¹ Table 2.1 shows a review of previously published studies on sexual adjustment and functioning of hypospadias patients. Only four investigations^{8-10, 18} have reported actual data on the ages at which hypospadias patients had reached several sexual milestones. The studies listed in table 2.1 revealed equivocal results; some reported that sexual adjustment was not retarded^{8, 9} or that sexual functioning was quite normal,^{6, 8, 11, 12, 15} whereas other researchers reported that sexual adjustment was delayed^{10, 18} or that fewer hypospadias patients were sexually active.^{3, 4, 10, 19} The percentages of sexually active patients who reported difficulties with sexual intercourse because of small penile size, penile curvature, dyspareunia, or impaired ejaculation ranged from 0 to 19.^{2, 4, 6-11, 15, 17-20}

Table 2.1. Review of published follow-up studies on the sexual adjustment and sexual functioning of hypospadias patients

Authors	Year	N	Mean age (range)	Sexual adjustment: age at first			Comparison data	Sexual experience	Sexual functioning
				Masturbation	French kiss	Sexual intercourse			
Pompino <i>et al.</i> ²	1969	164	-(6-25)	-	-	-	No	Of 20 patients (> 16 yrs), 9 had sexual intercourse experience.	Impaired ejaculation in 1, dyspareunia in 2 patients.
Farkas & Hynie ³	1970	130	-(≥18)	-	-	-	No	Of 96 patients, 31 abstained from and 25 had sexual intercourse rarely.	-
Ericsson & Von Hedenberg ⁴	1971	62	21 (≥16)	-	-	-	Yes	Of 62 patients, 38 had sexual intercourse experience, which was less than in the normal population.	Of 37 patients, 7 had sexual difficulties (smaller penile size, inability to achieve orgasm).
Heiss & Helmig ⁵	1974	201	19 (7-36)	-	-	-	No	Of 139 patients, 40 had sexual intercourse, and 70 masturbated.	-
Helmig ⁶	1974	103	20 (8-36)	-	-	-	No	Of 89 patients (≥ 14 yrs), 30 had sexual intercourse, and 55 masturbated.	Extravaginal ejaculation due to fistulas in 1 patient.
Sommerlad ⁷	1975	60	-(≥17)	-	-	-	No	Over 66% had sexual intercourse experience.	Sexual difficulties in 6 patients (smaller penile size, penile curvature, dyspareunia).

Table 2.1. (Continued)

Authors	Year	N	Mean age (range)	Sexual adjustment: age at first			Comparison data	Sexual experience	Sexual functioning
				Masturbation	French kiss	Sexual intercourse			
Avellán ⁸	1976	220	17 (6-30)	12.9 ^{a, b}	-	16.9 ^b	Yes	No retarded sexual adjustment.	Retrograde ejaculation in 3 patients.
Kenawi ⁹	1976	82	23 (18-35)	-	-	19.4 ^{c, d}	No	Of 82 patients, 63 had sexual intercourse experience. No delayed sexual adjustment.	Sexual difficulties in 10 patients (unsatisfactory erection, penile curvature, impaired ejaculation).
Berg <i>et al.</i> ¹⁰	1981	34	27 (21-34)	13.8 ^{a, c}	14.7 ^c	17.7 ^c	Yes	Retarded sexual adjustment, less extensive sexlife.	Sexual difficulties in 5 patients (weak or painful ejaculation or erection).
Maier & Tewes ¹¹	1984	118	-(19-30)	-	-	-	No	-	Sexual intercourse impossible due to penile curvature in 1 patient.
Maier ¹²	1986	188	-(19-28)	-	-	-	No	-	Patients with slight residual penile curvature complained about an unpleasant feeling of tension during the first few sexual relations.

^aFirst ejaculation. ^bMedian value. ^cMean value. ^dComputed from raw data.

Table 2.1. (Continued)

Authors	Year	N	Mean age (range)	Sexual adjustment: age at first			Comparison data	Sexual experience	Sexual functioning
				Masturbation	French kiss	Sexual intercourse			
Beretta <i>et al.</i> ¹³	1986	15	25 (≥ 19)	12-16	-	16-24	No	Of 15 patients, 10 had sexual intercourse, and 14 had masturbation experience.	-
Schwöbel <i>et al.</i> ¹⁴	1987	27	18 (16-28)	-	-	-	No	Of 27 patients, 6 patients had sexual intercourse experience.	-
Leuthardt & Morger ¹⁵	1987	22	24 (18-31)	-	-	-	No	Of 22 patients, 19 patients had sexual intercourse experience.	Dyspareunia in 1 patient.
Cracco <i>et al.</i> ¹⁶	1989	100	-(15-22)	-	-	-	No	Of 100 patients, 32 had sexual intercourse experience at the age of 17 years.	Of 32 sexually experienced patients, 22% had difficulties with sexual intercourse due to fear of failure or pain.
Schubert <i>et al.</i> ¹⁷	1989	81	-(7-47)	-	-	-	No	Of 59 patients (≥ 18 yrs), 45 had sexual intercourse experience.	Sexual difficulties in 5 patients (penile curvature, no somatic cause).

Table 2.1. (Continued)

Authors	Year	N	Mean age (range)	Sexual adjustment: age at first			Comparison data	Sexual experience	Sexual functioning
				Masturbation	French kiss	Sexual intercourse			
Bracka ¹⁸	1989	213	22 (15-24)	13.5 ^a ^b	-	17.3 ^a	No	Of 213 patients, 133 had sexual intercourse experience. Delayed sexual adjustment. Sexual drive was not diminished. A relation between sexual adjustment and severity of hypospadias, surgical procedure, and cosmetic result was found.	Sexual difficulties due to penile curvature in 12, due to impaired ejaculation in 4 patients. Other reported sexual difficulties: psychological impotence, skin shortage, phimosis, soft glans, tender scars, and meatal discomfort.
Eberle <i>et al.</i> ¹⁹	1993	42	26 (≥ 12)	-	-	-	No	The majority of patients did not have sexual intercourse. They masturbated regularly.	In 11 patients sexual abstinence was related to persistent deformities of the genitals (mainly micropenis).
Kumar & Harris ²⁰	1994	35	-(≥ 13)	-	-	-	No	Of 35 patients, 5 reported sexual intercourse, and all patients reported masturbation experience.	Patients reported no sexual problems.

^aMean value. ^bFirst ejaculation.

2.3 SATISFACTION WITH COSMETIC AND FUNCTIONAL RESULT OF SURGERY

The reported satisfaction with post-operative penile appearance of patients with hypospadias was also quite equivocal. The percentages of patients who were dissatisfied varied from 0 to 72.^{6, 7, 9-12, 14, 15, 18-20} The most mentioned motives for dissatisfaction were small penile size, residual penile curvature, skin redundancy, too proximal meatus, and flattened glanular shape.^{3, 4, 6, 7, 9, 10, 15, 18, 19} In addition to dissatisfaction with penile appearance, a major complaint of hypospadias patients was spraying of urine.^{5, 7, 10, 18, 19}

2.4 PENILE APPEARANCE IN RELATION TO SELF-CONSCIOUSNESS AND SEXUAL INHIBITIONS

Farkas and Hynie found that 58% of 96 adult hypospadias patients aged 18 years and older abstained from sexual intercourse.³ They noted that "the main factor preventing patients from developing normal sexual relationships appeared to be a feeling of inadequacy and inferiority due to fixed impressions of abnormality." However, they failed to find any anatomic differences between patients having regular sexual intercourse and patients abstaining from sexual activity.³ Although no detailed data were presented, another study on 119 hypospadias patients between 12 and 37 years of age indicated that they were self-conscious about their penile appearance with particular intensity in relation to the opposite sex.²¹ Most of them feared making contact with girls for fear of failure during possible sexual intercourse and consequently preferred not to take any interest in the female sex at all. Berg and colleagues¹⁰ suggested that the differences in sexual adjustment between 34 adult hypospadias patients aged 21 to 34 years and 36 age-matched comparison subjects were of a psychological rather than of a physical nature, since they could not find differences in biological sexual maturation. This supposition was supported by later studies. Beretta and co-workers¹³ reported that the retarded sexual adjustment of 15 adult hypospadias patients (aged ≥ 19 yrs) was related to inferiority feelings and fear of failure. Schubert and associates¹⁷ found that 18.5% of 59 adult hypospadias patients aged 18 to 47 years suffered from inhibitions in sexual life. Cracco and collaborators¹⁶

reported that 22% of 32 sexually experienced hypospadias patients between 15 and 22 years of age suffered from fear of failure or pain during sexual intercourse. More recently, it was found in a study with 42 patients with proximal hypospadias (aged ≥ 12 yrs) that persistent deformities of the genitals (predominantly micropenis) were important reasons for abstaining from sexual intercourse in the patients aged 17 years and older.¹⁹

Self-consciousness of hypospadias patients about their penile appearance may also affect social life, for instance during changing publicly after sport activities. Sommerlad⁷ found that about one third of 60 patients avoided changing in public as a result of self-consciousness about their penis. Another study from a European country where circumcision is *uncommon* reported that the circumcised appearance of the penis was a motive for embarrassment during changing in public.⁶ The results of a recent study by Kumar and Harris²⁰ differed from the above mentioned studies; they found that 80% of 35 patients considered their penis to be normal looking and felt confident to use public toilets and communal changing rooms.

2.5 PSYCHOSEXUAL AND PSYCHOSOCIAL ADJUSTMENT OF HYPOSPADIAS PATIENTS

Table 2.2 shows a review of investigations which have focused on psychosocial and psychosexual consequences of hypospadias surgery. All studies indicated that hypospadias patients are at risk for developing psychosocial or psychosexual problems. In a study with 11 boys with hypospadias between 2 and 11 years of age, Robertson and Walker²² reported that pre-operatively these boys had anxieties which were related to their penis. Such anxieties did not exist in 11 comparison boys with a cleft palate; these boys presumably had other anxieties. Caution must be taken with interpreting these results, since the number of patients was small and the presence of anxieties was inferred by the investigators, scoring the occurrence of phallic symbols through a projective drawing test.

In a study of 3 adolescent boys (aged 14, 14½, and 15½ yrs) who had undergone surgery for diverse urogenital anomalies, all three demonstrated feelings of incompetence, inadequacy, and fears about their futures in terms

Table 2.2. Review of published follow-up studies on the psychosexual and psychosocial adjustment of hypospadias patients

Authors	Year	N	Mean age (range)	Comparison data	Research objective	Instruments	Results
Robertson & Walker ²²	1975	11	- (2-11)	Yes	Anxiety reactions prior to surgery.	Robertson Auditory Projective Test.	Parents had anxieties about the future maleness of their son with hypospadias, whereas the children had anxieties about urinating and separation. In cases of hypospadias, families had a secret related to the imperfect penis which was not shared with friends or relatives. The more the secret was hidden, the more anxious the family became that someone would find out the secret.
Cogan <i>et al.</i> ²³	1975	3*	-(14-16)	No	Body image, psychosexual development.	Draw a Person Test.	All 3 cases had profound problems with body image identity and sexuality. All drawings illustrated feelings of inadequacy, incompleteness, incompetence, and impotence, all together with extreme confusion with gender identity. All cases had fears and questions about their futures in terms of masculinity, health and role definition. They sought peer group acceptance but could not approach too closely for fear of discovery of their secret anatomical and sexual deficiencies.
Blotcky & Grossman ²⁴	1978	15 ^b	-(7-17)	Yes	Emotional reactions following urogenital surgery.	Interview.	An association was found between childhood genital surgery and the occurrence of emotional disturbance. A high occurrence (33%) of emotional disturbance was found. 17% of children seeking psychiatric help had undergone genital surgery during childhood. Genital surgery may have particular meaning to the child, contrary to surgery of other bodily parts.
Lepore & Kesler ²⁵	1979	10	-(2-6)	No	Post-operative behavior following hypospadias surgery.	Clinical observations.	Post-operative behavior was characterized by anger, aggression and negative interactions in stead of more typical reactions such as clinging behavior, long-term withdrawal or regression. Parental attitudes towards hypospadias surgery seemed to be different from those towards other types of surgery.

*One boy had hypospadias, one had epispadias and extrophy of the bladder, and one had bilateral ureterovesical obstruction. ^b8 boys and 7 girls with several urogenital anomalies (hypospadias, chordee, meatal stenosis, cryptorchidism, urinary tract infection).

Table 2.2. (Continued)

Authors	Year	N	Mean age (range)	Comparison data	Research objective	Instruments	Results
Kipikaša <i>et al.</i> ²¹	1979	119	24 (12-37)	No	Psychosocial adjustment.	Interview.	Many psychosocial disorders in a great number of patients. Most of them had been fearful and reticent already as children. They grew into solitary reclusive adults. By exaggerating their condition many of them tried to escape in a world of their own. The subjective experiencing of their defect was felt with particular intensity in relation to the opposite sex. Most of them feared establishing contact with girls for fear of failure during possible sexual intercourse.
Berg <i>et al.</i> ^{16, 26, 27}	1981 1982 1983	34	27 (21-34)	Yes	Psychosocial adjustment.	Semi-structured interview. Rorschach test. Intelligence test.	Retrospectively, hypospadias patients reported more childhood psychopathology; extreme shyness and enuresis had been more common. They had been more timid, isolated, and mobbed. As adults hypospadias patients reported more depression and anxiety. Although no differences in cognitive functioning were found, hypospadias patients had less qualified professions. They showed less capacity for social and emotional relations, higher hostility levels, less self-esteem, and less activity. No relation with hypospadias severity was found.
Berg & Berg ²⁸	1983	34	27 (21-34)	Yes	Gender identity. Sexual orientation.	Rorschach test. Franck Drawing Completion Test. Gough Femininity Scale.	Hypospadias patients were less secure in their maleness, but had a similar sexual orientation as comparison subjects. Hypospadias patients tended to prefer more feminine sex roles. Patients with more severe hypospadias tended to prefer more feminine sex roles than patients with less severe hypospadias. Less secure masculine gender identity was more common among patients with less severe hypospadias.
Berg <i>et al.</i> ²⁹	1983	34	27 (21-34)	Yes	Relation between androgens and personality characteristics.	Semi-structured interview. Rorschach test.	Hypospadias patients had lower levels of dihydrotestosterone than comparison subjects. High testosterone levels were related to low hostility scores in hypospadias patients as well as comparison subjects. Relations with androgens and other personality characteristics could not be found.

Table 2.2. (Continued)

Authors	Year	N	Mean age (range)	Comparison data	Research objective	Instruments	Results
Sandberg <i>et al.</i> ³⁰	1989	69	- (6-10)	Yes	Behavioral/emotional problems, social competence, academic achievement, gender role behavior, and family adjustments.	Child Behavior Checklist. Child Behavior and Attitude Questionnaire. Child Game Participation Questionnaire. Impact on Family Scale.	Boys with hypospadias showed more behavioral problems: higher scores on the Internalizing scale, Schizoid or Anxious scale, and Depressed scale. Proportion of hypospadias patients who fell in the clinical range for behavioral problems did not differ from a nonclinical sample. Boys with hypospadias showed lower social competence. There were no differences in academic achievement. There was a positive correlation between hypospadias severity and CBCL scores. Boys with hypospadias tended to show increased cross-gender behavior. There was a positive correlation between the number of operations and school problems, cross-gender behavior and social competence. The impact of hypospadias on families was considerable less than the impact of chronic illness.
Purschke & Standke ³¹	1993	47	?	Yes	Psychosocial functioning.	?	In comparison with healthy children hypospadias patients showed strong feelings of inferiority and an increased sensibility, were socially less adapted and more difficult to train. There was a negative correlation between psychosocial functioning and hypospadias severity. More mental problems were noted with increasingly frequent repetition of operations. Children who were operated between 5 and 6 years of age were psychologically less strained.*

*Adapted from the abstract (Medline), since the original paper could not be obtained.

of their masculinity and health.²³ These latter findings cannot be extrapolated to patients with urogenital anomalies in general, because of the small number of patients and the fact that these patients were selected because they had problems.

In 1978, Blotcky and Grossman²⁴ reported a higher occurrence of emotional disturbance in a group of 15 children (both sexes) after urogenital surgery than in a group of 33 children after ear, nose, and throat surgery. Unfortunately, they did not report the methods used for assessing emotional disturbance and they made no distinction between boys and girls.

In 1979, Lepore and Kesler²⁵ clinically observed 10 boys with hypospadias between 2 and 6 years of age and found that they showed in the first days after surgery a distinct pattern of behavior, characterized by anger, aggression, and negative interactions, in stead of more typical reactions to hospitalizations such as clinging behavior, long-term withdrawal or regression. The reliability and validity of their research methods are uncertain, since they did not use standardized assessment techniques. Besides, they did not investigate a comparison group and the number of patients was small.

Kipikaša, Longauer, and Urbanová²¹ reported many psychosocial disorders in 119 hypospadias patients aged 12 to 37 years; most of them had been fearful and reticent already as children, they grew into solitary, reclusive, and shy adults and tried to stay away from team life. A major drawback of the latter study is that it was descriptive; no detailed data based on quantifiable standardized assessments with known psychometric properties were presented.

In the early eighties the first *comparative* studies about psychosocial and psychosexual adjustment of hypospadias patients showed that 34 adult hypospadias patients between 21 and 34 years of age had a poorer psychosocial adjustment than 36 age-matched comparison subjects treated for verified appendicitis.^{10, 26, 27} Hypospadias patients were more timid, isolated, and mobbed as children, and during adulthood they were more fearful and socially isolated, showed less self-esteem, had less capacity for interpersonal social and emotional relations, and had less qualified professions. Drawbacks of the latter studies are the inference of psychiatric symptoms by the

investigators following a semi-structured interview and a projective drawing test (Rorschach test), and the retrospective assessment of the problems during childhood.

More recently, Sandberg and co-workers³⁰ investigated 69 boys with hypospadias between 6 and 10 years of age. They found that hypospadias patients showed more behavioral problems and lower social competence than boys from a nonclinical sample. Although the investigators used questionnaires with good validity and reliability, their results cannot be extrapolated to hypospadias patients older than 10 years. Sandberg and associates³⁰ also found that hypospadias patients showed somewhat more cross-gender behavior during childhood and Berg and co-workers²⁸ reported that hypospadias patients had a less masculine gender identity during adulthood.

2.6 CONCLUDING COMMENTS

Although the previously mentioned studies seem to indicate that hypospadias patients are at risk for retarded sexual adjustment, are self-consciousness about or dissatisfied with their penile appearance, and have poorer psychosocial functioning, several difficulties impede proper interpretation of their results. (1) In most studies it was unclear how the patients were selected and what the response rates were. In other words, selection bias could have occurred. (2) Some studies included children and adolescents as well as adults, without a distinction between the two, whereas other studies investigated adults only. (3) In many studies the number of patients investigated was rather small. (4) Few studies used standardized questionnaires with known reliability and validity. (5) Few investigations used comparison data collected in a similar manner as the patient data. (6) Few studies investigated the effects of severity of hypospadias, number of operations, timing of surgery, and types of surgical procedures.

In summary, there is still a dearth of proper *empirical* knowledge about the psychosexual and psychosocial adjustment of hypospadias patients at different ages as compared to "normal" subjects.

2.7 AIMS OF THE STUDY

In the present thesis a study will be described on the psychosexual adjustment, genital perception, and psychosocial adjustment of children and adolescents as well as adults following hypospadias surgery. The study aimed to answer the following research questions:

1. Do hypospadias patients have a disturbed *psychosexual* adjustment as compared to age-matched normal subjects?
2. Do hypospadias patients have a disturbed *psychosocial* adjustment as compared to age-matched normal subjects?
3. Do hypospadias patients have a more negative genital perception than age-matched normal subjects?
4. What is the effect of various medical characteristics (i.e., severity of hypospadias, age at final surgery, number of operations, surgical procedure) on questions 1 to 3?
5. What is the effect of subjects' age on questions 1 to 3?
6. What is the effect of coping with hypospadias on questions 1 to 3?

2.8 REFERENCES

1. Hynie, J. (1966). The sexuological aspects of hypospadias. *Acta Chirurgicae Plasticae*, 8 (3), 232-235.
2. Pompino, H. J., Ziekgraf, Th., Pietschmann, J. H., & Schmidt, W. (1969). Langzeitkatamnesen von 164 Hypospadien [Long-term results of 164 hypospadias patients]. *Zeitschrift für Kinderchirurgie*, 7 (3), 519-533.
3. Farkas, L. G., & Hynie, J. (1970). Aftereffects of hypospadias repair in childhood. *Postgraduate Medicine*, 47, 103-105.
4. Ericsson, N. O., & Von Hedenberg, C. (1971). Sexualfunktion hos patienter operade för hypospadi [Sexual functioning of patients operated for hypospadias]. *Läkartidningen*, 68 (21), 2480-2484.
5. Heiss, W. H., & Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadi-Operationen [Long-term results following hypospadias surgery]. *Zeitschrift für Kinderchirurgie*, 14 (4), 445-451.
6. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadi-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, 186, 421-425.

7. Sommerlad, B. C. (1975). A long-term follow-up of hypospadias patients. *British Journal of Plastic Surgery*, 28, 324-330.
8. Avellán, L. (1976). The development of puberty, the sexual début and sexual function of hypospadiacs. *Scandinavian Journal of Plastic and Reconstructive Surgery*, 10, 29-44.
9. Kenawi, M. M. (1976). Sexual function of hypospadiacs. *British Journal of Plastic Surgery*, 47, 883-890.
10. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology*, 125, 313-317.
11. Maier, W. A., & Tewes, G. (1984). Sexual function after operations for hypospadias according to Ombrédanne. *Progress in Pediatric Surgery*, 17, 79-82.
12. Maier, W. A. (1986). Die sexuelle Funktion nach einer Harnröhrenplastik im Kindesalter nach dem Verfahren von Ombrédanne [Sexual function after Ombrédanne-urethroplasty in childhood]. *Aktuelle Urologie*, 17, 295-297.
13. Beretta, G., Mazzucchelli, S., Zanollo, A., Negri, L., & Catanzaro, F. (1986). Ipospadias: Comportamento sessuale in maschi adulti sottoposti a trattamento chirurgico correttivo in età pediatrica [Sexual aspects of men operated for hypospadias during childhood]. *Minerva Urologica e Nefrologica*, 38, 17-20.
14. Schwöbel, M. G., Sacher, P., & Stauffer, U. G. (1987). Die Dennis-Browne-Korrektur der Hypospadias: Langzeitergebnisse [The Dennis Browne method for hypospadias repair: Long term results]. *Zeitschrift für Kinderchirurgie*, 42, 157-160.
15. Leuthardt, R., & Morger, R. (1987). Langzeitergebnisse der Hypospadiasoperationen nach Ombrédanne [Long-term results after Ombrédanne's operation for hypospadias]. *Zeitschrift für Kinderchirurgie*, 42, 153-156.
16. Cracco, A., Dettin, C., Cordaro, S., Angriman, A., Donadio, P., & Belloli, G. (1989). Rilevazioni psicologiche su soggetti adulti operati in età pediatrica di ipospadia [Psychological examination of adult subjects operated for hypospadias in childhood]. *Pediatria Medica e Chirurgica*, 11, 447-450.
17. Schubert, J., Kelly, L. U., & Trinckauf, H. H. (1989). Der einfluß plastischer Korrekturmaßnahmen bei Hypospadias penis auf das Sexualverhalten im fertilen Lebensalter [The effects of hypospadias surgery on sexual functioning in adulthood]. *Zeitschrift für Urologie und Nephrologie*, 82, 121-125.
18. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
19. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, 150, 1474-1477.

20. Kumar, M. V. K., & Harris, D. L. (1994). A long term review of hypospadias repaired by split preputial flap technique (Harris). *British Journal of Plastic Surgery*, 47, 236-240.
21. Kipikaša, A., Longauer, F., & Urbanová, E. (1979). Hypospadias in relation to some clinical and psychosocial problems in adulthood. *Acta Chirurgicae Plasticae*, 21, 228-235.
22. Robertson, M., & Walker, D. (1975). Psychological factors in hypospadias repair. *Journal of Urology*, 113, 698-700.
23. Cogan, S. F., Becker, R. D., & Hofmann, A. D. (1975). Adolescent males with urogenital anomalies: Their body image and psychosexual development. *Journal of Youth and Adolescence*, 14 (4), 359-373.
24. Blotcky, M. J., & Grossman, I. (1978). Psychological implications of childhood genitourinary surgery. *Journal of the American Academy of Child Psychiatry*, 17, 488-497.
25. Lepore, A. G., & Kesler, R. W. (1979). Behavior of children undergoing hypospadias repair. *Journal of Urology*, 122, 68-70.
26. Berg, R., Berg, G., & Svensson, J. (1982). Penile malformation and mental health. A controlled psychiatric study of men operated for hypospadias in childhood. *Acta Psychiatrica Scandinavica*, 66, 398-416.
27. Berg, G., & Berg, R. (1983). Castration complex. Evidence from men operated for hypospadias. *Acta Psychiatrica Scandinavica*, 68, 143-153.
28. Berg, R., & Berg, G. (1983). Penile malformation, gender identity and sexual orientation. *Acta Psychiatrica Scandinavica*, 68, 154-166.
29. Berg, R., Berg, G., Edman, G., Svensson, J., & Åström, G. (1983). Androgens and personality in normal men and men operated for hypospadias in childhood. *Acta Psychiatrica Scandinavica*, 68, 167-177.
30. Sandberg, D. E., Meyer-Bahlburg, H. F. L., Aranoff, G. S., Sconzo, J. M., & Hensle, T. W. (1989). Boys with hypospadias: A survey of behavioral difficulties. *Journal of Pediatric Psychology*, 14 (4), 491-514.
31. Purschke, C., & Standke, M. (1993). Psychische Besonderheiten bei Knaben mit Hypospadien [Psychological characteristics of boys with hypospadias]. *Pädiatrie und Grenzgebiete*, 31 (3), 175-185.

CHAPTER 3

METHOD

3.1 SELECTION OF THE SAMPLES

3.1.1 HYOSPADIAS PATIENTS

All patients were selected from the Department of Plastic and Reconstructive Surgery at the University Hospital "Dijkzigt" and the Department of Pediatric Urology at the Sophia Children's Hospital in Rotterdam, The Netherlands. Subjects aged 9 years and older were included in the study. A distinction was made between children and adolescents (aged 9 to 18 years) and adults (aged 18 years and older), and between patients treated with ventralizing and terminalizing repairs.

Selection of patients aged 18 years and older. Between 1960 and 1990, 571 consecutive patients were treated for hypospadias with ventralizing repairs^{1, 2} at the Department of Plastic and Reconstructive Surgery at the "Dijkzigt" Hospital in Rotterdam. At the time when the patient samples were selected (December 1992), 423 patients were 18 years and older. A random sample of 100 patients was drawn from these 423 patients. Of the 100 patients who were included in the present study, 80 had distal and 20 had proximal hypospadias.

Selection of patients aged 9 to 18 years. Of the above mentioned 571 patients, 104 were between 9 and 18 years and were included in the present study. Seventy-two patients had distal and 32 had proximal hypospadias. Of all 458 patients treated for hypospadias between 1980 and 1992 at the Department of Pediatric Urology of the Sophia Children's Hospital in Rotterdam, 113 boys aged 9 to 18 years at the onset of the present study had undergone terminalizing repairs.³⁻⁸ Of these 113 boys, 86 had distal and 27 had proximal hypospadias. The latter 27 patients were all included in the present study. From the 86 patients with distal hypospadias, 33 were randomly selected and included in the study.

Thus, the target group of the study comprised 100 adult patients (ventralizing repairs) + 104 boys (ventralizing repairs) + 60 boys (terminalizing repairs) = 264 patients.

3.1.2 COMPARISON SUBJECTS

Comparison subjects treated for an inguinal hernia without concomitant surgery on the genitals (e.g., orchiopexy, circumcision) were selected from the files of the Department of Pediatric Surgery at the Sophia Children's Hospital in Rotterdam. These subjects were chosen as comparison subjects because they were also hospitalized for a surgical intervention, but not on the genitals. Therefore, differences between psychosexual adjustment, genital perception, or psychosocial adjustment of patients with hypospadias and the comparison subjects will be more likely to be related to the hypospadias and subsequent surgery on the genitals than to a surgical intervention per se.

Selection of subjects aged 18 years and older. In order to obtain at least 100 adult comparison subjects, 3 random samples (total 218 subjects) matched for current age, and age at the first operation had to be drawn from a pool of 1,305 men. A group of 102 adult comparison subjects proved to have been treated for an inguinal hernia only.

Selection of subjects aged 9 to 18 years. To obtain a comparison group of at least 164 children and adolescents treated for an inguinal hernia only, 2 random samples (total 228 subjects) matched for current age, and age at the first operation had to be drawn from a pool of 2,239 boys. Consequently, a group of 165 comparison boys remained.

3.2 PROCEDURE

After the present study was approved by the Medical Ethics Committee of the Erasmus University and the University Hospital in Rotterdam, a letter was mailed to all subjects between January 1993 and May 1994 explaining the study and asking them to participate. About one week later, they were phoned to inquire about possible participation. Subjects who could not be reached by phone received a second and, if necessary, a third letter with the request to contact the investigators. When subjects did not want to partici-

pate this was accepted, and the motive for refusal was asked. All consenting subjects were invited to come to the hospital. Subjects, who did not want to come to the hospital were visited at home, or they completed the questionnaires at home and returned them by mail. The participating subjects received a poster or a gift coupon for their co-operation.

3.3 RESPONSE

Table 3.1 shows the numbers of subjects who participated in the present study. Subjects who were mentally retarded were excluded from the target population.

3.4 SOCIOECONOMIC SAMPLE CHARACTERISTICS

Occupational status of the adult subjects and of the children and adolescents' parents was assessed and was scored on a 6-point scale, where 6 = highest status occupation.⁹ Furthermore, religion and urbanization were assessed. Urbanization was scored on a 3-point scale (1 = <20,000 inhabitants; 2 = 20,000-100,000 inhabitants; and 3 = >100,000 inhabitants). Religion of the adult subjects was scored as follows: 1 = no religion; 2 = Roman Catholic; and 3 = Protestant. Parental religion of the children and adoles-

Table 3.1. Response rates of hypospadias patients and comparison subjects

Subjects	Target population	Response	Refusers	Untraceable/ excluded subjects
<i>Adults</i>	<i>N</i>	<i>n (%)^a</i>	<i>n (%)^a</i>	<i>n</i>
Hypospadias patients	100	73 (80.2)	18 (19.8)	9
Comparison subjects	102	50 (52.1)	46 (47.9)	6
<i>Children and adolescents</i>	<i>N</i>	<i>n (%)^a</i>	<i>n (%)^a</i>	<i>n</i>
Hypospadias patients	164	116 (74.8)	39 (25.2)	9
Comparison subjects	165	88 (57.5)	65 (42.5)	12

^aAdjusted for untraceable and excluded subjects.

Chapter 3

cents was scored similarly with 4 = Islamic as additional religion. No statistically significant differences in these socioeconomic characteristics were found (see table 3.2).

3.5 STRUCTURE OF THE THESIS

Table 3.3 gives an overview of the the assessment procedures used in the present thesis. In chapters 4 to 8 of this thesis, the different assessments will

Table 3.2. Socioeconomic sample characteristics

Socioeconomic variables	Subjects aged 9 to 18 years		Subjects aged 18 to 38 years	
	Hypospadias patients (<i>N</i> = 116)	Comparison subjects (<i>N</i> = 88)	Hypospadias patients (<i>N</i> = 73)	Comparison subjects (<i>N</i> = 50)
<i>Current age (yrs)</i>				
Mean (<i>SD</i>)	14.4 (2.7)	13.9 (2.5)	25.6 (5.7)	25.3 (3.7)
<i>SES (1-6)^a</i>				
Mean (<i>SD</i>)	4.1 (1.6)	4.3 (1.6)	3.1 (1.4) ^b	3.2 (1.8) ^b
<i>Religion</i>				
No religion	42.5%	47.1%	57.7%	68.0%
Roman Catholic	23.6%	16.5%	22.5%	12.0%
Protestant	29.2%	29.4%	19.7%	20.0%
Islamic	3.8%	4.7%	-	-
Unknown	0.9%	2.4%	-	-
<i>Urbanization</i>				
< 20,000 inhabitants	33.7%	29.4%	37.0%	28.6%
20,000 - 100,000 inhabitants	42.3%	40.0%	28.8%	24.5%
> 100,000 inhabitants	24.0%	30.6%	34.2%	46.9%

Note. There were no statistically significant differences between hypospadias patients and comparison subjects (Student's *t* tests, Mann-Whitney *U* tests, chi-square tests; *p*'s > .05).

^aSocioeconomic status (1 = lowest status occupation; 6 = highest status occupation³). ^bSubjects who were not yet professionals (*N* = 36) were excluded from analysis.

Table 3.3. Assessment procedures

Research objective	Instruments		Chapter
	Children and adolescents (9 to 18 years)	Adults (18 to 38 years)	
Sexual adjustment, sexual behavior and functioning	Semi-structured interview*	Semi-structured interview*	4 & 5
Genital perception	GPS-J*	GPS*	6
Agreement between patients' and surgeon's satisfaction with surgical results	GPS, physical examination	GPS, physical examination	7
Psychosocial functioning	DPQ-J, ¹⁰ SAS-C, ¹¹ CBCL/YSR ¹²	DPQ, ¹³ COI, ¹⁴ YASR ¹⁵	8

GPS-J = Junior Genital Perception Scale; GPS = Genital Perception Scale; DPQ-J = Junior Dutch Personality Questionnaire; DPQ = Dutch Personality Questionnaire; SAS-C = Social Anxiety Scale for Children; COI = Contact with Others Inventory; CBCL = Child Behavior Checklist; YSR = Youth Self-Report; YASR = Young Adult Self-Report.

be described in detail and consequences of the findings will be discussed. The clinical consequences of the findings, some methodological considerations, and recommendations for future research will be given in chapter 9.

3.6 REFERENCES

1. Van der Meulen, J. C. (1977). The correction of hypospadias. *Plastic & Reconstructive Surgery*, 59 (2), 206-215.
2. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
3. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
4. Mathieu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabalanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.
5. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.

* For description of the questionnaires, see Appendices A₁, A₂, B₁, and B₂.

6. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
7. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
8. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadie [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.
9. Van Westerlaak, J. M., Kropman, J. A., & Collaris, J. W. M. (1975). *Beroepenklapper* [Professional index]. Nijmegen, The Netherlands: Instituut voor Toegepaste Sociologie.
10. Luteijn, F., Van Dijk, H., & Van der Ploeg, F. A. E. (1989). *Handleiding bij de Junior Nederlandse Persoonlijkheds Vragenlijst, herziene uitgave* [Manual for the Junior Dutch Personality Questionnaire, revised edition]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
11. Dekking, Y. M. (1982). *Handleiding bij de Sociale Angstschaal voor Kinderen* [Manual for the Social Anxiety Scale for Children]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
12. Achenbach, T. M. (1991). *Integrative guide for the 1991 CBCL/4-18, YSR, and TRF profiles*. Burlington, VT: University of Vermont, Department of Psychiatry.
13. Luteijn, F., Starren, J., & Van Dijk, H. (1975). *Handleiding bij de Nederlandse Persoonlijkheds Vragenlijst* [Manual for the Dutch Personality Questionnaire]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
14. Van Dam-Baggen, C. M. J., & Kraaimaat, F. W. (1987). *Handleiding bij de Inventarisatielijst Omgaan met Anderen* [Manual for the Contact with Others Inventory]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
15. Achenbach, T. M. (1990). *The Young Adult Self-Report*. Burlington, VT: University of Vermont, Department of Psychiatry.

CHAPTER 4

PSYCHOSEXUAL ADJUSTMENT OF MEN WHO UNDERWENT HYPOSPADIAS REPAIR: A NORM-RELATED STUDY*

Marc A.M. Mureau,¹ Froukje M.E. Slijper,¹ Jacques C. van der Meulen,²
Frank C. Verhulst,¹ A. Koos Slob.³

¹Department of Child & Adolescent Psychiatry, ²Department of Plastic & Reconstructive Surgery, ³Department of Endocrinology & Reproduction, Sophia Children's Hospital Rotterdam/University Hospital Rotterdam/Erasmus University Rotterdam.

ABSTRACT

Purpose: To study psychosexual adjustment, sexual functioning and genital appraisal of adult hypospadias patients.

Materials and methods: A total of 73 hypospadias patients and 50 comparison subjects received a semi-structured interview.

Results: More hypospadias patients (32.8%) than comparison subjects (12.8%) had been inhibited with seeking sexual contacts. Hypospadias patients reported a more negative genital appraisal than comparison subjects, but did not have a different sexual adjustment. Severity of hypospadias negatively affected genital appraisal. Age at final surgery positively correlated with sociosexual development. Many patients (37%) wished functional or cosmetic penile improvement.

* Accepted for publication, Journal of Urology, 1995.

Conclusions: The majority of hypospadias patients experienced a normal adult sex life. They were reluctant to seek advice for problems. We recommend to follow patients through adolescence.

4.1 INTRODUCTION

Although hypospadias is a relatively common urogenital anomaly, with a reported incidence ranging from 0.8 to 8.2 per 1,000 live male births,¹ few studies exist on its psychosexual consequences in adulthood.²⁻¹⁴ These studies do not report unequivocal results. Some authors found that sexual development was not delayed and sexual functioning was quite normal,⁴⁻⁷ whereas others reported less positive results.^{2, 3, 8, 12, 13} An example of contradictory results is the patient's satisfaction with penile appearance. Some investigators reported that all patients were satisfied with the cosmetic result,⁹⁻¹¹ while others found that 38% still felt deformed,¹² or that as much as 72% regarded their penis as abnormal.⁵

Most of these studies suffer from methodological imperfections, such as low response rates, different age ranges, and the lack of comparison data. Ericsson and Von Hedenberg,³ just as Avellán,⁶ tried to overcome the latter problem and used results of a study on sexuality in the general population of Sweden as comparison data. To our knowledge, only Berg and associates⁸ investigated a comparison group of men in a similar manner as the hypospadias patients for psychosexual and social adjustment in adulthood after (hypospadias) surgery during childhood.

Because of the contradictory results of the previous studies, and because of the scarcity of norm-related studies on psychosexual and psychosocial adjustment after hypospadias surgery, the present study was carried out. The aims of our study were: (1) to compare the psychosexual and psychosocial adjustment of children, adolescents and adults, who underwent surgery for hypospadias, with that of age-matched normal subjects; (2) to determine the impact of medical treatment of hypospadias patients on the psychosexual and psychosocial adjustment. This paper will only report the results of psychosexual adjustment of *adult* men after hypospadias surgery.

4.2 SUBJECTS AND METHODS

Patients. A random sample of 100 men was drawn from 423 consecutive patients aged 18 years and older, who underwent surgery for hypospadias between 1964 and 1990 at the University Hospital "Dijkzigt" in Rotterdam (Department of Plastic and Reconstructive Surgery).

Comparison group. A comparison group was selected of subjects who were treated for an inguinal hernia without concomitant surgery on the genitals (e.g. orchiopexy, circumcision). In order to obtain a group of at least 100 comparison subjects, 3 random samples (total 218 subjects) matched for current age, and age at the first operation had to be drawn from a pool of 1,305 men. Consequently, a group of 102 comparison subjects remained.

Instruments. A semi-structured interview with pre-formulated questions was used, including questions on the ages at which different sexual milestones were reached; sexual behavior; sexual desire; satisfaction with sexual functioning; the occurrence of sexual problems; inhibitions with seeking sexual contacts; genital appraisal; and satisfaction with the surgical result.

Frequencies of sexual behavior were scored per month. Sexual desire and satisfaction with sexual functioning were measured by means of Visual Analogue Scales (VAS). On a line of 100 mm the subjects expressed what their mean sexual desire had been (0 = very low desire; 100 = very high desire) and what the mean satisfaction with their sexual functioning had been in the 3 months preceding the interview (0 = very dissatisfied; 100 = very satisfied).

A sexual problem score was calculated by adding up the answers of 9 questions dealing with sexual problems (inhibitions during sexual activity; fear of failure; problems with sexual intercourse due to: erectile failure, a small penis or a curvature of the penis; retrograde ejaculation; painful erections/orgasms). The sexual problem score ranges from 0 to 13 (0 = no sexual problems).

Genital appraisal was assessed by asking the subjects whether they thought their penile appearance was the same as in other men, whether they were satisfied with their penile appearance, and whether they were satisfied with penile size in flaccid and in erect state. Also the motives for perceiving

a different penile appearance, or for dissatisfaction were asked. A genital appraisal score was computed by adding up the answers of these 4 questions. A score of one was attributed to the answers "yes." The genital appraisal score varies from 0 to 4 (0 = very negative genital appraisal; 4 = very positive genital appraisal). Satisfaction with the result of surgery was assessed on a 7-point scale (1 = very dissatisfied; 7 = very satisfied).

Religion was scored as follows: 1 = no religion; 2 = Roman Catholic; 3 = Protestant, and urbanization was scored on a 3-point scale (1 = <20,000 inhabitants; 2 = 20,000-100,000 inhabitants; 3 = >100,000 inhabitants). Occupational level was scored on a 6-point scale (6 = highest status occupation).¹⁵

Invitation to participate. From January 1993 to April 1994 all subjects received a letter explaining the study and asking to participate. About one week after they had received the letter, they were phoned to inquire whether or not they were willing to participate.

Statistical methods. Student's *t* tests, Mann-Whitney *U* tests (corrected for ties), stratified Wilcoxon tests, chi-square tests, and Spearman's rank correlation coefficients (corrected for ties, one-tailed probabilities) were used.¹⁶ Probabilities < .05 were accepted as significant.

4.3 RESULTS

Response. Of the 100 selected patients, 6 were untraceable, and 3 were mentally retarded. Seventy-three patients, or 80.2% (73/91), participated in our study. Of the comparison group, 5 subjects were untraceable, and one subject had died. Of the remaining 96 men, 50 (52.1%) were interviewed.

Socioeconomic characteristics. There was no significant difference in the occupational level of the subjects (Mann-Whitney *U* test, $z = -0.07$, $N = 87$, $p = .943$). Thirty-six cases were excluded from analysis because 34 subjects were not yet professionals and 2 values were missing. There was also no significant difference in religion ($\chi^2_{[2]} = 2.29$, $p = .318$) and urbanization ($\chi^2_{[2]} = 2.02$, $p = .365$). The mean age at the time of the interview was 25.6 years ($SD = 5.7$) in the patients versus 25.3 years ($SD = 3.7$) in the comparison subjects (Student's *t* test, $t(120.9) = -0.40$, $p = .688$). Because no signifi-

Psychosexual Adjustment of Adult Hypospadias Patients

Table 4.1. Mean ages in years (SD) of adult hypospadias patients (N = 73) and comparison subjects (N = 50) at which sexual milestones were reached (Student's t tests)

Mean age at:	Hypospadias patients		Comparison subjects		<i>t</i>	<i>df</i>	<i>p</i>
		<i>N</i>		<i>N</i>			
First time in love	14.0 (3.7)	69	12.6 (3.4)	50	-2.07	117	.041
First time dating	15.3 (2.2)	64	15.0 (2.6)	48	-0.67	110	.502
First time French kissing	14.8 (2.7)	67	15.1 (2.8)	48	0.56	113	.576
First time dressed necking and genital fondling	15.6 (2.7)	65	15.8 (2.6)	47	0.49	110	.626
First time undressed necking and genital fondling	17.0 (2.5)	63	16.8 (2.7)	46	-0.34	107	.738
First time sexual intercourse	17.9 (2.3)	57	17.3 (2.6)	43	-1.27	98	.206
First time masturbating	13.9 (2.0)	66	13.4 (1.7)	47	-1.39	111	.169

cant differences in the socioeconomic characteristics were found, statistical correction for these variables was not necessary.

Sexual adjustment. There were no differences in the mean ages at which sexual milestones were reached, except that the hypospadias patients fell in love for the first time somewhat later than the comparison men (see table 4.1). Of the patients, 20.5% had not yet experienced sexual intercourse, versus 12% of the comparison men; this difference is not statistically significant ($\chi^2_{[1]} = 1.53, p = .216$).

Sexual behavior and sexual functioning. From table 4.2 it is clear that no significant differences existed in sexual behavior and sexual functioning between the hypospadias patients and comparison men. Although there was no statistically significant difference in the occurrence of sexual problems, there seemed to be a discrepancy in the reported difficulties during sexual intercourse related to the original penis defect which were only reported by the patients. Three patients (5.2%) reported difficulties during sexual intercourse due to a residual curvature of their penis and 3 due to a small penis. However, the mean flaccid penile length of the latter patients (4.8 cm) did

Table 4.2. Sexual behavior and sexual functioning, mean values (SD), of adult hypospadias patients ($N = 73$) and comparison subjects ($N = 50$) (Mann-Whitney U tests, corrected for ties)

	Hypospadias		Comparison		z	p
	patients	N	subjects	N		
Number of coitus partners	4.1 (6.9)	72	3.9 (4.7)	50	-0.73	.467
Sexual activity with a partner, frequency per month	7.6 (7.4)	58	6.8 (6.3)	44	-0.22	.828
Masturbation, frequency per month	6.8 (6.9)	66	5.0 (5.3)	47	-1.09	.274
Sexual problems (0-13)	0.5 (1.2)	45	0.4 (0.6)	37	-0.76	.445
Sexual desire (0-100) ^a	64.9 (20.2)	73	69.7 (21.7)	47	-1.80	.071
Satisfaction with sexual functioning (0-100) ^a	75.8 (23.7)	73	83.7 (14.5)	50	-1.51	.130

^avisual analogue scale.

not differ significantly from those patients who did not report problems with sexual intercourse (5.8 cm; Mann-Whitney U test, $z = -1.30$, $N = 46$, $p = .195$; J.F.A. van der Werff, unpublished results). One patient reported to suffer from dyspareunia probably due to skin tethering on the ventrum of his penis.

Inhibitions with seeking sexual contacts. Of the subjects with sexual experience, more hypospadias patients (32.8%) than comparison subjects (12.8%) reported that they had been inhibited with seeking sexual contacts before they had obtained experience with genital fondling (table 4.3). This had been caused by embarrassment about the penile appearance in all 21 patients and in half of 6 comparison men. After obtaining coitus experience, still more hypospadias patients than comparison men reported such inhibitions (see table 4.3). Noteworthy is that 18 of 21 patients and 5 of 6 comparison subjects who reported inhibitions prior to their first sexual experience, had experience with sexual intercourse at the time of the interview. So, apparently these feelings of inhibition did not prevent them from getting sexual intercourse experience.

Psychosexual Adjustment of Adult Hypospadias Patients

Table 4.3. Number of adult hypospadias patients (N = 73) and comparison subjects (N = 50), who reported to have been inhibited with seeking sexual contacts (Chi-square tests)

	Hypospadias patients (N = 64)		Comparison subjects (N = 47)		χ^2	df	p
	N	%	N	%			
Prior to sexual experience: ^a							
Inhibited	21	32.8	6	12.8	5.92	1	.015
Not inhibited	43	67.2	41	87.2			

	Hypospadias patients (N = 57)		Comparison subjects (N = 44)		χ^2	df	p
	N	%	N	%			
After obtaining coitus experience:							
Never been inhibited	33	57.9	38	86.4	10.42	2	.005
Inhibitions have disappeared	1	1.8	1	2.3			
Inhibitions have diminished, have remained, or have worsened	23	40.4	5	11.4			

^aat least dressed necking and genital fondling.

Genital appraisal. Hypospadias patients reported a more negative genital appraisal (mean = 2.4, *SD* = 1.1) than the comparison men (mean = 3.2, *SD* = 0.8; Mann-Whitney *U* test, *z* = -4.43, *N* = 121, *p* < .0001). About twice as many hypospadias patients (84.7%) as comparison men (40.0%) considered their penile appearance to be different from other men ($\chi^2_{[1]} = 26.45$, *p* < .00001). More hypospadias patients (25.0%) than comparison subjects (12.0%) tended to be dissatisfied with their penile appearance ($\chi^2_{[1]} = 3.16$, *p* = .078). Table 4.4 shows the spontaneously reported motives for perceiving a different penile appearance, and for being dissatisfied with the penile appearance. The main motive in the patients (59.7%) for perceiving a different penile appearance was the absence of a prepuce. Although 20 comparison subjects (40.0%) did not consider their penis to look the same as in other

Chapter 4

Table 4.4. Motives for perceiving a different penile appearance, and for being dissatisfied with the penile appearance, reported by adult hypospadias patients ($N = 73$) and comparison subjects ($N = 50$) (More motives per subject are possible)

	Reported motives for:							
	Perceiving a different penis				Dissatisfaction			
	Hypospadias patients ($N = 72$)		Comparison subjects ($N = 50$)		Hypospadias patients ($N = 72$)		Comparison subjects ($N = 50$)	
	N	%	N	%	N	%	N	%
Circumcised appearance	43	59.7	-	-	2	2.8	-	-
Smaller penile size	11	15.3	2	4.0	8	11.1	4	8.0
Position of the meatus	8	11.1	-	-	1	1.4	-	-
Penile appearance all together	6	8.3	-	-	4	5.6	-	-
Glanular shape	5	6.9	-	-	3	4.2	-	-
Scars	4	5.6	-	-	2	2.8	-	-
Prepuce skin on ventral side of glans as a result of surgery	4	5.6	-	-	-	-	-	-
Penile color	2	2.8	-	-	3	4.2	-	-
Ventrally tilted glans	2	2.8	-	-	1	1.4	-	-
Thicker penis	1	1.4	2	4.0	-	-	-	-
Hair on shaft of the penis	-	-	-	-	1	1.4	-	-
Penile curvature	-	-	1	2.0	1	1.4	2	4.0
(Prepuce) skin redundancy	-	-	1	2.0	4	5.6	-	-
Everybody is different	-	-	10	20.0	-	-	-	-
More beautiful penile appearance	-	-	2	4.0	-	-	-	-
Unknown reason	-	-	2	4.0	-	-	1	2.0
Total subjects	61	84.7	20	40.0	18	25.0	6	12.0

men, the reported motives were not related to a penis malformation. Although the circumcised appearance of the penis was the main motive in the hypospadias patients for being self-conscious, it was not an important motive for being dissatisfied with their penile appearance. In both groups the main motive for dissatisfaction was penile size. In addition to these spontaneously reported motives for dissatisfaction (see table 4.4), all subjects were explicitly asked whether or not they were satisfied with penile size. It appeared that more patients (28, 38.4%) than comparison subjects (10, 20.0%) considered their flaccid penis to be too small ($\chi^2_{[1]} = 4.68, p = .030$). These percentages diminished to 13.7% and 4.0% respectively, for penile size in erection ($\chi^2_{[1]} = 3.17, p = .075$).

Although hypospadias patients had a more negative genital appraisal, they did not report significantly more often to have had difficulties with undressing publicly (32.9% versus 22.0% of the comparison subjects; $\chi^2_{[1]} = 1.72, p = .189$). However, most hypospadias patients (18 of 24) reported that self-consciousness about their penis was the motive for having difficulties with undressing in public, whereas most comparison men (7 of 11) reported that prudishness was the motive for not undressing in public.

Effect of the severity of hypospadias. Patients with proximal hypospadias had a more negative genital appraisal than patients with distal hypospadias (see table 4.5). The main explanation for this difference might be that more patients with proximal hypospadias than with distal hypospadias complained about a too small penis (62.5% versus 31.6%; $\chi^2_{[1]} = 5.05, p = .025$). Also, patients with proximal hypospadias reported to be more inhibited with seeking sexual contacts. Patients with proximal hypospadias did not show a different sexual adjustment and sexual behavior from men with distal hypospadias, with the exception of having a higher sexual desire (see table 4.5).

Effect of surgical procedure. Sixty-one patients (83.6%) were treated according to the Van der Meulen repair,¹⁷ and 10 patients (13.7%) according to the Byars/Browne repair.¹⁸ In 2 patients the surgical procedure used was unknown. There were no significant differences in sexual adjustment, sexual behavior, sexual problems, sexual inhibitions, genital appraisal and satisfac-

tion with the surgical result, between these two repairs, after statistical correction for the severity of hypospadias (stratified Wilcoxon tests, p 's $> .10$).

Effect of the number of operations. In half of the patients treatment was completed in 2 operations (mean = 3.2; range: 1-20). Spearman's rank correlation coefficients (r_s) were computed to investigate a possible relation be-

Table 4.5. Sexual behavior and functioning, sexual inhibitions, genital appraisal and satisfaction with the surgical result, mean values (SD), of adult hypospadias patients (N = 73), by severity of hypospadias (Mann-Whitney U tests, corrected for ties)

	Distal hypospadias (N = 57)	N	Proximal hypospadias (N = 16)	N	z	p
Number of different sexual experiences (0-6) ^a	5.3 (1.6)	56	5.6 (0.9)	16	-0.14	.887
Number of coitus partners	4.5 (7.7)	56	2.9 (2.9)	16	-0.21	.832
Sexual problems (0-13)	0.3 (0.6)	33	1.0 (2.1)	12	-0.43	.670
Sexual activity with a partner, frequency per month	7.2 (7.5)	43	8.5 (7.3)	15	-0.71	.478
Masturbation, frequency per month	6.8 (7.0)	52	6.9 (6.5)	14	-0.20	.843
Satisfaction with sexual functioning (0-100) ^b	76.4 (22.7)	57	73.5 (27.7)	16	-0.07	.947
Sexual desire (0-100) ^b	62.1 (19.6)	57	75.1 (19.4)	16	-2.16	.031
Inhibitions prior to experience with genital fondling (0-2)	0.5 (0.7)	51	1.2 (0.8)	13	-2.76	.006
Inhibitions after obtaining coitus experience (0-6)	0.9 (1.5)	44	3.0 (2.3)	14	-3.28	.001
Genital appraisal (0-4)	2.6 (1.0)	56	1.7 (1.1)	15	-2.67	.008
Satisfaction with surgical result (1-7)	5.3 (1.4)	57	4.8 (1.6)	16	-1.45	.148

^abeing in love; dating; French kissing; dressed necking and genital fondling; undressed necking and genital fondling; sexual intercourse. ^bvisual analogue scale.

Psychosexual Adjustment of Adult Hypospadias Patients

tween the number of operations and sexual adjustment. There were positive correlations between the level of being sexually inhibited before, as well as after obtaining experience with sexual intercourse, and the number of operations: $r_s = 0.31$ ($N = 70, p = .004$) and $r_s = 0.49$ ($N = 57, p < .001$) respectively. There were no statistically significant correlations between the number of operations and the other interview topics listed in table 4.5.

Effect of the age at which surgery was completed. Half of the patients had undergone their first surgery at the age of 4 years (mean = 5.2 yrs; range: 0.9-22.0 yrs). The median value for the age at which surgery was completed was 8.7 years (mean = 9.7 yrs; range: 1.2-33.5 yrs). High positive correlations were found between the age at which surgery was completed and the extent of being sexually inhibited before, as well as after obtaining sexual experience: $r_s = 0.45$ ($N = 65, p < .001$) and $r_s = 0.62$ ($N = 53, p < .001$) respectively, and a small negative correlation was found with satisfaction with sexual functioning ($r_s = -0.24, N = 66, p = .026$). Table 4.6 shows Spearman's rank correlation coefficients between the ages at which sexual milestones were

Table 4.6. Spearman's rank correlation coefficients between sexual adjustment and the age at which surgery was completed in adult hypospadias patients (N = 73)

	Age at which surgery was completed		
	r_s	N^a	$P_{statist}$
First time in love	.14	62	.141
First time dating	.01	58	.462
First time French kissing	.35	61	.003
First time dressed necking and genital fondling	.33	60	.005
First time undressed necking and genital fondling	.35	57	.004
First time sexual intercourse	.31	54	.011
First time masturbating	.23	60	.040

^aIn 7 cases the age at which surgery was completed was not known: $N_{total} = 66$.

Chapter 4

reached, and the age at which surgery was completed. The later treatment was completed, the later the patients began with making their first sexual contacts.

Wishes for improvement. Twenty-seven patients (37%) expressed the wish for a functional or cosmetic penile improvement (table 4.7). Following the interview, 5 patients (6.8%) were re-operated. For spraying of urine in 3 cases, for a fistula in 1 case, and in another case for penile skin tethering which caused dyspareunia. Noteworthy is that all of these patients stated that they would not have readily consulted a physician for their problems on their own initiative.

Table 4.7. Motives for wishing a functional or cosmetic improvement of the surgical result, reported by adult hypospadias patients (N = 73)

Reported motive for wishing improvement:	N
Too small penile size	6
Spraying of urine	4
Skin redundancy	3
Position of the meatus	3
Fistula	2
Glanular shape	2
Circumcised appearance	1
Scars	1
Penile skin tethering causing dyspareunia	1
Hair on shaft of the penis	1
Shape of the penis	1
Wanting a normal appearance of the penis	1
More intensive orgasms	1
Total number of patients	27

Counseling of hypospadias patients. After the patients had been interviewed, the opportunity was given to ask questions. Many questions concerned the incidence of hypospadias, heredity, and (in)fertility. A major complaint was the lack of proper guidance and explanation during treatment. Many patients did not know why they had been operated exactly. The majority appreciated the possibility to ask questions and to talk about their anxieties. For some patients it was the first time in their life they had ever talked so frankly about their (sex)lives. Most of the patients seemed satisfied with the explanations given.

4.4 DISCUSSION

In the present study, hypospadias patients reported more often to feel inhibited with seeking sexual contacts and reported a more negative genital appraisal than comparison subjects. These feelings did not lead to retarded sexual adjustment or to different sexual functioning. Patients with proximal hypospadias were less satisfied with their penile appearance and more inhibited with seeking sexual contacts than patients with distal hypospadias. A positive correlation was found between age at which surgery was completed and psychosexual adjustment. These findings confirm the results of earlier investigations which reported relatively normal sexual behavior and functioning in hypospadias patients.⁴⁻⁷

The rather big difference in response rate between the hypospadias patients (80.2%) and the comparison subjects (52.1%) needs some comment. The main motive for refusal by comparison men was a reluctance to talk about sexual matters. Thus, it seems likely that sexually more permissive comparison subjects participated. This could mean that the mean ages at which sexual milestones were reached by the comparison subjects were more likely underestimates, rather than overestimates.

About one third of the patients felt inhibited with seeking sexual contacts prior to their first experience with genital fondling as a result of embarrassment about their penile appearance. This corroborates other studies, which reported that 50% of the hypospadias patients experienced anxieties during

adolescence,⁵ or that 18.5% of the hypospadias patients suffered from inhibitions.¹⁴

Almost 60% of the patients felt self-conscious about the circumcised appearance of their penis. We believe that such feelings exist probably because many hypospadias patients fear that their circumcised penis reveals their hypospadias to others. In The Netherlands circumcision is uncommon among white Dutch boys. Therefore, patients (and their parents) should receive proper information and learn that also after circumcision of a normal penis, the prepuce is lacking and the glans is exposed. Thus, a circumcised hypospadiac and normal penis look very similar. This information should help the patient in accepting the appearance of his circumcised penis.

Similar to other studies,^{3, 5, 8, 13} the main reason for dissatisfaction with penile appearance in hypospadias patients was penile size. More patients with proximal than patients with distal hypospadias regarded their penis to be too small. This corroborates earlier findings by Berg and associates.⁸ The relation with severity of hypospadias is in line with the negative correlation between the degree of hypospadias and penile development.¹² Since surgeons pay much attention to reconstruct the urethra and penile curvature as perfectly as possible, they should realize that many patients worry about the size of their penis, in spite of being reasonably satisfied with the surgical result. Patients and parents should be informed that surgery will not enlarge penile size.

Although patients with proximal hypospadias had a more negative genital appraisal and felt more inhibited with seeking sexual contacts than men with distal hypospadias, they did not differ in their sexual activities. This is in contrast with the findings of Eberle and associates,¹³ who reported that most men with proximal hypospadias did not have sexual intercourse. A possible explanation for this discrepancy could be the relatively high number of patients (13/42) with sexual ambiguity (micropenis, undescended testes, no prostate and gynecomastia) in the study of Eberle and associates.¹³ Bracka¹² also reported retarded sexual adjustment of men with proximal hypospadias, but unfortunately he did not present any statistical analyses to support his findings.

A very important factor affecting psychosexual adjustment seems to be the age at which surgery was completed. The later the patients underwent surgery, the higher their inhibitions were with seeking sexual contacts, and the later they made their first sexual contacts. This latter finding was also reported by Avellán.⁶ This could be explained by a relatively large number of men who underwent final surgery at a much later age than the often recommended maximum age of 18 months.¹⁹ This supposition is supported by results from our study with 116 hypospadias patients aged 9 to 18 years (including 41 patients with one stage terminalizing repairs) who had been treated at much earlier ages (median age: 5.2 yrs), which did not show significant correlations between age at final surgery and psychosexual adjustment.²⁰ Unfortunately, from our data we are unable to provide a maximum age at which treatment should be completed, but we feel confident to suggest that treatment should be completed as early as possible, because then patients will be less inhibited with seeking sexual contacts. A study, which compares psychosexual adjustment of patients who underwent surgery between 6 and 18 months, between 18 months and 3 years, and patients who underwent surgery at a later age, while keeping variables as surgical procedure and severity of hypospadias constant, will be necessary to determine the optimal timing of hypospadias surgery.

Of our patients, 37% expressed the wish for improvement of the surgical result. Bracka¹² reported that 44% requested further surgery, while they had not sought advice on their own accord. Like other investigators,¹²⁻¹⁴ we also want to advocate the importance of following hypospadias patients into adolescence as a standard procedure, since hypospadias patients are reluctant to seek medical advice on their own initiative, even when they are experiencing considerable difficulties. On the other hand, it is reassuring to discover through the present investigation, that the vast majority of men with hypospadias experience relatively normal sexual adjustment and lead an adult sexlife that does not differ greatly from that of men with normal genitalia.

ACKNOWLEDGEMENTS

We gratefully acknowledge the collaboration of Manon Bos and Martine Biewenga, for interviewing the comparison subjects, and Jan van der Ende for his help and advice with statistical analyses. This project was financially supported by the Sophia Foundation for Medical Research.

4.5 REFERENCES

1. Levitt, S. B., & Reda, E. F. (1988). Hypospadias. *Pediatric Annals*, *17* (1), 48-57.
2. Farkas, L. G., & Hynie, J. (1970). Aftereffects of hypospadias repair in childhood. *Postgraduate Medicine*, *47*, 103-105.
3. Ericsson, N. O., & Von Hedenberg, C. (1971). Sexuallfunktioner hos patienter operade för hypospadi [Sexual functioning of patients operated for hypospadias]. *Läkartidningen*, *68* (21), 2480-2484.
4. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadi-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, *186*, 421-425.
5. Sommerlad, B. C. (1975). A long-term follow-up of hypospadias patients. *British Journal of Plastic Surgery*, *28*, 324-330.
6. Avellán, L. (1976). The development of puberty, the sexual début and sexual function of hypospadiacs. *Scandinavian Journal of Plastic and Reconstructive Surgery*, *10*, 29-44.
7. Kenawi, M. M. (1976). Sexual function of hypospadiacs. *British Journal of Plastic Surgery*, *47*, 883-890.
8. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology*, *125*, 313-317.
9. Maier, W. A., & Tewes, G. (1984). Sexual function after operations for hypospadias according to Ombrédanne. *Progress in Pediatric Surgery*, *17*, 79-82.
10. Schwöbel, M. G., Sacher, P., & Stauffer, U. G. (1987). Die Dennis-Browne-Korrektur der Hypospadi-Operation: Langzeitergebnisse [The Dennis Browne method for hypospadias repair: Long term results]. *Zeitschrift für Kinderchirurgie*, *42*, 157-160.
11. Leuthardt, R., & Morger, R. (1987). Langzeitergebnisse der Hypospadi-Operationen nach Ombrédanne [Long-term results after Ombrédanne's operation for hypospadias]. *Zeitschrift für Kinderchirurgie*, *42*, 153-156.
12. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, *42*, 251-255.
13. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, *150*, 1474-1477.

14. Schubert, J., Kelly, L. U., & Trinckauf, H. H. (1989). Der einfluß plastischer Korrekturmaßnahmen bei Hypospadias penis auf das Sexualverhalten im fertilen Lebensalter [The effects of hypospadias surgery on sexual functioning in adulthood]. *Zeitschrift für Urologie und Nephrologie*, 82, 121-125.
15. Van Westerlaak, J. M., Kropman, J. A., & Collaris, J. W. M. (1975). *Beroepenklapper* [Professional index]. Nijmegen, The Netherlands: Instituut voor Toegepaste Sociologie.
16. Altman, D. G. (1991). *Practical statistics for medical research*. London: Chapman & Hall.
17. Van der Meulen, J. C. (1977). The correction of hypospadias. *Plastic & Reconstructive Surgery*, 59 (2), 206-215.
18. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
19. Manley, C. B. (1982). Elective genital surgery at one year of age: Psychological and surgical considerations. *Surgical Clinics of North America*, 62 (6), 941-953.
20. Mureau, M. A. M., Slijper, F. M. E., Nijman, R. J. M., Van der Meulen, J. C., Verhulst, F. C., & Slob, A. K. (1995). Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study. *Journal of Urology*, in press.

CHAPTER 5

PSYCHOSEXUAL ADJUSTMENT OF CHILDREN AND ADOLESCENTS AFTER DIFFERENT TYPES OF HYPOSPADIAS SURGERY: A NORM-RELATED STUDY*

Marc A.M. Mureau,¹ Froukje M.E. Slijper,¹ Rien J.M. Nijman,² Jacques C. van der Meulen,³ Frank C. Verhulst,¹ A. Koos Slob.⁴

¹Department of Child & Adolescent Psychiatry, ²Department of Pediatric Urology, ³Department of Plastic & Reconstructive Surgery, ⁴Department of Endocrinology & Reproduction, Sophia Children's Hospital Rotterdam/ University Hospital Rotterdam/Erasmus University Rotterdam.

ABSTRACT

Purpose: To study psychosexual adjustment, sexual behavior, and genital appraisal of hypospadias patients aged 9 to 18 years.

Materials and methods: 116 hypospadias patients and 88 comparison boys received a semi-structured interview.

Results: Hypospadias patients had a more negative genital appraisal and anticipated more ridicule by a partner because of their penile appearance than comparison boys, but did not have a different sexual adjustment. No significant impact of medical treatment (surgical procedures, number of operations, age at final surgery) was found. Many hypospadias patients (38.8%) wished functional or cosmetic penile improvement.

* Accepted for publication, Journal of Urology, 1995.

Conclusions: Besides terminalizing techniques, ventralizing procedures can also be performed without negatively affecting psychosexual adjustment. Hypospadias patients were reluctant to seek advice for problems: they should be followed through adolescence.

5.1 INTRODUCTION

Studies on psychosexual adjustment of children and adolescents after hypospadias surgery are scarce. In some that included children, only adolescents and adults were asked questions concerning sexuality.^{1, 2} Avellán³ reported data on masturbation from the age of 10 years and data on sexual intercourse from the age of 13 years in a longitudinal study on the development of puberty of hypospadias patients; the data did not reveal retarded sexual development.

Interpreting the results of these studies is impeded by two major problems. Firstly, these studies included children and adolescents as well as adults. Secondly, no comparison data were used collected similarly as the patient data.

Thus, because detailed knowledge about psychosexual adjustment of hypospadias patients in childhood and adolescence is lacking, we conducted a norm-related study. Besides, we wanted to study the effects of different surgical hypospadias repairs on psychosexual adjustment.

The present study is part of a larger study in which the psychosexual adjustment of adult hypospadias patients was investigated as well. This paper will only report the results of psychosexual adjustment of *children and adolescents* who underwent surgery for hypospadias in the past.

5.2 SUBJECTS AND METHODS

Patients. Of all 571 patients treated for hypospadias between 1960 and 1990 (Department of Plastic and Reconstructive Surgery) with *ventralizing* techniques^{4, 5} which bring the meatus onto the underside of the glans or coronal sulcus,⁶ 104 were between 9 and 18 years and were included in the present study. Of all 458 patients treated for hypospadias between 1980 and 1992 (Department of Pediatric Urology), 113 were between 9 and 18 years at

Psychosexual Adjustment of Children and Adolescents with Hypospadias

Table 5.1. Number of hypospadias patients in the sample by surgical procedure and severity of hypospadias

Patients with distal hypospadias (N = 105)			Patients with proximal hypospadias (N = 59)		
Ventralizing repairs		Terminalizing repairs	Ventralizing repairs		Terminalizing repairs
Van der Meulen ⁴	Byars/Browne ⁵	Mathieu, ⁷ Koff, ⁸ GAP, ⁹ MAGPI ¹⁰	Van der Meulen ⁴	Byars/Browne ⁵	TPIF, ¹¹ Perović ¹²
27 (38)	26 (34)	21 (33)	18 (26)	4 (6)	20 (27)

Note. Numbers between brackets are target numbers.

the onset of the present study and had undergone terminalizing repairs⁷⁻¹² which split, core, or tunnel through the glans to create a true terminal glanular meatus.⁶ Of these 113 patients, 86 had distal and 27 proximal hypospadias. The latter were all included in the present study. From the 86 patients with distal hypospadias, only 33 were randomly selected to obtain approximately equal groups of patients. Thus, the total target group comprised 60 (terminalizing repairs) + 104 (ventralizing repairs) = 164 boys (table 5.1).

Comparison group. Boys who were treated for an inguinal hernia without concomitant surgery on the genitals (e.g., orchiopexy, circumcision) served as a comparison group. In order to obtain a comparison group of at least 164 boys, 2 random samples (total 228 subjects) matched for current age, and age at the first operation had to be drawn from a pool of 2,239 boys. Finally, a group of 165 boys remained.

Instruments. A semi-structured interview was used consisting of five parts with pre-formulated questions. The first part included general questions about friends, school, hobbies, and sports. The second part comprised questions on hospitalization memories. The third part assessed how the subject dealt with his condition in several social situations. Genital appraisal was assessed in the fourth part. In the last part the ages at which different sexual milestones were reached, future or present inhibitions with seeking sexual

contacts, sexual behavior and the occurrence of sexual problems were assessed.

Subjects were asked if they considered their penile appearance similar to other boys (yes or no), if they were satisfied with their penile appearance (yes or no) and if they were satisfied with penile size in flaccid and erect state (yes or no). A genital appraisal score, calculated by summing the answers of these 4 questions (yes = 1; no = 0), could range from 0 to 4 (0 = negative genital appraisal; 4 = positive genital appraisal). Motives for perceiving a different appearance, and for being dissatisfied were also asked. The extent of being afraid of ridicule by a partner because of the penile appearance was scored on a 6-point scale (1 = never afraid; 6 = always afraid).

Parental religion (1 = no religion; 2 = Roman Catholic; 3 = Protestant; 4 = Islamic/Muslim), urbanization (1 = <20,000 inhabitants; 2 = 20,000-100,000 inhabitants; 3 = >100,000 inhabitants), and parental occupational level (1 = lowest status occupation; 6 = highest status occupation) were assessed as well.¹³

Invitations to participate. From November 1993 to May 1994 all patients and comparison subjects received a letter explaining the study and asking to participate. About one week later they were phoned to inquire whether or not the parents and children would participate.

Statistical methods. Student's *t* tests, Mann-Whitney *U* tests, corrected for ties (*MWU*), Kruskal Wallis one-way ANOVAs (corrected for ties), stratified Wilcoxon tests, chi-square tests, Fischer's exact tests (one-tailed probabilities), Mantel Haenszel tests, Spearman's rank correlation coefficients (r_s , corrected for ties, one-tailed probabilities) and partial correlation coefficients (r_p , one-tailed probabilities) were used.¹⁴ Probabilities < .05 were accepted as significant.

5.3 RESULTS

Response. Eight of the hypospadias patients were untraceable and one was mentally retarded. Of the remaining 155 boys, 116 (74.8%) participated in our study (see table 5.1). Eight of the 165 comparison subjects were untraceable and 4 were mentally disabled, leaving 88 boys (57.5%) as a comparison group.

Psychosexual Adjustment of Children and Adolescents with Hypospadias

Socioeconomic characteristics. Between the patients and comparison subjects, there were no significant differences in the parents' occupational level (*MWU*, $z = -0.71$, $N = 185$), nor in the parents' religion ($\chi^2_{[3]} = 1.49$) and urbanization ($\chi^2_{[2]} = 1.07$). The mean age at the time of the interview was 14.4 years ($SD = 2.7$) in the hypospadias patients and 13.9 years ($SD = 2.5$) in the comparison boys ($t(202) = -1.20$). Because no significant differences in the socioeconomic characteristics were found, statistical controlling for these variables was not necessary.

Sexual adjustment and behavior. There were no significant differences in the mean ages at which sexual milestones were reached, with the exception that hypospadias patients tended to fall in love later than the comparison subjects (see table 5.2). Sexual behavior was similar in both groups (table 5.3).

Anxieties and inhibitions with seeking sexual contacts. As is evident from table 5.4, more hypospadias patients than comparison boys reported to anticipate to become inhibited (9 to 12 yrs), or reported to be actually inhibited (13 to 18 yrs) with seeking sexual contacts because of their penile appearance. Even at early ages boys with hypospadias were more concerned about

Table 5.2. Mean ages in years (*SD*) of hypospadias patients ($N = 116$) and comparison subjects ($N = 88$) aged 9 to 18 years, at which different sexual milestones were reached (*Student's t tests*)

Mean age at:	Hypospadias patients	<i>N</i>	Comparison subjects	<i>N</i>	<i>t</i>	<i>df</i>	<i>p</i>
First time in love	9.5 (3.2)	92	8.7 (2.6)	79	-1.84	168.3	.068
First time French kissing	13.2 (1.7)	55	12.6 (2.4)	40	-1.54	93	.128
First time dressed necking and genital fondling	14.8 (1.3)	22	14.2 (1.5)	13	-1.21	33	.235
First time undressed necking and genital fondling	15.1 (1.0)	14	15.0 (1.3)	10	-0.30	22	.769
First time sexual intercourse	15.3 (1.1)	12	14.8 (1.6)	6	-0.79	16	.440
First time masturbating	11.7 (2.6)	63	12.0 (2.3)	42	0.49	103	.629

Table 5.3. Sexual behavior, mean values (SD), of hypospadias patients ($N = 116$) and comparison subjects ($N = 88$) aged 9 to 18 years (Mann-Whitney U tests, corrected for ties)

	Hypospadias patients		Comparison subjects		z	p
		N		N		
Number of coitus partners	0.2 (1.0)	113	0.3 (1.3)	86	-0.79	.430
French kissing, frequency per month	6.1 (10.7)	52	4.5 (8.3)	39	-0.62	.536
Sexual activity with partner, frequency per month	3.2 (4.2)	19	3.3 (4.3)	9	-0.18	.853
Masturbation, frequency per month	7.2 (8.0)	63	6.6 (6.0)	45	-0.31	.759
Sexual desire (0-10) ^a	6.4 (1.7)	53	6.0 (2.0)	30	-0.92	.355
Satisfaction with sexual functioning (0-10) ^a	8.0 (1.8)	22	8.0 (1.3)	9	-0.20	.840

^a0 = very low sexual desire/very dissatisfied with sexual functioning; 10 = very high sexual desire/very satisfied with sexual functioning.

their sexuality than comparison boys; the latter less often thought about being or becoming inhibited with seeking sexual contacts (see table 5.4).

Of the sexually experienced boys (at least French kissing), hypospadias patients more often were afraid of being ridiculed by a partner because of their penile appearance than the comparison subjects ($M = 2.0$, vs. $M = 1.1$; MWU , $z = -4.06$, $N = 86$, $p < .0001$).

Genital appraisal. Boys with hypospadias reported a more negative genital appraisal than the comparison boys ($M = 2.7$, vs. $M = 3.6$; MWU , $z = -6.84$, $N = 197$, $p < .0001$). Of the hypospadias patients, about 78% considered their penile appearance to be different from other boys, versus about 13% of the comparison boys. The most reported motives for perceiving a different penile appearance were the absence of a prepuce, scars, smaller size, and glanular shape (see table 5.5). Approximately 25% of the patients stated to be dissatisfied with their penile appearance, compared to only about 5% of the comparison subjects. Noteworthy is that the circumcised appearance of the penis was no motive in any of the boys for being dissatisfied. Scars, penile

Psychosexual Adjustment of Children and Adolescents with Hypospadias

Table 5.4. Number of hypospadias patients ($N = 116$) and comparison subjects ($N = 88$) aged 9 to 18 years, who reported to anticipate to become inhibited (9 to 12 yrs) or reported to be actually inhibited (13 to 18 yrs) with seeking sexual contacts due to their penile appearance (Chi-square tests)

Subjects aged 9 to 12 years ($N = 64$)							
	Hypospadias patients ($N = 37$)		Comparison subjects ($N = 16$)		χ^2	df	p
	N	%	N	%			
Inhibited	6	16.2	-	-	9.95	2	.007
Not inhibited	20	54.1	4	25.0			
Do not know	11	29.7	12	75.0			

Subjects aged 13 to 18 years ($N = 140$)							
	Hypospadias patients ($N = 75$)		Comparison subjects ($N = 56$)		χ^2	df	p
	N	%	N	%			
Inhibited	18	24.0	1	1.8	13.78	2	.001
Not inhibited	46	61.3	40	71.4			
Do not know	11	14.7	15	26.8			

size, and glanular shape were the most frequent motives for dissatisfaction (table 5.5). All reported motives for dissatisfaction were related to the original penile defect.

Twenty-eight patients (24.6%) versus 10 comparison subjects (11.6%) considered their flaccid penis to be too small and one comparison boy found his penis too large ($\chi^2_{[2]} = 6.48, p = .039$). Significantly more comparison subjects than hypospadias patients (10 versus 4) were dissatisfied with the size of their penis in erection ($\chi^2_{[2]} = 6.04, p = .049$). This difference was caused by the fact that more comparison boys than patients (6 versus 1) judged their penis too large when erected.

Table 5.5. Motives for perceiving a different penile appearance, and for being dissatisfied with the penile appearance, reported by hypospadias patients ($N = 116$) and comparison subjects ($N = 88$) aged 9 to 18 years (More motives per subject are possible)

	Reported motives for:							
	Perceiving a different penis				Dissatisfaction			
	Hypospadias patients ($N = 113$)		Comparison subjects ($N = 85$)		Hypospadias patients ($N = 114$)		Comparison subjects ($N = 86$)	
	N	%	N	%	N	%	N	%
Circumcised appearance	54	47.8	-	-	-	-	-	-
Scars	23	20.4	-	-	12	10.5	-	-
Smaller penile size	20	17.7	4	4.7	11	9.6	2	2.3
Glanular shape	19	16.8	-	-	7	6.1	-	-
Penile appearance all together	11	9.7	3	3.5	3	2.6	-	-
Position of the meatus	7	6.2	-	-	1	0.9	-	-
Thicker penis	4	3.5	-	-	-	-	-	-
Penile color	2	1.8	-	-	1	0.9	-	-
Appearance of the meatus	2	1.8	-	-	1	0.9	-	-
Skin redundancy	2	1.8	-	-	-	-	-	-
Scrotum	1	0.9	-	-	-	-	-	-
No pubic hair	1	0.9	-	-	-	-	-	-
Spraying of urine	-	-	-	-	1	0.9	-	-
Penile curvature	-	-	-	-	1	0.9	-	-
Larger penile size	-	-	2	4.0	-	-	-	-
Veins on shaft of the penis	-	-	1	1.2	-	-	-	-
Everybody is different	-	-	1	1.2	-	-	-	-
Phimosis	-	-	-	-	-	-	1	1.2
Do not know/unknown motive	-	-	1	1.2	-	-	1	1.2
Total subjects	88	77.9 ^a	11	12.9 ^a	29	25.4 ^b	4	4.7 ^b

^aSignificant difference: $\chi^2_{(1)} = 81.82, p < .00001$. ^bSignificant difference: $\chi^2_{(1)} = 15.37, p = .00009$.

Psychosexual Adjustment of Children and Adolescents with Hypospadias

More hypospadias patients (14; 12.1%) than comparison boys (2; 2.3%) reported to have a (slight) ventral penile curvature in erection ($\chi^2_{[1]} = 6.43$, $p = .011$). Hypospadias patients with a ventral penile curvature were just as often dissatisfied ($\chi^2_{[1]} = 0.12$) or self-conscious ($\chi^2_{[1]} = 0.53$) about their penile appearance as those without a ventral curvature.

Dealing with hypospadias in social situations. More patients (49.1%) than comparison boys (34.9%) reported to hide their genitals in public lavatories due to embarrassment ($\chi^2_{[1]} = 4.06$, $p = .044$). In situations where they had to undress in public (i.e., after sport activities), more patients than comparison subjects (40.7% versus 14.1%) had received comments on the appearance of their penis ($\chi^2_{[2]} = 19.82$, $p < .00001$). Five patients (4.4%) and one comparison boy (1.2%) stated that nobody had ever seen them undressed. Of the 44 patients who ever had received a remark about their penis, 16 (36.4%) had been mobbed, mainly because of the circumcised appearance or the size of their penis. Thirty-seven patients (32.7%) had never told anyone that they had been operated on their penis; usually because of fear of ridicule.

Effect of the severity of hypospadias. Seventy-four patients (63.8%) had distal and 42 patients (36.2%) had proximal hypospadias. Since there was an overrepresentation of older patients (13 to 18 yrs) with distal hypospadias ($\chi^2_{[1]} = 7.91$, $p = .005$), comparisons of the number of patients with different sexual experiences (as in table 5.6) between patients with distal and patients with proximal hypospadias were statistically controlled for current age; these analyses revealed no significant differences (Mantel Heanszel tests). Patients with distal and proximal hypospadias also did not differ in sexual behavior (topics listed in table 5.3; *MWUs*), in genital appraisal (*MWU*, $z = -0.01$, $N = 112$), and in satisfaction with the surgical result (*MWU*, $z = -0.51$, $N = 113$). Approximately the same proportion of patients with distal (26.0%) and proximal hypospadias (22.0%) were dissatisfied with penile size ($\chi^2_{[1]} = 0.24$, $p = .628$). However, of the sexually experienced patients (at least French kissing), more patients with proximal hypospadias (7; 50.0%) than patients with distal hypospadias (7; 18.9%) reported to be inhibited with seeking sexual contacts (Fischer's exact test, $p = .033$).

Effect of surgical procedure. Seventy-five patients (64.7%) underwent a ventralizing procedure^{4, 5} and 41 (35.3%) a terminalizing procedure⁷⁻¹² for correction of the hypospadias. Table 5.6 shows that there were no differences in sexual adjustment between the two surgical procedures after statistical controlling for the severity of hypospadias. Also no significant differences were found in genital appraisal (stratified Wilcoxon test, $\chi^2_{[1]} = 0.62$) and satisfaction with the surgical result (stratified Wilcoxon test, $\chi^2_{[1]} = 2.11$) after statistical controlling for the severity of hypospadias.

Effect of the number of operations. The mean number of operations was 2.4 ($SD = 1.4$, range: 1-8). There were no statistically significant correlations between the number of operations and sexual experience and genital appraisal. A small positive correlation ($p = .074$) between fear of ridicule by a partner and a small negative correlation ($p = .086$) between satisfaction with the surgical result and the number of operations were found (see table 5.7).

Effect of the age at which surgery was completed. The mean age at the first operation was 4.0 years ($SD = 2.9$; range: 0.3-12.3 yrs) and the mean age at which surgery was completed was 6.3 years ($\bar{SD} = 3.4$; range: 1.1-13.8 yrs). Because of a positive correlation between age at which treatment was completed and current age ($r_s = 0.30$, $N = 115$, $p = .001$), correlation coeffi-

Table 5.7. Spearman's rank correlation coefficients (r_s) and partial correlation coefficients (r_p), controlling for current age, between sexual experience, fear of ridicule, genital appraisal, satisfaction with surgical result, and number of operations as well as age at which treatment was completed in hypospadias patients aged 9 to 18 years (one-tailed p -values)

	Number of operations			Age at which treatment was completed		
	r_s	N	p	r_p	df	P
Number of different sexual experiences ^a	.00	112	.500	-.06	107	.260
Fear of ridicule by a partner ^b	.20	55	.074	-.08	48	.299
Genital appraisal	-.05	111	.304	.10	107	.150
Satisfaction with surgical result	-.13	112	.086	-.01	107	.445

^adating; going steady; being in love; French kissing; necking and genital fondling, dressed; necking and genital fondling, undressed; sexual intercourse. ^bonly subjects with at least French kissing experience.

Table 5.6. Number of hypospadias patients ($N = 116$) aged 9 to 18 years with sexual experience, by surgical procedure, controlling for severity of hypospadias (Mantel Haenszel tests)

	Patients with distal hypospadias ($N = 74$)				Patients with proximal hypospadias ($N = 42$)				χ^2	df	p
	Ventralizing repairs ($N = 52$)		Terminalizing repairs ($N = 21$)		Ventralizing repairs ($N = 20$)		Terminalizing repairs ($N = 20$)				
	N	%	N	%	N	%	N	%			
Being in love	44	84.6	16	76.2	16	80.0	16	80.0	0.41	1	.521
French kissing	30	57.7	10	47.6	8	40.0	7	35.0	0.65	1	.419
Dressed necking and genital fondling	13	25.0	4	19.1	3	15.0	2	10.0	0.51	1	.477
Undressed necking and genital fondling	9	17.3	3	14.3	1	5.0	1	5.0	0.08	1	.778
Sexual intercourse	8	15.4	2	9.5	1	5.0	1	5.0	0.34	1	.562
Masturbating	35	67.3	13	61.9	9	45.0	9	45.0	0.11	1	.740

cients including age at which surgery was completed were statistically controlled for current age. No significant correlations were found between the age at which treatment was completed and sexual experience, genital appraisal, fear of being ridiculed by a partner and satisfaction with the surgical result after statistical controlling for current age (see table 5.7).

Effect of timing surgery. Surgery was performed in 20 patients (17.2%) before the age of 3 years, in 26 patients (22.4%) between the ages of 3 and 6 years, and 22 patients (19.0%) were older than 6 years when they were treated. Since there was an overrepresentation of older children (13 to 18 yrs) who were treated after the age of 6 years ($\chi^2_{[2]} = 7.17, p = .028$), statistical analyses were controlled for current age. The older patients who were treated between the ages of 3 and 6 years tended to be less satisfied with the surgical result and tended to have a more negative genital appraisal than patients who were operated at a younger or an older age (table 5.8). There was no significant difference between the sexually experienced patients (at least French kissing), who were operated before the age of 3 years, between the ages of 3 and 6 years and patients older than 6 years in the fear of being ridiculed by a partner (Kruskal Wallis one-way ANOVA, $H = 2.11, N = 31$).

Wishes for improvement. Patients who had undergone a ventralizing repair tended to express more often the wish for a cosmetic or functional penile improvement (43.1% of patients with distal and 55.0% of patients with proximal hypospadias) than patients who had undergone terminalizing repairs (28.6% of patients with distal and 30.0% of patients with proximal hypospadias), after statistical controlling for the severity of hypospadias (Mantel Heanszel test, $\chi^2_{[1]} = 3.58, p = .059$). In table 5.9 the motives for wishing penile improvement are listed by surgical procedure. The main motives for wishing improvement were penile size, circumcised appearance, and scars. Interestingly, only 3 patients who underwent a ventralizing procedure, wanted an improvement of the position of the meatus (table 5.9).

Effect of current age. Of the patients aged 9 to 12 years, 13.2% were not satisfied with their penile appearance versus 31.6% of the patients aged 13 to 18 years ($\chi^2_{[1]} = 4.53, p = .033$). A small positive correlation ($r_s = 0.26, N = 52, p = .029$) was found between current age and the extent of fear of being

Table 5.8. Sexual experience, genital appraisal and satisfaction with the surgical result, mean values (SD), by timing of hypospadias surgery, controlling for current age (Stratified Wilcoxon tests)

	Current age: 9-12 years						Current age: 13-18 years						χ^2	df	p
	Patients who were treated between:						Patients who were treated between:								
	0-3 yrs	N	3-6 yrs	N	6-14 yrs	N	0-3 yrs	N	3-6 yrs	N	6-14 yrs	N			
Number of different sexual experiences (0-7) ^a	1.4 (1.1)	8	1.0 (0.9)	12	1.7 (2.1)	3	3.5 (1.7)	10	3.5 (1.9)	13	4.5 (2.2)	19	3.96	2	.138
Genital appraisal (0-4) ^b	2.9 (0.8)	8	2.6 (0.8)	12	3.3 (0.6)	3	2.6 (1.0)	9	2.2 (1.0)	13	2.8 (1.0)	19	5.09	2	.079
Satisfaction with surgical result (1-7) ^c	5.6 (1.1)	8	5.5 (1.2)	12	6.7 (0.6)	3	5.5 (0.8)	11	4.8 (1.3)	12	5.3 (1.1)	19	4.84	2	.089

^adating; going steady; being in love; French kissing; necking and genital fondling, dressed; necking and genital fondling, undressed; sexual intercourse. ^b0 = negative genital appraisal; 4 = positive genital appraisal. ^c1 = very dissatisfied with surgical result; 7 = very satisfied with surgical result.

Chapter 5

Table 5.9. Motives for wishing a cosmetic or functional improvement, by surgical procedure, reported by hypospadias patients ($N = 116$) aged 9 to 18 years (More motives per subject possible)

	Patients who had ventralizing repairs ($N = 75$)		Patients who had terminalizing repairs ($N = 41$)		Total ($N = 116$)	
	N	%	N	%	N	%
Penile size	11	14.7	5	12.2	16	13.8
Circumcised appearance	11	14.7	2	4.9	13	11.2
Scars	9	12.0	3	7.3	12	10.3
Glanular shape	4	5.3	3	7.3	7	6.0
Penile appearance all together	5	6.7	-	-	5	4.3
Position of meatus	3	4.0	1	2.4	4	3.4
Spraying of urine	2	2.7	1	2.4	3	2.6
Penile curvature	1	1.3	1	2.4	2	1.7
Appearance of meatus	-	-	2	4.9	2	1.7
Dribbling of urine	1	1.3	-	-	1	0.9
Wetting bed	1	1.3	-	-	1	0.9
Total subjects	33	44.0	12	29.2	45	38.8

ridiculed by a partner. Of the patients aged 13 to 18 years, 36 (48.6%) wanted an improvement of the functional or cosmetic result versus 9 (23.7%) of the patients aged 9 to 12 years ($\chi^2_{(1)} = 6.51, p = .011$).

5.4 DISCUSSION

The present study shows that 9 to 18 year old boys with hypospadias did not have a retarded sexual adjustment or different sexual behavior as compared with age-matched "normal" boys. The results of the present investigation corroborate findings of studies in *adult* hypospadias patients,^{1, 3, 15, 16} which also did not show retarded sexual adjustment or different sexual behavior.

About 16% of the younger patients (9 to 12 yrs) reported that they anticipated to become inhibited, and 24% of the older patients (13 to 18 yrs) repor-

ted to be actually inhibited with seeking sexual contacts because of their penile appearance. In our previous study on adult hypospadias patients,¹⁶ one third reported to have been inhibited during seeking sexual contacts. Therefore, it seems that the more sexually mature hypospadias patients become, the more they will anticipate inhibitions.

About 80% of the hypospadias patients felt self-conscious about their penile appearance and approximately 25% of the patients was dissatisfied with their penile appearance, which corroborates findings of our study on adult hypospadias patients.¹⁶ Scars and penile size were the most reported motives for dissatisfaction and the circumcised appearance was the most reported motive for being self-conscious. There are a few possible explanations for this latter finding. Firstly, Dutch patients with hypospadias might have perceived the *loss* of foreskin as a *defect* of their penis. Secondly, it is our impression that many hypospadias patients considered their circumcised penis smaller than an uncircumcised penis. The absence of a foreskin produces optically a shorter penis. Interestingly, our results differ from an American study which reported that adolescent "normal" boys who were circumcised as a routine were more satisfied with their status than uncircumcised boys, presumably because the latter perceived themselves as being a minority.¹⁷ We assume that in the present study this latter supposition could also be one of the explanations why hypospadias patients were more self-conscious about their penile appearance than (uncircumcised) comparison subjects, since circumcision is uncommon among white Dutch boys. Therefore, especially in countries where circumcision is uncommon, parents and patients should be informed properly that after surgery the penis will have a circumcised appearance, i.e. the foreskin is lacking and the glans is permanently exposed. It is further important to inform the patients and their parents that a circumcised hypospadiac penis looks almost identical as a circumcised normal penis. There might be another possibility. Recently, a surgical procedure has been described to reconstruct the prepuce, if feasible (e.g., Kropfl, Schardt, and Fey¹⁸). Especially in countries where circumcision is uncommon, such foreskin saving surgery should be offered to those parents and patients who pre-operatively have extreme difficulties with circumcision.

One quarter of the hypospadias patients was dissatisfied with penile size. This was, similar to studies in adult hypospadias patients,^{15, 16, 19-20} a major motive for dissatisfaction. In the present study, no relation was found between dissatisfaction with penile size and severity of hypospadias, contrary to what is reported for adult patients.^{6, 16, 19}

Although patients with proximal hypospadias were more often afraid of ridicule by a partner because of their penile appearance, no difference was found in sexual activities of patients with distal and proximal hypospadias. This is similar to findings in our study on adult hypospadias patients,¹⁶ but different from Eberle and associates²⁰ who reported that most men with proximal hypospadias did not have sexual intercourse. A possible explanation for this discrepancy is the relatively high number of patients with sexual ambiguity (micropenis, undescended testes, no prostate, and gynecomastia) in the study of Eberle and associates.²⁰ Bracka⁶ also reported retarded sexual adjustment of men with proximal hypospadias, but unfortunately he did not provide statistical analyses to support his findings.

No significant correlations were found between psychosexual adjustment and the number of operations or the age at which treatment was completed. Other investigations^{3, 16} reported a positive correlation between age at which treatment was completed and sexual adjustment. The reason why in the present study no significant correlation was found could be explained by the fact that the maximum age at which treatment was finished was much lower: 13.8 years in the present study versus 30 years and older in other investigations.^{3, 16}

Older patients (13 to 18 yrs), who had undergone surgery between the ages of 3 and 6 years, tended to have a more negative genital appraisal and tended to be less satisfied with the surgical result than patients who were operated at an earlier or a later age. This finding seems to support the idea that one should not operate between the ages of 3 to 5 years,²¹ when children are too young to deal with fears that are aroused by surgery of the genitals, and when they are too old to undergo the operation unconsciously. Thus, we believe it would be best to treat hypospadias patients before the age of 3 years.

No differences were found in sexual adjustment, sexual behavior, and genital appraisal between patients operated on either by ventralizing or terminalizing procedures, except that patients who were treated with ventralizing procedures more often tended to express the wish for an improved surgical result. Bracka⁶ reported that patients who underwent ventralizing procedures showed more retarded sexual adjustment than patients who were treated with terminalizing procedures. As Sommerlad¹⁵ found in adult hypospadias patients, few patients in the present study showed concern about the position of the meatus, regardless of the type of repair. Since an important goal for hypospadias surgeons is to correct the hypospadias as perfectly as possible, a terminalizing procedure is preferred. However, the results of the present study and of our study in adult hypospadias patients¹⁶ seem to indicate that a ventralizing procedure can be performed safely (e.g., in countries with less specialized health care), without negatively affecting psychosexual adjustment.

From a psychosexual point of view, treatment of proximal hypospadias can be done safely with a two-stage procedure, since the number of operations proved to have no significant effect on psychosexual adjustment or genital appraisal.

When specifically asked for, about 39% of the patients expressed the wish for cosmetic or functional improvement of the surgical result. It is virtually impossible to ascertain how many of these patients had an objective reason for such a wish, since there does not exist a "gold standard" for an excellent surgical result. We recently found that there hardly existed any agreement between 35 patients treated with terminalizing repairs (part of the present study) and their surgeon in their respective satisfaction with the surgical result.²² Furthermore, the patients were less satisfied than their surgeon.²² We would emphasize that hypospadias surgeons should *explicitly* ask whether or not the patient is satisfied with his penile appearance. If the patient is not satisfied, the surgeon should inquire about the motive for dissatisfaction and inform the patient whether or not it is feasible to further surgically improve penile appearance.

Older patients (13-18 yrs) were less satisfied with their penile appearance than younger patients (9-12 yrs). Also, they more often expressed the wish for an improvement of the surgical result. These findings, together with the knowledge that hypospadias patients are reluctant to seek advice on their own initiative, even when they are experiencing considerable difficulties,^{6, 16} indicate that it might be worthwhile to follow hypospadias patients into adolescence. Such guidance should be offered to all patients following hypospadias surgery and ideally, it should be part of the standard therapeutic procedures of such patients.

ACKNOWLEDGEMENTS

We gratefully acknowledge the collaboration of Johan van Hattem, for interviewing the comparison boys, and Jan van der Ende for his help and advice with statistical analyses. This project was financially supported by the Sophia Foundation for Medical Research.

5.5 REFERENCES

1. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadiie-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, 186, 421-425.
2. Schubert, J., Kelly, L. U., & Trinckauf, H. H. (1989). Der einfluß plastischer Korrekturmaßnahmen bei Hypospadiia penis auf das Sexualverhalten im fertilen Lebensalter [The effects of hypospadias surgery on sexual functioning in adulthood]. *Zeitschrift für Urologie und Nephrologie*, 82, 121-125.
3. Avellán, L. (1976). The development of puberty, the sexual début and sexual function of hypospadiacs. *Scandinavian Journal of Plastic and Reconstructive Surgery*, 10, 29-44.
4. Van der Meulen, J. C. (1977). The correction of hypospadias. *Plastic & Reconstructive Surgery*, 59 (2), 206-215.
5. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
6. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
7. Mathieu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabalanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.
8. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
9. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.

Psychosexual Adjustment of Children and Adolescents with Hypospadias

10. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
11. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
12. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadie [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.
13. Van Westerlaak, J. M., Kropman, J. A., & Collaris, J. W. M. (1975). *Beroepenklapper* [Professional index]. Nijmegen, The Netherlands: Instituut voor Toegepaste Sociologie.
14. Altman, D. G. (1991). *Practical statistics for medical research*. London: Chapman & Hall.
15. Sommerlad, B. C. (1975). A long-term follow-up of hypospadias patients. *British Journal of Plastic Surgery*, 28, 324-330.
16. Mureau, M. A. M., Slijper, F. M. E., Van der Meulen, J. C., Verhulst, F. C., & Slob, A. K. (1995). Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study. *Journal of Urology*, in press.
17. Schlossberger, N. M., Turner, R. A., & Irwin, C. E. Jr. (1991). Early adolescent knowledge and attitudes about circumcision: Methods and implications for research. *Journal of Adolescent Health*, 13 (4), 293-297.
18. Kropfl, D., Schardt, M., & Fey, S. (1992). Modified meatal advancement and glanuloplasty with complete foreskin reconstruction. *European Urology*, 22 (1), 57-61.
19. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology*, 125, 313-317.
20. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, 150, 1474-1477.
21. Manley, C. B. (1982). Elective genital surgery at one year of age: Psychological and surgical considerations. *Surgical Clinics of North America*, 62 (6), 941-953.
22. Mureau, M. A. M., Slijper, F. M. E., Slob, A. K., Verhulst, F. C., & Nijman, R. J. M. (1995). Satisfaction with penile appearance following hypospadias surgery: The patients' and the surgeon's view. *Journal of Urology*, in press.

CHAPTER 6

GENITAL PERCEPTION OF CHILDREN, ADOLESCENTS, AND ADULTS OPERATED ON FOR HYPOSPADIAS: A COMPARATIVE STUDY*

Marc A.M. Mureau,^{1,2} Froukje M.E. Slijper,¹ A. Koos Slob,³
Frank C. Verhulst.¹

¹Department of Child & Adolescent Psychiatry, ²Department of Plastic & Reconstructive Surgery, ³Department of Endocrinology & Reproduction, Sophia Children's Hospital/University Hospital Rotterdam/Erasmus University Rotterdam.

ABSTRACT

Genital perception of patients operated on for hypospadias, a congenital anomaly of the penis, was studied through a standardized self-report questionnaire. Genital perception of 73 adults (18 to 38 years) and 116 children and adolescents with hypospadias (9 to 18 years) was compared with that of 50 and 88 age-matched comparison males, respectively, treated for an inguinal hernia. The relationships of patient age, coping with genital appearance, severity of hypospadias, number of operations, age at final surgery, or type of surgical treatment for hypospadias with genital perception of hypospadias patients were also investigated. Hypospadias patients had a more negative genital perception than comparison males, predominantly because they were less satisfied with size and shape of their penis and with the position of the meatus. Genital perception was not related to patient age

* Accepted for publication, Journal of Sex Research, 1995.

or type of surgical treatment for hypospadias, but was significantly more negative when penile appearance was judged to be different from that of other males and when dissatisfaction with circumcised status occurred as a result of surgery. We recommend informing parents and patients that hypospadias surgery will not enlarge penile size, and that after surgery the penis will have a circumcised appearance. In countries where circumcision is uncommon, hypospadias repairs that save the foreskin should be offered as an alternative.

6.1 INTRODUCTION

Hypospadias is a common urogenital anomaly with a reported incidence ranging from 0.8 to 8.2 per 1,000 live male births.¹ It is caused by a disturbance in the development of the urethra, resulting in an abnormal position of the urethral opening. Several degrees of hypospadias exist, characterized by a different position of the urethral meatus. In a dichotomized classification, *distal* hypospadias refers to conditions in which the position of the urethral meatus is situated on the glans penis, the coronal sulcus, or just proximal to the coronal sulcus, and *proximal* hypospadias refers to conditions in which the urethra opens at some point along the penile shaft, at the penoscrotal junction, or on the perineum. Penile curvature, penile underdevelopment, and occasionally penoscrotal transposition, cryptorchidism, as well as an enlargement of the prostatic utricle may also be observed. The latter concomitant anomalies are found more often in proximal than in distal hypospadias.² Surgical reconstruction of the urethra and straightening of the penis are necessary to ensure urinating in the standing position and unhampered adult sexual functioning. In addition to restoring function, another important goal of hypospadias surgery is the achievement of a cosmetic appearance of the penis which is as "normal" as possible.

Although more than 150 surgical techniques to correct hypospadias have been described,¹ studies on genital perception of patients with hypospadias are scarce. Plastic surgeons and pediatric urologists continuously try to improve the cosmetic surgical results to make the penis look as "natural" as possible. For instance, one effort to improve the cosmetic results was the

development of *terminalizing* surgical techniques that split, core, or tunnel through the glans to create a true terminal meatus, replacing earlier *ventralizing* surgical techniques, which bring the meatus onto the underside of the glans or corona.³ Another example of improvement of medical treatment is that surgery is performed with increasingly fewer operations at earlier ages. Whether these changes in treatments also have a positive effect on genital perception of patients with hypospadias has not been thoroughly investigated.

Studies about the satisfaction with post-operative penile appearance of patients with hypospadias showed equivocal results. The percentages of patients who were dissatisfied varied from 0 to 72.³⁻⁸ Several difficulties concern the interpretation of the results of these studies. In many studies it is unclear how the patients were selected and what the response rates were. Some studies included children and adolescents, as well as adults, whereas other researchers investigated adult patients only. In the studies that included children, no distinction was made between adults and children. To measure patients' satisfaction, some studies used satisfaction with penile appearance, whereas other investigators used satisfaction with the surgical result. These two measures of satisfaction are not necessarily the same or comparable: a patient can be satisfied with the results of surgery, although he is not satisfied with his penile appearance (e.g., penile size). Finally, none of the previously mentioned studies used *comparison* data collected in a similar manner as the patient data.

In summary, genital perception of patients with hypospadias has never been investigated systematically, although there are several reasons to speculate that such patients might develop a more negative genital perception. Factors that could play a role in the development of a more negative genital perception of patients with hypospadias are (a) perception of body stimuli, (b) stimuli from the environment in the form of comparison with others, and (c) response from others. These latter factors are derived from Belfer, Harrison, Pillemer, and Murray in an article on the development of body image⁹ and will be explained next.

6.1.1 PERCEPTION OF BODY STIMULI

Before surgery, the perception of sensory and tactile stimuli during either urinating or an erection might be disturbed. Before surgery, many boys with hypospadias are not able to direct their urine stream properly which forces them to sit down during urinating. After surgery, they are able to urinate in a standing position with the possibility of directing the urine stream actively. The sensations during erection of the penis, which is curved downward, might also differ from those after surgery when the erect penis is straight.

In the first weeks after hypospadias surgery, urinating may be very painful, and the penis is reddish, swollen, and painful. Therefore, boys with hypospadias might develop an anxiety or reluctance to urinate or touch their penis.

6.1.2 STIMULI FROM THE ENVIRONMENT IN THE FORM OF COMPARISON WITH OTHERS

Very young boys with hypospadias are unconscious of their abnormal penis. When they grow older and cognitive function develops, they gradually become aware of the different appearance of their penis as compared to others, such as their father or peers. Even after surgery, they perceive differences in penile appearance between themselves and others, because hypospadias surgery never produces a perfectly normal penile appearance. For instance, surgical hypospadias repairs usually produce a circumcised appearance of the penis. Circumcision is uncommon among Dutch boys in The Netherlands. This difference could make patients with hypospadias self-conscious about their penile appearance. Furthermore, patients with hypospadias may be different with regard to penile size. Penile underdevelopment is sometimes associated with hypospadias, especially in the severe cases.² Investigations about the long-term effects of hypospadias surgery during childhood have indicated that small penile size was a major reason for being dissatisfied with penile appearance in adult patients.^{3, 8, 10, 11}

Because most boys with hypospadias are not able to urinate in the male manner until after surgery, they refrain from demonstrating their prowess at

urinating at certain distances in competition with other boys,¹² which could lead to competence anxieties related to their penis.

6.1.3 RESPONSE FROM OTHERS

Parents of children with hypospadias have anxieties about their son's future masculine capabilities.¹² Because parents' attitudes and feelings about a deformity may affect their child's ability to cope with his body,⁹ boys with hypospadias may develop a more negative genital perception. Also the pride about their genitals, which children already may begin to develop during the second year of life when there is an increase in genital orientation and awareness,¹³ could diminish or disappear as a result of ridicule by peers.

Two additional factors that might be negatively related to genital perception of patients with hypospadias are the timing of surgery and the parents' and patients' unrealistic expectation of the cosmetic result. In many patients, surgery is performed between the ages of three and five years. At these ages, surgery of the genitals could be of particular significance for the child, in contrast to surgery of other bodily parts.^{14, 15} Boys who have been told that their penis will be "normal" after surgery could be very disappointed with the cosmetic result. The boys' wish to have a penile appearance similar to other boys is not fulfilled after correction, which might lead to disappointment about and fixation on their penile appearance.

The current study was carried out to investigate genital perception of hypospadias patients in a standardized manner, compared to that of age-matched males who were treated for an inguinal hernia only. Comparison males who previously had been circumcised or treated for cryptorchidism were excluded. Thus, differences in genital perception between patients with hypospadias and the comparison males would be more likely to be related to the hypospadias and subsequent surgery on the genitals than to a surgical intervention per se. We investigated whether (a) patients with hypospadias had a more negative genital perception than age-matched males, who did not undergo surgery on their genitals; and (b) whether genital perception of patients with hypospadias was related to current age, comparison of the genitals with others, the response from others, the severity of hypospadias,

and different surgical treatments (i.e., number of operations, age at final surgery, timing of surgery, and type of surgical procedure).

6.2 METHOD

6.2.1 SAMPLE SELECTION

Selection of patients with hypospadias. Of 571 consecutive patients who were treated for hypospadias between 1960 and 1990 with ventralizing surgical repairs^{2, 16} at the Department of Plastic and Reconstructive Surgery of the University Hospital Rotterdam, 423 patients were 18 years and older at the time of the current study. A random sample of 100 participants was drawn from these 423 adult patients. Of the previously mentioned 571 patients, 104 boys were between 9 and 18 years and were included in the current study. Of 458 consecutive patients treated for hypospadias between 1980 and 1992 at the Department of Pediatric Urology of the Sophia Children's Hospital in Rotterdam, 113 boys underwent terminalizing repairs¹⁷⁻²² and were between 9 and 18 years at the time of this study. Of these 113 boys, 86 had distal and 27 had proximal hypospadias. The latter 27 patients were all included in the current study. From the 86 patients with distal hypospadias, 33 were randomly selected and included in the study. The target group of the study comprised 100 adult patients (ventralizing repairs) + 104 boys (ventralizing repairs) + 60 boys (terminalizing repairs) = 264 patients (table 6.1).

Response. Of the 100 selected adult patients with hypospadias, 6 were untraceable, and 3 were mentally retarded. Seventy-three patients participated in our study. Of the 164 selected boys with hypospadias, 8 were untraceable and one boy was mentally retarded. Of the remaining 155 boys, 116 participated in our study (table 6.1).

Selection of comparison males. Comparison males treated for an inguinal hernia *without* concomitant surgery on the genitals (i.e., circumcision, orchiopexy) were selected from the files of Pediatric Surgery of the same medical center. To obtain at least 100 *adult* comparison men, 3 random samples (218 participants) matched for current age, and age at the first operation were

Genital Perception following Hypospadias Surgery

Table 6.1. Number of patients in the sample, by age, severity of hypospadias, and surgical procedure

Patients aged 18 years and older (<i>n</i> = 100)		
Surgical procedures	Distal hypospadias	Proximal hypospadias
Ventralizing repairs		
Van der Meulen procedure ²	49 (71)	14 (18)
Byars/Browne procedure ¹⁶	8 (9)	2 (2)
Patients aged 9 to 18 years (<i>n</i> = 164)		
Ventralizing repairs		
Van der Meulen procedure ²	27 (38)	18 (26)
Byars/Browne procedure ¹⁶	26 (34)	4 (6)
Terminalizing repairs		
Glans approximation procedure ¹⁷		
Meatoplasty and glanuloplasty incorporated ¹⁸	21 (33)	-
Urethral advancement ¹⁹		
Mathieu procedure ²⁰		
Transverse preputial island flap ²¹	-	20 (27)
Onlay preputial island flap ²²		

Note: Numbers between parentheses are target numbers.

drawn from a pool of 1,305 men. Finally, a group of 102 adult comparison men remained. To obtain a comparison group of at least 164 *children and adolescents*, 2 random samples (228 participants) matched for current age, and age at the first operation were drawn from a pool of 2,239 boys. A group of 165 comparison boys remained.

Response. Of the adult comparison men, 5 were untraceable and 1 had died. Fifty of the remaining 96 men were willing to participate. Of the 165

comparison boys, 8 were untraceable and 4 were mentally disabled. Eighty-eight comparison boys participated in the current study.

6.2.2 SOCIOECONOMIC SAMPLE CHARACTERISTICS

Occupational status of the adult men and of the children and adolescents' parents was assessed and was scored on a six-point scale, where 6 = highest status occupation.²³ Furthermore, religion and urbanization were assessed. Urbanization was scored on a three-point scale (1 = <20,000 inhabitants; 2 = 20,000-100,000 inhabitants; and 3 = >100,000 inhabitants). Religion of the adult participants was scored as follows: 1 = no religion; 2 = Roman Catholic; and 3 = Protestant. Parental religion of the children and adolescents was scored similarly with 4 = Islamic as additional religion.

6.2.3 MEASURES

Although several standardized instruments which measure body perception have been developed,²⁴ items on the perception of the genitals are usually lacking. Therefore, we developed our own two *standardized* self-report questionnaires (child/adolescent and adult version) with several items about different aspects of the genitals and the body. Items about different body aspects were included to investigate whether genital perception is related to body perception and to minimize the occurrence of embarrassment and socially desirable answers.

Genital perception scale for adults. In a questionnaire with 20 items about different aspects of the genitals and body, participants were asked to express their satisfaction with each genital or body aspect by means of a five-point scale ranging from *very dissatisfied* to *very satisfied*. For a description of the items, see tables 6.2 and 6.4 in the Results section. At the end of the questionnaire a schematic drawing of a penis was depicted (see figure 6.1).

Structure of the genital perception scale for adults. After principal component analysis with varimax rotation,²⁵ two factors (factor loadings of items ≥ 0.35) appeared to be most interpretable. By adding the raw scores of the 9 items that loaded on the first factor, a Genital Perception Score (GPS) was

calculated ranging from 9 to 45 (*very negative genital perception to very positive genital perception*). Cronbach's α of this GPS was 0.89. A Body Perception Score (BPS) was calculated by adding the 11 items of the second factor. The value of this BPS (Cronbach's $\alpha = 0.85$) can vary from 11 to 55 (*very negative body perception to very positive body perception*).

Genital perception scale for children and adolescents. We developed a questionnaire that slightly differed from the adult version, consisting of 18 items about different aspects of the body and genitals. For a description of items, except for the item "chest hair," see tables 6.3 and 6.4 in the Results section. This time we opted for a 4-point scale ranging from *very dissatisfied* to *very satisfied*. The "neutral" answer was omitted to force boys to choose between satisfied and dissatisfied (see reasoning and discussion in Van Dongen-Melman, Koot, & Verhulst²⁶). A schematic drawing of a naked boy was depicted at each left page of the questionnaire (figure 6.1).

Structure of the genital perception scale for children and adolescents. Just as was the case for the adult version, two interpretative factors could be

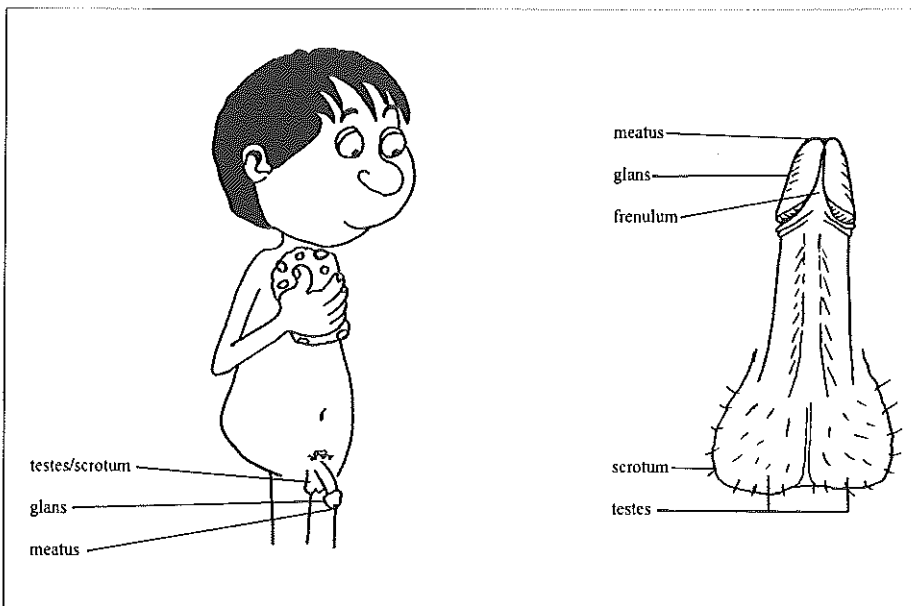


Figure 6.1. Schematic drawings used in Genital Perception Questionnaires. (Left: Genital Perception Scale for boys. Right: Genital Perception Scale for men)

discerned after principal component analysis with varimax rotation.²⁵ By adding the raw scores of the 8 items that loaded on the first factor, a GPS was calculated ranging from 8 to 32 (*very negative genital perception* to *very positive genital perception*). Cronbach's α of this GPS was 0.86. A BPS was calculated by adding the 10 items of the second factor. The value of this BPS (Cronbach's $\alpha = 0.78$) can vary from 10 to 40 (*very negative body perception* to *very positive body perception*).

6.2.4 ADDITIONAL QUESTIONS

Similarly to all other body and genital aspects, patients were asked to rate their satisfaction with their circumcised status; adults on a five-point scale and children and adolescents on a four-point scale. They were also asked if they had ever received comments on their penile appearance (yes or no) and if they considered their penile appearance to be similar to that of other males (yes or no).

6.2.5 PROCEDURE

Between January 1993 and May 1994 a letter was mailed to all participants, explaining the purpose of the study and asking them to participate. Comparison males were informed that they served as a comparison group in a study about the genital perception of patients with an anomaly of the genitals. They were informed that many questions would concern their satisfaction about the appearance of their genitals. About one week after the participants had received the letter, they were phoned to inquire about possible participation. Persons who could not be reached by phone received a second and, if necessary, a third letter with the request to contact us. All consenting participants were invited to come to the hospital. Persons who did not want to come to the hospital were visited at home, or they completed the questionnaires at home and returned them by mail. The participants received a poster or a gift coupon for their cooperation.

6.3 RESULTS

6.3.1 DATA ANALYSIS

Student's *t* tests, one-way ANOVAs, ANCOVAs, MANOVAs, and Spearman's rank correlation coefficients (r_s , corrected for ties) were used. Probabilities < .05 are reported as significant. All probabilities are two-tailed, unless otherwise indicated.

6.3.2. SOCIOECONOMIC SAMPLE CHARACTERISTICS

No statistically significant differences between patients and comparison males were found in their mean age ($M = 14.4$ yrs vs. $M = 13.9$ yrs in the

Table 6.2. Genital perception of adult men with hypospadias (N = 73) and comparison men (N = 50) aged 18 to 38 years

Satisfaction with: ^a	Hypospadias patients		Comparison men		Statistics	
	Mean (SD)	N ^b	Mean (SD)	N	<i>t</i>	<i>p</i>
Flaccid penile size	3.1 (1.2)	71	3.7 (0.9)	50	3.15	.002
Erected penile size	3.7 (0.9)	71	4.1 (0.7)	50	3.18	.002
Penile thickness	3.9 (0.9)	71	4.2 (0.6)	50	1.61	.110
Position of meatus	3.7 (0.9)	68	4.3 (0.7)	50	4.13	<.001
Glans shape	3.7 (0.8)	69	4.2 (0.7)	50	3.39	.001
Glans size	3.9 (0.9)	71	4.1 (0.6)	50	1.95	.054
Penile appearance in general	3.4 (0.9)	69	4.1 (0.7)	50	4.69	<.001
Position of penis in erection	3.8 (1.0)	69	4.0 (0.8)	50	1.56	.121
Penile color	3.8 (0.6)	71	4.1 (0.5)	50	2.65	.009
Genital Perception Score (9-45) ^c	33.0 (6.0)	68	36.9 (5.0)	50	4.01	<.001

^aSatisfaction was scored on a 5-point scale (1 = very dissatisfied; 5 = very satisfied). ^bDiscrepancy with 73 because of missing values. ^c9 = very negative genital perception; 45 = very positive genital perception.

Chapter 6

Table 6.3. Genital perception of boys with hypospadias ($N = 116$) and comparison boys ($N = 88$) aged 9 to 18 years

Satisfaction with: ^a	Hypospadias patients		Comparison boys		Statistics	
	Mean (SD)	N	Mean (SD)	N^b	t	p
Flaccid penile size	2.9 (1.1)	116	3.4 (0.7)	86	3.87	<.001
Erected penile size	3.5 (0.8)	116	3.6 (0.6)	85	1.06	.291
Penile thickness	3.3 (0.9)	116	3.5 (0.7)	86	1.99	.048
Position of meatus	3.5 (0.9)	116	3.7 (0.7)	85	1.98	.049
Glans shape	3.2 (0.9)	116	3.6 (0.6)	86	3.88	<.001
Glans size	3.3 (0.9)	116	3.6 (0.7)	86	2.62	.009
Penile appearance in general	3.0 (1.0)	116	3.5 (0.7)	86	4.89	<.001
Testes/scrotum	3.4 (0.8)	116	3.5 (0.6)	86	0.42	.672
Genital Perception Score (8-32) ^c	26.0 (5.1)	116	28.4 (4.1)	84	3.57	<.001

^aSatisfaction was scored on a 4-point scale (1 = very dissatisfied; 4 = very satisfied). ^bDiscrepancy with 88 because of missing values. ^c8 = very negative genital perception; 32 = very positive genital perception.

children and adolescents; and $M = 25.6$ yrs vs. $M = 25.3$ yrs in the adults), occupational level, religion, and urbanization. Because no differences in these socioeconomic characteristics were found, statistical controls for these variables were not performed.

6.3.3 HYOSPADIAS AND GENITAL PERCEPTION

Adult men with hypospadias had a lower GPS than comparison men, which means that they were less satisfied with their penile appearance (see table 6.2). A MANOVA with all 9 genital items as dependent variables and group (patients vs. comparison men) as independent factor indicated a significant overall group effect, $F(9, 108) = 3.17, p = .002$. Separate t tests showed that adult patients with hypospadias were significantly less satisfied

Genital Perception following Hypospadias Surgery

Table 6.4. Body perception of adult men with hypospadias (N = 73) and comparison men (N = 50) aged 18 to 38 years

Satisfaction with: ^a	Hypospadias patients		Comparison men		Statistics	
	Mean (SD)	N ^b	Mean (SD)	N	t	p
Chest hair	3.7 (1.0)	71	3.6 (0.8)	46		
Chest size	3.9 (0.8)	71	3.5 (1.0)	50		
Face	3.9 (0.7)	71	3.8 (0.7)	50		
Body hair	3.9 (0.8)	71	3.8 (0.7)	50		
Depth of voice	4.0 (0.7)	71	4.0 (0.7)	50		
Muscles	4.1 (0.7)	69	3.7 (0.9)	50		
Body appearance in general	4.1 (0.6)	69	3.8 (0.8)	50		
Legs	4.2 (0.8)	71	3.6 (1.1)	50		
Arms	4.2 (0.7)	71	3.8 (0.9)	50		
Hands	4.2 (0.7)	71	3.9 (0.8)	50		
Eyes	4.2 (0.8)	70	4.2 (0.8)	50		
Body Perception Score (11-55) ^c	44.5 (5.4)	68	41.5 (5.8)	46	-2.87	.005

^aSatisfaction was scored on a 5-point scale (1 = very dissatisfied; 5 = very satisfied). ^bDiscrepancy with 73 because of missing values. ^cDiscrepancy with 50 due to missing values. ^d11 = very negative body perception; 55 = very positive body perception.

with their penile size in flaccid as well as in erect state, penile appearance in general, glans shape, the position of the meatus, and penile color (table 6.2).

Also children and adolescents with hypospadias were less satisfied with their penile appearance than comparison boys, as indicated by a lower GPS (table 6.3). Because a MANOVA with all 8 genital items as dependent variables and group as independent factor indicated a significant overall group effect, $F(8, 191) = 3.77, p < .001$, eight separate t tests were conducted. They revealed that boys with hypospadias were less satisfied with their penile size

in flaccid state, penile appearance in general, its thickness, the size and shape of their glans, and the position of the meatus.

6.3.4 HYPOSPADIAS AND BODY PERCEPTION

Adult men with hypospadias received a significantly higher BPS than comparison men (table 6.4). A MANOVA with all 11 body items as dependent variables and group as independent factor did not show a significant overall group effect, $F(11, 102) = 1.66, p = .092$.

Children and adolescents with hypospadias did not differ in body perception from comparison boys as illustrated by a similar BPS ($M = 34.5, SD = 4.2$ vs. $M = 33.6, SD = 5.1$), $t(147.8) = -1.39$.

6.3.5 CURRENT AGE AND GENITAL AND BODY PERCEPTION

Four analyses of covariance with group (patients vs. comparison males) as a factor and current age as a covariate were performed. These analyses revealed no statistically significant effect of current age on the GPS of children and adolescents, the GPS of adults, nor on the BPS of adults, except for the BPS of children and adolescents ($\beta = -0.15$), $F(1, 191) = 4.21, p = .042$.

6.3.6 SATISFACTION WITH CIRCUMCISED STATUS AND GENITAL PERCEPTION

Adult patients with hypospadias who were satisfied with their circumcised status had a higher GPS ($M = 34.8$) than patients who were neither satisfied nor dissatisfied ($M = 30.3$) or than patients who were dissatisfied with their circumcised status, ($M = 32.9$), one-way ANOVA, $F(2, 65) = 4.41, p = .016$. Also, children and adolescents with hypospadias who were satisfied with their circumcised status had a higher GPS ($M = 27.7$) than those who were not satisfied with their circumcised status ($M = 21.7$), $t(112) = -5.56, p < .001$.

6.3.7 COMPARISON WITH OTHERS AND GENITAL PERCEPTION

Of the adult patients with hypospadias, 61 patients (84.7%), and of the children and adolescents, 88 (77.9%) considered their penile appearance to be different from other males'. Adults as well as children and adolescents with hypospadias who considered their penile appearance to be similar to other males' had a higher GPS than those who perceived a penile appearance different from other males', ($M = 37.7$ vs. $M = 32.2$ for the men), $t(66) = 2.80, p = .007$; ($M = 29.2$ vs. $M = 25.1$ for the boys), $t(111) = 3.64, p < .001$. Patients who considered their penile appearance similar to other males' received mean GPSs approximately equal to comparison males ($M = 37.7$ vs. $M = 36.9$, respectively, for the adults, and $M = 29.2$ vs. $M = 28.4$, respectively for the children and adolescents).

6.3.8 RESPONSE FROM OTHERS AND GENITAL PERCEPTION

Of the adult patients with hypospadias, 24 patients (33.3%), and of the children and adolescents, 46 (40.7%) had received comments on their penile appearance during undressing publicly (i.e., after sport activities). Adult patients who had ever received comments on their penile appearance, had a lower GPS ($M = 30.0$) than those who had never received comments ($M = 34.6$), $t(66) = -3.15, p = .002$. This was not the case for the children and adolescents with hypospadias ($M = 25.2$ vs. $M = 26.4$ respectively), $t(106) = -1.20$.

6.3.9 HYPOSPADIAS SEVERITY AND GENITAL PERCEPTION

Of the adult patients, 57 patients (78.1%) had distal hypospadias, and 16 patients (21.9%) had proximal hypospadias. Patients with proximal hypospadias had a significantly lower GPS ($M = 29.3$); they were less satisfied than were patients with distal hypospadias ($M = 34.2$), $t(66) = 3.04, p = .003$. In the children and adolescents with hypospadias, no significant difference was found in the mean GPS between the boys with distal ($M = 26.6$) and proximal hypospadias ($M = 25.1$), $t(114) = 1.52$.

6.3.10 SURGICAL TREATMENT FOR HYPOSPADIAS AND GENITAL PERCEPTION

Number of operations. The mean number of operations to correct the hypospadias was 3.2 ($SD = 3.6$, range: 1-20) in the adult patients and 2.4 ($SD = 1.4$, range:1-8) in the patients between 9 and 18 years. There was a small negative correlation between the number of operations and the GPS ($r_s = -0.28$, $N = 66$, $p = .012$, one-tailed) in the adult men with hypospadias, but there existed no such correlation in the children and adolescents ($r_s = -0.10$, $N = 115$).

Age at which surgery was completed. The mean age at which surgical hypospadias treatment was completed was 9.7 years ($SD = 6.4$, range: 1.2-33.5 yrs) in the adult men and 6.3 years ($SD = 3.4$, range: 1.1-13.8 yrs) in the children and adolescents. There was a non-significant trend in adult patients that, with increasing age at which surgery was completed, the GPS became lower ($r_s = -.18$, $N = 62$, $p = .078$, one-tailed). No such correlation was found in the children and adolescents ($r_s = 0.06$, $N = 116$).

Surgical procedure. No significant differences were found in the GPS of the adult patients treated with the Van der Meulen and the Byars/Browne repair, both ventralizing techniques, leaving the meatus at the undersurface of the glans or corona, $t(64) = -0.20$. Also, no difference emerged in the GPS of the children and adolescents treated with ventralizing or terminalizing techniques that produce an intraglanular terminal meatus, $t(114) = 0.56$. Children and adolescents treated with terminalizing techniques were equally satisfied with the position of their meatus ($M = 3.6$) as were children and adolescents treated with ventralizing techniques ($M = 3.4$), $t(106.4) = -1.05$.

6.4 DISCUSSION

From the current study it is clear that children and adolescents as well as adults with hypospadias had a more negative genital perception than age-matched comparison males, predominantly because they were less satisfied with the size and shape of their penis and the position of their meatus. Although patients with hypospadias had a more negative genital perception, they did not have a more negative body perception than age-matched comparison males. Genital perception of patients with hypospadias was not related

to surgical treatments, but it was significantly more negative when penile appearance was judged to be different from that of other males and when dissatisfaction with circumcised status occurred. The design of our study gives us reasons to have confidence in the results, because genital perception was assessed in a standardized manner; comparison males were used; the number of persons investigated was fairly large; the Cronbach's α s indicated good reliability of the instruments used; and patients who were treated with different surgical procedures were investigated.

Patients with hypospadias in the current study were less satisfied with penile size than comparison males. This corroborates earlier findings^{27, 28} and other researchers who reported that penile size was a major motive for dissatisfaction in patients with hypospadias.^{3, 8, 10, 11} Several investigators have reported data on flaccid penile length of adult hypospadias patients; the reported mean values varied from 6.0 to 9.7 cm.^{3, 29-31} However, because none of the researchers investigated a comparison group of normal males and because they used different norms for normal flaccid penile length, they reached different conclusions about whether penile underdevelopment was encountered more often among patients with hypospadias. In a recent study on 35 boys with hypospadias who are part of the current group of boys,³² we found that significantly more patients fell below the 10th percentile for stretched penile length of normal age-matched boys as reported by Schonfeld and Beebe.³³ However, we also found that patients who fell below the 10th percentile for stretched penile length were equally (dis)satisfied with their penile appearance and size as patients who fell above the 10th percentile.³² Apparently, there does not seem to exist a clear relationship between dissatisfaction with penile size or appearance and actual penile length in these boys with hypospadias.

Because surgical hypospadias repairs never affect penile size, we believe that it is important for plastic surgeons and pediatric urologists to give proper information to parents *and* patients (if they are old enough) that surgery will not enlarge the penis. Patients who post-operatively have psychological difficulties with accepting their (small) penis should be offered professional psychological or sexological help. Adolescent or adult patients with hypo-

spadias with a micropenis (< 2.5 *SDs* below the mean) should be counseled properly: they can be informed that it is possible for men with a micropenis to have a satisfactory sexual relationship.^{34, 35} If psychological or sexological counseling does not help and the patient keeps suffering from severe psychological distress, phallic reconstruction³⁶ or penile lengthening might be suggested.

A second interesting result of the current study was that dissatisfaction with the circumcised status was an important factor that was negatively associated with genital perception. There are two possible explanations for this finding. First, patients with hypospadias might have perceived the *loss* of their foreskin as a *defect* of their penis. Second, it is our impression that many patients with hypospadias, especially children, considered their circumcised penis smaller than an uncircumcised penis. This is understandable, because the absence of a foreskin produces optically a shorter penis. (It should be kept in mind that in The Netherlands circumcision is uncommon among Dutch males.) We found earlier that the circumcised appearance was the most reported motive for perceiving a different penile appearance and for being self-conscious: it was reported by 60% of the adult patients, and by 48% of the children and adolescents with hypospadias.^{27, 28} To our knowledge only two other researchers on the long-term follow-up of hypospadias patients reported that they felt embarrassed about the circumcised appearance of their penis.^{5, 29} Interestingly, our results differ from the American study by Schlossberger, Turner, and Irwin,³⁷ who reported that adolescent boys who were circumcised as a routine were more satisfied with their status than were uncircumcised boys, presumably because the latter perceived themselves as being a minority. We assume that in the current study this latter supposition could also be one explanation why patients with hypospadias were less satisfied with their penile appearance than were the uncircumcised comparison males. Researchers who compare genital perception of patients with hypospadias with that of males who were circumcised for non-medical reasons (e.g., hygiene, religion) should investigate this hypothesis.

Parents and patients should be informed properly that after surgery the penis will have a circumcised appearance; the foreskin is lacking, and the

glans is exposed permanently. We got the impression that many patients feared that the circumcised appearance could reveal their hypospadias. In other words, they should be informed that, in fact, a circumcised normal and a circumcised hypospadiac penis look very similar. This information may help patients to accept the appearance of their circumcised penis. However, there might be another possibility. Recently, a surgical procedure has been described to reconstruct the prepuce, if feasible (e.g., Kropfl, Schardt, & Fey³⁸). Especially in countries where circumcision is uncommon, such foreskin saving surgery should be offered as an alternative to the parents.

A third factor that was significantly associated with genital perception was the patients' ability to cope with their penile appearance. Patients with hypospadias who considered their penile appearance to be different from that of other males showed a more negative genital perception. On the other hand, patients who considered their penile appearance similar to other males received mean GPSs approximately equal to comparison males. The ability of patients with hypospadias to cope with their somewhat different penile appearance is positively related to genital perception.

Patients with hypospadias did not have a more negative body perception than age-matched comparison males. Adult patients with hypospadias even had a more positive body perception than age-matched comparison men. These findings, supported by the results of factor analysis that revealed two distinct factors (GPS and BPS), indicate that genital perception and body perception are two separate dimensions.

No significant relationship of current age was found with genital perception of children and adolescents as well as with genital perception of adult patients with hypospadias. A small relationship of current age was found with body perception of children and adolescents only: older boys had a somewhat more negative body perception than younger boys. These findings differ from the results of a longitudinal study with adolescent boys aged 11 to 18 years, in which the investigator reported that boys had a higher body image satisfaction at 18 years than at earlier ages.³⁹

Small but statistically significant relationships of severity of hypospadias, the number of operations, and the age at which surgery was finished with

genital perception were found only in adult patients. A possible explanation for this difference between children and adolescents with hypospadias on the one hand and adult patients with hypospadias on the other hand could be the differences in medical histories. The mean number of operations was smaller and the mean age at which surgery was finished was earlier in the children and adolescents than in the adult patients. Thus, fewer operations at earlier ages seem preferable. From our data we are unable to provide a maximum age at which treatment should be completed, but we agree with Schultz, Klykylo, and Wacksman⁴⁰ that, from a psychosexual point of view, it is best to treat boys with hypospadias before the age of 30 months. Comparisons of genital perception and psychosexual adjustment of patients who underwent surgery between 6 and 18 months, between 18 months and 3 years, and at a later age will be necessary to determine the optimal timing of hypospadias surgery.

Although no significant differences in genital perception of patients treated with ventralizing and terminalizing procedures were found, it should not be concluded that this improvement of medical care has not been worthwhile. From this study it remains unknown whether adult patients treated with ventralizing procedures are equally satisfied with their penile appearance as adult patients treated with terminalizing procedures, because of a lack of the latter patients in our study. Maybe adult patients consider the position of their meatus to be of more importance than do children and adolescents with hypospadias. A possible explanation why no difference was found in the satisfaction with the position of the meatus between patients treated with ventralizing and terminalizing procedures could be that the urethral meatus, which is reconstructed to the tip of the penile glans by a terminalizing procedure may retract in the first years after surgery to the coronal side of the glans.⁴¹ Thus, the difference in the cosmetic results between a ventralizing procedure and a terminalizing procedure could gradually diminish in a few years after surgery. A support for this idea is that patients in the current study treated with terminalizing procedures who had a glanular meatus at physical examination were significantly more satisfied

with the position of the meatus than patients treated with terminalizing procedures who had a retracted coronal meatus at physical examination.³²

Other factors that might negatively affect genital perception of patients with hypospadias, such as unrealistic expectations of the cosmetic results of surgery, the parents' anxieties about their son's masculine capabilities (e.g., sexual development and functioning, fertility), or the parents' negative perception of their son's penile appearance were not investigated in the current study. It would be worthwhile to carry out a prospective study which investigates these factors in patients and parents pre- and post-operatively, so that information will be obtained to improve the counseling of parents and patients with hypospadias.

Although patients with hypospadias had a more negative genital perception, we found earlier that they did not experience a retarded sexual adjustment or different sexual behavior as compared to normal males.^{27, 28} Parents (and patients) can now be informed that they can expect a normal sexual development and normal sexual functioning. We believe this information should help patients to cope with their hypospadias and accept their penile appearance.

ACKNOWLEDGEMENTS

We gratefully acknowledge the collaboration of Dr. R. J. M. Nijman, pediatric urologist and H. M. Koot, Ph.D., psychologist for their help in preparing the manuscript. This project was financially supported by the Sophia Foundation for Medical Research.

6.5 REFERENCES

1. Levitt, S. B., & Reda, E. F. (1988). Hypospadias. *Pediatric Annals*, 17 (1), 48-57.
2. Van der Meulen, J. C. (1985). Hypospadias. In F. K. Muir (Ed.), *Current operative surgery: Plastic and reconstructive* (pp. 119-146). London: Ballière and Tindall.
3. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
4. Farkas, L. G., & Hynie, J. (1970). Aftcreffects of hypospadias repair in childhood. *Postgraduate Medicine*, 47, 103-105.
5. Heiss, W. H., & Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadiie-Operationen [Long-term results following hypospadias surgery]. *Zeitschrift für Kinderchirurgie*, 14 (4), 445-451.

6. Sommerlad, B. C. (1975). A long-term follow-up of hypospadias patients. *British Journal of Plastic Surgery*, 28, 324-330.
7. Leuthardt, R., & Morger, R. (1987). Langzeitergebnisse der Hypospadioperationen nach Ombrédanne [Long-term results after Ombrédanne's operation for hypospadias]. *Zeitschrift für Kinderchirurgie*, 42, 153-156.
8. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, 150, 1474-1477.
9. Belfer, M. L., Harrison, A. M., Pillemer, F. C., & Murray, J. E. (1982). Appearance and influence of reconstructive surgery on body image. *Clinics in Plastic Surgery*, 9 (3), 307-315.
10. Ericsson, N. O., & Von Hedenberg, C. (1971). Sexualfunktion hos patienter operade för hypospadi [Sexual functioning of patients operated for hypospadias]. *Läkartidningen*, 68 (21), 2480-2484.
11. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology*, 125, 313-317.
12. Robertson, M., & Walker, D. (1975). Psychological factors in hypospadias repair. *Journal of Urology*, 113, 698-700.
13. Galenson, E. (1990). Observations of early infantile sexual and erotic development. In M. E. Perry (Ed.), *Handbook of sexology. Vol. 7: Childhood and adolescent sexology* (pp. 169-178). New York: Elsevier.
14. Blotcky, M. J., & Grossman, I. (1978). Psychological implications of childhood genitourinary surgery. *Journal of the American Academy of Child Psychiatry*, 17, 488-497.
15. Manley, C. B. (1982). Elective genital surgery at one year of age: Psychological and surgical considerations. *Surgical Clinics of North America*, 62 (6), 941-953.
16. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
17. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.
18. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
19. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
20. Mathieu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.

Genital Perception following Hypospadias Surgery

21. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
22. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadie [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.
23. Van Westerlaak, J. M., Kropman, J. A., & Collaris, J. W. M. (1975). *Beroepenklapper* [Professional index]. Nijmegen, The Netherlands: Instituut voor Toegepaste Sociologie.
24. Lacey, J. H., & Birchnell, S. A. (1986). Body image and its disturbances. *Journal of Psychosomatic Research*, 30 (6), 623-631.
25. Gorsuch, R. L. (1983). *Factor analysis*. Hillsdale, N. J.: Erlbaum.
26. Van Dongen-Melman, J. E. W. M., Koot, H. M., & Verhulst, F. C. (1993). Cross-cultural validation of Harter's self-perception profile for children in a Dutch sample. *Educational and Psychological Measurement*, 53, 739-753.
27. Mureau, M. A. M., Slijper, F. M. E., Van der Meulen, J. C., Verhulst, F. C. & Slob, A. K. (1995). Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study. *Journal of Urology*, in press.
28. Mureau, M. A. M., Slijper, F. M. E., Nijman, R. J. M., Van der Meulen, J. C., Verhulst, F. C., & Slob, A. K. (1995). Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study. *Journal of Urology*, in press.
29. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadie-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, 186, 421-425.
30. Figalova, P., Farkas, L. G., & Hajnis, K. (1968). Eine Studie zur Erforschung somatischer Merkmale bei Patienten mit Hypospadie [A study to investigate physical characteristics of hypospadias patients]. *Zeitschrift für Kinderchirurgie*, 14, 445-451.
31. Pompino, H. J., Zickgraf, Th., Pietschmann, J. H., & Schmidt, W. (1969). Langzeitkatamnesen von 164 Hypospadien [Long-term results of 164 hypospadias patients]. *Zeitschrift für Kinderchirurgie*, 7, 519-533.
32. Mureau, M. A. M., Slijper, F. M. E., Slob, A. K., Verhulst, F. C., & Nijman, R. J. M. (1995). Penile appearance satisfaction following hypospadias surgery: The patients' and the surgeon's view. *Journal of Urology*, in press.
33. Schonfeld, W. A., & Beebe, G. W. (1942). Normal growth and variation in the male genitalia from birth to maturity. *Journal of Urology*, 48, 759-777.
34. Reilly, J. M., & Woodhouse, C. R. J. (1989). Small penis and the male sexual role. *Journal of Urology*, 142, 569-571.
35. Van Seters, A. P., & Slob, A. K. (1988). Mutually satisfying heterosexual relationship with micropenis of husband. *Journal of Sex & Marital Therapy*, 14, 98-107.

Chapter 6

36. Gilbert, D. A., Jordan, G. H., Devine, C. J. Jr., Winslow, B. H., & Schlossberg, S. M. (1993). Phallic reconstruction in prepubertal and adolescent boys. *Journal of Urology*, *149*, 1521-1526.
37. Schlossberger, N. M., Turner, R. A., & Irwin, C. E. Jr. (1991). Early adolescent knowledge and attitudes about circumcision: Methods and implications for research. *Journal of Adolescent Health*, *13* (4), 293-297.
38. Kropfi, D., Schardt, M., & Fey, S. (1992). Modified meatal advancement and glanuloplasty with complete foreskin reconstruction. *European Urology*, *22* (1), 57-61.
39. Rauste-von Wright, M. (1989). Body image satisfaction in adolescent girls and boys: A longitudinal study. *Journal of Youth and Adolescence*, *18*, 71-83.
40. Schultz, J. R., Klykylo, W. M., & Wacksman, J. (1983). Timing of elective hypospadias repair in children. *Pediatrics*, *71*, 342-351.
41. Unluer, E. S., Miroglu, C., Ozdiler, E., & Ozturk, R. (1991). Long-term follow-up results of the MAGPI (meatoplasty and glanuloplasty) operations in distal hypospadias. *International Urology & Nephrology*, *23* (6), 581-587.

CHAPTER 7

SATISFACTION WITH PENILE APPEARANCE FOLLOWING HYPOSPADIAS SURGERY: THE PATIENTS' AND THE SURGEON'S VIEW*

Marc A.M. Mureau,^{1,2} Froukje M.E. Slijper,¹ A. Koos Slob,³
Frank C. Verhulst,¹ Rien J.M. Nijman.⁴

¹Department of Child & Adolescent Psychiatry, ²Department of Plastic & Reconstructive Surgery, ³Department of Endocrinology & Reproduction, ⁴Department of Pediatric Urology, Sophia Children's Hospital Rotterdam/ University Hospital Rotterdam/Erasmus University Rotterdam.

ABSTRACT

Purpose: To study: (1) the degree of agreement between hypospadias patients' and surgeon's satisfaction with the cosmetic surgical result; (2) the relation between penile length, meatal position, and patients' satisfaction.

Materials and methods: Cosmetic and functional results of 35 boys with hypospadias were assessed and a standardized questionnaire was completed by patients and surgeon.

Results: There existed hardly any agreement between hypospadias patients' and surgeon's satisfaction with the patients' penile appearance; patients were less satisfied than their surgeon. No significant correlation between penile satisfaction and penile length was found. Patients with a retracted meatus were less satisfied with the meatal position than patients with

* Accepted for publication, Journal of Urology, 1995.

a glanular meatus. Of the 35 patients, 4 were re-operated following the present study.

Conclusions: Hypospadias surgeons should explicitly ask whether or not patients are satisfied, and they should follow their patients through adolescence.

7.1 INTRODUCTION

Corrective surgery of hypospadias, i.e. reconstruction of the urethra and straightening of the penis is necessary to ensure voiding in the standing position and unhampered adult sexual functioning. Another important goal of hypospadias surgery is the achievement of a cosmetic penile appearance which is as normal as possible. Some studies on the long-term results of hypospadias surgery reported *patients'* satisfaction with their penile appearance,^{1,4} while other investigations reported *surgeons'* satisfaction with the surgical result.^{5,6} To our knowledge only Schwöbel and collaborators⁷ investigated both hypospadias patients and surgeons with regard to their satisfaction with the cosmetic result. Of the 27 patients, 92% judged the cosmetic result to be satisfactory or excellent, and all patients had a satisfactory or excellent cosmetic result as judged by the investigator.⁷ Unfortunately, from their data it is impossible to determine the degree of agreement between patient's and surgeon's satisfaction and only the position of the urethral meatus was considered in the judgment of the surgical result.

The above mentioned studies made no distinction between satisfaction with functional result and satisfaction with cosmetic penile appearance, and it was not known whether or not hypospadias patients were (dis)satisfied with the same aspects of penile appearance as the surgeon. It seems essential to know whether or not hypospadias patients and surgeons consider the same aspects of penile appearance to be important, in order to find out whether or not treatment or counseling of hypospadias patients should be adjusted. Therefore, we performed the present study and investigated in a standardized manner (1) the level of agreement between hypospadias patients' and surgeon's satisfaction with different aspects of the patients' penile appearance; and (2) the relation between physical genital characteristics (penile length,

meatal position) and satisfaction with penile appearance in hypospadias patients.

7.2 SUBJECTS AND METHODS

Patients. Of all 458 patients, treated for hypospadias between 1980 and 1992 at the Sophia Children's Hospital in Rotterdam (Department of Pediatric Urology), 113 boys (aged 9 to 18 years at the onset of the present study) were previously treated with a terminalizing technique.⁸⁻¹³ Of these 113 boys, 86 had distal and 27 had proximal hypospadias. The latter 27 patients were all included in the present study. To obtain an approximately equal group of patients with distal hypospadias and because of a lack of time, only 33 patients were randomly selected from the patients with distal hypospadias. Thus, 60 boys were included in the present study.

Invitations to participate. From November 1993 to May 1994 the patients (and parents) received a letter explaining the purpose of the study (which is part of a larger study on psychosexual and psychosocial adjustment of hypospadias patients) and inviting them to participate. About one week later they were phoned to inquire whether or not they were willing to come to the hospital.

Instruments. A standardized questionnaire was used which consisted of 8 items about different aspects of the genitals (for description of the items, see table 7.2). Satisfaction with each genital aspect was rated on a 4-point scale ranging from very dissatisfied (1) to very satisfied (4). A Genital Perception Score (GPS) was calculated by summing the raw scores for each item. The GPS ranges from 8 to 32 (very dissatisfied to very satisfied with genital appearance). In addition to the hypospadias patients who completed this questionnaire in the *absence* of their surgeon, the latter - a pediatric urologist - rated *his* satisfaction with the patient's penile appearance through an identical questionnaire.

Physical examination. Penile length was measured from the pubic bone to the tip of the glans with the patient in a supine position. Penile length of the hypospadias patients was measured with a cotton swab which was held along the dorsal side of the *stretched* penis and which was marked at the

level of the penile top. Subsequently, the length of the cotton swab was measured with a ruler.

The position of the meatus was assessed and marked in a schematic drawing of a penis. From these markings, two meatal positions were scored: 1 = glanular position; 2 = coronal position.

Statistical methods. Student's *t* tests, Mann-Whitney *U* tests (corrected for ties), Wilcoxon matched-pairs signed-rank tests, Spearman's rank correlation coefficients (r_s , corrected for ties), Pearson's correlation coefficients (r), partial correlation coefficients (r_p), and binomial tests were used.¹⁴ Probabilities (two-tailed, unless otherwise indicated) $< .05$ were accepted as significant.

Table 7.1. Sample characteristics of the responders and non-responders

	Hypospadias patients aged 9 to 18 years ($N = 60$)		Statistics	<i>p</i>
	Responders ($N = 35$)	Non-responders ($N = 25$)		
Mean (<i>SD</i>) current age	13.2 (2.7)	14.0 (2.7)	$t = -1.17$.250
<i>Severity of hypospadias</i>	<i>n</i> (%)	<i>n</i> (%)		
Distal hypospadias	18 (51.4)	15 (60.0)	$\chi^2 = 0.43$.514
Proximal hypospadias	17 (48.6)	10 (40.0)		
<i>Surgical procedure</i>	<i>n</i> (%)	<i>n</i> (%)		
MAGPI, ⁸ GAP, ⁹ Koff, ¹⁰ Mathieu ¹¹	18 (51.4)	15 (60.0)	$\chi^2 = 0.43$.514
TPIF, ¹² Perović ¹³	17 (48.6)	10 (40.0)		
Mean (<i>SD</i>) age at final surgery	7.3 (3.2)	7.6 (3.2)	$z = -0.37$.711
Mean (<i>SD</i>) number of operations	2.3 (1.3)	2.4 (1.4)	$z = -0.22$.827

7.3 RESULTS

Response. Of the 60 children and adolescents with hypospadias, 3 were untraceable and 35 (61.4%) visited the hospital and were physically examined. For these 35 boys each questionnaire was completed.

Non-response bias. Since the percentage of patients for whom questionnaires were completed was rather low, a selection bias could have occurred. Therefore, several sample characteristics of the responders and non-responders were compared. The participating patients did not significantly differ from the non-responders in mean age, severity of hypospadias, surgical procedure, mean age at final surgery, and mean number of operations (see table 7.1). Thus, we assume that the participating patients are a representative sample.

Table 7.2. Satisfaction with penile appearance, mean values (SD), of children and adolescents with hypospadias aged 9 to 18 years (N=35), judged by patients and pediatric urologist (Wilcoxon matched-pairs signed-rank tests)

Satisfaction with: ^a	Hypospadias patients	Pediatric urologist	N ^b	z	p
Flaccid penile size	3.1 (1.1)	3.9 (0.4)	35	-3.34	.0008
Penile thickness	3.1 (0.9)	3.9 (0.3)	35	-4.01	.0001
Glanular size	3.1 (1.0)	3.9 (0.3)	35	-3.64	.0003
Glanular shape	3.1 (0.9)	3.5 (0.6)	35	-2.31	.0207
Position of meatus	3.6 (0.7)	3.4 (0.7)	35	-1.27	.2049
Scars	2.8 (1.3)	3.2 (0.7)	33	-1.82	.0680
Scrotum/testes	3.3 (0.8)	3.9 (0.2)	35	-3.70	.0002
Penile appearance in general	3.0 (1.0)	3.4 (0.6)	35	-1.72	.0853
Genital Perception Score (8-32) ^c	25.1 (6.1)	29.1 (2.5)	33	-3.26	.0011

^aScored on a 4-point scale, where 1 = very dissatisfied and 4 = very satisfied. ^bNumber of pairs. ^c8 = very negative overall genital perception; 32 = very positive overall genital perception.

Differences in satisfaction between patients and surgeon. Table 7.2 shows that children and adolescents with hypospadias were less satisfied with their genital appearance than the pediatric urologist. They were less satisfied mainly with penile and glanular size and shape. Interestingly, hypospadias patients were also less satisfied than the surgeon with the appearance of their scrotum and testes. After exclusion of the patients who were also treated for cryptorchidism (3 of 35 patients), the remaining patients still were less satisfied with the appearance of their scrotum and testes than their surgeon (Wilcoxon matched-pairs signed-rank test, $z = -3.68$, $N = 32$, $p < .001$).

Agreement of satisfaction between hypospadias patients and surgeon. In table 7.3 Spearman's rank correlation coefficients between hypospadias patients' and surgeon's satisfaction with several genital aspects of the patients are listed. In general, there was hardly any agreement between hypospadias patients' and surgeon's satisfaction, except for satisfaction with scars (table 7.3).

Table 7.3. Agreement between hypospadias patients' and surgeon's satisfaction with several genital aspects of the patients (Spearman's rank correlation coefficients)

Surgeon	Hypospadias patients aged 9 to 18 years ($N = 35$)		
	r_s	N	p
Flaccid penile size	.17	35	.340
Penile thickness	.28	35	.106
Glanular size	.08	35	.644
Glanular shape	.30	35	.084
Position of meatus	.29	35	.088
Scars	.53	33	.002
Scrotum/testes	-.23	35	.186
Penile appearance in general	-.07	35	.686
Genital Perception Score	.31	33	.078

Hypospadias Patients and Surgeon: Disagreement with Surgical Results

Table 7.4. Stretched penile length of 35 hypospadias patients, compared to normative data, by age

Hypospadias patients (N = 35)			Percentiles of stretched penile length (cm), Schonfeld and Beebe, 1942 ¹⁵	
Age (yrs)	N	Stretched penile length (cm)	P ₁₀	P ₉₀
9	5	5; 5.5; 6; 8; 8; 8	4.9	7.6
10	3	6.5; 7; 9		
11	6	<u>3</u> ; 5; 5.5; 6; 7; 8	4.7	8.7
12	2	7.5; 8	4.9	11.3
13	3	<u>6</u> ; 7; 10	6.1	12.2
14	4	8; 10; 11; 12	6.6	13.5
15	3	<u>9</u> ; 10; 10.5	9.1	14.8
16	4	<u>10</u> ; <u>10</u> ; <u>10</u> ; 11	10.8	15.3
17	2	12; 15		
18	1	<u>10</u>	10.8	15.5

Underlined figures indicate that penile length is below the 10th percentile.

Physical characteristics. The mean *stretched* penile length of the children and adolescents was 8.4 cm (*SD* = 2.5 cm; range: 3.0 - 15.0 cm). Significantly more patients (21.2%; binomial test, $p_{1-tailed} = .032$) fell below the 10th percentile for stretched penile length, as reported by Schonfeld and Beebe¹⁵ (see table 7.4). A high correlation existed between patients' age and penile length ($r = 0.76$, $N = 33$, $p < .001$). Since there were no statistically significant differences in mean age between patients with distal and proximal hypospadias (Student's *t* test, $t(35) = 0.25$, $p = .808$), the comparison of penile length between patients with distal and proximal hypospadias was not statistically controlled for age. This analysis revealed no significant differences between patients with distal (mean = 8.6 cm) and proximal hypospadias (mean = 8.2 cm; Student's *t* test, $t(31) = 0.42$, $p = .679$). The urethral meatus was situated at a glanular position in 24 patients (68.6%) and at a coronal position in 11 patients (31.4%).

Relation between physical characteristics and satisfaction with penile appearance. Because there was a significant correlation between age and penile length, the correlation between the Genital Perception Score and penile length was controlled for age. Unexpectedly, there did not exist a positive and statistically significant correlation between penile length and the patients' Genital Perception Score ($r_p = -0.31$, 29 *df*, $p = .091$). Patients who fell below the 10th percentile for stretched penile length were equally satisfied with flaccid penile size as patients who fell above the 10th percentile (Mann-Whitney *U* test, $z = 0.00$). Patients with a glanular meatal position reported to be more satisfied with the position of the meatus than patients with a coronal meatal position (Mann-Whitney *U* test, $N = 35$, $z = -2.39$, $p = .017$).

Re-operations. Following the physical examination in this study 4 boys (11%) were re-operated, because of a (iatrogenic) lateral penile curvature in 2 cases, a fistula in 1 case, and a retracted coronal meatus in another case. It is of particular interest that all of these patients stated that they would not have readily consulted a physician for their problems on their own initiative.

7.4 DISCUSSION

In the present study there existed hardly any agreement between hypospadias patients' and surgeon's satisfaction with the patients' penile appearance. These results corroborate our clinical impression that "excellent" cosmetic results as judged by surgeons do not have to lead to satisfied patients. Conversely, patients with "poor" cosmetic results can nevertheless be satisfied and not wanting further surgery, even when cosmetic improvement is feasible. Therefore, hypospadias surgeons should *explicitly* ask whether or not the patient is satisfied with his penile appearance. If the patient is not satisfied, the surgeon should inquire about the motive for dissatisfaction, and should inform the patient whether or not it would be feasible to further surgically improve penile appearance.

A striking result is that hypospadias patients were less satisfied with the appearance of their scrotum and testes than the surgeon, even after exclusion of patients who were also treated for cryptorchidism. Patients were less

Hypospadias Patients and Surgeon: Disagreement with Surgical Results

satisfied than their surgeon with 5 of the 8 different genital aspects. These findings suggest that hypospadias patients have a more negative overall genital perception. This latter supposition is supported by the findings of our study on genital perception of 116 boys (including the 35 boys of the present study) as well as 73 adults with hypospadias, which showed that they had a significantly more negative overall genital perception than age-matched comparison males.¹⁶

In the present study the mean number of operations and the mean age at which treatment was finished were quite high, which could have negatively affected genital perception of the patients. However, in our previous study we found that there did not exist significant relationships between genital perception of 116 boys with hypospadias (including the 35 boys of the present study) and the number of operations or the age at which treatment was finished.¹⁶

Approximately one third of the patients treated with terminalizing repairs had a retracted coronal meatus. This corroborates the results of an earlier study which showed partial or complete meatal retraction in 37% of 41 patients treated with a MAGPI procedure,¹⁷ but it contradicts results of another study which reported meatal retraction in only 0.6% of 1,111 cases treated with a MAGPI procedure.¹⁸ However, these results are hard to compare, since the mean follow-up was much longer in the present study (5.8 yrs, range: 2.3-9.2 yrs) than in the previous studies (29 months¹⁷ and 2.3 months¹⁸), and the present study also comprised patients with proximal hypospadias treated with different surgical techniques. Nevertheless, meatal retraction remains a possible long-term complication to look for when following hypospadias patients.

Patients with a terminal meatus were more satisfied with the position of the meatus than patients with a (retracted) coronal meatus. This seems to indicate that it is important for the surgeon to try to achieve a terminal meatus. However, since the position of the meatus was not a major motive for concern in children and adolescents as well as adults with hypospadias,^{19, 20} we recommend to re-advance a retracted meatus *only* when the patient wishes a terminal meatus or when the retracted meatus causes spraying of urine.

Given the 10th and 90th percentiles are the ranges of normal variation,¹⁵ in the present study penile underdevelopment was encountered about twice as more often among hypospadias patients as among age matched normal boys. *Stretched* instead of *flaccid* penile length was measured, since the latter varies appreciably in response to tactile, thermal, and other environmental changes,¹⁵ which makes measuring flaccid penile length less reliable than measuring stretched penile length. Therefore, we advise future investigators who want to study penile length of hypospadias patients to measure *stretched* penile length from the pubic bone to the tip of the glans for which reliable normative data exist from birth to maturity.¹⁵

No difference in penile length was found between patients with distal and proximal hypospadias, contrary to earlier studies which found a negative correlation between flaccid^{2, 21} or stretched² penile length and severity of hypospadias. Although penile size was the most reported motive for dissatisfaction in children and adolescents as well as adults with hypospadias,^{19, 20} in the present study no significant correlation was found between penile length and the patients' satisfaction.

Although one cannot exclude the possibility that hypospadias patients with problems more often tended to participate in the present study than patients without problems, 11% of the children and adolescents with hypospadias were re-operated following the present study because of a poor long-term functional result of surgery. We strongly advocate that hypospadias surgeons should follow their patients during adolescence as a standard therapeutic procedure, since: (1) hypospadias patients may be reluctant to seek medical advice on their own initiative, even when they are experiencing considerable difficulties;^{2, 19, 20} (2) boys with hypospadias are treated at increasingly earlier ages (often before 18 months) and therefore are not able to express their (dis)satisfaction with the surgical result; (3) our earlier study with children and adolescents showed that adolescents (13-18 yrs) more often wanted cosmetic or functional penile improvement than children (9-12 yrs);²⁰ and (4) complications, such as meatal retraction or stenosis, residual penile curvature, or adult sexual dysfunction often become apparent many years after surgical treatment is finished.

Hypospadias Patients and Surgeon: Disagreement with Surgical Results

ACKNOWLEDGEMENTS

This project was financially supported by the Sophia Foundation for Medical Research.

7.4 REFERENCES

1. Sommerlad, B. C. (1975). A long-term follow-up of hypospadias patients. *British Journal of Plastic Surgery*, 28, 324-330.
2. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
3. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, 150, 1474-1477.
4. Kumar, M. V. K., & Harris, D. L. (1994). A long term review of hypospadias repaired by split preputial flap technique (Harris). *British Journal of Plastic Surgery*, 47, 236-240.
5. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadiie-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, 186, 421-425.
6. Beretta, G., Mazzucchelli, S., Zanollo, A., Negri, L., & Catanzaro, F. (1986). Ipospadias: Comportamento sessuale in maschi adulti sottoposti a trattamento chirurgico correttivo in età pediatrica [Sexual aspects of men operated for hypospadias during childhood]. *Minerva Urologica e Nefrologica*, 38, 17-20.
7. Schwöbel, M. G., Sacher, P., & Stauffer, U. G. (1987). Die Dennis-Browne-Korrektur der Hypospadias: Langzeitergebnisse [The Dennis Browne method for hypospadias repair: Long term results]. *Zeitschrift für Kinderchirurgie*, 42, 157-160.
8. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
9. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.
10. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
11. Mathieu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabalanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.
12. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
13. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadias [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.
14. Altman, D. G. (1991). *Practical statistics for medical research*. London: Chapman & Hall.
15. Schonfeld, W. A., & Beebe, G. W. (1942). Normal growth and variation in the male genitalia from birth to maturity. *Journal of Urology*, 48, 759-777.

16. Murcau, M. A. M., Slijper, F. M. E., Slob, A. K., & Verhulst, F. C. (1995). Genital perception of children, adolescents, and adults operated on for hypospadias: A comparative study. *Journal of Sex Research, in press.*
17. Unluer, E. S., Miroglu, C., Ozdifer, E., & Ozturk, R. (1991). Long-term follow-up results of the MAGPI (meatoplasty and glanuloplasty) operations in distal hypospadias. *International Urology & Nephrology, 23* (6), 581-587.
18. Duckett, J. W., & Snyder, H. M. (1992). Meatal advancement and glanuloplasty hypospadias repair after 1,000 cases: Avoidance of meatal stenosis and regression. *Journal of Urology, 147*, 665-669.
19. Mureau, M. A. M., Slijper, F. M. E., Van der Meulen, J. C., Verhulst, F. C. & Slob, A. K. (1995). Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study. *Journal of Urology, in press.*
20. Mureau, M. A. M., Slijper, F. M. E., Nijman, R. J. M., Van der Meulen, J. C., Verhulst, F. C., & Slob, A. K. (1995). Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study. *Journal of Urology, in press.*
21. Figalova, P., Farkas, L. G., & Hajnis, K. (1968). Eine Studie zur Erforschung somatischer Merkmale bei Patienten mit Hypospadie [A study to investigate physical characteristics of hypospadias patients]. *Zeitschrift für Urologie, 61*, 313-317.

CHAPTER 8

PSYCHOSOCIAL FUNCTIONING OF CHILDREN, ADOLESCENTS, AND ADULTS FOLLOWING HYPOSPADIAS SURGERY: A COMPARATIVE STUDY*

Marc A.M. Mureau,^{1,2} Froukje M.E. Slijper,¹ A. Koos Slob,³
Frank C. Verhulst.¹

¹Department of Child & Adolescent Psychiatry, ²Department of Plastic & Reconstructive Surgery, ³Department of Endocrinology & Reproduction, Sophia Children's Hospital Rotterdam/University Hospital Rotterdam/Erasmus University Rotterdam.

ABSTRACT

Psychosocial functioning of patients operated for hypospadias, a congenital anomaly of the penis, was studied through standardized self-report questionnaires on personality, social anxiety, and emotional/behavioral problems. Psychosocial functioning of 73 adult patients (18 to 38 yrs) and 116 children and adolescents (9 to 18 yrs) with hypospadias was compared with that of 50 and 88 age-matched comparison subjects, respectively, treated for an inguinal hernia. The relationships of coping with penile appearance, subject age, severity of hypospadias, number of operations, age at final surgery, and type of surgical procedure with psychosocial functioning of hypospadias patients were also investigated. Psychosocial adjustment of hypospadias patients did not differ significantly from that of age-matched comparison subjects, except 25- to 38-year-old hypospadias patients repor-

*Submitted for publication, 1995.

ted more vague somatic complaints and anxieties, more feelings of inadequacy, and more social anxieties. No significant relationships of various medical characteristics with psychosocial functioning could be discerned. Genital/body perception of hypospadias patients correlated positively with psychosocial functioning, albeit with low values. These findings are important for pediatric urologists and plastic surgeons in the counseling process of hypospadias patients and their parents.

8.1 INTRODUCTION

Hypospadias is a common urogenital anomaly with a reported incidence ranging from 0.8 to 8.2 per 1,000 live male births.¹ It is caused by a disturbance in the development of the urethra resulting in an abnormal position of the urethral opening. *Distal* hypospadias refers to conditions in which the position of the urethral meatus is situated on the glans penis, the coronal sulcus, or just proximal to the coronal sulcus. *Proximal* hypospadias refers to conditions in which the urethra opens at some point along the penile shaft, at the penoscrotal junction, or on the perineum. Penile curvature, penile underdevelopment, and occasionally penoscrotal transposition, cryptorchidism, as well as an enlargement of the prostatic utricle may be observed; these latter concomitant anomalies are found more often in proximal than in distal hypospadias.² Surgical reconstruction of the urethra and straightening of the penis is necessary to ensure voiding in a standing position and unhampered adult sexual functioning. Another important goal of hypospadias surgery is the achievement of a cosmetic appearance of the penis which is as "normal" as possible.

Although more than 150 surgical techniques to correct hypospadias have been described,¹ studies on psychosocial functioning of hypospadias patients are scarce. Plastic surgeons and pediatric urologists continuously try to improve the cosmetic surgical techniques to make the penis look as "natural" as possible. For instance, one improvement was the development of *terminalizing* surgical techniques which split, core, or tunnel through the glans to create a true terminal glanular meatus, replacing earlier *ventralizing* surgical techniques, which bring the meatus onto the underside of the glans or coro-

na.³ Another example of medical improvement is that surgery is performed with increasingly fewer operations at earlier ages. Whether these improvements also have a positive effect on psychosocial functioning of the patients has not been thoroughly investigated. One could assume that hypospadias patients are at risk for poorer psychosocial functioning than normal males. Most boys with hypospadias are not able to urinate in the standing position until after surgery. Robertson and Walker⁴ noted that this could keep such boys from entering the competition with other boys, demonstrating their prowess at urinating at certain distances. This could play a role in the acquisition of feelings of incompetence, inadequacy, or loss of self-confidence. The experience of childhood genital surgery could play a role in the development of poorer psychosocial functioning. Blotcky and Grossman⁵ reported a higher occurrence of emotional disturbance in a group of 15 children (both sexes) after urogenital surgery than in a group of 33 children after ear, nose, and throat surgery. Lepore and Kesler⁶ reported in a study with 10 hypospadias patients between 2 and 6 years, that they showed a distinct pattern of behavior in the first days after surgery characterized by anger, aggression, and negative interactions. After hypospadias surgery the penile appearance of patients with hypospadias remains different from normal males. For example, after hypospadias surgery the appearance of the penis is circumcised, which is uncommon among Dutch boys in The Netherlands. Sometimes there is penile underdevelopment which is associated with hypospadias, especially in the severe cases.² Such different penile appearance could make hypospadias patients more self-conscious about or less satisfied with their penile appearance, which might negatively affect their general psychosocial functioning.

In a study of 3 adolescent boys who had undergone surgery for different urogenital anomalies,⁷ all 3 showed feelings of incompetence, inadequacy, and fears about their future masculinity and health. These latter findings cannot be extrapolated to patients with urogenital anomalies in general, because of the small number of patients and the fact that these patients were selected because they had problems. Kipikaša, Longauer, and Urbanová⁸ reported many psychosocial disorders in 119 adult hypospadias patients;

most of them had been fearful and reticent already as children, they grew into withdrawn adults and were shy, diffident, and tried to stay away from team life. A major drawback of the latter study is that it was only descriptive; no detailed data based on quantifiable standardized assessments with known psychometric properties were presented. The first comparative studies on psychosocial and psychosexual adjustment of hypospadias patients showed that 34 adult hypospadias patients had a poorer psychosocial adjustment than 36 age-matched comparison subjects operated for verified appendicitis.^{9, 10} Hypospadias patients were more timid, isolated and mobbed as children, and during adulthood they were more fearful and socially isolated, showed less self-esteem, had less capacity for interpersonal social and emotional relations, and had less qualified professions. Caution must be taken with interpreting these results, since a rather small number of adult patients was investigated, psychiatric problems were inferred by the investigators through a projective test and a semi-structured interview, and the problems during childhood were assessed retrospectively. More recently, Sandberg and co-workers investigated 69 boys with hypospadias between the ages of 6 and 10 years and reported that they showed more behavioral problems and lower social competence than boys from a nonclinical sample.¹¹ Although the investigators used questionnaires with good validity and reliability, their results cannot be extrapolated to hypospadias patients older than 10 years.

In summary, there is still a dearth of *systematic* knowledge about the psychosocial functioning of hypospadias patients at different ages, compared to normal subjects. Besides, it has never been investigated to what extent patients' perception of their penile appearance is related to their psychosocial functioning. In other words, do hypospadias patients who have a positive genital perception fare better psychosocially than those who have a negative genital perception? Therefore, we carried out a study which compared the psychosocial functioning of children and adolescents as well as of adults treated for hypospadias in the past, with age-matched normal subjects. The main research questions were: (1) do hypospadias patients have a poorer psychosocial adjustment than age-matched normal subjects?; (2) what is the relationship of subject age with psychosocial functioning?; (3) what are the

relationships of various medical characteristics (severity of hypospadias, number of operations, age at final surgery, surgical procedure) with psychosocial functioning?; and (4) does there exist a positive relationship between genital/body perception and psychosocial functioning of hypospadias patients?

8.2 METHOD

8.2.1 SAMPLE SELECTION

Hypospadias patients. Of all 571 hypospadias patients treated with ventralizing surgical repairs^{2, 12} between 1960 and 1990 at the Department of Plastic and Reconstructive Surgery of the University Hospital Rotterdam, 423 patients were 18 years and older at the time of the present study. A random sample of 100 subjects was drawn from these 423 adult patients. Of the above mentioned 571 patients, 104 boys were between 9 and 18 years and were included in the present study. Of all 458 patients treated for hypospadias between 1980 and 1992 at the Department of Pediatric Urology of the Sophia Children's Hospital in Rotterdam, 113 boys had undergone terminalizing repairs¹³⁻¹⁸ and were between 9 and 18 years at the time of the present study. Of these 113 boys, 86 had distal and 27 had proximal hypospadias. The latter 27 patients were all included in the present study. From the 86 patients with distal hypospadias, 33 were randomly selected and included in the study. The target group of the study comprised 100 adult patients (ventralizing repairs) + 104 boys (ventralizing repairs) + 60 boys (terminalizing repairs) = 264 patients (table 8.1).

Response. Of the 100 selected adult patients with hypospadias, 6 were untraceable and 3 were mentally retarded. Seventy-three patients (80.2%) participated in our study. Of the 164 selected boys with hypospadias, 8 were untraceable and one boy was mentally retarded. Of the remaining 155 boys, 116 (74.8%) participated in our study (table 8.1).

Comparison subjects. Comparison subjects treated for an inguinal hernia without concomitant genital surgery (i.e., circumcision, orchiopexy), were selected from the files of Pediatric Surgery of the same medical center. To obtain at least 100 *adult* comparison subjects, 3 random samples (218 sub-

Chapter 8

Table 8.1. Number of patients in the sample, by age, severity of hypospadias, and surgical procedure

Patients aged 18 years and older ($n = 100$)		
Surgical procedures	Distal hypospadias	Proximal hypospadias
Ventralizing repairs		
Van der Meulen procedure ²	49 (71)	14 (18)
Byars/Browne procedure ¹²	8 (9)	2 (2)
Patients aged 9 to 18 years ($n = 164$)		
Ventralizing repairs		
Van der Meulen procedure ²	27 (38)	18 (26)
Byars/Browne procedure ¹²	26 (34)	4 (6)
Terminalizing repairs		
Glans approximation procedure ¹³		
Meatoplasty and glanuloplasty incorporated ¹⁴	21 (33)	-
Urethral advancement ¹⁵		
Mathieu procedure ¹⁶		
Transverse preputial island flap ¹⁷	-	20 (27)
Onlay preputial island flap ¹⁸		

Note: Numbers between brackets are target numbers.

jects) matched for current age, and age at the first operation had to be drawn from a pool of 1,305 men. Finally, a group of 102 adult comparison subjects remained. To obtain a comparison group of at least 164 *children and adolescents*, 2 random samples (228 subjects) matched for current age, and age at the first operation had to be drawn from a pool of 2,239 boys. A group of 165 comparison boys remained.

Response. Of the adult comparison group, 5 subjects were untraceable and one had died. Fifty (52.1%) of the remaining 96 subjects participated. Of

the 165 comparison boys, 8 were untraceable and 4 were mentally disabled. Eighty-eight comparison boys (57.5%) participated in this study.

8.2.2 SOCIOECONOMIC VARIABLES

Occupational status of the adult subjects and of the parents of the children and adolescents was scored on a 6-point scale, where 6 = highest status occupation.¹⁹ Furthermore, religion and urbanization were assessed. Urbanization was scored on a 3-point scale (1 = <20,000 inhabitants; 2 = 20,000-100,000 inhabitants; and 3 = >100,000 inhabitants). Religion of the adult subjects was scored as follows: 1 = no religion; 2 = Roman Catholic; and 3 = Protestant; parental religion of the children and adolescents was scored similarly with 4 = Islamic as additional religion.

8.2.3 MEASURES

To assess emotional functioning the Dutch Personality Questionnaire (DPQ)²⁰ was used with adults, and the Junior Dutch Personality Questionnaire (DPQ-J)²¹ was used with children and adolescents. From the DPQ as well as the DPQ-J the scales Neuroticism (21 and 28 items respectively), Social Inadequacy (15 and 13 items respectively), and Dominance (17 and 15 items respectively) were used. The Neuroticism scale measures vague somatic complaints and anxieties, and feelings of inadequacy; the Social Inadequacy scale measures social anxiety and avoidance of social contacts; and the Dominance scale measures self-confidence and the preference to take initiative and direct other persons. The response format of the items of the DPQ and DPQ-J are: 2 = yes, 1 = do not know, and 0 = no. The higher the scores on Neuroticism and Social Inadequacy the poorer emotional functioning is. Subjects who receive high scores on Dominance prefer to take initiative and to direct others. The reported reliability (coefficients α varied from 0.64 to 0.87) and validity of the DPQ and DPQ-J scales were acceptable.^{20, 21}

To assess social anxiety and social competence, the Contact with Others Inventory (COI)²² was used with adults, and the Social Anxiety Scale for Children (SAS-C)²³ was used with children and adolescents. From the COI the Total Frequency Scale consisting of 35 items about different social

situations, and the Total Anxiety Scale consisting of 35 identical items were used. Frequencies of and the extent of anxiety in 35 different social situations are scored on 5-point scales, where 1 = never/not nervous at all, and 5 = always/very nervous. From the SAS-C the Total Social Anxiety Score (46 items) assessing cognitive, physiological, and emotional anxiety reactions in different social situations was used. The response possibilities of the separate items are binomial (yes or no, nervous or not nervous). Since the SAS-C was originally developed for children between the ages of 9 and 12 years,²³ the use of language was slightly adapted in order to make the questionnaire more appropriate for adolescents between the ages of 13 and 18 years (e.g., "other children" was changed into "peers"). Furthermore the item "I am nervous, when I have to go to swimming lesson" was changed into "I am nervous, when I have to go to gym." The original version was used for children aged 9 to 12 years, the adapted version was used for adolescents aged 13 to 18 years. The reported reliability (coefficients α varied from 0.89 to 0.96) and validity of the COI and the SAS-C were acceptable.^{22, 23}

To assess behavioral/emotional problems, the Child Behavior Checklist (CBCL)²⁴ and the Youth Self-Report (YSR)²⁴ were used with children and adolescents, and the Young Adult Self-Report (YASR)²⁵ was used with adults. The CBCL, YSR, and YASR have roughly the same format and consist of respectively 20, 17, and 14 competence items and respectively 120, 103, and 110 problem items, covering a broad range of emotional and behavioral problems. In the present study only the problem items were used. The CBCL, YSR, and YASR consist of eight empirically derived "syndromes" (Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior), two broad band syndromes (Internalizing and Externalizing), and a Total Problem Score. The Internalizing scale consists of the Withdrawn, Somatic Complaints, and Anxious/Depressed syndromes. The Externalizing scale consists of the Aggressive Behavior and Delinquent Behavior syndromes. The CBCL is completed by parents, the YSR and YASR are self-report questionnaires. The response format of the items is: 0 if the problem item is not true; 1 if the problem item is somewhat or sometimes

true; and 2 if the problem item is very true or often true. Verhulst and collaborators translated the CBCL, YSR, and YASR into Dutch and derived norms for the Dutch population; they reported good reliability, validity, and stability of the syndromes.²⁶⁻²⁹

To assess genital and body perception, we developed two standardized self-report questionnaires (child/adolescent and adult version) consisting of 18 and 20 items respectively, about different aspects of the genitals and body. Satisfaction with each genital or body aspect was rated on a 4-point scale (child/adolescent version) or a 5-point scale (adult version), ranging from very dissatisfied to very satisfied. After principal component analysis with varimax rotation, two factors could be discerned in both questionnaires: a Genital Perception Score (GPS), and a Body Perception Score (BPS).³⁰ The GPS consists of 8 and 9 items, and the BPS consists of 10 and 11 items for the child/adolescent and adult version respectively. Cronbach's α of the GPS is 0.89 for the child/adolescent and 0.86 for the adult version, and Cronbach's α of the BPS is 0.85 for the child/adolescent and 0.78 for the adult version.³⁰

8.2.4 PROCEDURE

Between January 1993 and May 1994, a letter was mailed to all subjects explaining the study and asking them to participate. About one week later, they were phoned to inquire about possible participation. Subjects who could not be reached by phone received a second and, if necessary, a third letter with the request to contact us. When subjects did not want to participate this was accepted, and the motive for refusal was asked. All consenting subjects were invited to come to the hospital. Subjects, who did not want to come to the hospital were visited at home, or they completed the questionnaires at home and returned them by mail. The participating subjects received a poster or a gift coupon for their cooperation.

8.3 RESULTS

8.3.1 STATISTICAL ANALYSIS

Student's *t* tests, 2 x 2 ANOVAs, Mann-Whitney *U* tests (corrected for ties), chi-square tests, Fischer's exact tests, Pearson's correlation coefficients, partial correlation coefficients, and Spearman's rank correlation coefficients (corrected for ties) were used. Probabilities < .05 were accepted as significant. Except for probabilities of the correlation coefficients, all probabilities are two-tailed.

8.3.2 SOCIOECONOMIC SAMPLE CHARACTERISTICS

No statistically significant differences between patients and comparison subjects were found in their mean age ($M = 14.4$ yrs vs. $M = 13.9$ yrs in the children and adolescents; and $M = 25.6$ yrs vs. $M = 25.3$ yrs in the adults),

Table 8.2. Occupational level of adult hypospadias patients ($N = 73$) and comparison subjects ($N = 50$)

Occupational level ¹⁹	Hypospadias patients ($N = 71$)		Comparison subjects ($N = 50$)	
	<i>N</i>	%	<i>N</i>	%
Unskilled manual professions	3	4.2	5	10.0
Skilled manual professions	19	26.8	11	22.0
Lower administrative and technical professions	17	23.9	6	12.0
Owners of small businesses	4	5.6	-	-
Higher administrative and technical professions	4	5.6	7	14.0
Higher professions, executives, owners of large businesses	6	8.5	5	10.0
Pupils, university students	18	25.4	16	32.0

Note. Difference between occupational level of hypospadias patients and comparison subjects is not statistically significant (pupils and university students excluded from analysis; Mann-Whitney *U* test, $z = -.07$).

religion, and urbanization. Also no significant differences were found between the parents' occupational level of patients and comparison subjects or between the occupational level of adult hypospadias patients and comparison subjects (table 8.2). Since no differences in these socioeconomic characteristics were found, statistical controlling for these variables was not performed.

8.3.3 GROUP EFFECTS

Tables 8.3 and 8.4 show the mean scores of respectively the DPQ, COI, and YASR for the adult subjects and the DPQ-J, SAS-C, YSR, and CBCL for the children and adolescents. ANOVAs with a 2, age (9-12 vs. 13-18 years or 18-24 vs. 25-38 years) x 2, group (hypospadias patients vs. comparison subjects) factorial design, revealed neither statistically significant overall group effects for the children and adolescents nor for the adults, except for the Total Anxiety Score from the COI and the Dominance scale from the DPQ-J (tables 8.3 and 8.4). The significant interactions between age and group for the Neuroticism and Social Inadequacy scales from the DPQ, and the Total Anxiety scale from the COI with the adult subjects (table 8.3) were analysed with Student's *t* tests. These analyses revealed that 25- to 38-year-old hypospadias patients had significantly higher scores than comparison subjects of the same age on the Neuroticism, Social Inadequacy, and Total Anxiety scales (p 's < .05). These scores did not differ between the 18- to 24-year-old hypospadias patients and comparison subjects. No statistically significant group effects were found on the eight CBCL/YSR/YASR syndromes (Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior).

8.3.4 AGE EFFECTS

The significant interactions between age and group for the Neuroticism, Social Inadequacy, and Total Anxiety scales with the adult subjects (table 8.3), were further analysed with Student's *t* tests. It was shown that there were significant age effects for the adult comparison subjects only (p 's <

Table 8.3. Mean scores (SD) of the DPQ, COI, and YASR measuring emotional functioning, social anxiety, and behavioral problems of adult hypospadias patients ($N = 73$) and comparison subjects ($N = 50$), by subjects' age (2×2 ANOVAs)

Instrument Scale (range)	Subjects aged 18 to 24 years		Subjects aged 25 to 38 years		age <i>F</i>	group <i>F</i>	age x group <i>F</i>
	Hypospadias patients ($N = 36$)	Comparison subjects ($N = 24$)	Hypospadias patients ($N = 37$)	Comparison subjects ($N = 26$)			
<i>DPQ</i>	$N = 31$	$N = 24$	$N = 35$	$N = 26$			
Dominance (0-34) ^a	17.4 (6.7)	17.1 (5.6)	17.2 (5.9)	19.1 (7.4)	0.55	0.44	0.84
Neuroticism (0-42) ^b	11.2 (8.8)	14.5 (8.3)	9.8 (6.0)	6.3 (5.2)	12.67*	0.01	6.50*
Social Inadequacy (0-30) ^b	8.6 (6.9)	9.7 (6.9)	8.9 (7.1)	4.8 (5.5)	3.34	1.32	4.19*
<i>COI</i>	$N = 30$	$N = 24$	$N = 35$	$N = 26$			
Total Frequency Score (35-175) ^a	112.4 (17.0)	108.5 (10.7)	119.1 (21.2)	114.0 (16.2)	3.54	1.94	0.04
Total Anxiety Score (35-175) ^b	74.8 (23.6)	66.4 (14.1)	66.7 (20.0)	56.6 (14.3)	5.68*	6.15*	3.91*
<i>YASR</i>	$N = 30$	$N = 21$	$N = 37$	$N = 25$			
Internalizing (0-62) ^b	9.6 (8.1)	10.0 (4.6)	8.2 (4.8)	6.6 (6.5)	4.03*	0.23	0.71
Externalizing (0-52) ^b	9.5 (4.5)	10.7 (4.6)	7.6 (4.0)	8.8 (4.7)	4.93*	2.04	0.00
Total Problem Score (0-220) ^b	34.8 (20.6)	38.4 (12.4)	29.1 (14.4)	27.7 (14.8)	6.61*	0.11	0.63

DPQ = Dutch Personality Questionnaire; COI = Contact with Others Inventory; YASR = Young Adult Self-Report.

^aHigher scores indicate better psychosocial functioning. ^bLower scores indicate better psychosocial functioning. *Significant effect ($p < .05$).

Table 8.4. Mean scores (SD) of the DPQ-J, SAS-C, CBCL, and YSR measuring emotional functioning, social anxiety, and behavioral problems of hypospadias patients aged 9 to 18 years ($N = 116$) and comparison subjects ($N = 88$), by subjects' age (2×2 ANOVAs)

Instrument Scale (range)	Subjects aged 9 to 12 years		Subjects aged 13 to 18 years		age <i>F</i>	group <i>F</i>	age x group <i>F</i>
	Hypospadias patients ($N = 39$)	Comparison subjects ($N = 25$)	Hypospadias patients ($N = 77$)	Comparison subjects ($N = 63$)			
<i>DPQ-J</i>	$N = 39$	$N = 24$	$N = 77$	$N = 63$			
Dominance (0-30) ^a	13.3 (4.7)	12.2 (4.9)	15.9 (4.6)	13.9 (4.7)	8.94*	4.35*	0.36
Neuroticism (0-56) ^b	12.4 (7.8)	11.8 (7.2)	12.8 (8.9)	11.1 (8.7)	0.01	0.79	0.13
Social Inadequacy (0-26) ^b	11.2 (4.9)	11.3 (5.5)	10.2 (5.5)	9.7 (5.0)	2.43	0.08	0.16
<i>SAS-C</i>	$N = 39$	$N = 25$	$N = 77$	$N = 62$			
Total Social Anxiety Score (0-46) ^b	6.5 (5.7)	4.7 (7.1)	6.7 (5.7)	7.5 (7.2)	2.38	0.22	1.72
<i>CBCL</i> ^c	$N = 37$	$N = 24$	$N = 77$	$N = 60$			
Internalizing (0-62) ^b	5.5 (4.9)	6.8 (5.4)	5.4 (5.3)	5.6 (5.3)	0.63	0.71	0.41
Externalizing (0-66) ^b	8.2 (7.5)	7.5 (5.8)	7.1 (7.0)	8.3 (8.1)	0.02	0.04	0.70
Total Problem Score (0-236) ^b	22.7 (16.9)	23.0 (14.6)	20.2 (17.6)	21.2 (17.7)	0.65	0.06	0.02
<i>YSR</i> ^d	$N = 20$	$N = 9$	$N = 77$	$N = 63$			
Internalizing (0-62) ^b	8.8 (6.9)	6.1 (3.5)	7.3 (4.5)	7.8 (6.6)	0.01	0.76	1.63
Externalizing (0-60) ^b	11.6 (6.3)	9.6 (4.9)	11.1 (6.0)	9.4 (6.5)	0.06	1.85	0.02
Total Problem Score (0-202) ^b	32.8 (17.6)	25.3 (8.7)	30.9 (15.2)	29.0 (17.6)	0.07	1.78	0.62

DPQ-J = Junior Dutch Personality Questionnaire; SAS-C = Social Anxiety Scale for Children; CBCL = Child Behavior Checklist; YSR = Youth Self-Report.

^aHigher scores indicate better psychosocial functioning. ^bLower scores indicate better psychosocial functioning. ^cCBCL was completed by the parents. ^dYSR was completed by subjects older than 10 years only. *Significant effect ($p < .05$).

.05): 18- to 24-year-old comparison subjects had higher scores than 25- to 38-year-old comparison subjects on the Neuroticism, Social Inadequacy, and Total Anxiety scales. Age (of adult patients as well as adult comparison subjects) was significantly related to the Internalizing, Externalizing and Total Problem Score from the YASR: older adult subjects had lower scores, which means that they had less behavioral/emotional problems than younger adult subjects (table 8.3). In the children and adolescents, age was significantly related to the Dominance scale from the DPQ-J: older subjects had higher scores, which means that they were more self-confident and they more often preferred to take initiative than younger subjects (table 8.4).

8.3.5 "SEX PROBLEMS" AND ENURESIS

The CBCL, YSR, and YASR contain several so-called "sex problem" items (6, 3, and 4 respectively; table 8.5). The CBCL also contains two items on enuresis. Table 8.5 shows the percentage of subjects who reported these problems to be at least sometimes or somewhat true. Chi-square tests and Fischer's exact tests revealed no significant differences in these separate sex problem and enuresis items between hypospadias patients and comparison subjects. A "sex problem syndrome" was computed by adding up the raw scores of the 6 sex problem items of the CBCL. Children and adolescents with hypospadias tended to have a higher mean score on this sex problem syndrome than the comparison boys ($M = 0.29$ vs. $M = 0.13$, $t(169.8) = -1.88$, $p = .061$).

8.3.6 PSYCHOSOCIAL FUNCTIONING OF HYPOSPADIAS PATIENTS RELATED TO MEDICAL CHARACTERISTICS

Of the adult patients, 57 (78.1%) had distal hypospadias and 16 patients (21.9%) had proximal hypospadias. These figures were 74 (63.8%) and 42 (36.2%) respectively for the children and adolescents with hypospadias. The mean number of operations to correct the hypospadias was 3.2 ($SD = 3.6$, range: 1-20) in the adult patients, and 2.4 ($SD = 1.4$, range: 1-8) in the patients between 9 and 18 years. The mean age at which surgical treatment was

Table 8.5. Percentage of subjects, who reported the sex problem and enuresis items of the CBCL, YSR, and YASR to be at least somewhat or sometimes true

Item	Subjects aged 9 to 18 years				Subjects aged 18 to 38 years	
	CBCL		YSR		YASR	
	Hypospadias patients (N = 115)	Comparison subjects (N = 85)	Hypospadias patients (N = 96)	Comparison subjects (N = 73)	Hypospadias patients (N = 67)	Comparison subjects (N = 48)
	%	%	%	%	%	%
Behaves like opposite sex	0.9	2.4	3.1	4.1	3.0	0.0
Wishes to be of opposite sex	0.0	0.0	2.1	2.8	3.0	4.2
Plays with own sex parts in public	4.3	2.4	-	-	-	-
Plays with own sex parts too much	5.3	3.6	-	-	-	-
Sexual problems	3.5	0.0	-	-	-	-
Thinks about sex too much	9.7	6.0	37.5	39.7	65.7	72.9
Wets self during the day	4.4	1.2	-	-	-	-
Wets the bed	9.6	5.9	-	-	-	-
Worries about relations with the opposite sex	-	-	-	-	29.9	25.0

CBCL = Child Behavior Checklist; YSR = Youth Self-Report; YASR = Young Adult Self-Report.

Note. There are no statistically significant differences between hypospadias patients and comparison subjects (chi-square tests, Fischer's exact tests).

Chapter 8

completed was 9.7 years ($SD = 6.4$, range: 1.2-33.5 yrs) in the adult patients and 6.3 years ($SD = 3.4$, range: 1.1-13.8 yrs) in the children and adolescents.

Table 8.6. Partial correlation coefficients, controlling for subjects' age between genital and body perception, and psychosocial functioning

Instrument Scale	Hypospadias patients aged 9 to 18 years ($N = 116$)		Hypospadias patients aged 18 to 38 years ($N = 73$)	
	GPS	BPS	GPS	BPS
<i>DPQ-J/DPQ</i>	$df = 113$	$df = 112$	$df = 61$	$df = 61$
Dominance	-.21**	-.22**	-.24**	-.02
Neuroticism	-.25**	-.28**	-.04	-.28**
Social Inadequacy	-.05	-.28**	.00	-.22**
<i>COI</i>			$df = 59$	$df = 60$
Total Frequency Score	-	-	.20*	.34**
Total Anxiety Score	-	-	-.04	-.15
<i>SAS-C</i>	$df = 113$	$df = 112$		
Total Social Anxiety Score	-.13*	-.29**	-	-
<i>YSR/YASR</i>	$df = 94$	$df = 94$	$df = 61$	$df = 61$
Internalizing	-.29**	-.53**	-.19*	-.35**
Externalizing	-.32**	-.36**	.03	-.09
Total Problem Score	-.38**	-.48**	-.17*	-.31**
<i>CBCL</i>	$df = 107$	$df = 106$		
Internalizing	-.10	-.29**	-	-
Externalizing	-.07	-.19**	-	-
Total Problem Score	-.11	-.25**	-	-

GPS = Genital Perception Score; BPS = Body Perception Score; DPQ = Dutch Personality Questionnaire; DPQ-J = Junior Dutch Personality Questionnaire; COI = Contact with Others Inventory; SAS-C = Social Anxiety Scale for Children; CBCL = Child Behavior Checklist; YSR = Youth Self-Report; YASR = Young Adult Self-Report.

* $p < .10$. ** $p < .05$ (one-tailed probabilities).

Student's *t* tests indicated no statistically significant differences in the mean scores of the questionnaires used between patients with distal and proximal hypospadias. No statistically significant Spearman's rank correlation coefficients were found between the number of operations, the age at which surgical treatment was finished, and the scores of the questionnaires used. Also no significant differences emerged in mean scores of the questionnaires used between patients treated with ventralizing and terminalizing operation techniques. In summary, no statistically significant relationships of the previously mentioned medical characteristics with psychosocial functioning of hypospadias patients could be discerned.

8.3.7 PSYCHOSOCIAL FUNCTIONING OF HYPOSPADIAS PATIENTS RELATED TO GENITAL/BODY PERCEPTION

Mureau and colleagues³⁰ reported that children and adolescents as well as adults with hypospadias had significantly lower Genital Perception Scores (GPS) than comparison subjects (i.e., they were less satisfied with their penile appearance). Besides, adult hypospadias patients had a significantly higher Body Perception Score (BPS), they were more satisfied with their body appearance than comparison subjects. Children and adolescents with hypospadias did not differ from comparison boys with respect to the BPS.

In table 8.6 partial correlation coefficients, controlling for subjects' age, between the GPS/BPS and the DPQ/DPQ-J, COI/SAS-C, and CBCL/YSR/YASR are listed. All significant correlation coefficients are small to moderate. Correlation coefficients between the GPS and the other questionnaires are smaller than those between the BPS. Except for the correlation coefficients between the Dominance scale of the DPQ and DPQ-J, all significant correlations seem to indicate that the less negative the genital or body perception of hypospadias patients was, the better their psychosocial functioning.

8.4 DISCUSSION

The main results of this study can be summarized as follows. Hypospadias patients did not have a poorer psychosocial adjustment than age-matched comparison subjects, although 25- to 38-year-old hypospadias patients reported more vague somatic complaints and anxieties, more feelings of inadequacy, and more social anxieties. Neither the severity of hypospadias nor the various medical characteristics were significantly related to psychosocial functioning. Hypospadias patients who were satisfied with their genitals and body reported a somewhat better psychosocial functioning than patients who were less satisfied. The results of the present study differ in several aspects from other comparative investigations which reported poorer psychosocial functioning of patients following urogenital surgery.^{5, 9-11}

Adult hypospadias patients in the present study did not have less qualified professions than comparison subjects, which contradicts the results from Berg and associates who found that adult hypospadias patients tended to be less competitive in their choice of profession than comparison subjects.³¹ Furthermore, hypospadias patients in the present study (except the 25- to 38-year-old patients) showed similar psychosocial functioning as comparison subjects. Berg and collaborators^{9, 10} reported that adult hypospadias patients aged 21 to 34 years were more anxious, had less self-esteem, and had reduced capacity for social and emotional relationships than comparison subjects. The latter finding corroborates our findings where 25- to 38-year-old hypospadias patients had more vague somatic complaints and anxieties, had more feelings of inadequacy, and were more socially anxious. The above mentioned difference between 25- to 38-year-old hypospadias patients and comparison subjects need some discussion, since age did not significantly affect the Neuroticism, Social Inadequacy, and Social Anxiety scales in the *normative samples*.^{20, 22} Maybe in the present study the 25- to 38-year-old comparison subjects are a somewhat biased group, since only 43% of the selected older comparison subjects actually participated in the present study. Thus, one could hypothesize that these subjects were less prone to have vague somatic complaints and anxieties, feelings of inadequacy, and social anxieties, and therefore had lower scores.

In the present study no differences in psychosocial functioning between 9 to 18 year old hypospadias patients and comparison boys were found, which differs from the results of Berg and co-workers who found that adult hypospadias patients reported they had been more timid, isolated, and mobbed as children.^{9, 10} These results are hard to compare, however, since they were retrospectively inferred by the investigators in the latter studies, contrary to the present study in which children and adolescents completed questionnaires themselves.

Hypospadias patients (children and adolescents as well as adults) of the present study did not report more behavioral or emotional problems than comparison subjects, contrary to Sandberg and colleagues who reported that hypospadias patients (6 to 10 years) had higher mean scores on the Child Behavior Checklist (CBCL) than subjects from a normative sample.¹¹ Sandberg and associates used *normative* data for the CBCL and there was no match for socioeconomic background between hypospadias patients and nonclinical subjects, contrary to the present study. In a meta-analytic review of 87 investigations about children's psychological adjustment to physical disorders, Lavigne and Faier-Routman³² found that comparisons with *norms* for outcome measures tended to produce larger effect sizes than did comparisons with *study controls* (as in the present study). Furthermore, since any case that received mental health service for at least the year preceding the completion of the CBCL was excluded from the normative sample which was used in the study of Sandberg and co-workers,¹¹ the reported differences between hypospadias patients and the normative sample might have been inflated. However, Sandberg and collaborators concluded that hypospadias is not generally associated with a marked increase of significant behavioral problems *during childhood*, since they could not find a statistically significant difference in the number of patients and comparison subjects that fell within the clinical range for the CBCL.¹¹ The results of the present study confirm their conclusion and indicate further that generally hypospadias patients do not have more behavioral/emotional problems than comparison subjects during adolescence or adulthood.

Although a substantial number of children and adolescents with hypospadias still suffered from diurnal (4.4%) or nocturnal enuresis (9.7%), this did not differ significantly from the comparison subjects in the present study. This finding contradicts results of previous investigations which reported that hypospadias patients more often suffered from enuresis during childhood.^{9, 11} Maybe the latter results differ from the present study because the occurrence of enuresis was retrospectively assessed in the study from Berg and colleagues⁹ and the hypospadias patients investigated by Sandberg and associates¹¹ were much younger (between 6 and 10 years) than the young patients of the present study (between 9 and 18 years).

Although sex problems were not encountered significantly more often in children and adolescents with hypospadias than in comparison boys, the first tended to have higher mean scores on the "sex problem syndrome" of the CBCL than the latter. Since the CBCL was completed by the parents, the latter finding might indicate that parents of hypospadias patients worried more about their son's sexuality than parents of comparison boys.

No significant effects of severity of hypospadias could be found. These findings corroborate the results of the study from Berg and collaborators,^{9, 10} but differ from Sandberg and colleagues¹¹ who found that patients with the more severe forms of hypospadias tended to have higher scores on the CBCL Hyperactive, Delinquent Behavior, and Externalizing scales. No significant relationships of the number of operations, the age at which surgery was finished, and different surgical procedures with general psychosocial functioning were found in the present study. These latter findings do not necessarily mean that improvements of medical treatment have not been worthwhile, since we previously found that surgical treatment with fewer operations at earlier ages was positively associated with genital perception of hypospadias patients,³⁰ and that patients with a terminal glanular meatus were more satisfied with the meatal position than patients with a coronal meatus.³³

Although all significant correlations between psychosocial functioning and genital/body perception (except for the Dominance scales) were in the expected direction, they were rather small. Nonetheless, they seem to indica-

te that the less negative genital/body perception was in hypospadias patients, the better their psychosocial functioning. In other words, coping with the somewhat different penile appearance seems to be positively associated with psychosocial functioning of hypospadias patients. These findings are in line with another study which showed that males who were satisfied with their physical appearance had more favorable psychological qualities (they had a better self-concept, were less neurotic, and more extravert) than males who were dissatisfied with their physical appearance.³⁴

We found previously that hypospadias patients were more dissatisfied with their penile appearance,³⁰ more often had received comments on their penile appearance, and were more self-conscious and embarrassed about their penile appearance, which caused them to be more inhibited with seeking sexual contacts than comparison subjects.^{35, 36} On the other hand, our previous results also showed that hypospadias patients did not report a psychosexual adjustment, sexual behavior, and sexual functioning different from comparison subjects.^{35, 36} These earlier findings, together with the results of the present study suggest that *in general* males who were operated for hypospadias can expect to have a normal psychosexual and psychosocial development. Like Sandberg and co-workers,¹¹ however, we want to emphasize that these conclusions do not rule out that there are individual cases who are vulnerable to problems of adjustment and to whom professional psychological help should be given. It would be worthwhile to carry out a prospective study which investigates patients and parents pre- and postoperatively, so that risk factors for poor adjustment following hypospadias surgery can be determined. In conclusion, the finding that generally hypospadias patients can expect a normal psychosexual and psychosocial development is important for pediatric urologists and plastic surgeons in the counseling process of hypospadias patients and their parents.

ACKNOWLEDGEMENTS

This project was financially supported by the Sophia Foundation for Medical Research.

8.5 REFERENCES

1. Levitt, S. B., & Reda, E. F. (1988). Hypospadias. *Pediatric Annals*, 17 (1), 48-57.
2. Van der Meulen, J. C. (1985). Hypospadias. In F. K. Muir (Ed.), *Current operative surgery: Plastic and reconstructive* (pp. 119-146). London: Ballière and Tindall.
3. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
4. Robertson, M., & Walker, D. (1975). Psychological factors in hypospadias repair. *Journal of Urology*, 113, 698-700.
5. Blotcky, M. J., & Grossman, I. (1978). Psychological implications of childhood genitourinary surgery. *Journal of the American Academy of Child Psychiatry*, 17, 488-497.
6. Lepore, A. G., & Kesler, R. W. (1979). Behavior of children undergoing hypospadias repair. *Journal of Urology*, 122, 68-70.
7. Cogan, S. F., Becker, R. D., & Hofmann, A. D. (1975). Adolescent males with urogenital anomalies: Their body image and psychosexual development. *Journal of Youth and Adolescence*, 14 (4), 359-373.
8. Kipikaša, A., Longauer, F., & Urbanová, E. (1979). Hypospadias in relation to some clinical and psychosocial problems in adulthood. *Acta Chirurgicae Plasticae*, 21, 228-235.
9. Berg, R., Berg, G., & Svensson, J. (1982). Penile malformation and mental health. A controlled psychiatric study of men operated for hypospadias in childhood. *Acta Psychiatrica Scandinavica*, 66, 398-416.
10. Berg, G., & Berg, R. (1983). Castration complex. Evidence from men operated for hypospadias. *Acta Psychiatrica Scandinavica*, 68, 143-153.
11. Sandberg, D. E., Meyer-Bahlburg, H. F. L., Aranoff, G. S., Sconzo, J. M., & Hensle, T. W. (1989). Boys with hypospadias: A survey of behavioral difficulties. *Journal of Pediatric Psychology*, 14 (4), 491-514.
12. Tolhurst, D. E. (1989). A standard method for the correction of hypospadias. *British Journal of Plastic Surgery*, 42, 638-644.
13. Zaontz, M. R. (1989). The GAP (glans approximation procedure) for glanular/coronal hypospadias. *Journal of Urology*, 141, 359-361.
14. Duckett, J. W. (1981). MAGPI (meatoplasty and glanuloplasty). A procedure for subcoronal hypospadias. *Urologic Clinics of North America*, 8 (3), 513-519.
15. Koff, S. A. (1981). Mobilisation of the urethra in surgical treatment of hypospadias. *Journal of Urology*, 125, 394-397.
16. Mathicu, P. (1932). Traitement en un temps l'hypospadias balanique et juxtabalanique [One-stage correction of glanular and coronal hypospadias]. *Journal de Chirurgie*, 39, 481-486.

17. Duckett, J. W. (1981). The island flap technique for hypospadias repair. *Urologic Clinics of North America*, 8 (3), 503-511.
18. Perović, S. (1981). Operationsprinzip bei der penilen Hypospadie [Principles for the treatment of penile hypospadias]. *Aktuelle Urologie*, 12 (suppl.), 78-83.
19. Van Westerlaak, J. M., Kropman, J. A., & Collaris, J. W. M. (1975). *Beroepenklapper* [Professional index]. Nijmegen, The Netherlands: Instituut voor Toegepaste Sociologie.
20. Luteijn, F., Starren, J., & Van Dijk, H. (1975). *Handleiding bij de Nederlandse Persoonlijkheds Vragenlijst* [Manual for the Dutch Personality Questionnaire]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
21. Luteijn, F., Van Dijk, H., & Van der Ploeg, F. A. E. (1989). *Handleiding bij de Junior Nederlandse Persoonlijkheds Vragenlijst, herziene uitgave* [Manual for the Junior Dutch Personality Questionnaire, revised edition]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
22. Van Dam-Baggen, C. M. J., & Kraaimaat, F. W. (1987). *Handleiding bij de Inventarisatielijst Omgaan met Anderen* [Manual for the Contact with Others Inventory]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
23. Dekking, Y. M. (1982). *Handleiding bij de Sociale Angstschaal voor Kinderen* [Manual for the Social Anxiety Scale for Children]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
24. Achenbach, T. M. (1991). *Integrative guide for the 1991 CBCL/4-18, YSR, and TRF profiles*. Burlington, VT: University of Vermont, Department of Psychiatry.
25. Achenbach, T. M. (1990). *The Young Adult Self-Report*. Burlington, VT: University of Vermont, Department of Psychiatry.
26. Verhulst, F. C., Akkerhuis, G. W., & Althaus, M. (1985). Mental health in Dutch children: (I) A cross-cultural comparison. *Acta Psychiatrica Scandinavica*, 72, (suppl.323).
27. Verhulst, F. C., Prince, J., Vervuurt-Poot, C., & De Jong, J. (1989). Mental health in Dutch adolescents: self-reported competencies and problems for ages 11-18. *Acta Psychiatrica Scandinavica*, 80, (suppl. 356).
28. Ferdinand, R. F., & Verhulst, F. C. (1994). The prediction of poor outcome in young adults: Comparison of the Young Adult Self-Report, the General Health Questionnaire and the Symptom Checklist. *Acta Psychiatrica Scandinavica*, 89, 405-410.
29. Ferdinand, R. F., Verhulst, F. C., & Wiznitzer, M. (1995). Continuity and change of self-reported problem behaviors from adolescence into young adulthood. *Journal of the American Academy of Child and Adolescent Psychiatry*, 34, 680-690

30. Mureau, M. A. M., Slijper, F. M. E., Slob, A. K., & Verhulst, F. C. (1995). Genital perception of children, adolescents, and adults operated on for hypospadias: A comparative study. *Journal of Sex Research, in press.*
31. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology, 125*, 313-317.
32. Lavigne, J. V., & Faier-Routman, J. (1992). Psychological adjustment to pediatric physical disorders: A meta-analytic review. *Journal of Pediatric Psychology, 17*, 133-157.
33. Mureau, M. A. M., Slijper, F. M. E., Slob, A. K., Verhulst, F. C., & Nijman, R. J. M. (1995). Satisfaction with penile appearance following hypospadias surgery: The patients' and the surgeon's view. *Journal of Urology, in press.*
34. Tucker, L. A. (1984). Physical attractiveness, somatotype, and the male personality: A dynamic interactional perspective. *Journal of Clinical Psychology, 40*, 1226-1234.
35. Mureau, M. A. M., Slijper, F. M. E., Nijman, R. J. M., Van der Meulen, J. C., Verhulst, F. C., & Slob, A. K. (1995). Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study. *Journal of Urology, in press.*
36. Mureau, M. A. M., Slijper, F. M. E., Van der Meulen, J. C., Verhulst, F. C. & Slob, A. K. (1995). Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study. *Journal of Urology, in press.*

CHAPTER 9

EVALUATION AND RECOMMENDATIONS

9.1 INTRODUCTION

The objective of the present investigation was to study the psychosexual and psychosocial adjustment, and genital perception of children, adolescents, and adults following hypospadias surgery, compared to age-matched subjects operated for an inguinal hernia. In addition, effects of various medical characteristics (severity of hypospadias, age at final surgery, number of operations, surgical procedures), subjects' age, and coping with hypospadias on psychosexual and psychosocial adjustment, and genital perception of hypospadias patients were also studied.

The clinical consequences of the findings of the present study, some methodological considerations, and recommendations for future research will be presented in the next six paragraphs.

9.2 CLINICAL CONSEQUENCES

9.2.1 COUNSELING OF PATIENTS AND PARENTS

All surgical techniques performed on the hypospadias patients in the present study resulted in a circumcised penile appearance. Because the surgical hypospadias corrections not always produced a glanular terminal meatus, it is a striking and important finding that the circumcised appearance had a greater impact on genital perception than the (abnormal) position of the meatus. About 48% of the children and adolescents and 60% of the adults were self-conscious about the circumcised appearance of their penis. Patients who were satisfied with their circumcised status had a more positive genital perception (approximately the same as that of comparison subjects) than patients who were dissatisfied with their circumcised status. Only two other European studies on the long-term effects of hypospadias surgery reported that hypospadias patients felt embarrassed about the circumcised appearance

of their penis.^{1, 2} There are several explanations for the patients' self-consciousness about the circumcised penile appearance. (1) Since in The Netherlands circumcision is uncommon among Dutch boys, patients may have feared that the circumcised appearance reveals their hypospadias. (2) Hypospadias patients may have perceived the *loss* of foreskin following surgery as a *defect* of their penis. (3) Many patients, especially children, may have considered their circumcised penis smaller than an uncircumcised penis; presumably because the absence of a foreskin produces optically a shorter penis. Therefore, we advocate to inform parents and patients (from the age of about 4 years) that following hypospadias surgery the glans will be exposed permanently because the prepuce is missing; that circumcision is a normal procedure in many cultures and is usually performed for non-medical reasons (e.g., religion, hygiene); and that a circumcised *hypospadiac* penis looks almost identical as a circumcised *normal* penis. We believe that this information will help patients and/or parents to accept the somewhat different appearance of the circumcised penis. However, there might be a new possibility. Recently, a surgical procedure has been developed to reconstruct the prepuce, if feasible (e.g., Kropfl, Schardt, & Fey³). Especially in countries where circumcision is uncommon, such foreskin preserving surgery should be offered to those parents and patients who pre-operatively have (extreme) difficulties with circumcision. Although it is not possible in all cases, it is a feasible alternative especially in young children with distal hypospadias.

In the present study, more patients than comparison subjects were dissatisfied with flaccid penile size. Actually, flaccid penile size was a major motive for dissatisfaction in hypospadias patients. This is in line with the results of earlier investigations.⁴⁻⁷ Significantly more children and adolescents with hypospadias (21%) fell below the 10th percentile for stretched penile length as reported by Schonfeld and Beebe.⁸ There was neither a significant correlation between penile length and penile satisfaction, nor a significant difference in satisfaction with penile size between patients who fell below the 10th percentile and patients who fell above the 10th percentile. Because penile size was a major motive for concern, parents and patients

should be informed that hypospadias surgery will not enlarge penile size. Patients who post-operatively have psychological difficulties with accepting their (small) penis should be offered professional psychological or sexological help.

Many adult hypospadias patients and parents had questions or worries about the incidence and inheritance of hypospadias, and about the probability of being infertile. Hypospadias surgeons should always inform parents and patients properly about these matters, even when patients or parents do not (dare to) bring up these subjects.

Neither diurnal nor nocturnal enuresis was encountered significantly more often among the 9- to 18-year-old hypospadias patients than among age-matched comparison boys. Parents may relate the occurrence of enuresis to the hypospadias. They can now be informed and reassured that this is not necessarily the case.

In the present study, no clear evidence was found that in general hypospadias patients (children and adolescents as well as adults) had a poorer psychosexual or psychosocial adjustment than age-matched comparison subjects. We believe this is very relevant information for plastic surgeons and pediatric urologists when counseling parents and patients. The information that this study does not provide indications to expect poorer sexual development or functioning, nor poorer psychosocial functioning than males without hypospadias, should diminish parental anxieties and should help patients to cope better with their hypospadias. However, this does not rule out the possibility that a small number of patients might have problems with psychosexual or psychosocial adjustment, to whom professional psychological or sexological help should be given.

In conclusion, we advocate to inform parents and patients, if they are old enough, about the previously mentioned topics during the first visit prior to the (first) operation. Ideally, parents and patients should be given a leaflet in which all information is described in comprehensible (i.e., non-medical) language, so that they can read all information once again at leisure. Furthermore, photographs of *average* post-operative results (next to the pre-opera-

tive penile appearance) should be depicted in the leaflet, so that parents and patients will also be able to *see* what they can expect.

9.2.2 TREATMENT OF HYPOSPADIAS PATIENTS WITH A MICROPENIS

The recommendations of the present paragraph are mainly based upon the results of earlier investigations of other scientists.⁹⁻¹⁸ Treatment and counseling of hypospadias patients with a micropenis (< 2.5 *SDs* below the mean) is dependent on the age of the patient. If a patient with a micropenis is presented shortly after birth, human chorionic gonadotropin intramuscularly should be given first to assess androgen sensitivity to penile growth.⁹ If there is no significant growth of the penis, *early* surgical sex reassignment should be considered seriously.⁹ If the patient is sensitive to HCG stimulation, he should be treated with hormones (locally or intramuscularly administered testosterone enanthate or propionate^{10, 11} or locally administered dihydrotestosterone¹²). Presumably it is best to start hormonal treatment during puberty. Follow-up studies on patients with a micropenis showed that early (pre-pubertal) hormonal therapy only brought about premature penile growth and that the final adult penile length remained underdeveloped.^{13, 14} Recent studies with humans¹² and with rats¹⁵ revealed that hormonal therapy during puberty produced a better increase in penile size than hormonal treatment before puberty.

Adolescent or adult patients with hypospadias with a micropenis should be counseled properly: they can be informed that it is possible for men with a micropenis to have a satisfactory sexual relationship.^{14, 16} If psychological or sexological counseling should not help and the patient keeps suffering from severe psychological distress, it might be considered to offer phallic reconstruction^{17, 18} or penile lengthening.

9.2.3 NEED TO FOLLOW-UP HYPOSPADIAS PATIENTS

When specifically asked for, about 38% of the hypospadias patients in the present study (children and adolescents as well as adults) expressed a wish for a functional or cosmetic penile improvement. Of the 73 adult patients, 5 (7%) were re-operated following the interview. For spraying of urine in 3

cases, for a fistula in 1 case, and in another case for penile skin tethering that caused dyspareunia. Of the 35 children and adolescents treated with terminalizing repairs, 4 (11%) were re-operated because of a poor result. Because of a (iatrogenic) lateral penile curvature in 2 cases, a fistula in 1 case, and a retracted coronal meatus in another case. It is of particular interest that all of these patients stated that they would not have readily consulted a physician for their problems on their own initiative. Embarrassment or fear for (another) operation may have accounted for this reluctance to seek professional help.

Although one cannot exclude the possibility that somewhat more hypospadias patients with problems participated in the present study than patients without problems, we strongly advocate - like other investigators^{4, 5, 19} - that hypospadias surgeons should see their patients during adolescence at least once as a standard therapeutic procedure. (1) Hypospadias patients may be reluctant to seek medical advice on their own initiative, even when they are experiencing considerable difficulties; (2) boys with hypospadias are treated at increasingly earlier ages (often before 18 months) and therefore are not able to express their (dis)satisfaction with the surgical result; (3) in the present study adolescents with hypospadias (13-18 yrs) more often wanted cosmetic or functional penile improvement than children with hypospadias (9-12 yrs); (4) complications, such as meatal retraction or stenosis, residual penile curvature, or adult sexual dysfunction often become apparant many years after surgical treatment is finished; and (5) in the present study many adult hypospadias patients had questions or worries, mainly about the incidence and heredity of hypospadias, and the probability of being infertile.

During the follow-up visit of the adolescent patient, the surgeon should explicitly ask whether or not the patient is satisfied with his penile appearance. If the patient is not satisfied, the surgeon should inquire about the motive for dissatisfaction, and should inform the patient whether or not it would be feasible to further surgically improve penile appearance. In addition, the surgeon should explicitly inquire about the patient's questions or worries concerning sexuality, since the patient himself might not dare to bring up the subject. If the patient has worries concerning his sexuality, the surgeon

should inform him that it is not abnormal to have such worries, and he or she should answer the patient's questions properly. The results of this study support the surgeon who now can reassure and inform the patient that his hypospadias does not (need to) have negative consequences for his sexual development or sexual functioning.

9.2.4 IMPLICATIONS FOR SURGICAL TREATMENT OF HYPOSPADIAS PATIENTS

Only in adult patients the age at final surgery negatively affected sexual adjustment and the level of inhibition with seeking sexual contacts. Small positive correlations were found between the number of operations and the level of inhibition with seeking sexual contacts in adults as well as children and adolescents with hypospadias. The number of operations and the age at final surgery correlated negatively with the Genital Perception Score only in adult patients. No statistically significant effects of the number of operations and the age at final surgery on psychosocial functioning were found. The negative effects of surgical treatment found in adult patients, could be derived from the later age at final surgery ($M = 9.7$ yrs; range 1.2-33.5 yrs vs. $M = 6.3$ yrs; range 1.1-13.8 yrs) and the higher number of operations ($M = 3.2$; range: 1-20 vs. $M = 2.4$; range: 1-8). It seems preferable that surgery is performed at an early age with as few operations as possible.

No differences were found in psychosexual adjustment, genital perception, and psychosocial functioning of patients treated with different surgical procedures (Van der Meulen vs. Byars/Browne procedure with the adults, and ventralizing vs. terminalizing techniques with the children and adolescents). Children and adolescents treated with ventralizing procedures tended to express more often the wish for penile improvement than those treated with terminalizing techniques. Patients who had a glanular terminal meatus were more satisfied with their meatal position than patients with a retracted coronal meatus. Because of these latter findings and because an important goal for hypospadias surgeons is to correct the hypospadiac penis as perfectly as possible, a terminalizing procedure is preferred. However, the results of the present study also indicate that a ventralizing procedure can be perfor-

med safely (e.g., in countries with less specialized health care), without negatively affecting psychosexual and psychosocial adjustment.

9.3 METHODOLOGICAL CONSIDERATIONS

The response rates of the comparison subjects might seem rather low (52.1% in the adults, and 57.5% in the children and adolescents), but are in fact very similar to the response rates of four representative national surveys on sexuality in the Dutch population (46.9% to 58.3%).²⁰

The validity and reliability of the semi-structured interviews used in chapters 4 and 5 were unknown. Since the results on genital appraisal of the semi-structured interviews (chapters 4 and 5) corroborate the results on genital perception, which were derived from standardized questionnaires with good reliability (chapter 6), we have confidence in the interview results. Also, the interview data on psychosexual adjustment of the adult subjects (chapter 4) very much resembled those on psychosexual adjustment of children and adolescents (chapter 5). Finally, because 87% of the children and adolescents and 74% of the adult subjects were personally interviewed (the others filled out all questionnaires at home and returned them by mail), it was possible to clarify contradictory answers of most subjects.

The genital perception scale items for the children and adolescents were 4-point scales, in which a neutral answer (neither satisfied nor dissatisfied) was omitted. In the genital perception scale for the adults 5-point scales were used including a neutral answer. This methodological imperfection makes it impossible to study the age effect on genital perception by comparing the genital perception scores of the children and adolescents with those of the adults.

In light of the above mentioned methodological considerations, we have confidence in our results, since psychosexual adjustment, genital perception, and psychosocial functioning were assessed in a standardized manner; comparison subjects were used; fairly large number of subjects were investigated; validity and reliability of most instruments used were acceptable;²¹⁻²⁹ and a sufficiently heterogeneous group of patients treated with different surgical procedures was investigated.

9.4 RECOMMENDATIONS FOR FUTURE RESEARCH

In the Sophia Children's Hospital terminalizing procedures to correct hypospadias were not performed until the early eighties. It was therefore impossible to select a group of adult patients treated with these techniques. A study which investigates psychosexual adjustment, genital perception, and psychosocial adjustment of adult patients treated with terminalizing repairs will be necessary to determine whether or not our findings in children and adolescents will also be true for adult patients.

Although the results from the present study seem to indicate that it is preferable to perform genital surgery before the age of 3 years (see chapter 5), proper empirical knowledge on the optimal timing of hypospadias surgery is still lacking. Therefore, a study that compares psychosexual adjustment and genital perception of patients who underwent surgery between 6 and 18 months, between 18 months and 3 years, and patients who underwent surgery at a later age, keeping variables as surgical procedure and severity of hypospadias constant, will be necessary to determine the optimal timing of hypospadias surgery.

Factors which might be negatively associated with genital perception of hypospadias patients, such as unrealistic expectations of the cosmetic results of surgery, the parents' anxieties about their son's masculine capabilities, or the parents' negative perception of their son's penile appearance, were not investigated in the present study. It would be worthwhile to carry out a prospective study which investigates these factors in patients and parents pre- and post-operatively, so that information will be obtained to improve the counseling of parents and patients with hypospadias.

Because a very important motive for dissatisfaction with penile appearance appeared to be penile size and reliable information on penile development of hypospadias patients is still lacking at the present, a longitudinal study which investigates penile development of hypospadias patients and age-matched normal comparison subjects should be carried out. Ideally, stretched penile length should be measured from the pubic bone to the tip of the penis with the subject in a supine position.

9.5 CONCLUDING REMARKS

The results of this work should be implemented by surgeons, psychologists, and sexologists in their care of hypospadias patients. Then, a major objective of this thesis has been accomplished and the hypospadias patients' and comparison subjects' efforts in participating in the present study have been worthwhile.

9.5 REFERENCES

1. Heiss, W. H., & Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadiie-Operationen [Long-term results following hypospadias surgery]. *Zeitschrift für Kinderchirurgie*, 14 (4), 445-451.
2. Helmig, F. J. (1974). Langzeitergebnisse nach Hypospadiie-Operationen [Long-term results following hypospadias surgery]. *Klinische Pädiatrie*, 186, 421-425.
3. Kropfl, D., Schardt, M., & Fey, S. (1992). Modified meatal advancement and glanuloplasty with complete foreskin reconstruction. *European Urology*, 22 (1), 57-61.
4. Bracka, A. (1989). A long-term view of hypospadias. *British Journal of Plastic Surgery*, 42, 251-255.
5. Eberle, J., Überreiter, S., Radmayr, C., Janetschek, G., Marberger, H., & Bartsch, G. (1993). Posterior hypospadias: Long-term followup after reconstructive surgery in the male direction. *Journal of Urology*, 150, 1474-1477.
6. Ericsson, N. O., & Von Hedenberg, C. (1971). Sexualfunktion hos patienter operade för hypospadi [Sexual functioning of patients operated for hypospadias]. *Läkartidningen*, 68 (21), 2480-2484.
7. Berg, R., Svensson, J., & Åström, G. (1981). Social and sexual adjustment of men operated for hypospadias during childhood: A controlled study. *Journal of Urology*, 125, 313-317.
8. Schonfeld, W. A., & Beebe, G. W. (1942). Normal growth and variation in the male genitalia from birth to maturity. *Journal of Urology*, 48, 759-777.
9. Aaronson, I. A. (1994). Micropenis: Medical and surgical implications. *Journal of Urology*, 152, 4-14.
10. Burstein, S., Grumbach, M. M., & Kaplan, S. L. (1979). Early determination of androgen-responsiveness is important in the management of micropallus. *The Lancet*, 2, 83-986.
11. Guthrie, R. D., Smith, D. W., & Graham, C. B. (1973). Testosterone treatment for micropenis during early childhood. *Journal of Pediatrics*, 83 (2), 247-252.

12. Choi, S. K., Han, S. W., Kim, D. K., & De Lignieres, B. (1993). Transdermal dihydrotestosterone therapy and its effects on patients with microphallus. *Journal of Urology*, *150*, 657-660.
13. Money, J., Lehne, G. K., & Pierre-Jerome, F. (1984). Micropenis: Adult follow-up and comparison against new norms. *Journal of Sex & Marital Therapy*, *10* (2), 105-116.
14. Reilly, J. M., & Woodhouse, C. R. J. (1989). Small penis and male sexual role. *Journal of Urology*, *142*, 569-571.
15. Husmann, D. A., & Cain, M. P. (1994). Microphallus: Eventual phallic size is dependent on the timing of androgen administration. *Journal of Urology*, *152*, 734-739.
16. Van Seters, A. P., & Slob, A. K. (1988). Mutually satisfying heterosexual relationship with micropenis of husband. *Journal of Sex & Marital Therapy*, *14*, 98-107.
17. Gilbert, D. A., Jordan, G. H., Devine C. J. Jr., Winslow, B. H., & Schlossberg, S. M. (1993). Phallic construction in prepubertal and adolescent boys. *Journal of Urology*, *149*, 1521-1526.
18. Byun, J. S., Cho, B. C., & Baik, B. S. (1994). Results of one-stage penile reconstruction using an innervated radial osteocutaneous flap. *Journal of Reconstructive Microsurgery*, *10*, 321-331.
19. Schubert, J., Kelly, L. U., & Trinckauf H. H. (1989). Der einfluß plastischer Korrekturmaßnahmen bei Hypospadias penis auf das Sexualverhalten im fertilen Lebensalter [The effects of hypospadias surgery on sexual functioning in adulthood]. *Zeitschrift für Urologie und Nephrologie*, *82*, 121-125.
20. Van Zessen, G., & Sandfordt, Th. G. M. (1991). *Seksualiteit in Nederland. Seksueel gedrag, risico en preventie van AIDS* [Sexuality in The Netherlands. Sexual behavior, risk, and AIDS prevention]. Lisse, The Netherlands: Swets & Zeitlinger, B. V.
21. Luteijn, F., Starren, J., & Van Dijk, H. (1975). *Handleiding bij de Nederlandse Persoonlijkheds Vragenlijst* [Manual for the Dutch Personality Questionnaire]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
22. Luteijn, F., Van Dijk, H., & Van der Ploeg, F. A. E. (1989). *Handleiding bij de Junior Nederlandse Persoonlijkheds Vragenlijst, herziene uitgave* [Manual for the Junior Dutch Personality Questionnaire, revised edition]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
23. Van Dam-Baggen, C. M. J., & Kraaimaat, F. W. (1987). *Handleiding bij de Inventarisatielijst Omgaan met Anderen* [Manual for the Contact with Others Inventory]. Lisse, The Netherlands: Swets & Zeitlinger B. V.
24. Dekking, Y. M. (1982). *Handleiding bij de Sociale Angstschaal voor Kinderen* [Manual for the Social Anxiety Scale for Children]. Lisse, The Netherlands: Swets & Zeitlinger B. V.

25. Verhulst, F. C., Akkerhuis, G. W., & Althaus, M. (1985). Mental health in Dutch children: (I) A cross-cultural comparison. *Acta Psychiatrica Scandinavica*, 72, (suppl. 323).
26. Verhulst, F. C., Prince, J., Vervuurt-Poot, C., & De Jong, J. (1989). Mental health in Dutch adolescents: self-reported competencies and problems for ages 11-18. *Acta Psychiatrica Scandinavica*, 80, (suppl. 356).
27. Ferdinand, R. F., & Verhulst, F. C. (1994). The prediction of poor outcome in young adults: Comparison of the Young Adult Self-Report, the General Health Questionnaire and the Symptom Checklist. *Acta Psychiatrica Scandinavica*, 89, 405-410.
28. Ferdinand, R. F., Verhulst, F. C., & Wiznitzer, M. (1995). Continuity and change of self-reported problem behaviors from adolescence into young adulthood. *Journal of the American Academy of Child and Adolescent Psychiatry*, 34, 680-690.
29. Mureau, M. A. M., Slijper, F. M. E., Slob, A. K., & Verhulst, F. C. (1995). Genital perception of children, adolescents, and adults operated on for hypospadias: A comparative study. *Journal of Sex Research*, in press.

Subject no.: Interviewed at hospital
 Date: ./. /. Interviewed at home
 Filled out at home

1. Have you been ill in the last 3 months?
2. Do you have problems with your health (e.g., high blood pressure, diabetes mellitus)?
3. Have you ever used medication in the past?
4. Do you use medication at this moment?
5. How many glasses of alcohol containing drinks (e.g., beer, wine) do you consume each day on average?

<input type="radio"/> > 10	<input type="radio"/> 7-8	<input type="radio"/> 3-4	<input type="radio"/> 0-1
<input type="radio"/> 9-10	<input type="radio"/> 5-6	<input type="radio"/> 1-2	<input type="radio"/> none
6. Do you smoke? yes no
- A. What do you smoke? pipe cigarettes cigars
- B. How much do you smoke?
7. Do you use other drugs/stimulants? no yes,
8. What is your marital status?

<input type="radio"/> never married	<input type="radio"/> living together	<input type="radio"/> separated
<input type="radio"/> married	<input type="radio"/> divorced	<input type="radio"/>
9. Do you live with someone else at this moment? yes no
- A. How long do you live together with this person?
- B. Do you have sex with this person? yes no
- C. How do you judge your relationship with this person?

<input type="radio"/> very unhappy	<input type="radio"/> a little unhappy	<input type="radio"/> a little happy	<input type="radio"/> very happy
<input type="radio"/> unhappy	<input type="radio"/> neither unhappy, nor happy	<input type="radio"/> happy	

DEVELOPMENT

10. What kind of friends did you have at primary school?

<input type="radio"/> mainly boys	<input type="radio"/> mainly girls
<input type="radio"/> about as many boys as girls	<input type="radio"/> I did not have any friends
11. Were they close friends or casual acquaintances?

<input type="radio"/> close friends	<input type="radio"/> casual acquaintances	<input type="radio"/> I did not have any friends
-------------------------------------	--	--
12. What kind of friends did you have during secondary education?

<input type="radio"/> mainly boys	<input type="radio"/> mainly girls
<input type="radio"/> about as many boys as girls	<input type="radio"/> I did not have any friends
13. Were they close friends or casual acquaintances?

<input type="radio"/> close friends	<input type="radio"/> casual acquaintances	<input type="radio"/> I did not have any friends
-------------------------------------	--	--
14. What kind of friends do you have at this moment?

<input type="radio"/> mainly men	<input type="radio"/> mainly women
<input type="radio"/> about as many men as women	<input type="radio"/> I don't have any friends
15. Are they close friends or casual acquaintances?

<input type="radio"/> close friends	<input type="radio"/> casual acquaintances	<input type="radio"/> I did not have any friends
-------------------------------------	--	--

16. When did you go out for the first time (in a group)?

When I was years old I have never been out

17. Did you go out with boys or with girls

mainly with boys

mainly with girls

with about as many boys as girls

I have never been out

18. Where did you go to?

to a bar or disco

to

to a film

I have never been out

19. Please, indicate with a vertical mark on the horizontal line what your sexual orientation is:

I am only attracted
to males

I am attracted to males
as well as females

I am only attracted to
to females

20. Have you ever been in love? yes no I don't know

21. How old were you when you fell in love for the first time? years old

22. Have you ever been in love more often, after the first time? yes no

23. When you were in love, what did you do to get her/his attention?

nothing, she/he never knew I was in love with her/him

I took the initiative to make a date

I hinted that I was in love, but I never took the initiative

I did something else,

24. When were you interested in girls/boys for the first time? When I was years old

25. What did you think about that?

26. When did you have a date for the first time?

When I was years old

I have never dated someone

27. Have you ever tried to make sexual advances to your date? yes no

A. If yes, has it ever gone too far for you? yes no

B. If yes, why did it go too far for you?

28. How old were you, when you French kissed for the first time?

..... years old

I have never French kissed

29. How old were you, when you masturbated (jerked/jacked off) for the first time?

..... years old

I have never masturbated

30. How old were you, when you necked and fondled each others genitals for the first time with your clothes on?

..... years old

I have never had such an experience

31. How old were you, when you necked and fondled each others genitals for the first time with your clothes off (in the nude)?

..... years old

I have never had such an experience

32. Do you have the feeling that, before you had these sexual experiences, you were inhibited with seeking sexual contacts because of embarrassment about your penile appearance?

33. Have these feelings changed after you obtained these sexual experiences?

34. Do you have experience with sexual intercourse? yes no

35. If yes, how old were you, when you had sexual intercourse for the first time? years old

36. Have you ever been afraid to be rejected by a partner because of your penile appearance?

37. Have these feelings changed after you obtained experience with sexual intercourse?

SEXUAL FUNCTIONING

38. Keep in mind an average month of the past 3 months (thus no vacation, no sickness, no extra business). How often did you have sexual activity (sexual intercourse, oral sex, genital fondling) with a partner?

with orgasm per month
without orgasm per month

39. Some men come always quickly, when they have sexual intercourse, whereas other men come always slowly. Do you have the feeling that you came quickly or slowly during sexual intercourse in the past 3 months?

o I came always quickly o I came almost always slowly
o I came rather quickly o I came always slowly
o sometimes quickly, sometimes slowly o I never had an orgasm at all

40. Some men come always quickly, when they have sex (oral sex, genital fondling), whereas other men come always slowly. Do you have the feeling that you came quickly or slowly during sexual activity in the past 3 months?

o I came always quickly o I came almost always slowly
o I came rather quickly o I came always slowly
o sometimes quickly, sometimes slowly o I never had an orgasm at all

41. Please, indicate with a vertical mark on the horizontal line the average quality of your erections during sexual activity (oral sex, genital fondling):

totally flaccid totally erected

42. Please, indicate with a vertical mark on the horizontal line the average quality of your erections during sexual intercourse:

totally flaccid totally erected

43. Please, indicate with a vertical mark on the horizontal line your average satisfaction with your erections during sexual intercourse:

very dissatisfied very satisfied

44. How often were you afraid to fail (fear of failure) during sexual activity in the last 3 months?

o always o often o seldom
o almost always o now and then o never

45. Could you explain why you had these feelings?

46. Do you have the feeling that you are inhibited during sexual activity because of your feelings about your penile appearance?

47. With how many partners did you have sexual intercourse?

48. Did you ever have problems with sexual intercourse (more than one answer possible)?
- no
 - yes:
 - because of tiredness
 - because I was too nervous
 - because I used a condom
 - because I did not get an erection
 - because my penis is too small
 - because my penis is curved when erect
 - because of something else,

49. How much of your sexual activity in the past 3 months did you enjoy?
- nothing
 - almost nothing
 - less than half
 - about half
 - more than half
 - almost everything
 - everything

MASTURBATION

50. Please, indicate with a vertical mark on the horizontal line your average sexual desire during the last 3 months:

very low very high

51. Please, indicate with a vertical mark on the horizontal line the average quality of your erections during masturbation:

totally flaccid totally erected

52. Please, indicate with a vertical mark on the horizontal line your average satisfaction with your erections during masturbation:

very dissatisfied very satisfied

53. Keep an average month of the past 3 months in mind (thus no vacation, no sickness, no extra business). How often did you masturbate?

with orgasm per month
without orgasm per month

54. Some men come always quickly, when they masturbate, whereas other men come always slowly. Do you have the feeling that you came quickly or slowly during sexual intercourse in the past 3 months?

- I came always quickly
- I came rather quickly
- sometimes quickly, sometimes slowly
- I came almost always slowly
- I came always slowly
- I never had an orgasm at all

55. Some men do not produce any fluid, when they have an orgasm. They have a so-called dry orgasm. Have you ever had such dry orgasms?

- I don't know
- I don't know
- almost always
- often
- now and then
- seldom
- never
- I never have orgasms

56. Keep an average month of the past 3 months in mind. How often did you have a sexual dream?
 per month never
57. Keep an average month of the past 3 months in mind. How often did you have an erection just after you woke up (morning erection)?
 per month never
58. Keep an average month of the past 3 months in mind. How often did you have sexual thoughts or feelings of sexual desire?
 per month never
59. Keep an average month of the past 3 months in mind. How often did you have a more or less spontaneously erected penis (not during sexual activity and not during waking up)?
 per month never
60. Please, indicate with a vertical mark on the horizontal line your average satisfaction with your sexual functioning:

very dissatisfied very satisfied

61. How important is sex for you?
 totally not important for me, I don't care
 I do not know whether it is important, although I don't want to be without it
 it is sometimes important
 it is usually important
 it is important
 it is very important
 it is extremely important, it would be very difficult to live without it
62. Have you ever had painful erections?
 always often seldom I have never erections
 almost always now and then never
63. Have you ever had painful orgasms?
 always often seldom I have never orgasms
 almost always now and then never
64. Is your penis straight in erection?
 yes
 no
- A. Is there a dorsal curvature? yes no
 Is there a ventral curvature? yes no
 Is there a curvature to the left? yes no
 Is there a curvature to the right? yes no
 Is there an axial torsion? yes no
- B. Does the penile curvature cause difficulties during sexual intercourse?
 yes no

GENITAL APPRAISAL

- 65. Do you feel your penis looks similar to that of other men?
- 66. Are you satisfied with your penile appearance?
 - o yes
 - o no, I am dissatisfied because
- 67. Have these feelings/satisfaction changed over time (compared to the past)?
- 68. Are you satisfied with the length of your flaccid penis?
 - o yes
 - o no, it is too small
 - o no, it is too large
- 69. Are you satisfied with the length of your erected penis?
 - o yes
 - o no, it is too small
 - o no, it is too large
- 70. Do you have difficulties with undressing in public (e.g., showering after sport activities, sauna)?
.....
- 71. Have these feelings/difficulties changed over time (compared to the past)?
- 72. Have you ever had comments from others about your penile appearance?
- 73. Have you ever had comments from a (sex)partner about your penile appearance?
- 74. How satisfied are you with the final surgical result?
 - o very satisfied
 - o a little satisfied
 - o a little dissatisfied
 - o very dissatisfied
 - o satisfied
 - o neither satisfied, nor dissatisfied
 - o dissatisfied
- 75. What do you want to be improved?
- 76. Do you have any other comments or remarks about your sexual functioning during the last 3 months, which were not mentioned in the interview?
- 77. Do you have any questions or remarks following this interview?

Circle the number that corresponds the most with your feelings about the indicated part of your body.
 Don't think too long about it. Please, do not forget to answer all questions!

- 1 I am very dissatisfied with it
- 2 I am dissatisfied with it
- 3 I am neither dissatisfied, nor satisfied with it
- 4 I am satisfied with it
- 5 I am very satisfied with it

For example, legs:

-If you are dissatisfied with the appearance of your legs, circle the number 2.

-If you are very satisfied with your legs, circle the number 5.

	I am very dissatisfied with it	I am dissatisfied with it	I am neither dissatisfied, nor satisfied with it	I am satisfied with it	I am very satisfied with it
1 Arms	1	2	3	4	5
2 Testes	1	2	3	4	5
3 Scrotum	1	2	3	4	5
4 Legs	1	2	3	4	5
5 Chest hair	1	2	3	4	5
6 Chest size	1	2	3	4	5
7 Depth of Voice	1	2	3	4	5
8 Penile thickness	1	2	3	4	5
9 Face	1	2	3	4	5
10 Glans size	1	2	3	4	5
11 Hands	1	2	3	4	5
12 Penile skin distribution around corona	1	2	3	4	5
13 Penile color	1	2	3	4	5
14 Penile size (flaccid)	1	2	3	4	5
15 Penile size (erect)	1	2	3	4	5
16 Body hair	1	2	3	4	5
17 Scars	1	2	3	4	5
18 Eyes	1	2	3	4	5

Genital Perception Scale

	I am very dissatisfied with it	I am dissatisfied with it	I am neither dissatisfied, nor satisfied with it	I am satisfied with it	I am very satisfied with it
19 Missing of prepuce	1	2	3	4	5
20 Missing of frenulum	1	2	3	4	5
21 Position of meatus	1	2	3	4	5
22 Pubic hair	1	2	3	4	5
23 Muscles	1	2	3	4	5
24 Penile position in erection	1	2	3	4	5
25 Body appearance in general	1	2	3	4	5
26 Penile appearance in general	1	2	3	4	5
27 Glans shape	1	2	3	4	5

Subject no.: Interviewed at hospital
 Date: ./. /. Interviewed at home
 Filled out at home

The following interview is about your daily activities, your friends, love and sex, your hospitalization, and about your feelings with these items.

1. How old are you? years old
2. What kind of school do you go to?
 - primary school middle general education
 - lower technical and vocational training secondary education
 - middle technical and vocational training
3. Which class do you attend?
4. How are you doing in school?
 - very good neither good, nor bad very bad
 - good bad
5. Do you get along with other boys and girls?
 - I get along with everybody I get along with almost nobody
 - I get along with almost everybody I get along with nobody
 - I get along with a few children
6. Do you have any friends? yes no
7. How many friends do you have?
8. Are they mainly boys or girls?
 - mainly boys about as many boys as girls mainly girls
9. Are they friends with whom you play every day (close friends) or are they friends you see only occasionally (casual friends)?
 - they are all close friends most are casual friends
 - most are close friends they are all casual friends
 - about as many are close friends as casual friends
10. What do you do with your friends (you may mark more than one answer)?
 - we play hide and seek we skip rope we sport
 - we shoot marbles we play computer games we go out
 - we play soldier/cowboy we watch television we sit and talk together
 - we play doctors and nurses we play board game we do something else,
11. What do you do when you are alone?
 - I do creative things (e.g., drawing, making kits) I sport
 - I solve puzzles I play computer games
 - I read I watch television
 - I play games I do something else,

Now I want to talk with you about your hospitalization(s) and what you can remember of it.

12. A. Why have you been in the hospital?
- B. How many times were you in the hospital?

- C. How many times have you been operated?
- D. How many times have you been in the hospital for your willy?
13. Could you tell me what you remember of your stayings in the hospital because of your willy?
14. How many times have you been operated on your willy?
15. How old were you, when you were operated on your willy for the 1st time?
 ... years old o I don't know
16. How old were you, when you were operated on your willie for the 2nd time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 3rd time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 4th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 5th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 6th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 7th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 8th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 9th time?
 ... years old o I don't know
- How old were you, when you were operated on your willy for the 10th time?
 ... years old o I don't know
17. Could you tell me why you have been operated on your willy (you may mark more than one answer)?
- | | |
|---|---|
| <input type="checkbox"/> the pee hole was not on the right place | <input type="checkbox"/> peeing was very painful |
| <input type="checkbox"/> my willie was curved | <input type="checkbox"/> because of something else, |
| <input type="checkbox"/> I could not pee in the standing position | <input type="checkbox"/> I don't know |
| <input type="checkbox"/> I had to force during peeing | |
18. Could you describe the appearance of your willy just after the first operation?
19. How did you feel about the surgical result or the appearance of your willy just after the first operation (you may mark more than one answer)?
- | | | | | |
|--------------------------------|-------------------------------------|---------------------------------------|---|---------------------------------------|
| <input type="checkbox"/> happy | <input type="checkbox"/> angry | <input type="checkbox"/> relieved | <input type="checkbox"/> I felt nothing | <input type="checkbox"/> I don't know |
| <input type="checkbox"/> sad | <input type="checkbox"/> frightened | <input type="checkbox"/> disappointed | <input type="checkbox"/> | |
20. Did your willy look like what you had expected after the first operation?
- | | |
|--|--|
| <input type="checkbox"/> he looked exactly like what I had expected | <input type="checkbox"/> he did not look what I had expected |
| <input type="checkbox"/> he looked a little like what I had expected | <input type="checkbox"/> I don't know |
| <input type="checkbox"/> he looked hardly like what I had expected | |
21. What did you expect he would look like?
22. Did you have any pain during peeing after the operation(s)?
- | | | |
|--|--|---|
| <input type="checkbox"/> yes, I had very much pain | <input type="checkbox"/> yes, I had a littled pain | <input type="checkbox"/> no, I had no pain at all |
| <input type="checkbox"/> yes, I had much pain | <input type="checkbox"/> no, I had hardly pain | <input type="checkbox"/> I don't know |

23. Did you have any pain after the operation(s) when you were not peeing?
 yes, I had a lot of pain yes, I had a littled pain no, I had no pain at all
 yes, I had much pain no, I had hardly any pain I don't know
24. Did the doctors/your parents tell you how your willy would look like after the operation?
 they told me exactly how my willy would look like
 they told me a little how my willy would look like
 they hardly told me how my willy would look like
 they did not tell me at all how my willy would look like
 I don't know
25. Did the doctors/your parents explain to you what was going to happen? Did they prepare you for the operation?
 they explained exactly what was going to happen and why it was necessary
 they explained a little what was going to happen and why it was necessary
 they hardly explained what was going to happen and why it was necessary
 they did not explain at all what was going to happen and why it was necessary
 I don't know
26. Many children are afraid to be operated. Were you afraid before your first operation?
 I was very afraid I was hardly afraid I don't know
 I was a little afraid I was not afraid at all
27. Why did you have to be operated (you may mark more than one answer)?
 because the doctors told me I had to (go to question 29) because I wanted it myself
 because my parents wanted it (go to question 29) I don't know
29. Why did you want to be operated?
 I wanted to be able to pee in the standing position
 I wanted that the pee hole was on the right place
 I wanted my willy to be straight
 I wanted my willy to be normal
 I wanted
30. Why did you have to be operated the second time (you may mark more than one answer)?
 because the doctors told me I had to (go to question 32) because I wanted it myself
 because my parents wanted it (go to question 32) I don't know
31. Why did you want to be operated?
 I wanted to be able to pee in the standing position
 I wanted that the pee hole was on the right place
 I wanted my willy to be straight
 I wanted my willy to be normal
 I wanted
32. Were you afraid for the 3rd operation? I have been operated 2 x (go to 56)
33. Why did you have to be operated then?
34. Why did you want to be operated?
35. Were you afraid for the 4th operation?.... I have been operated 3 x (go to 56)
36. Why did you have to be operated then?

37. Why did you want to be operated?
38. Were you afraid for the 5th operation?... I have been operated 4 x (go to 56)
39. Why did you have to be operated then?
40. Why did you want to be operated?
41. Were you afraid for the 6th operation?... I have been operated 5 x (go to 56)
42. Why did you have to be operated then?
43. Why did you want to be operated?
44. Were you afraid for the 7th operation?... I have been operated 6 x (go to 56)
45. Why did you have to be operated then?
46. Why did you want to be operated?
47. Were you afraid for the 8th operation?... I have been operated 7 x (go to 56)
48. Why did you have to be operated then?
49. Why did you want to be operated?
50. Were you afraid for the 9th operation?... I have been operated 8 x (go to 56)
51. Why did you have to be operated then?
52. Why did you want to be operated?
53. Were you afraid for the 10th operation?... I have been operated 9 x (go to 56)
54. Why did you have to be operated then?
55. Why did you want to be operated?
56. Do you think the result is improved following these operations?
 yes, the surgical result is improved (go to question 57)
 no, the surgical result is worsened (go to question 58)
 I don't know
57. What do you think is improved (you may mark more than one answer)?
 position of the pee hole penile curvature penile appearance in general
 scars penile length something else,
- glans shape skin around glans
- Go to question 59
58. What do you think is worsened?
 position of the pee hole penile curvature penile appearance in general
 scars penile length something else,
- glans shape skin around glans
59. Did you find the one time less annoying to be operated than the other time?
 yes I have been operated only once (go to question 64)
 no (go to question 64) I don't know (go to question 64)
60. Which time was least annoying?
61. Why?
62. Which time was most annoying?
63. Why?

Now I want to talk with you about peeing, undressing in public, and how you feel your willy looks.

64. Were you afraid of peeing just after the operation (you may mark more than one answer)?
 o yes, because of the pain o yes, because
 o yes, I was afraid of forcing o I don't know
 o yes, because I was afraid the stitches would break
65. -
66. Do you know when you were toilet trained?
 o yes, when I was years old o I am not toilet trained yet
 o no, I don't know
67. Young children (toddlers) often walk around naked. Did you walk around naked when you were very young?
 o yes, it happened regularly
 o no, my parents never let me walk around naked, because they think it is not done
 o no, my parents never let me walk around naked, because they were afraid others would see my willy
 o no, I did not dare to walk around naked
 o no,
68. Do you go to public lavatories or to private lavatories, when you have to pee?
 o I always go to a private lavatory
 o I only go to a public lavatory, when there is nobody else
 o I go to a public lavatory, but I always try to hide my willy
 o I always go to a public lavatory, without hiding my willy
 o I
69. And when there are only public lavatories? Do you hold your water untill you are home or do go to the public lavatories anyway?
 o I hold my water untill I am home
 o I go to the public lavatory, but I wait first untill everybody is gone
 o I go to a public lavatory, but I always try to hide my willy
 o I always go to a public lavatory, without hiding my willy
 o I
70. Every child went to swimming lessons. Did you find it difficult to undress in public before your swimming lesson?
 o yes, therefore I always put on my swimming trunks at home
 o yes, therefore I always tried to hide my willy by standing in a corner or by turning myself around
 o yes, but there were private changing rooms, so I did not have to undress in public
 o no, but I put on my swimming trunks at home anyway, because I found it more practice
 o no, I always undressed in public without hiding my willy
 o no, but there were only private changing rooms
71. And after swimming lesson, how did you change?
 o together with the other boys, because I have no difficulties with undressing in public
 o together with the other boys, but I tried to hide my willy by turning myself around
 o in a private changing room, because I did not dare to undress in public
 o in a private changing room, because there were only private changing rooms
 o I put on my trousers over my (wet) swimming trunks

72. Did you undress in public more easily after your operation(s)?
 yes
 no, I have the same difficulties with undressing in public as before the operation(s)
 no, I undress in public just as easily as before the operation(s)
 no, I got more difficulties with undressing in public after the operation(s)
 I don't know
73. Have you ever received comments on the appearance of your willy?
 yes no, because others never have seen me naked
 no (go to question 76)
74. Have you ever been teased, when you received comments?
 yes no
75. What did they say (you may mark more than one answer)?
 they made comments about the position of the pee hole
 they made comments on the scars
 they said I had a small willy
 they made comments about the circumcised appearance
 they made comments about something else,
76. Have you ever told anyone (e.g., your friends) that you have been operated on your willy?
 yes no, because it is nobody's business
 no, because I am afraid of ridicule no, because
77. When you look at your willy, do you find it looks the same as that of other boys?
 yes (go to question 79) no I don't know
78. What do you think looks differently (you may mark more than one answer)?
 the missing of the foreskin the appearance of my willy in general
 my willy is smaller the position of the pee hole
 the color of my willy the scars
 the shape of the glans something else,
79. Are you satisfied with the appearance of your willy?
 yes (go to question 81) no
80. About what are you dissatisfied (you may mark more than one answer)?
 the missing of the foreskin the appearance of my willy in general
 my willy is smaller the position of the pee hole
 the color of my willy the scars
 the shape of the glans something else,
81. Compared to the past, are you now more satisfied with the appearance of your willy?
 I am more satisfied than in the past I am equally dissatisfied as in the past
 I am equally satisfied as in the past I am less dissatisfied than in the past
 I am less satisfied than in the past I don't know
82. Are you satisfied with the length of your flaccid willy?
 yes no, I think it is too small no, I think it is too large
83. Are you satisfied with the length of your erected willy?
 yes no, I think it is too small no, I think it is too large
84. What do you think of the number of operations you underwent?
 too many enough too few I don't know

85. How satisfied are you with the final result of surgery?
 very satisfied neither satisfied, nor dissatisfied very dissatisfied
 satisfied a little dissatisfied
 a little satisfied dissatisfied
86. Would you like to have something improved (you may mark more than one answer)?
 no
 yes
 the missing of my foreskin the appearance of my willy in general
 the small length of my willy the position of the pee hole
 the color of my willy the scars
 the shape of the glans something else,

We have talked a while about your stayings in the hospital and about your satisfaction with the appearance of your willy, but now I would like to talk about sex

87. Have you ever talked about sex(ual matters) with your friends?
 yes no (go to question 90)
88. How many times? times per month
89. How does that go (you may mark more than one answer)?
 everyone brags about it and I too (go to question 91)
 everyone talks seriously about their (sexual) experiences (go to question 91)
 everyone tells each other with whom they want to date/go steady (go to question 91)
 I usually keep my mouth and listen to the others (go to question 91)
 (go to question 91)
90. Why not?
 my friends don't talk about sex(ual matters)
 I don't like talking about sex(ual matters)
 I can do without talking about sex(ual matters)
 I am not allowed to talk about sex(ual matters) by my parents

91. When you are watching a film on television in which a couple is making love/kissing;how do you feel about that (you may mark more than one answer)?
 I think it is stupid I have to blush I find it sexy
 I find it annoying I think it is exciting I find it
92. Have you ever fantasized about girls (or boys)?
 no, never yes, per week
93. Many boys like to touch their willy and play with it (jerking off). Have you also ever done this?
 yes
 no, I have never done this (go to question 102)
94. How old were you when you did it for the first time? years old
95. Were you already operated on your willy?
 yes no I don't know

96. How did it go just after your operation(s). Were you afraid to touch your willy?
 o yes o I don't know (go to question 100) o no (go to question 100)
97. Where were you afraid of?
98. How long did this fear last? months o I don't know
99. About how many times per month do you play with your willy (do you jerk off)?
 per month
100. Some boys get a very pleasant feeling when they jerk off (= coming). Then, fluid comes out their willy (= sperm). Does this also happens to you?
 o always o often o seldom
 o almost always o now and then o never
101. Some boys always come quickly when they jerk off, in other boys it always takes a long time. Do you have the feeling you come quickly or does it take a long time?
 o I always come quickly o it always takes a long time
 o I come rather quickly o I never come when I jerk off
 o sometimes quickly, sometimes slowly o I never jerk off
 o it almost always takes a long time

Now I want to talk about being in love, going out, etc.

102. Do you already go out sometimes (e.g., to the cinema or parties) (you may mark more than one answer)?
 o yes, to the cinema o yes, to discotheques o yes, to
 o yes, to parties o yes, to bars o no, I never go out (go to question 105)
103. How often do you go out? times per month
104. With whom do you go out?
 o mainly with boys (go to question 106)
 o with both boys and girls (go to question 106)
 o mainly with girls (go to question 106)
105. Why don't you go out?
 o I am not (yet) allowed to go out by my parents o I am too young to go out
 o I don't have any friends to go out with o because
 o I don't have a need for going out (yet)
106. Have you ever been in love?
 o yes o no (go to question 113) o I don't know (go to question 113)
107. How old were you the first time? years old
108. Have you ever been in love more often?
 o yes o no
109. Were you in love with girls or boys?
 o always with girls, never with boys
 o almost always with girls, seldom with boys
 o often with girls, now and then with boys
 o now and then with girls, often with boys
 o seldom with girls, almost always with boys
 o never with girls, always with boys

110. Where did you meet the girls (boys) with whom you were in love?
o in the neighbourhood o at parties o in bars
o at school o in discotheques o somewhere else,
111. How do you feel about being in love (you may mark more than one answer)?
o it is fun o it is scary o it is
112. What do you do when you are in love (you may mark more than one answer)?
o nothing, I don't let it show and she (he) will never find out
o I let her know through my/her friends and try to find out what she (he) feels about me
o I chat with her (him) and try to make a date
o I
113. Have you ever had a date?
o yes, times o no
114. Have you ever had a girlfriend (going steady)?
o yes, times o no (go to question 116)
115. Do you have a girlfriend at this moment?
o no o yes, I am going steady for weeks
116. Have you ever French kissed with someone?
o no (go to question 124) o yes, the first time I was years old
117. Have you tried to French kiss your date?
o always o often o seldomly
o almost always o now and then o never (go to question 119)
118. Have you ever gone any further (e.g., fondling each other)?
o always o often o seldomly
o almost always o now and then o never
119. How often have you French kissed with someone in the past month? times
120. When you are French kissing with someone, do you fondle each other?
o always o often o seldomly
o almost always o now and then o never (go to question 124)
121. Which body parts do you fondle (you may mark more than one answer)?
o I fondle her arms o I fondle her bottom o I fondle her pussy
o she fondles my arms o she fondles my bottom o she fondles my willy

o I fondle her breasts o I fondle her belly o I fondle her
o she fondles my chest o she fondles my belly o she fondles my

o I fondle her thighs o I fondle her back
o she fondles my thighs o she fondles my back
122. Have you ever necked and fondled each other's genitals with your clothes on?
o no (go to question 124) o yes, the first time I was years old
123. Have you ever necked and fondled each other's genitals with your clothes off?
o no o yes, the first time I was years old

- 124.A. Do you have the feeling you are inhibited in your contact with girls, because of the appearance of your willy?
 yes no I don't know
- 124.B. And are you inhibited with seeking sexual contacts because of the appearance of your willy?
 yes no I don't know
125. How inhibited are you with seeking sexual contacts?
 very inhibited a little inhibited totally not inhibited
 inhibited hardly inhibited I don't know
126. Have you ever anticipated of being afraid of ridicule by the one with whom you will make love for the first time, and who will see your willy?
 always often seldom
 almost always now and then never
 When question 116 was answered "no" go to question 149.
127. Do you have the feeling that your inhibitions with seeking sexual contacts have changed after your first sexual experiences?
 yes, my inhibitions have worsened
 yes, my inhibitions have diminished, although not disappeared
 yes, my inhibitions, have disappeared
 yes, with my present girlfriend my inhibitions have disappeared, but with a possible new future girlfriend they might return
 no, my inhibitions have remained
 no, I have never had inhibitions with seeking sexual contacts
128. Have you ever been afraid to be rejected by a girl after she has seen your willy?
 always often seldom
 almost always now and then never
129. Have you ever gone to bed with someone (= sexual intercourse)?
 no (go to question 131) yes, the first time I was years old
130. With how many girls have you gone to bed?
131. Has the fear for rejection changed after you obtained experience with undressed necking?
 yes, it has disappeared yes, it has worsened
 yes, it has diminished I have never been afraid to be rejected
 no, it has remained the same I don't have experience with undressed necking

The next questions deal with the past month

132. How often did you have sexual activity (genital fondling, sexual intercourse) with a partner?
 with orgasm per month
 without orgasm per month
133. Some boys always come quickly when they have sexual intercourse, in other boys it always takes a long time. Do you have the feeling you came quickly, or did it take a long time?
 I always came quickly it always took a long time
 I came rather quickly I never came during intercourse
 sometimes quickly, sometimes slowly I never had sexual intercourse in the past month
 it almost always took a long time month

134. Some boys always come quickly when they have sexual activity (genital fondling, oral sex, not sexual intercourse), in other boys it always takes a long time. Do you have the feeling you came quickly, or did it took a long time?
 I always came quickly it always took a long time
 I came rather quickly I never came during sexual activity
 sometimes quickly, sometimes slowly I never had sexual activity in the past month
 it almost always took a long time
135. Circle a number (0 to 10), which indicates the average quality of your erections during sexual activity:
 0 1 2 3 4 5 6 7 8 9 10
 0=totally flaccid very hard=10
136. Circle a number (0 to 10), which indicates the average quality of your erections during sexual intercourse:
 0 1 2 3 4 5 6 7 8 9 10
 0=totally flaccid very hard=10
137. Circle a number (0 to 10), which indicates your average satisfaction with the quality of your erections:
 0 1 2 3 4 5 6 7 8 9 10
 0=very dissatisfied very satisfied=10
138. How often were you afraid to fail during sexual activity in the past month?
 always seldom I didn't have any sexual activity
 almost always now and then in the past month (go to question 140)
 often never (go to question 140)
139. Could you explain why?
140. Have you ever had difficulties with sexual intercourse (now or in the past) (you may mark more than one answer)?
 yes because of tiredness because my willy became flaccid
 because of nervousness because my willy is too small
 because I used a condom because my willy is curved
 because I did not get an erection because,
- no
141. Have you ever received comments on the appearance of your willy by your (ex)girlfriends (you may mark more than one answer)?
 yes comments on the position of the pee hole cuddling comments
 comments on the scars she asked what happened
 comments on the length comments on
- comments on the missing of my foreskin
 no

142. When you are making love to someone, do you tell her you have been operated on your willy?
 always often seldomly
 almost always now and then never
143. Do you tell her about it before or after you have sex?
 always before I have sex, never afterwards
 almost always before I have sex, seldomly afterwards
 often before I have sex, now and then afterwards
 now and then before I have sex, often afterwards
 seldomly before I have sex, almost always afterwards
 never before I have sex, always afterwards
 I never talk about it
144. How many of your sexual activity did you enjoy in the past month?
 I enjoyed nothing I enjoyed more than half
 I enjoyed almost nothing I enjoyed almost everything
 I enjoyed less than half I enjoyed everything
 I enjoyed about half
145. Circle a number (0 to 10), which indicates your average sexual desire in the past month:

0 1 2 3 4 5 6 7 8 9 10
0=very low very high=10
146. Circle a number (0 to 10), which indicates your average satisfaction with your sexual functioning:

0 1 2 3 4 5 6 7 8 9 10
0=very dissatisfied very satisfied=10
147. Have you ever had painful orgasms?
 always often seldom
 almost always now and then never
148. How often did you have sexual thoughts or feelings of sexual desire (lust for sex) in the past month?
 never per week
 per month per day
149. How important is sex to you?
 totally not important, it does not interest me
 I don't know whether it is important to me, but I wouldn't want be without it
 it is now and then important to me
 it is important to me
 it is very important to me
 it is extremely important to me

150. How often did you have an exciting (sexual) dream in the past month?
o never o per week
o per month o per day
151. How often did you awake with an erection in the past month?
o never o per week
o per month o per day
152. How often did you have more or less spontaneous daytime erections in the past month?
o never o per week
o per month o per day
153. Have you ever had painful erections?
o always o often o seldomly
o almost always o now and then o never
154. Is your willy straight in erection?
o yes
o no
If no, is it curved upward? o yes o no
is it curved downward? o yes o no
is it curved to the left? o yes o no
is it curved to the right? o yes o no
is it rotated around the axis? o yes o no
Do you have difficulties with sexual intercourse because of the curvature of your willy?
o yes o no o not applicable
155. Do you have any questions or remarks following this interview?
.....

This is a list of sentences about other boys' satisfaction with various parts of their body. Please indicate for every sentence how satisfied you are with that particular part of your boy. Every line is made up of two sentences. Below every sentence there are two boxes, thus per line there are four boxes. For every sentence you have to mark one of these four boxes.

There are no wrong or right answers, what you feel matters.

For some sentences it may be a little scary to give an answer. That is all right, but try to answer them as honestly as possible.

Below you see an example on how the questions have to be filled out:

Some boys are very satisfied
with their arms

BUT

other boys are very dissatis-
fied with their arms

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

or

Some boys are very satisfied
with their arms

BUT

other boys are very dissatis-
fied with their arms

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

or

Some boys are very satisfied
with their arms

BUT

other boys are very dissatis-
fied with their arms

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

Junior Genital Perception Scale

1. Some boys are very satisfied
with their arms

BUT

other boys are very dissatisfied
with their arms

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

2. Some boys are very satisfied
with their balls/scrotum

BUT

other boys are very dissatisfied
with their balls/scrotum

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

3. Some boys are very satisfied
with their legs

BUT

other boys are very dissatisfied
with their legs

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

4. Some boys are very satisfied
with their chest size

BUT

other boys are very dissatisfied
with their chest size

What boys are you like?

I am just like
these boys

I am a little like
these boys

I am just like
these boys

I am a little like
these boys

5. Some boys are very satisfied with the depth of their voice

BUT

other boys are very dissatisfied with the depth of their voice

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

6. Some boys are very satisfied with the thickness of their willy

BUT

other boys are very dissatisfied with the thickness of their willy

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

7. Some boys are very satisfied with their face

BUT

other boys are very dissatisfied with their face

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

8. Some boys are very satisfied with their glans

BUT

other boys are very dissatisfied with their glans

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

Junior Genital Perception Scale

9. Some boys are very satisfied with their hands

BUT

other boys are very dissatisfied with their hands

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

10. Some boys are very satisfied with the length of their flaccid willy

BUT

other boys are very dissatisfied with the length of their flaccid willy

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

11. Some boys are very satisfied with the length of their erect willy

BUT

other boys are very dissatisfied with the length of their erect willy

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

12. Some boys are very satisfied with their body hair

BUT

other boys are very dissatisfied with their body hair

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

13. Some boys are very satisfied with the scars of the operations

BUT

other boys are very dissatisfied with the scars of the operations

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

14. Some boys are very satisfied with their eyes

BUT

other boys are very dissatisfied with their eyes

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

15. Some boys are very satisfied with the missing of their foreskin

BUT

other boys are very dissatisfied with the missing of their foreskin

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

16. Some boys are very satisfied with the position of their peehole

BUT

other boys are very dissatisfied with the position of their peehole

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

Junior Genital Perception Scale

17. Some boys are very satisfied with their muscles

BUT

other boys are very dissatisfied with their muscles

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

18. Some boys are very satisfied with their body in general

BUT

other boys are very dissatisfied with their body in general

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

19. Some boys are very satisfied with the appearance of their willy

BUT

other boys are very dissatisfied with the appearance of their willy

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

20. Some boys are very satisfied with the shape of their glans

BUT

other boys are very dissatisfied with the shape of their glans

What boys are you like?

I am just like these boys

I am a little like these boys

I am just like these boys

I am a little like these boys

SUMMARY

The objective of the present thesis was to investigate psychosexual and psychosocial adjustment, and genital perception of children, adolescents, and adults following hypospadias surgery, compared to age-matched subjects treated for an inguinal hernia. In addition, effects of various medical characteristics (severity of hypospadias, age at final surgery, number of operations, surgical procedures), subject age, and coping with hypospadias on psychosexual adjustment and psychosocial, and genital perception of hypospadias patients were also studied.

In chapter 1, an overview of the embryology, etiology, morphology, and of surgical treatment of hypospadias is presented. Hypospadias is caused by a disturbance in the development of the urethra, resulting in an abnormal position of the meatus. The reported incidence of hypospadias ranges from 0.8 to 8.2 per 1,000 live male births. Several etiological factors as (sex) chromosomal aberrations, autosomal (dominant/recessive) inheritance, multifactorial inheritance, endocrinopathies, and environmental factors have been proposed. However, until now the exact cause of isolated hypospadias remains unknown. Surgical reconstruction of the urethra and straightening of the penis is necessary to ensure voiding in the standing position and unhampered adult sexual functioning.

In chapter 2, an extensive literature review is given on studies about the long-term psychosexual and psychosocial consequences of hypospadias. Most of the studies seem to indicate that hypospadias patients are at risk for retarded sexual adjustment, are self-conscious about or dissatisfied with their penile appearance, and have poorer psychosocial functioning. Because several methodological shortcomings impede proper interpretation of the results of the reviewed studies, the present study was carried out.

In chapter 3, the selection of patients and comparison subjects, procedure, response, sample characteristics, and questionnaires used are described. One hundred-and-sixteen children and adolescents (aged 9 to 18 years) and 73 adults (aged 18 to 38 years) with hypospadias, and 88 and 50 age-matched

Summary

comparison subjects, respectively, treated for an inguinal hernia participated in the present study.

In chapter 4, the psychosexual adjustment of adult hypospadias patients is described. No significant differences were found between hypospadias patients and comparison subjects in the mean ages at which they had reached their first sexual milestones. Also no significant differences were found in sexual behavior and sexual functioning of hypospadias patients and comparison subjects. Of the sexually experienced subjects, more patients (33%) than comparison subjects (13%) reported that they had been inhibited with seeking sexual contacts. Hypospadias patients reported a significantly more negative genital appraisal than comparison subjects. More patients (38%) than comparison subjects (20%) considered their flaccid penis to small. Penile size was the most mentioned motive for dissatisfaction. Most hypospadias patients (60%) reported that the circumcised appearance of their penis was a motive for perceiving a different penile appearance. Although hypospadias severity was negatively related to genital appraisal and the level of inhibition with seeking sexual contacts, it was not significantly related to sexual adjustment or sexual behavior. Type of surgical procedure was neither significantly related to sexual adjustment nor to genital appraisal of hypospadias patients. Positive correlations were found between the number of operations, the age at final surgery, and the level of inhibition with seeking sexual contacts. The later treatment was completed, the later patients began with making their first sexual contacts. Thirty-seven percent expressed the wish for functional or cosmetic penile improvement. Following the interview, 5 patients (7%) were re-operated because of a poor functional or cosmetic surgical result.

In chapter 5, the psychosexual adjustment of children and adolescents with hypospadias is described. No significant differences were found between hypospadias patients and comparison subjects in the mean ages at which they had reached their first sexual milestones. Also no significant differences were found in the sexual behavior and sexual functioning of hypospadias patients and comparison subjects. Of the 9- to 12-year-old boys, 16% of the hypospadias patients and none of the comparison boys anticipa-

ted to become inhibited with seeking sexual contacts. More 13- to 18-year-old hypospadias patients (24%) than comparison subjects (2%) reported to be actually inhibited with seeking sexual contacts. Of the boys with at least French kissing experience, hypospadias patients more often were afraid of being ridiculed by a partner because of their penile appearance than comparison boys. Hypospadias patients had a significantly more negative genital appraisal than comparison boys. Significantly more patients (25%) than comparison boys (12%) were dissatisfied with their flaccid penile size. In children and adolescents with hypospadias, scars, penile size, and glanular shape were the most spontaneously mentioned motives for dissatisfaction, whereas the circumcised appearance was the most mentioned motive (48%) for perceiving a different penile appearance. Although hypospadias severity was negatively related to the level of inhibition with seeking sexual contacts, it was neither related to sexual adjustment, nor to genital appraisal. Type of surgical procedure was neither significantly related to sexual adjustment, nor to genital appraisal of hypospadias patients. Patients treated with ventralizing procedures (44%) more often tended to express the wish for functional or cosmetic improvement of their penile appearance than patients treated with terminalizing procedures (29%). No significant correlations were found between the age at final surgery and psychosexual adjustment. There did not exist significant correlations between the number of operations, the age at final surgery, and psychosexual adjustment of hypospadias patients. More 13- to 18-year-old patients (32%) than 9- to 12-year-old patients (13%) were dissatisfied with their penile appearance, and more of the first (49%) than of the latter patients (24%) wanted improvement of their penile appearance.

In chapter 6, the genital perception of children, adolescents, and adults following hypospadias surgery is described. Genital perception was measured through two standardized self-report questionnaires (child/adolescent and adult version). Hypospadias patients had a more negative genital perception than age-matched comparison subjects. There did not exist a significant correlation between subject age and genital perception of hypospadias patients. Hypospadias patients who were satisfied with their circumcised status had a significantly more positive genital perception than patients who were

Summary

dissatisfied with their circumcised status. Hypospadias patients who considered their penile appearance to be similar to that of other males had a more positive genital perception than patients who perceived their penis to be different from that of other males. Adult hypospadias patients who had ever received comments on their penile appearance had a more negative genital perception than patients who never had encountered comments. This difference did not exist with the children and adolescents with hypospadias. Small negative correlations (p 's $< .10$) were found between the number of operations, the age at final surgery, and genital perception of adult patients only. No statistically significant differences were found in the genital perception of hypospadias patients treated with different surgical procedures.

In chapter 7, the agreement between hypospadias patients' and the surgeon's satisfaction with the cosmetic surgical result, as well as the relationships between penile length, meatal position, and patients' satisfaction are investigated. Through a standardized questionnaire, 35 hypospadias patients (aged 9 to 18 years) and their pediatric urologist each expressed their level of satisfaction with the cosmetic surgical result. Hypospadias patients were significantly less satisfied than their surgeon with penile and glanular size and shape, and with the appearance of their testes and scrotum. In general, there was a poor agreement between patients' and surgeon's satisfaction with the cosmetic result, except for satisfaction with scars. Mean stretched penile size was 8.4 cm (range: 3.0-15.0 cm). Significantly more patients (21%) fell below the 10th percentile for stretched penile length of normal age matched boys. A high correlation existed between penile length and subject age. No statistically significant difference between penile length of patients with distal and proximal hypospadias was found. The meatus was situated at a glanular position in 69% and at a coronal position in 31%. There did not exist a positive correlation between penile length and the patients' genital perception. Patients who fell below the 10th percentile for stretched penile length were equally satisfied with flaccid penile size as patients who fell above the 10th percentile. Patients with a glanular meatus were more satisfied with their meatal position than patients with a coronal meatus. Follow-

ing the physical examination, 4 boys (11%) were re-operated because of a poor surgical result.

In chapter 8, the psychosocial adjustment of children, adolescents, and adults following hypospadias surgery is described. Psychosocial functioning was assessed through standardized self-report questionnaires on personality (DPQ/DPQ-J), social anxiety (COI/SAS-C), and emotional/behavioral problems (CBCL/YSR/YASR). Also, the effects of subjects' age, various medical characteristics, and coping with hypospadias on psychosocial functioning were investigated. Hypospadias patients had a similar psychosocial adjustment as age-matched comparison subjects, except 25- to 38-year-old hypospadias patients who reported more vague somatic complaints and anxieties, more feelings of inadequacy, and more social anxieties. Adult hypospadias patients had similar qualified professions as adult comparison subjects. The occurrence of "sex problems" (derived from the CBCL/YSR/YASR) and enuresis (only 9- to 18-year-olds) was approximately similar in hypospadias patients as in comparison subjects. Neither the severity of hypospadias nor the various medical characteristics affected psychosocial functioning significantly. Hypospadias patients who were satisfied with their genitals and body reported a somewhat better psychosocial functioning than patients who were less satisfied.

In chapter 9, the major findings of the present thesis are discussed and clinical as well as research recommendations are given. Following the results of the present study, we advocate to give parents and patients proper information. It is important to explain to parents that after hypospadias surgery the penis is circumcised, that circumcision is a normal routine procedure in many cultures and is also performed for non-medical reasons, and that a circumcised *normal* penis looks almost identical as a circumcised *hypospadiac* penis. We believe that this information will help in accepting the different appearance of the circumcised penis. However, if foreskin preserving surgery is feasible, it should be offered as an alternative to those parents and patients who pre-operatively have extreme difficulties with circumcision.

Because penile size was a major motive for concern, parents and patients should be informed that surgery will not enlarge penile size. Patients who

have extreme difficulties with accepting their (small) penis should be offered professional psychological help. Adolescent or adult patients with hypospadias with a micropenis should be offered proper counseling. If psychological or sexological counseling does not help and the patient keeps suffering from severe psychological distress, it might be considered to offer phallic reconstruction or penile lengthening.

Hypospadias surgeons can now inform and reassure parents and patients that there are no indications to expect poorer sexual development or functioning, nor poorer psychosocial functioning than males without hypospadias. This should diminish parental anxieties about their son's masculine capabilities, and could help patients to cope with their hypospadias. However, this does not rule out the possibility that a small number of patients might have problems with psychosexual or psychosocial adjustment, to whom professional psychological or sexological help should be given. We strongly advocate that hypospadias surgeons should see their patients during adolescence at least once as a standard therapeutic procedure, since: hypospadias patients may be reluctant to seek medical advice for their problems on their own initiative; boys with hypospadias are treated at increasingly earlier ages and therefore are not able to express their (dis)satisfaction with the surgical result; in the present study adolescents with hypospadias more often wanted cosmetic or functional penile improvement than children with hypospadias; complications, such as meatal retraction or stenosis, residual penile curvature, or adult sexual dysfunction often become apparant many years after surgical treatment is finished; and in the present study many adult hypospadias patients had questions or worries.

From the present research it may be concluded that it is preferable to operate with a terminalizing procedure at an early age with as few operations as possible. However, the results of the present study also seem to indicate that a ventralizing procedure can be performed safely (e.g., in countries with less specialized health care), without negatively affecting psychosexual and psychosocial adjustment.

The results of this work should be implemented by surgeons, psychologists, and sexologists in their care of hypospadias patients. Then, a major

objective of this thesis has been accomplished and the hypospadias patients' and comparison subjects' efforts in participating in the present study have been worthwhile.

SAMENVATTING

Het doel van dit proefschrift was de psychoseksuele en psychosociale ontwikkeling, en genitaalwaardering van kinderen, adolescenten en volwassenen na chirurgische hypospadie correctie te bestuderen, in vergelijking met die van op leeftijd gematchte vergelijkingspersonen (geopereerd vanwege een liesbreuk). Daarnaast werden ook relaties bestudeerd van verscheidene medische factoren (hypospadie ernst, leeftijd waarop de behandeling werd afgerond, aantal operaties, operatietype), huidige leeftijd en het omgaan met het hebben van hypospadie met de psychoseksuele en psychosociale ontwikkeling, en genitaalwaardering ontwikkeling.

In hoofdstuk 1 wordt een overzicht beschreven van de embryologie, etiologie, morfologie en chirurgische behandeling van hypospadie. Hypospadie wordt veroorzaakt door een stoornis in de ontwikkeling van de pisbuis (urethra) met als gevolg een abnormale positie van het plasgaatje (meatus). De gerapporteerde incidentie van hypospadie varieert van 0,8 tot 8,2 per 1.000 levend geboren jongens. Verscheidene factoren zoals (geslachts) chromosoom afwijkingen, autosomale (dominant/recessief) overerving, multifactoriële overerving, endocrinopathieën en omgevingsfactoren zijn geopperd hypospadie te veroorzaken. Echter, de exacte oorzaak van geïsoleerde hypospadie is tot op heden nog onbekend. Chirurgische reconstructie van de urethra en strekking van de penis is noodzakelijk, zodat staand urineren en ongestoord seksueel functioneren mogelijk is.

In hoofdstuk 2 wordt een uitgebreid literatuuroverzicht gegeven van studies over de psychoseksuele en psychosociale gevolgen van hypospadie. De meeste studies lijken aan te geven dat hypospadie patiënten een verhoogd risico hebben voor een vertraagde seksuele ontwikkeling, dat zij zich schamen voor of ontevreden zijn over het uiterlijk van hun penis en dat zij slechter psychosociaal functioneren. Omdat verscheidene methodologische tekortkomingen een juiste interpretatie van de onderzoeksresultaten bemoeilijken, is het onderhavige onderzoek uitgevoerd.

In hoofdstuk 3 worden de selectie van de patiënten en vergelijkingspersonen, de procedure, de respons, de kenmerken van de onderzoeksgroep en de vragenlijsten beschreven. Honderd zestien kinderen en adolescenten (9 t/m 18 jaar) en 73 volwassenen (18 t/m 38 jaar), en respectievelijk 88 en 50 leeftijd gematchte vergelijkingspersonen deden mee aan het huidige onderzoek.

In hoofdstuk 4 wordt de psychosociale ontwikkeling van volwassen hypospadie patiënten beschreven. Er werden geen significante verschillen gevonden tussen hypospadie patiënten en vergelijkingspersonen in de gemiddelde leeftijden waarop de seksuele mijlpalen werden bereikt. Ook bestonden er geen significante verschillen tussen het seksueel gedrag en functionaliteiten van hypospadie patiënten en vergelijkingspersonen. Meer seksueel actieve patiënten (33%) dan vergelijkingspersonen (13%) rapporteerden zich geremd te hebben gevoeld tijdens het zoeken van seksuele contacten. Hypospadie patiënten hadden een significant negatievere genitaalwaardering dan vergelijkingspersonen. Meer hypospadie patiënten (38%) dan vergelijkingspersonen (20%) vonden hun slappe penis te klein. Penislengte was de voornaamste reden voor ontevredenheid over het penis uiterlijk. De meeste hypospadie patiënten (60%) meldden dat het besneden uiterlijk van hun penis de reden was waarom zij vonden dat het uiterlijk van hun penis verschilde met dat van andere mannen. Ofschoon de ernst van de hypospadie een negatieve invloed had op genitaalwaardering en remmingen in het zoeken van seksuele contacten, had het geen significant effect op de seksuele ontwikkeling en gedrag. De operatie procedures hadden geen invloed op de seksuele ontwikkeling, noch op de genitaalwaardering van hypospadie patiënten. Positieve correlaties werden gevonden tussen het aantal operaties, de leeftijd bij de laatste operatie en de mate van remmingen in het zoeken van seksuele contacten. Hoe later de behandeling beëindigd werd, des te later patiënten begonnen met het maken van seksuele contacten. Zevenendertig procent van de patiënten wenste een verbetering van het cosmetisch of functioneel operatie resultaat. Naar aanleiding van het onderzoeksinterview zijn 5 patiënten (7%) opnieuw geopereerd vanwege een slecht functioneel of cosmetisch resultaat.

In hoofdstuk 5 wordt de psychoseksuele ontwikkeling van kinderen en adolescenten met hypospadie beschreven. Er werden geen significante verschillen gevonden tussen de seksuele ontwikkeling van patiënten en vergelijkingsjongens. Er bestonden ook geen significante verschillen tussen het seksueel gedrag en functioneren van hypospadie patiënten en vergelijkingsjongens. Zestien procent van de 9 tot 12 jarige hypospadie patiënten en niemand van de vergelijkingsjongens verwachtte geremd te worden in het zoeken van seksuele contacten. Meer 13 tot 18 jarige hypospadie patiënten (24%) dan vergelijkingsjongens (2%) rapporteerden zich geremd te voelen in het zoeken van seksuele contacten. Hypospadie patiënten met tenminste tongzoen-ervaring waren vaker bang uitgelachen te zullen worden door een partner vanwege het uiterlijk van hun penis dan vergelijkingsjongens. Hypospadie patiënten hadden een significant negatievere genitaalwaardering dan vergelijkingsjongens. Meer patiënten (25%) dan vergelijkingsjongens (12%) waren ontevreden over de lengte van hun slappe penis. Littekens, penislengte en vorm van de eikel waren de meest genoemde redenen voor ontevredenheid. Het besneden uiterlijk van de penis werd het vaakst genoemd (48%) als reden waarom hypospadie patiënten vonden dat het uiterlijk van hun penis anders was dan dat van andere jongens. Ofschoon de ernst van de hypospadie een negatieve invloed had op de mate van remmingen in het zoeken van seksuele contacten, had het geen significant effect op de seksuele ontwikkeling, noch op de genitaalwaardering. De operatie procedures hadden geen invloed op de seksueel ontwikkeling, noch op genitaalwaardering van hypospadie patiënten. Er bestond een tendens dat patiënten die geopereerd waren met een ventraliserende procedure (44%) vaker een verbetering van het cosmetisch of functioneel resultaat wilden dan patiënten die geopereerd waren met een terminaliserende procedure (29%). Er werden geen significante correlaties gevonden tussen het aantal operaties, de leeftijd waarop de behandeling beëindigd was en de psychoseksuele ontwikkeling van hypospadie patiënten. Meer 13 tot 18 jarige patiënten (32%) dan 9 tot 12 jarige patiënten (13%) waren ontevreden met het uiterlijk van hun penis, en meer van de 13 tot 18 jarigen (49%) dan van de 9 tot 12 jarigen (24%) wensten een cosmetische of functionele verbetering van het operatie resultaat.

In hoofdstuk 6 wordt de genitaalbeleving van kinderen, adolescenten en volwassenen beschreven die geopereerd waren vanwege een hypospadie. Genitaalbeleving werd gemeten door middel van twee gestandaardiseerde zelfrapportage vragenlijsten (kind/adolescent en volwassen versie). Hypospadie patiënten hadden een negatievere genitaalbeleving dan op leeftijd gematchte vergelijkingspersonen. Hypospadie patiënten waren voornamelijk minder tevreden over de grootte en vorm van hun penis en eikel, en over de plaats van de meatus. Er werden geen significante correlaties gevonden tussen genitaalbeleving en leeftijd van hypospadie patiënten. Patiënten die tevreden waren met het besneden uiterlijk van hun penis hadden een significant positievere genitaalbeleving dan patiënten die hierover ontevreden waren. Patiënten die vonden dat hun penis er hetzelfde uitzag als bij andere mannen hadden een positievere genitaalbeleving dan patiënten die zeiden dat hun penis er anders uitzag dan die van andere mannen. Volwassen patiënten die ooit opmerkingen hadden gekregen over het uiterlijk van hun penis hadden een negatievere genitaalbeleving dan patiënten die nog nooit opmerkingen hadden gekregen. Dit verschil in genitaalbeleving bestond niet bij de kinderen en adolescenten met een hypospadie. Er bestonden alleen negatieve correlaties tussen het aantal operaties, de leeftijd bij de laatste operatie en de genitaalbeleving van volwassen patiënten. De verschillende operatieprocedures hadden geen statistisch significante invloed op de genitaalbeleving van patiënten.

In hoofdstuk 7 worden de overeenstemming in tevredenheid over het cosmetische operatieresultaat tussen patiënten en hun chirurg, en de relatie tussen penislengte, plaats van de meatus en de tevredenheid van de patiënten bestudeerd. Door middel van een gestandaardiseerde vragenlijst werd zowel de tevredenheid met het operatieresultaat van 35 patiënten (9 t/m 18 jaar) als die van hun chirurg gemeten. De patiënten waren significant minder tevreden dan hun chirurg met de grootte van hun penis en eikel en met het uiterlijk van hun testikels en scrotum. In het algemeen bestond er nauwelijks enige overeenstemming tussen de tevredenheid van patiënten en chirurg, met uitzondering van de tevredenheid met de littekens. De gemiddelde uitgerekte penislengte bedroeg 8,4 cm (uitersten: 3,0-15,0 cm). Significant meer pa-

tiënten (21%) vielen beneden de 10^e percentiel voor gestrekte penislengte van normale naar leeftijd gematchte jongens. Er bestond een hoge correlatie tussen penislengte en leeftijd. Er werd geen significant verschil in penislengte gevonden tussen patiënten met een distale en een proximale hypospadie. Bij 69% van de patiënten lag de meatus glanulair (in de eikel) en bij 31% coronair (t.h.v. de kraag). Er bestond geen significante en positieve correlatie tussen penislengte en genitaalbeleving. Patiënten die beneden de 10^e percentiel voor penislengte vielen waren even tevreden over hun penislengte als patiënten die boven de 10^e percentiel vielen. Patiënten met een glanulaire meatus waren significant tevredener met de plaats van hun meatus dan patiënten met een coronaire meatus. Naar aanleiding van het lichamelijk onderzoek werden 4 patiënten (11%) opnieuw geopereerd vanwege een slecht operatieresultaat.

In hoofdstuk 8 wordt het psychosociaal functioneren van kinderen, adolescenten en volwassenen beschreven die geopereerd waren vanwege een hypospadie. Psychosociaal functioneren werd gemeten met behulp van gestandaardiseerde zelfrapportage vragenlijsten over persoonlijkheid (NPV-J/NPV), sociale angst (SAS-K/IOA) en emotionele problemen en gedragsproblemen (CBCL/YSR/YASR). Tevens werden de effecten van de huidige leeftijd, verscheidene medische variabelen en coping met hypospadie op het psychosociaal functioneren onderzocht. Hypospadie patiënten verschilden niet in hun psychosociaal functioneren, vergeleken met op leeftijd gematchte vergelijkingspersonen, behalve 25 to 38 jarige patiënten; zij rapporteerden meer vage lichamelijke klachten en vage angsten, meer inadequatie gevoelens en meer sociale angst. Volwassen patiënten verschilden niet van vergelijkingspersonen in hun beroepsniveau. "Seks problemen" (items van de CBCL/YSR/YASR) en enuresis (broek- en bedplassen) kwamen ongeveer even vaak voor bij hypospadie patiënten als bij vergelijkingspersonen. Noch de ernst van de hypospadie, noch de verscheidene medische variabelen hadden een significant effect op het psychosociaal functioneren. Hypospadie patiënten die tevreden waren met hun genitaal en lichaam functioneerden psychosociaal iets beter dan patiënten die ontevreden waren.

In hoofdstuk 9 worden de belangrijkste bevindingen van het gehele onderzoek bediscussieerd en worden zowel klinische aanbevelingen als suggesties voor toekomstig onderzoek gegeven. Naar aanleiding van de resultaten van het huidige onderzoek, raden wij aan zowel ouders als patiënten duidelijk voor te lichten dat na hypospadie chirurgie de penis een besneden uiterlijk heeft, dat een besnijdenis een normale routinematige procedure is in veel culturen en dat het vaak wordt uitgevoerd vanwege niet medische redenen, en dat een besneden *normale* penis zeer veel lijkt op een besneden *hypospade* penis. Wij denken dat ouders en patiënten door deze informatie gemakkelijker het besneden uiterlijk van de penis accepteren. Echter, indien een voorhuid reconstruerende operatie technisch mogelijk is, zou dit als alternatief moeten worden aangeboden aan diè ouders en patiënten die pre-operatief zeer veel problemen hebben met een besneden uiterlijk van de penis.

Omdat penislengte een voorname reden voor bezorgdheid was, moet aan ouders en patiënten uitgelegd worden dat een operatieve hypospadiecorrectie de penis niet zal verlengen. Patiënten die psychologische problemen hebben met het accepteren van hun (kleine) penis, moet professionele psychologische of seksuologische hulp worden aangeboden. Adolescente of volwassen hypospadie patiënten met een micropenis moeten goed begeleid worden. Indien professionele psychologische of seksuologische hulp niet mocht helpen en de patiënt gebukt blijft gaan onder psychologisch leed, zou chirurgische penisreconstructie of penisverlenging overwogen kunnen worden.

Naar aanleiding van dit onderzoek kunnen hypospadie chirurgen ouders en patiënten voorlichten dat er geen aanwijzingen bestaan om een gestoorde seksuele ontwikkeling of seksueel functioneren, noch een gestoord psychosociaal functioneren te verwachten. Deze informatie zou de zorgen van de ouders over de "mannelijkheid" van hun zoon kunnen verminderen en zou patiënten kunnen helpen in het omgaan met hun hypospadie. Echter, dit sluit niet uit dat een klein aantal patiënten problemen met de psychoseksuele of psychosociale ontwikkeling kan hebben, aan wie professionele psychologische of seksuologische hulp gegeven dient te worden. Wij raden hypospadie chirurgen aan om hypospadie patiënten routinematig tenminste één maal

terug te zien in hun adolescentie, omdat: hypospadie patiënten niet geneigd zijn om uit zichzelf hulp te zoeken voor problemen; hypospadie patiënten tegenwoordig steeds vroeger geopereerd worden, waardoor zij hun (on)tevredenheid met het operatie resultaat niet zelf kunnen uiten; in dit onderzoek 13 to 18 jarige hypospadie patiënten vaker aangaven een cosmetische of functionele verbetering van het operatie resultaat te wensen dan 9 tot 12 jarige patiënten; complicaties zoals terugtrekking of vernauwing van de meatus, (rest)kromstand van de penis of seksuele problemen vaak optreden vele jaren nadat de behandeling beëindigd is; en veel volwassen hypospadie patiënten in dit onderzoek vragen of zorgen hadden.

Naar aanleiding van dit onderzoek kan geconcludeerd worden dat het de voorkeur verdient hypospadie patiënten te opereren op een zo jong mogelijke leeftijd met zo weinig mogelijk operaties en met een terminaliserende procedure. De resultaten van dit onderzoek geven echter ook aan dat ventraliserende technieken uitgevoerd kunnen worden (b.v. in landen met een minder gespecialiseerde gezondheidszorg), zonder de psychoseksuele en psychosociale ontwikkeling van de patiënten negatief te beïnvloeden.

De resultaten van dit promotie-onderzoek moeten worden geïmplementeerd door chirurgen, psychologen en seksuologen in hun zorg van hypospadie patiënten. Dan is het voornaamste doel van dit onderzoek bereikt en is de deelname van de hypospadie patiënten en vergelijkingspersonen aan het onderzoek de moeite waard geweest.

DANKWOORD

Allereerst gaat mijn dank uit naar de hypospadie patiënten, de controle personen en hun ouders die hun medewerking verleenden aan dit onderzoek door openhartig te durven spreken over zeer intieme onderwerpen en gevoelens. Hun medewerking heeft geleid tot een beter inzicht in de psychoseksuele en psychosociale ontwikkeling van hypospadie patiënten en zal hopelijk resulteren in verbeteringen in het voorlichtings- en begeleidingsproces van hypospadie patiënten en hun ouders.

Voorts wil ik mijn promotoren, Frank Verhulst en Koos Slob, en mijn copromotor Froukje Slijper bedanken voor hun begeleiding en ondersteuning in de afgelopen drie jaren.

Beste Frank, jouw beslissing om "de zoveelste geneeskunde student met wat vage onderzoeksplannen" een kans te geven, is van cruciaal belang geweest voor het slagen van dit onderzoek. Nadat jij mij in januari 1993 een vaste werkplaats gaf op de afdeling Kinder- en Jeugdpsychiatrie, waar ik gebruik kon maken van de aanwezige faciliteiten en kon leren van de expertise van jouw afdeling, is het onderzoek in een stroomversnelling geraakt. Jouw aanvankelijke reserves maakten snel plaats voor je onvoorwaardelijke steun aan en enthousiasme over het onderzoek, wat onder meer heeft geresulteerd in een aantal subsidies. Naast iemand met wie het zeer plezierig is om samen te werken, heb ik je ook leren kennen als een buitengewoon gezellig persoon die op zijn tijd in is voor een geintje en een biertje (al koste dat wel een keer het achterwiel van je fiets!).

Beste Koos, jij was als geen ander in staat mij met "een goed gevoel" naar huis te laten gaan, zelfs wanneer ik naar je toe kwam en het even niet meer zag zitten. Ik heb zeer veel waardering voor jouw positieve en enthousiaste instelling, waaruit ik veel energie en vertrouwen putte, alsmede voor de manier waarop jij jouw altijd opbouwende kritiek op mij overbracht. Daarnaast ben ik je zeer dankbaar voor de hoeveelheid tijd en energie die jij (vaak 's avonds laat) in het onderzoek gestopt hebt. Hoeveel uren heb je niet met en zonder mij boven vragenlijsten of manuscripten gehangen, om daar je

kritische visie op te geven? De vaak lange discussies die ik met je voerde over argumenten voor of tegen mogelijke verklaringen, of over een betere manier om een zin in het Engels op te schrijven, waren voor mij zeer leerzaam en uitdagend! Jouw bijdrage aan het onderzoek heeft tot onmiskenbare verbeteringen geleid.

Beste Froukje, met name jouw adviezen om een vergelijkingsgroep als ook patiënten van de afdeling Kinderurologie in het onderzoek te betrekken, blijken "gouden zetten" geweest te zijn. Mede door deze uitbreidingen van het onderzoek, waarvoor ik in eerste instantie vanwege de tijdsdruk huiverig was, is het onderzoek drastisch verbeterd en een succes geworden. Tevens heb jij mij geleerd, door middel van jouw klinisch inzicht en praktische instelling, droge data en resultaten te vertalen in heldere implicaties en aanbevelingen voor de hulpverleners in de kliniek. Jouw klinisch inzicht en je aanbevelingen hebben me tevens enorm geholpen bij het structureren van mijn gedachten en verwachtingen over de psychoseksuele en psychosociale ontwikkeling van hypospadie patiënten, wat het schrijfproces een stuk vergemakkelijkt heeft.

Rien Nijman en Professor van der Meulen ben ik zeer erkentelijk voor hun bijdrage aan het onderzoek. Beste Rien, jouw pragmatische instelling maakt je iemand met wie het snel en plezierig samenwerken is. Ik ben je zeer dankbaar dat je bereid was om (zelfs in het weekend!) je schaarse tijd te investeren in het terugzien van de patiënten van jouw afdeling. De wijze waarop jij met de patiënten en hun ouders omging, vond ik zeer leerzaam en bewonderenswaardig. Daarnaast las jij met je kritische "chirurgenoog" mijn manuscripten, die je vaak al binnen een week gecorrigeerd terugstuurde. Hoe je dat zo snel voor elkaar kreeg naast je drukke baan en gezinsleven is voor mij nog steeds een raadsel.

Geachte Professor van der Meulen, u hebt op verschillende manieren aan de wieg gestaan van dit onderzoek. U hebt mij in eerste instantie de mogelijkheid geboden om mezelf op de afdeling Plastische en Reconstructieve Chirurgie te verdiepen in de problematiek rondom de behandeling van hypospadie patiënten. Daarnaast zag u in, toen het duidelijk werd dat ik me niet zozeer met de chirurgische aspecten alswel met de psychoseksuele en

psychosociale aspecten van hypospadie ging bezighouden, dat goede begeleiding door deskundigen in de betreffende vakgebieden nodig was. U hebt er mede voor gezorgd dat ik op de afdeling Kinder- en Jeugdpsychiatrie terecht kon komen. Graag wil ik u bedanken voor uw belangstelling en adviezen, die u vanaf het allereerste begin van het onderzoek gegeven heeft.

De medisch studenten Manon Bos en Martine Biewenga en Johan van Hattem, student psychologie, dank ik voor de enorme bergen werk die zij in korte tijd voor mij verzet hebben door de vergelijkingspersonen te interviewen en door de vragenlijsten in te voeren in de computer. Dankzij hun inspanningen heb ik het onderzoek zo snel en voorspoedig kunnen uitvoeren.

Jan van der Ende wil ik graag bedanken voor zijn nimmer afnemende bereidheid te hebben willen luisteren naar mijn (statistische) problemen. Beste Jan, jij hebt mij als geen ander geleerd de resultaten van mijn (en andermans) onderzoek te relativiseren door mij te laten inzien dat ook de statistiek geen absolute zekerheden kan verschaffen. Daarnaast was het altijd zeer gezellig en ontspannend om even bij je binnen te lopen om met jou over de meest uiteenlopende onderwerpen van gedachten te wisselen. Je was een zeer plezierige collega, en het mag best weleens gezegd worden dat de afdeling enorm boft met zo iemand als jij.

De medewerkers van de patiëntencodering en archivering van het SKZ, en met name Maarten van de Lippe, dank ik voor hun snelle en gerichte zoekacties naar de honderden patiëntendossiers. Luca Incrocci ben ik zeer erkentelijk voor het vertalen van de Italiaanse publikaties. Frans Bentvelsen en Philippe Weijerman dank ik voor de keren dat zij de patiënten onderzochten wanneer Rien verhinderd was. Robert Ferdinand en mijn jaarclubgenoten Robert Schulte, Arthur Richardson en Ronald van den Mosselaar dank ik, omdat zij bereid waren als proefpersoon te dienen waardoor Manon en Martine zich het interviewen enigszins eigen konden maken alvorens zij "het veld" ingingen. Fotograaf Wil Hofman dank ik voor het kosteloos ter beschikking stellen van de coverfoto. Mijn clubgenoot Maarten van den Bosch ben ik zeer erkentelijk voor de schitterende tekeningen en voor het ontwerp van de kافت. Klasse gedaan Maarten!

Dankwoord

Mijn lieve ouders dank ik voor de mogelijkheden die zij mij geboden hebben om mijn studie en promotie-onderzoek te kunnen voltooien, en natuurlijk voor hun onvoorwaardelijke steun en liefde die zij mij gedurende mijn hele leven gegeven hebben.

Tot slot wil ik alle medewerkers van de afdeling Kinder- en Jeugdpsychiatrie bedanken voor drie jaar zeer plezierig samenwerken. Mede dankzij jullie ging ik elke dag met plezier naar mijn werk, wat mijn werklust zeer ten goede kwam. Het waren leuke, gezellige en leerzame jaren die zeer snel voorbij gegaan zijn. Ik zal er met een goed gevoel naar blijven terugkijken.

Rotterdam, juli 1995,
Marc Mureau.

CURRICULUM VITAE

Marc Mureau werd geboren op 31 januari 1970 te Prinsenbeek. In 1988 behaalde hij zijn V.W.O. diploma aan het Mencia de Mendoza Lyceum in Breda. In hetzelfde jaar begon hij met de studie geneeskunde aan de Faculteit der Geneeskunde en Gezondheidswetenschappen van de Erasmus Universiteit Rotterdam, waar hij in maart 1995 zijn doctoraalexamen aflegde. Van januari 1991 tot december 1992 inventariseerde hij op de afdeling Plastische en Reconstructieve Chirurgie van het Academisch Ziekenhuis Rotterdam de medische gegevens van 571 hypospadië patiënten. Deze inventarisatie diende als basis voor het in dit proefschrift beschreven onderzoek, dat van januari 1993 tot juni 1995 werd uitgevoerd op de afdeling Kinder- en Jeugdpsychiatrie, in nauwe samenwerking met de afdelingen Endocrinologie en Voortplanting, Plastische en Reconstructieve Chirurgie en Kinderurologie van het Academisch Ziekenhuis Rotterdam/Erasmus Universiteit Rotterdam. Sinds mei 1995 is de auteur bezig met zijn co-assistent-schappen teneinde zijn opleiding tot arts te voltooien.



