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Anomalies of the penis and scrotum in adults

Nugteren, Helena Madelinde

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Anomalies of the penis and scrotum in adults

A multidisciplinary approach

Helena Madeline Nugteren

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A multidisciplinary approach

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General introduction

1

There are many different scrotal and penile anomalies, some common and some rarely encountered. They affect men of all ages and symptoms can vary considerably depending on the particular condition involved. Despite their heterogeneous pathological background, they are all likely to cause psychological distress, sexual dysfunction, and may disrupt sexual life and thereby diminish quality of life. Given this broad variety of “problems”, it is not surprising that a broad variety of “solutions” are also available and used in clinical practice. Some of the anomalies require surgical treatment, while other times a more psychological or counseling approach is put into practice; however, very often it is a multidimensional approach that prevails. Human sexuality is an illustration of bio-psycho-social interaction, and, when problematic, professional help needs to encompass all three of these domains. Depending on the exact nature of the problem and its context, one or more treatment options may be chosen and/or combined. Just as scientists do, clinicians use the principle of Ockham’s razor: Keep it as simple as possible. If a simple, preferably non-invasive intervention suffices, then this should prevail.

In this thesis, four scrotal and penile anomalies are studied: the short penis, the acute erection angle, Peyronie’s disease, and genital lymph edema. Some of these pathologies are so rarely encountered in our daily practice that their treatment is mainly based on the personal experience of the surgeon or of his/her colleagues.

The first aim of this introduction is to provide general information about these four anomalies, including their prevalence, etiology, and treatment. The second aim of this introduction is to describe the two main research questions, a number of sub-questions, and the accompanying outline of this thesis. In the following chapters, each one of the four anomalies will be put into a urological and sexological perspective. In order to arrive at a clear insight into these kinds of urological anomalies and their treatment options, it is important to understand their various anatomical backgrounds. A short review of the embryologic aspects of the male urogenital tract, which form the basis for congenital anomalies, is provided.

EMBRYOLOGY OF THE MALE GENITALS

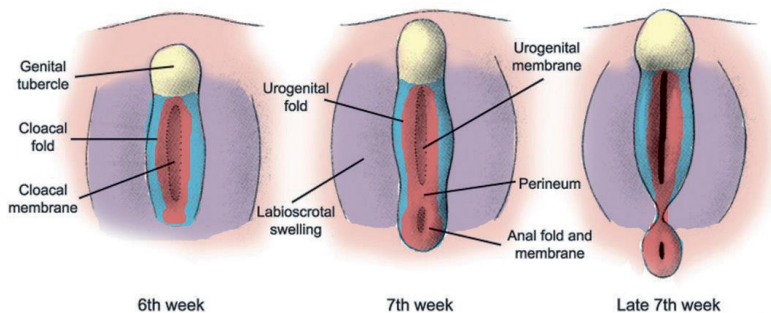
Early development of the external genitalia is similar in both genders. External genital differentiation for the male sex begins in the seventh week of gestation and is completed by the sixteenth to seventeenth week. It is a complex process involving SRY genetic programming, cell differentiation, hormonal signaling, enzyme activity, and tissue remodeling.

Under the influence of the SRY gene, the bipotential gonad differentiates into a testes ultimately resulting in androgen production. External genital development undergoes two stages that can be distinguished by independent hormone growth prior to testicular development and hormone dependent growth under the influence of androgens. Hormone

independent development occurs between conception and seven to eight weeks' gestation.¹ This takes place under the influence of a cascade of genes including sonic hedgehog, BMP4, Glia 123, and Wilms Tumor gene.

While the anorectal canal and urogenital sinus are being separated by the urorectal septum, the mesoderm anterior and cranial to the phallic segment of the urogenital sinus expands. This forms the genital tubercle, which will eventually form the phallic segment. In this indifferent stage around the third week of development, mesenchyme cells originating in the region of the primitive streak migrate around the cloacal membrane to form a pair of cloacal folds (or urogenital folds). Cranial to the cloacal membrane, the folds unite to form the genital tubercle, whereas in the caudal part cloacal folds are subdivided into the urethral folds anteriorly and into the anal folds posteriorly (figure 1).^{2,3} The urogenital (cloacal) membrane starts to resolve in the seventh week, forming a urogenital sinus. The formation of the penis starts by elongation of the genital tubercle to form the corpora cavernosa and glans penis. By the tenth week, the urethral folds start to fuse from the urogenital sinus orifice toward the tip of the phallus. At the fourteenth week, the fusion is complete and the penile urethra is formed. The corpus spongiosum results from the differentiation of the mesenchymal masses around the urethra. The glandular urethra

A. Indifferent stage



B. Male

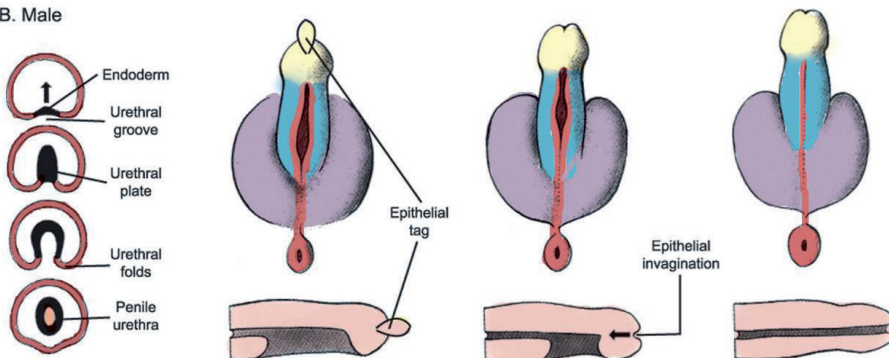


Figure 1. Human urogenital embryology. (A) Sex undifferentiated precursors prior to 7 weeks gestational age. (B) Male development in gestational weeks 7-17.³

develops as a result of canalization of an ectodermal epithelial cord that has grown through the glans. During the third month, the prepuce is formed by growing a fold at the base of the glans. Meanwhile, the genital swellings shift caudally and are recognizable as scrotal swellings. They meet and fuse, resulting in the formation of the scrotum.^{4,5}

During this complex developmental process, not only is the basis laid for a large variety of morphological appearances but also for anatomical and/or physiological anomalies. Sometimes the difference between “normal variation” and pathology is hard to discern. Therefore, we will now focus in more detail on the four anomalies, which together form the core of this thesis.

THE SMALL PENIS

The mechanism of growth cessation is still not completely clear, and therefore little is known about the cause of a penis that is too small. A small penis can occur as a separate abnormality, or it can be part of a series of anatomical abnormalities, such as bladder extrophy, epispadias, hypospadias, and disorders of sex development (DSD).

There are three anomalies that need further attention and will be discussed here: the micropenis, the small penis, and the webbed penis. Following on this, treatment for a small penis, which has a long history with broad cultural differences, will be briefly presented.

The micropenis

A small penis is not always a micropenis (figure 2). A micropenis refers to a specific disorder that has a specific set of causative factors and a different set of treatment modalities than does the small penis. The true micropenis is the result of a hormonal abnormality occurring after twelve weeks of gestation. There are three main causes for this condition: hypogonadotropic hypogonadism, hypergonadotropic hypogonadism (also known as primary hypogonadism), and idiopathic causes (table 1).^{6,7}



Figure 2. A micropenis

Table 1. Etiology of micropenis⁶

I	Deficient testosterone secretion
A	Hypogonadotropic hypogonadism
1	Isolated, including Kallmann's syndrome
2	Associated with other pituitary hormone deficiencies (e.g., CHARGE association)
3	Prader-Willi syndrome
4	Laurence-Moon syndrome
5	Bardet-Biedl syndrome
6	Rud's syndrome
B	Primary hypogonadism
1	Anorchia
2	Klinefelter's syndrome and poly X syndrome
3	Gonadal dysgenesis (incomplete)
4	Luteinizing hormone receptor defects (incomplete)
5	Genetic defects in testosterone steroidogenesis (incomplete)
6	Noonan's syndrome
7	Down syndrome
8	Robinow's syndrome
9	Bardet-Biedl syndrome
10	Laurence-Moon syndrome
II	Defects in testosterone action
A	Growth hormone/insulin-like growth factor-I deficiency
B	Androgen receptor defects (incomplete)
C	5 α -reductase deficiency (incomplete)
D	Fetal hydantoin syndrome
III	Development anomalies
A	Aphallia
B	Cloacal exstrophy
IV	Idiopathic causes
V	Associated with other congenital malformations

In a study by Nelson and colleagues the incidence of a micropenis was reported as 1.5 in 10,000 male children born in the United States of America (USA) between 1997 and 2000.⁸

To make an accurate diagnosis, the stretched penis length (SPL) should be measured.⁹ This length is measured by stretching the flaccid penis to its maximum length and to measure from the pubis to the tip of the stretched penis. Micropenis is defined as SPL 2.5 standard deviations less than the mean for a group without penile anomalies (table 2).¹⁰ The micropenis can be associated with cryptorchidism and small-volume testicles, as well as a hypoplastic scrotum, most likely due to the same causative factors that are responsible for the micropenis.⁶

The work up of the patient with a micropenis should be directed towards early diagnosis and therapy. Micropenis should be diagnosed in the neonatal period and differentiated from associated deformities and syndromes. Therefore, the patient should be referred to a (pediatric) endocrinologist. Endocrinologic assessment helps to determine at what level the cause of the micropenis is to be found, in terms of the hypothalamic-pituitary-testicular axis. Measurement of serum gonadotropins, testosterone, DHT, and precursors of testosterone are often helpful in evaluating the etiology. Sometimes genetic testing may be necessary to identify genetic sex and to rule out gonadal dysgenesis with a Y chromosome cell line. The most important concern is whether the child will have sufficient penile growth to allow sexual function as an adult.^{11,12}

Table 2. Normal Stretched Penis Length (SPL)¹⁰

Age	Mean \pm SD	Mean – 2.5 SD
Newborn, 60-week gestation	2.5 \pm 0.4	1.5
Newborn, 34-week gestation	3.0 \pm 0.4	2.0
0-5 months	3.9 \pm 0.8	1.9
6-12 months	4.3 \pm 0.8	2.3
1-2 years	4.7 \pm 0.8	2.6
2-3 years	5.1 \pm 0.9	2.9
3-4 years	5.5 \pm 0.9	3.3
4-5 years	5.7 \pm 0.9	3.5
5-6 years	6.0 \pm 0.9	3.8
6-7 years	6.1 \pm 0.9	3.9
7-8 years	6.2 \pm 1.0	3.7
8-9 years	6.3 \pm 1.0	3.8
9-10 years	6.3 \pm 1.0	3.8
10-11 years	6.4 \pm 1.1	3.7
adult	13.3 \pm 1.6	9.3

In the management of patients with micropenis, an assessment should be made as to the ability of the penis to respond to testosterone application. If the penis does not increase in length, there is probably an androgen resistance and therefore failure to virilize at puberty.

Is it important that the treatment with testosterone starts in early infancy? There is a natural decrease in androgen receptors during the early adulthood period and early

application of testosterone allows for penile androgen receptor concentration to increase.¹² In patients who do not respond to testosterone, application of 5- α dihydrotestosterone gel might be an alternative.¹³ If the micropenis does not show sufficient growth despite endocrine treatment, surgical treatment can offer an alternative. Different penile reconstructions are described, of which the radial forearm flap remains the most popular.¹⁴ Cosmetic and functional results are acceptable; the complication rate, however, is very high.⁶ A large study by Monstrey and colleagues showed a flap revision rate of 12%.¹⁵

In the patient with a micropenis, psychological counseling should be an integral part of management. It is important to minimize stigmatization and achieve greater self-acceptance as well as social acceptance of their condition.¹⁶

The small penis

According to the American sexologist Barry McCarthy, two out of three men believe that their penis is too small.¹⁷ He attributes the worries they have about the length of their penis to various factors. First, little boys see their father's penis for the first time at a "sensitive" age. Second, in a locker room, men usually see other men from the front. Apparently, the other man's penis is larger, because a man can only see his own penis from above. From above, one has an effect that artists refer to as "fore-shortening." The penis looks smaller than it really is. Third, there are considerable differences in the lengths of flaccid penises, while in erection there is not much difference. Fourth, men generally do not know all that much about the subject, because they do not like to talk openly about such intimate matters. To these factors a new factor can be added, which stems from the context of the Internet. The Worldwide Web has made pornography available to everybody, and young adult men especially see a lot of penises in action. These penises, however, usually belong to porn stars, who are selected for mega-sized penises. Like young women seeking "designer vulvas," young adult men think their penis should match the size of a porn star's penis.

The size of penises has a long history as a topic of scientific research and debate. In 1899, the German physician Loeb performed a study on fifty men aged between 18 and 35 years.¹⁸ The length of the visible part of the flaccid penis varied from 8 to 11 centimeters (average 9.4 cm) and the circumference from 8 to 10.5 cm. The Kinsey Report showed that only one quarter of men had an average-sized penis.¹⁹ However, extremes were fairly rare. Five percent of the men had an erection of less than 9 cm, and one percent was very well endowed with an erection of longer than 20 cm.

Doctor Jacobus X was the pseudonym of the surgeon in the French army who spent many years of his life examining and measuring hundreds of male and female genital organs from all corners of the world.²⁰ Africans had the longest penises: flaccid 12.5 to 15 cm, in erection 19 to 20 cm. According to this surgeon, the size of the penis was always closely linked to the size of the vagina of each particular race. "Hindu women whose men

have a short and thin penis, will have trouble with the average European,” wrote the army doctor. “The enormous penis of the African Negro would, in their view, be an instrument of torture.” Jacobs seems to be saying that Mother Nature has made sure that members of the same race seek each other out. The mixing of races would be unnatural. Nowadays, this notion is frowned upon, but in the 1930s such ideas were not uncommon. In 2007, Wylie and Eardley also reviewed measurements of penile sizes in different races (table 3).²¹

The webbed penis

A special form of small penis is the “webbed” penis (figure 3). It is in fact not small, but it does seem so, because the penis and the skin of the scrotum are joined together forming a so-called “web.” This may lead to problems with condom usage, because not only are standard diameters used but condoms also have the same diameter at the base as on the top. This may lead to a Procrustus dilemma: A condom that is too tight will lead to complaints, ranging from “it doesn’t feel comfortable” to “it is strangling me.” In contrast, a condom that is too loose may slip off accidentally. In this light, the study of a Dutch sexologist is of interest.²² Janssen performed a study on the circumference of the penis in erection and showed that, measured around the base, the average circumference of the Dutch penis in full erection was about 121 millimeters, with a range from 90 to 161 mm. In addition, in a quarter of the study subjects, the circumference of the penis in erection was less than 110 mm, in three quarters less than 130 mm, and in ninety percent less than 140 mm. The conclusion was that good instructions about condoms should always contain information about the circumference of the penis in relation to various sizes of condom.



Figure 3. The webbed penis

Table 3. A summary of reports of measurements of penile size²¹

Study	N	Mean or range age, years	Population	Raccid*			Erect*		
				length	stretched length	circumference	length	suprapubic fat depth	circumference
[13]	54	20-25	Caucasian	-	13.02	8.55	-	-	-
[54]	2770	20-59	-	9.7	-	-	15.5	-	-
[19]	156	-	Mostly Caucasian	-	-	-	16	13.5 (base)	-
[14]	80	54	White 67.5%, Black 20%, Asian 12.5%	8.85	12.5	9.7 (mid shaft)	12.89	2.85	12.3 (mid shaft)
[20]	184	-	Heterosexual 60%	10.4	-	-	15.71	-	13.2 (base)
[16]	813	30.8	All homosexual	10.4	-	9.8 (max)	16.4	-	12.6 (max)
	3417	30.5	All heterosexual	9.8	-	9.4 (max)	15.6	-	12.2 (max)
[15]	111	18-19	Potent German men	8.6	-	-	14.48	-	-
	32	40-68	German men with ED	9.22	-	-	14.18	-	-
[18]	3300	17-19	Italian men	9	12.5	10 (mid shaft)	-	-	-
[21]	200	20-22	Turkish men	6.8	8.98	-	12.7	-	-
[22]	104	54	British men	-	13 (median)	-	-	-	-
[17]	124	59	Before RP	9	13	9.5 (mid shaft)	-	2.5	-
	63	59	After RP	8	12.5	10 (mid shaft)	-	2	-
[11]	123	21.7	Korean men	6.9	9.6	8.5 (mid shaft)	-	1.1	-

*mean values, in cm, unless stated otherwise. RP, radical prostatectomy.

The malformation can be congenital or acquired. The acquired webbed penis is mainly iatrogenic, caused by the removal of too much skin on the ventral side during circumcision.

The etiology of the congenital webbed penis is unknown.²³ Some authors suggest that a webbed penis is secondary to aberrant preputial development, where there is insufficient ventral skin coverage of the penis and, as an alternative, the penis is covered by adjacent scrotal tissue.²⁴ A webbed penis can be associated with other malformations such as hypospadias, chordee, and micropenis.^{23,24}

In terms of surgical treatment of a webbed penis, it is usually sufficient to incise the skin in a transverse direction and stitch the wounds longitudinally. In patients with associated chordee and/or hypospadias, more extensive urethral reconstruction might be necessary.

A short history of penis enlargement

Throughout the centuries, men have been trying to make their penis longer. The most primitive way is by hanging a weight at the tip of the penis, such as stones. This may work, but also causes pain. In addition, there is the Polynesian stretching method using a moveable heavy pipe, or the Arabic “jelq” treatment (also known as “milking”). In the USA, one can buy equipment that performs massages according to the “jelq” method. The manufacturer claims that within twenty weeks, one can achieve an increase of two and a half centimeters in length. One can read all about it in a booklet by the American Gary Griffin.²⁵

According to Jolan Chang, writer of the book *The Tao of Love and Sex*, the answer lies particularly in practising.²⁶ Taoists believe that every part of the body can be trained and developed. The Tao method does not use artificial means or aids.

In the summer of 1993, the readers of the *Gaykrant*, a Dutch magazine for homosexuals, were surprised to read that in South Africa more than one hundred penile lengthening operations had been performed. When asked, the plastic surgeon from Johannesburg commented: “A month after the operation, the patients could resume their normal sex life. It leaves a scar that goes down as far as the scrotum. The patient receives an unbelievable amount of benefit, at such little inconvenience.” In the same article, it was said that it would not be long before the operation would be introduced in Western Europe. In August 1994, it was all set. A Dutch urologist, named Pieter Dik, told a national newspaper that he had ventured to perform the operation. This news coverage caused great upheaval. The hospital director, an influential professor of Andrology, a famous cosmetic surgeon, board members of the Dutch Society of Urologists, a health ethicist, and the chief editor of the *Gaykrant*, were all to have their say in various newspapers. Unfortunately, not only the urologist in question but all those who had commented did not seem to have done enough research in the scientific literature. The procedure was nothing more than a variation of a surgical procedure that has been known for many years in pediatric urology.²⁷

The essence of the surgical intervention is that the ligament which fixes the penis to the front of the pubic bone, the suspensory ligament, is cut. In this way, the “hanging” part of the penis becomes longer; it is optionally lengthened. At the same time, the surgeon cuts the skin in such a way that it can also slide towards the penis and performs a so-called V-Y plasty (figure 4). In other words, the incision has the form of a V, but after it has been sutured it forms a Y-shaped scar.

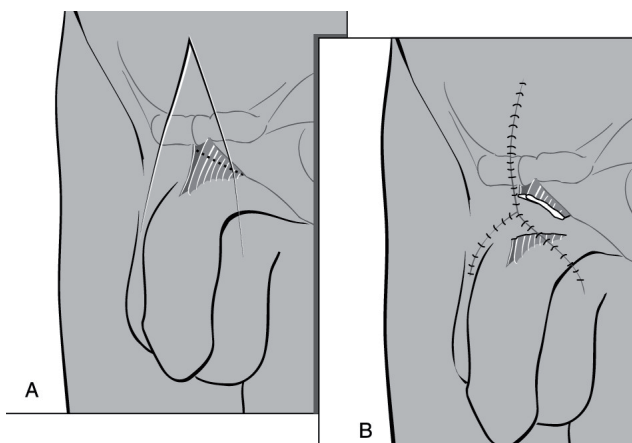


Figure 4. V-Y plasty

In most cases, however, an even older medical treatment may be successful: a good conversation between patient and physician about worries and facts. After listening and respecting the worries of the patient, this order is of eminent importance, the following facts can be discussed:

- The smaller the penis, the larger it becomes, proportionally, when erect
- Only friction in the outer part of the vagina is important during coitus
- The vagina can adapt itself to accommodate any size of penis
- Thickness is more important than length
- It is not a question of what he has, but how he uses it.

By keeping these general considerations in mind, the majority of men will be able to overcome most worries they may have about the length of their penis. And, as so often even nowadays, these recommendations are not new. In the seventeenth century, Reinier de Graaf (1641-1673) already wrote about the function of the vagina in the following way.²⁸

*Its function is simply to constrict,
when necessary, the part it embraces,
particularly at the time of coitus.
The woman's vagina in fact is so cleverly constructed
that it will accommodate itself
to each and every penis;*

*it will go out to meet a short one,
retire before a long one,
dilate for a fat one,
and constrict for a thin one.
Nature has taken account of every variety of penis
and so there is no need
to seek a scabbard the same size as your knife.*

In this thesis our experience in the management of men complaining of a small penis is described, including the associated emotional stress and physiological complaint that this “illness perception” can bring. In so doing, we will emphasize patient selection, outcome evaluation, and surgical techniques applied for penile enlargement.

THE ACUTE ERECTION ANGLE*

When the angle between the penis and the abdominal wall is too small, penetration may become difficult, if not impossible. In cases such as this the diagnosis of “acute erection angle” is applied. This problem is mostly caused by too much tension on the penile suspensory ligaments (PSL). It is this tension that determines the angle of elevation of the erect penis. Although many erect penises point upwards, most commonly at an angle of between 30 en 95 degrees down from vertical, it is also common and normal for the erect penis to point horizontally forwards or even vertically downwards. This variation in penile erection angle, along with the role of the PSL, has already been described by Kinsey and colleagues.¹⁹

In addition to excessive tension on the PSL, another cause of an acute erection angle can be a severe dorsal curvature at the peno-pubic level.²⁹

An acute erection angle can be treated by (partial) release of the suspensory ligament proper. A small suprapubic incision is made until the space between the penile base and the ligament area is reached. Because an acute erection angle is caused by shortness of the suspensory ligament proper, its partial release will decrease the erectile angle down from vertical. If partial release is not sufficient, checked by artificial saline erection, a complete transection may be done. An acute erection angle can also be treated by ventral corporoplasty according to Nesbit.^{29,30} During this operation ellipses are excised

* During our search of the literature, we found only very limited information about erection angles and the problem with regard to this angle. Because the term hyper-erection, as introduced by Daniel Yachia,²⁹ suggests a cause-effect relationship with the intensity of sexual arousal, we decided it would be more appropriate to use the term “acute erection angle”.

from Buck's fascia and the tunica albuginea at a point exactly opposite the site of greatest concavity. After this, the defects are closed again with interrupted sutures to straighten the deformity.³¹

In chapter 4, the clinical history and subsequent treatment of two men, who have had sexual problems due to their acute erection angle, are described in more detail.

PEYRONIE'S DISEASE

De la Peyronie (1693-1747) described an ailment in which the penis was curved or angulated in erection owing to the development of a dense fibrous plaque in the wall of one of the cavernous bodies.³² On the affected site, there is no elasticity, and the penis becomes curved dorsally and/or laterally (figure 5). The main symptoms are pain, penile shortening, along with difficult and painful vaginal intromission. Although the personal physician of the French King Louis XV gave his name to this penile anomaly, he was not the first to describe this phenomenon. Other investigators throughout history have reported on this abnormality, going as far back as Theodoric of Bologna in 1265.³³



Figure 5. Peyronie's disease; a severe curvature to the left (home-made picture taken from above)

Peyronie's disease is mainly encountered in men aged between fifty and sixty. The exact prevalence is unknown, due to the fact that many patients do not seek help.

A curved penis can also be the result of congenital asymmetry of the cavernous bodies, in which case the penis will nearly always be curved ventrally and/or laterally. The prevalence of this congenital asymmetry is estimated to be about six to eight per thousand male births. The pathogenesis of congenital penile curvature is unknown.³⁴

Until now the only treatment to correct penile curvature was surgery. If the curvature of the penis is stable one year after Peyronie's disease has presented, and there are difficulties with intercourse, and there is no pain when the penis is flaccid, there is an indication for surgical correction. There are two techniques available: (a) the fibrous

tissue is removed or incised and the defect is covered with a graft; and (b) nothing is done to the fibrous tissue, but the operation is performed on the healthy side of the penis where, with excisions of oval parts out of the *tunica albuginea*, the penis is pulled straight. A major disadvantage is that the penis becomes shorter, but a considerable advantage is that there is very little risk of rigidity decrease after the operation. This does not apply to the former technique, in which the fibrous tissue is removed or incised.

Another fibromatosis is Dupuytren's disease. This involves the palmar fascias in the hand, resulting in progressive contracture of one or more fingers.³⁵ Dupuytren's disease is thought to be the most common hereditary connective tissue disorder in Caucasians. In 1928 an association between Dupuytren's disease and Peyronie's disease was reported by Abernathy.³⁶ According to several studies, there is a three to fifteen percent chance that a man with Dupuytren's disease will have Peyronie's disease.^{37,38,39}

Chapter 6 describes the coexistence of Dupuytren's disease in a consecutive series of patients with Peyronie's disease, along with their clinical characteristics, as presenting at our outpatient urological clinic.

GENITAL LYMPH EDEMA

Another anomaly discussed in this thesis is genital lymph edema (figure 6). Acquired (secondary) causes of lymph edema may be classified as: 1) neoplastic, 2) infectious, 3) granulomatous, 4) reactive, 5) disorders of fluid balance, and 6) idiopathic.⁴⁰ If the cause is due to neoplasm, the inguinal and/or pelvic nodes are involved with metastatic cancer, most commonly with its origin in the prostate, penis, bladder, or lymphoma. Infections may be parasitic or bacterial. Worldwide, filariasis is the most common cause of genital edema, caused by the worms *Wuchereria bancrofti*, *Brugia malayi*, and *Brugia timori*. These worms occupy the lymphatic system, including the lymph nodes, and in chronic cases these worms lead to the disease elephantiasis.⁴¹ An estimated 120 million people in tropical and subtropical areas of the world are infected with lymphatic filariasis; of these, almost 25 million men have genital edema.⁴² Other causes of infectious genital lymph edema include the following microorganisms and diseases: streptococcus, lymphogranuloma venerium, tuberculosis, syphilis, chancroid, and leprosy. Genital edema rarely occurs due to hidradenitis suppurativa. Granulomatous diseases that may be associated with genital lymph edema include sarcoidosis, rheumatoid arthritis, and Crohn's disease.⁴³ Reactive causes include trauma, venous thrombosis, radiation therapy, and angioneurotic edema. Angioneurotic scrotal edema occurs most commonly in 4- to 6-year-old boys. It is sudden in onset, may be unilateral or bilateral, is non-tender, and extends onto the perineum and anterior abdominal wall. It does not require treatment and resolves in two to four days.⁴⁴ Disorders of fluid balance including cardiac and renal failure, fluid overload, and hypoproteinemia may also cause genital edema. If the cause is not clear, the condition is referred to as primary (or congenital) chronic lymph edema.



Figure 6. Filariasis with edema of the scrotum and leg involvement

In such a case the lymph vessels have intrinsic abnormalities; aplasia, hypoplasia, and hyperplasia have been mentioned in the literature.⁴⁵

The treatment for genital lymph edema is mostly conservative, particularly if the primary disease is self-limited and there is no permanent damage to the skin, lymphatic vessels, and subcutaneous tissue. Different pharmaceutical options in the treatment for lymphatic filariasis are described, but treatment with diethylcarbamazine (DEC), ivermectin, doxycycline, and/or albendazole is the most commonly used.⁴⁶⁻⁴⁹ Chronic edema of the external genitalia generally results in permanent pathological changes. The most effective therapy is surgical, and, when applied in properly selected patients, functional and cosmetic results are excellent. These patients may be rehabilitated so as to enjoy a normal sex life.

In this thesis the clinical history and surgical treatment of six patients with genital lymph edema are discussed.

RESEARCH QUESTIONS AND OUTLINE OF THIS THESIS

In contrast to only a few decades ago, nowadays the penile abnormalities sketched above are considered important as a medical topic. This is not only because they occur more frequently but also because of the combination and interaction of medical technological progression and cultural changes. As always, progress in medical treatment options goes hand in hand with the gradual elimination of taboos on the subject and diminished tolerance of “negative exceptions” or anomalies. With increasing awareness among the general public and reasonable to good results obtained with “repair” in selected cases, some surgical treatments of penile abnormalities have become established procedures in relatively little time. This high level of acceleration may also have some disadvantages; particularly in healthcare, hypes need to be prevented. For urologists, of course, it is important to remain in touch with innovative techniques both in the domain of human sexuality as well as in its problems. Given the primary medical ethical amendment, however, of doing no harm (*primum non nocere*), it is also important to take note of the limitations of these medical technical developments and of his/her profession. There will always be patients with penile abnormalities who are “surgically incurable,” or who are better off visiting a sexologist or psychotherapist. In all cases iatrogenic damage, big or small, emotional or financial, must be prevented. It was the awareness of this delicate balance that brought us to our first overarching research question:

1. *Is surgery a good, preferably the best, treatment option in case of penile or scrotal deformity?*

This main question was then differentiated into three sub-questions that refer to three morphological qualities and accompanying diagnoses:

- 1.1 *size*: the small penis
- 1.2 *angle*: acute erection angle
- 1.3 *expansion*: genital lymph edema.

These research questions will be answered, respectively, in:

- Chapter 2 18-Year experience in the management of men with a complaint of a small penis.
- Chapter 3 Penile enlargement: from medication to surgery.
- Chapter 4 Surgery for an “acute erection angle,” when counseling fails.
- Chapter 7 Chronic genital lymph edema: perineal skin flap plus graft reconstruction after excision.

The second main research question revealed itself along the way. While doing literature research, and based on clinical impressions, an association between Peyronie's and Dupuytren's disease became perceptible. The second research question is:

2. *Is there a common pathway to Peyronie's and Dupuytren's disease?*

This question is answered in:

Chapter 6 The association between Peyronie's and Dupuytren's disease.

Given the interrelatedness of discussing genital anomalies along with cultural perspectives, a separate chapter was written on myths and facts about the penis. Because it has not been submitted to a scientific journal, it was decided to add this chapter (5) as a "short intermezzo".

In science, it is not only important to describe the thing one does but also what one does not do. While in some patients erectile ability does not change as a result of deformity, the abnormal shape of the penis may cause severe functional sexual difficulties for the patient or couple. In some cases the shape of the penis may cause severe discomfort or even pain for the partner.¹⁸ The sexual dysfunction can become a psychosexual dysfunction, which may then lead to erectile dysfunction. In order to prevent confusion on a conceptual level, in this thesis we consciously did not discuss the erectile dysfunction that is sometimes associated with the described anomalies.

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**18-Year experience in the management
of men with a complaint
of a small penis**

Helena M. Nugteren¹, Mike T. Balkema²,
Astrid L. Pascal², Willibrord C. M. Weijmar Schultz²,
Rien J. M. Nijman¹, Mels F. van Driel¹

1. University of Groningen,
University Medical Center Groningen,
Department of Urology, Groningen, the Netherlands

2. University of Groningen,
University Medical Center Groningen,
Department of Obstetrics and Gynecology, Groningen,
the Netherlands

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INTRODUCTION

The erect penis has been in many cultures and throughout the millennia the ultimate symbol of masculine qualities, such as strength, endurance, bravery, potency, and power. Because of this symbolism, modern men are likely to feel insecure and even embarrassed if their penis is below average size.

Recent large-scale American research, amongst 25,594 men, indicated that 55% of interviewed males were content with their penis size versus 45% who stated that they would prefer a larger penis. A mere 0.2% stated they would prefer a smaller penis.¹ These figures suggest a profound lack of knowledge in the population about physical normality or average size of the male genitals. This result is likely to have been caused by exposure to pornographic material. Inaccurate perceptions about penis size may also be reinforced by the fact that men see the penis of other men from a sideways angle, whereas they only perceive their own penis from a top-down perspective, making it seem shorter than it really is; artists call this the *perception-shortening angle*. An additional problem is that men on the whole do not feel comfortable to raise the issue of penis size in conversation with others, resulting in little or no feedback on the perceived problem, and hence no opportunity to correct erroneous thought patterns.²

The aforementioned perception has been named *small penis syndrome*: a fear-fuelled subjective perception that a penis is considerably smaller than average. The emotional stress and the resulting avoiding behavior often lead to a larger problem than the irregularly sized penis actually warrants. A fear exists that other people can observe the small size of the penis, regardless of whether the man is dressed. According to Wylie and Eardly, this fear can become an obsession or part of a body dysmorphic disorder (BDD).³ BDD is a psychiatric disorder, described in the *Diagnostic and Statistical Manual of Mental Disorders IV* as a preoccupation with an imagined defect of appearance or an excessive concern about a slight physical anomaly. In order to diagnose BDD, it must be proven that the preoccupation causes clinically significant distress or impairment in social, occupational, or other important areas of functioning. Additionally, the preoccupation is not better accounted for by another mental disorder, such as dissatisfaction with body shape, as in anorexia nervosa.⁴

BDD has an inordinate degree of anguish. People with BDD may develop major depressive episodes and are at risk of suicide. There is embarrassment and fear of being scrutinized or mocked, which often causes these individuals to avoid social situations and intimate relationships.

The aim of this study was to report our experiences in the management of men complaining of a small penis.

PATIENTS AND METHODS

We compiled a list of men who presented themselves to the Urology Department at the University Medical Centre Groningen between 1989 and 2007, with the main complaint of having a 'too small penis size'. This list did not include patients who have been operated on because of hypospadias or epispadias. We collected the following data: age at time of first consultation, ethnic background, sexual preference, relational status, time when complaint was first perceived, and whether the complaint originated from a sense of embarrassment, insecurity or a different problem. We collected additional data around medicine use, psychiatric history, urological and trauma history, the measured size of the penis in maximum length, genetic and hormone profile, resulting treatment, complications from enhancing operation(s), and last, the satisfaction with the end result. After analysis, findings were entered into SPSS, using unweighted counts.

RESULTS

The sample comprised 60 male participants with an average age of 37 years (range=14–67 years). The majority of men (n=44) were native, 13 men originated from the Middle East, and 3 men were born in Surinam or the Dutch Antilles. Nearly half of the sample (n=25) had a long-term relationship, 6 men had different relationships over a set period of time, 10 were single, and 15 men stated that they had previously been in a long-term relationship, which had now ended. Of these 15 men, only 2 had found a new partner since the break-up. The relational status of 4 men was an unknown factor. Of the sample, 1 person stated his sexual preference as homosexual, 1 person stated his sexual preference as bisexual, and the remaining (n= 58) stated they were heterosexual.

The majority of the sample (n=40) indicated that they perceived shame and embarrassment about penis size as the biggest problem, not only in relation to women, but also in relation to men in a men-only social setting, such as communal shower after playing sports. Of the men, 8 named insecurity or fear of failure was named as the biggest problem. In addition, 9 men stated they experienced a coitus-related problem as a result of the small penis not being extended enough to insert into the vagina or not being able to maintain insertion into the vagina during thrusting movements. Three men reported problematic urinating, such as not being able to urinate from a standing position or urinating over their scrotum, as the main issue for presenting themselves to the hospital. Also, other men reported erection dysfunction (n=7) and premature ejaculation (n=3).

In response to the question of when in their lives they were first aware and worried about the size of their penis, 24 men answered that they experienced worrying thoughts on this matter from an early age onwards: 10 men stated that their peers subjected them to bullying behavior as a direct result of their small penis size. 6 men said they were first aware of the problem after their first genital sexual contact, and 7 men stated they had become worried after receiving derogative comments on the size of their penis.

It was not possible to ascertain from the notes in the patient-dossier the exact moment in time when the remaining 13 men first worried about the size of their penis.

With regard to psychopathology, a total of 14 men (23.4%) had received psychiatric care and/or were receiving psychiatric treatment during the time of their first urology consultation. Among these, 8 men suffered from depression, and a further 3 suffered from serious low self-esteem. Use of anti-depressants, antipsychotic drugs, or both was surprisingly low - only 4 of these men used one or both medications.

In our sample, the average size of the penis in maximum extension was 9.4 cm, with a total range of 5-15 cm. The patients' dossier did not specify the exact penis length, in centimeters, of 13 men. However, according to the physical examination of the urologist, each penis equaled a length that was 'within the range of normality'.

We determined the total serum testosterone for 20 of the 60 men, mostly because of obesity, erection dysfunction or both. We found 5 of the men to have testosterone levels <12 nmol/l and 3 of them received testosterone on prescription.

Initially, the urologist attended all patients. One third of the men (n=20) needed a mere one or two consultations. After receiving appropriate instructions, 5 men were given a stretch apparatus (the JES-Extender), which ultimately did not benefit any of them. One 16-year-old man was referred to a pediatrician for further hormonal research, and 15 men were referred on for additional counseling by a psychologist-sexologist.

Of the total sample of 60 men, 16 underwent surgery (table 1). Regarding the diagnosis, they formed a heterogenic group. Patients were primarily assigned for surgery if the penis was clearly short because of a distinct cause (e.g. an in fat sunken penis or a trauma), with a real perspective of a positive outcome. Thirteen received a VY-plasty, whilst two of them were given a double Z-plasty. In almost all cases, the surgery occurred alongside releasing the fundiform and suspensory ligaments, as well as the removal of suprapubic fat tissue.

The short penis of two of the patients was the direct result of a serious trauma: 1 man had a partial penis amputation because of cancer, and 1 man had suffered an inadequately treated pelvic fracture at 5 years of age, resulting in a sunken penis. The penises of 6 (very) obese men were buried in layers of fat tissue. In addition, 6 men had penoscrotal webbing, 4 of which were cleaved at the time of the VY-plasty.

Complications occurred in six cases; 3 men suffered a wound infection, 2 suffered from a hematoma, and 1 man came down with a wound infection in combination with scar-tissue hypertrophy. Of these 16 men who were operated on, 9 were experiencing functional complaints such as urinating problems, coitus problems, or both. Postoperatively, 7 of the 9 men were pleased with the final result, 1 man was dissatisfied, and 1 man's opinion was not noted down in the patients' dossier. Of the men without functional complaints, 2 were satisfied with the end result, 3 were dissatisfied and 1 reported being neither satisfied nor dissatisfied. Furthermore, it was unclear whether 1 man from this without-functional-complaints group was ultimately satisfied with the post-operative result (table 1).

Table 1. Operation results

Age, years	Initial length at extension, cm	Compliant(s)	Diagnosis	Technique	Length gain, cm	Complications	Satisfaction
18	7	shame	micropenis	VY-plasty	-	-	+/-
22	6	shame	in fat tissue sunken penis penoscrotal webbing frenulum breve	VY-plasty frenuloplasty	8	-	+
26	-	coitus-related problem	sunken penis due to a pelvic fracture at the age of 5 years old penoscrotal webbing	VY-plasty division penoscrotal webbing	-	-	+
31	8	shame	-	VY-plasty	2	wound infection	-
32	6	problematic urinating shame	in fat tissue sunken penis penoscrotal webbing	VY-plasty division penoscrotal webbing	2	hematoma	-
35	6	coitus-related problem	micropenis frenulum breve	VY-plasty frenuloplasty	2	-	++
38	7	coitus-related problem shame	micropenis due to Klinefelter disease	VY-plasty	-	hematoma	+
45	9	shame	in fat tissue sunken penis penoscrotal webbing	VY-plasty division penoscrotal webbing	-	wound infection scartissue hypertrophy	-
45	-	shame	UCHH* penoscrotal webbing	VY-plasty division penoscrotal webbing	-	-	+
47	10	coitus-related problem shame	frenulum breve	VY-plasty frenuloplasty	2-2.5	wound infection	+
51	7	coitus-related problem	micropenis due to UCHH* in fat tissue sunken penis	VY-plasty	-	wound infection	+
54	5	problematic urinating coitus-related problem	partial penis amputation due to cancer	VY-plasty	-	-	+
55	8	shame	in fat tissue sunken penis	VY-plasty	0.5	-	-
23	-	problematic urinating shame	in fat tissue sunken penis meatal stenosis	double Z-plasty meatus plasty	-	-	+
38	9	coitus-related problem shame	-	double Z-plasty	1	-	?
36	8.5	shame	penoscrotal webbing	division penoscrotal webbing	-	-	?

*UCHH, uncompensated hypergonadotropic hypogonadism

DISCUSSION

The group of men was highly heterogeneous in terms of their ages (range = 14 - 67 years), penis length, medical background, and received treatment. In 2007, 3% of the Dutch population originated from the Middle East, versus 2.8% originating from the Dutch Antilles or Suriname – both first and second generation inhabitants.⁵ However, the research sample (n=60) showed that 21.6% (n=13) were Middle Eastern in origin and 5% (n=3) originated from the Dutch Antilles or Suriname. Hence, an overrepresentation of Middle Eastern men had presented themselves to the University Medical Centre Groningen's Urology Department. One explanation for this result may be a significant cultural difference in applied meaning to penis-size, such as its relation to performance, masculinity, and status.

We considered it surprising that the studied population only included 2 homosexual or bisexual men because these men tend to spend more time, money and energy on their appearance than do heterosexual men, and homosexual/bisexual men are hence more likely than heterosexual men to be critical about their physical appearance.^{6,7} On this basis, it is fair to assume that homosexual or bisexual are more likely to be dissatisfied about their penis size, resulting in a more pronounced wish for intervention. According to Woertman et al.'s research, only 24.9% of homosexual men desire a larger penis.⁷ Firmness of the stomach, chest and buttocks as well as condition of the skin in general, are considered more important than penis length. This 24.9% is a small group compared to research findings on heterosexual men because 45% of this group would prefer more penis length.¹ In addition, homosexual men, just like heterosexual men, do consider their penis to be their most attractive body part.⁷

A male subjective idea that one's penis is too small or slightly smaller than average can have a most negative effect on self-confidence and the ability to lead a successful life. Because of the patient's feelings of shame, embarrassment, and insecurity about their genitals, their efforts in attempting to find a new partner are undermined, culminating in fears of an inability to sexually satisfy the potential new partner as a direct result of being under endowed. This article's sample included 13 men who had experienced a long-term relationship but who had not yet found a new partner. Furthermore, nearly all men who had stated their main reason for presenting themselves to the hospital as being insecure and/or suffering from fear of failure were single.

How do women perceive the size of their partner's penis? When asked, 84% of American women (n=26.437) stated they were most content with their partner's penis length, 14% wished for the length to be longer, and 2% wished for the length to be reduced.¹ Dutch research indicated that 71% of women are (very) satisfied with the length of their partner's penis.⁸ This warrants the question on whether size actually matters. Earlier research in Groningen showed that a mere 21% of women considered the length of the penis (very)

important, adding that the circumference of the penis was considered more important than length.⁹ Women never find the penis the most attractive body part of a man.

It was not surprising that 20% of the sampled men were, or had been, undergoing psychiatric treatment. Depressed men, as well as men with a low self-esteem and suffering from a negative self-image, are more likely to internalize the thought that their penis is too small. Psychosocial support from a psychologist-sexologist can help these men to change their negative spiral of self-perception in order to develop more confidence. Treatment is based on cognitive and behavioral therapies and techniques. Crippling thoughts, which produce undesirable behaviors and feelings, are replaced by enhancing self-affirming thoughts, backed up by adequate coping strategies. This enables patients to become more skilled in problem solving, as well as becoming emotionally less vulnerable. The emphasis lies on educating the patients on sexual knowledge, reformulating the perceived complaints, reducing restraining thoughts and behaviors, increasing positive thoughts and learning to focus on (and therefore to recognize) other physical sensations. Several researchers have shown the positive effect of a sex education-integrated treatment program. In the series of Shamloul 79 (86%) out of 92 patients found the combination of sex education with standard penile measurements helpful and relieving.¹⁰ Using a structured management and counseling protocol, Ghanem et al. found that only 9 (3.6%) out of 250 patients chose to seek further surgical intervention.¹¹

In Veale's study, cognitive behavioral therapy, in combination with a selective serotonin re-uptake inhibitor, appeared to work most effectively on BDD patients.¹² Patients who have suffered with serious psychopathology, a personality disorder and/or a serious psycho trauma in their medical history are encouraged to seek more extensive psychiatric care.

The literature cannot singularly point to an average penis length, which is likely to have been caused by the many ways in which the male genital has been subjected to different measuring methods. However, large-scale research in Caucasian males has yielded an average penis length of 12.5 cm in its extended condition. According to experts, a penis constitutes being 'too small' if the genital diverts more than 2.5 times the standard deviation of the average penis length.^{1,13,14} This translates as a length of less than 7.5 cm in extended condition. Length at maximum extended condition always equals length at erection. The aforementioned researched group of men averaged 9.5 cm in extended position, with a range of 5-15 cm. It is understandable that the average of this group was below the mean average of men in general at 12.5 cm because this group had presented themselves to the hospital with serious worries about their penis size. Of these researched men, 79% had an extended penis of more than 7.5 cm in length. This figure is likely to be higher still because 12 of the men did not have their penis specifically measured at the time of the consultation, but the examining urologist deemed their size as being 'normal'.

Previous research amongst Dutch men with a (perceived) small penis indicated that 67% may have suffered from BDD.¹⁵ Because our urological dossiers did not offer enough insight into possible significant suffering and limitations in social or occupational functioning, the authors feel they cannot make a statement on BDD in this context.

Of the 16 operated men, 6 had a penis length of 7.5cm or more in extension (table 1). In hindsight, it appeared that only 2 of the 6 men were pleased with the operation results. This meager improvement is the main reason that men with a penis length of 7.5 cm or more in extension are no longer treated by scalpel.

CONCLUSIONS

Men complaining of having a 'too small penis' form a heterogeneous group. They place their doctors before an ethical dilemma. It is hence of the utmost importance to explore the 'informed consent' at a multidisciplinary level. The urologist is to explore and to discuss all available options together with the patient, culminating in clarity on whether an operative intervention can or cannot bring relief to the experienced problem. Men with small penis syndrome do without fail benefit from several counseling sessions with a psychologist-sexologist. Depression and personality disorders are legitimate reasons for not proceeding with surgical treatment.

There are many different techniques used for (optically) lengthening the penis. However, research shows that those men without an anatomical deviancies, who already achieve a penis-length of no less than 7.5 cm in extension, have only very limited benefit from penis enhancing surgery. This particular patient-category should therefore be dissuaded from surgery.

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Penile enlargement: from medication to surgery

Helena M. Nugteren¹, Mike T. Balkema²,
Astrid L. Pascal², Willibrord C. M. Weijmar Schultz²,
Rien J. M. Nijman¹, Mels F. van Driel¹

1. University of Groningen,
University Medical Center Groningen,
Department of Urology, Groningen, the Netherlands

2. University of Groningen,
University Medical Center Groningen,
Department of Obstetrics and Gynecology, Groningen,
the Netherlands

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INTRODUCTION

The penis is central to cultural concepts of masculinity; its length, shape, appearance, and performance are held to be indicators of masculinity.¹ Equating penis size to masculinity makes under endowed men worry about the size of their penis. They are vulnerable in their search for a cure, which will often lead them to explore penis-enhancing possibilities on the Internet and elsewhere. Penis-lengthening pills, stretch apparatus, vacuum pumps, silicone injections, and lengthening and thickening operations are widely available in this unregulated world of self-medication. Advertisements overtly imply that there is a direct relationship between penis size and masculinity and suggest that women's sexual satisfaction greatly depends on penis size.

In this commentary, we describe these penis-enhancing possibilities, a sequel on our case series about how to deal with the complaint of a small penis.

NON-SURGICAL PENIS-ENHANCING METHODS

The Internet has a large market for penile/lengthening pills. The theory behind many of them is that the herbal blend will increase blood flow to the erect penis and therefore enhancing its size. The manufactures promise an increase in length of 2-10 cm. Many men believe and hope that this theory is true and are willing to pay a significant amount of money on such pills. There is, however, no evidence that this medication influences penis-size.

Penis-stretch apparatus and vacuum pumps are also popular choices for men wanting to enhance their penis size, sometimes in combination with aforementioned medication. Many types of penis-stretch apparatus are available nowadays. The Danish JES-Extender by DanaMedic Aps and the Spanish Andro-penis by Andromedical are best known. However, there is only poorly documented evidence to support the use of penile extenders or vacuum pumps.² It is, therefore, considered good advice to patients to neither purchase nor use such a device. However, should the patient undergo penis-lengthening surgery, the device may play a post-operative role.³ This matter still remains unclear and may warrant research.

Liquid injectable silicone (LIS) can be used for penile augmentation. To broaden the penis, the silicone is injected into the subdermal plane. However, the use of this product is highly controversial because of the possible complications. Short-term reactions include pain, ecchymosis, pigment change or, if the silicone is injected in the vascular system, embolism or pneumonitis. Direct injection in the corpora cavernosa could result in embolic events, priapism, or impotence. Long-term reactions include migration of silicone and silicone granulomas.⁴ Yacobi, Tsivian, Grinberg and Kessler (2007) reported their experience with 324 patients who received LIS to broaden the penis. They reported an increase in penile girth of 27% and only mild complications as bruising.⁵ A major drawback of this study is the short period before follow-up; most complications of LIS have a latency period of years after treatment.

SURGICAL PENIS-ENHANCING METHODS

When is it reasonable to consider surgery? Most urologists will draw the line at 7.5 cm in maximal extended condition, others will do so at 9 cm.^{6,7} It is undoubtable that psychosexual findings are most significant in reaching this decision. For example, if a man has the *idée fixe* that women are particularly convinced that 'size matters' and hence will not proceed in a relationship unless the nuptial equipment is adequate, then extensive counseling is most appropriate. The psychologist-sexologist needs to explore how the patient experiences himself, and the part his penis plays in this role. Previous sexual relationships will be minutely explored. Men with psychiatric disorder who have a pattern of expectation which is difficult to adjust in their minds are the worst possible candidates for operational intervention. There is no standard questionnaire yet that could be used to filter men who may be eligible for operation. However, an attempt to this purpose has been made by Spyropoulos et al. (2005).⁸

The urologist is responsible for detailed explanation of risks and complications of any operation and for putting this to paper. In addition, we believe that using random pictures of previous operations and their complications is of considerable importance in informing the patient.

The most widely used technique for optical lengthening is cutting the ligaments, in combination with removal of suprapubic fat tissue and by creating increased length in the penis shaft by performing a (double) Z-plasty or a VY-plasty. The fundiform ligaments are in fact a continuation of Scarpa's fascia. The suspensory ligament lies posterior to the fundiform ligaments. It is chunkier and is triangular. It splits in two around the base of the penis (figure 1).⁹ After cutting the ligaments, one can use fat tissue or a small silicone

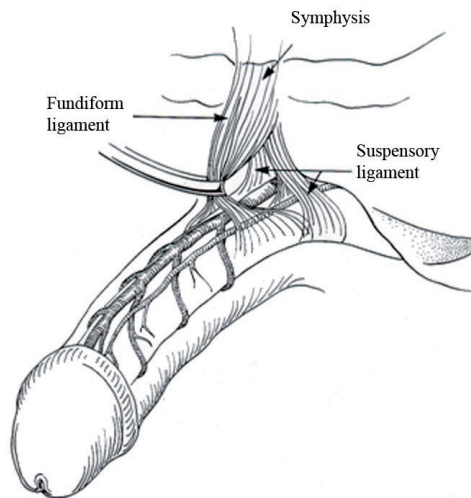


Figure 1. Ligaments of the penis

testis prosthesis to fill the cavity between the pubic bone and the cavernous bodies. In this way, the penis is kept as distal as possible, but it also ensures there can be no retracting scarring between corpora and pubic bone. Furthermore, it is recommended to attach the corpora to the lower rim of the pubic bone using two stitches. The next step is to remove fatty tissue from the pubic area. Excision of fat tissue by scalpel is preferred over liposuction, because the aforementioned method allows the septa to be included until it has become smoothed out. Last, the Z or VY-plasty is closed, leaving a vacuum-drain, if necessary.^{6,10,11}

A visual illusion of length gain of 2cm, in flaccid state, is considered an adequate result. However, men with body dysmorphic disorder (BDD) often have unrealistic expectations regarding the outcome of this surgical procedure.¹²

Complications in (double) Z-plasty and VY-plasty are scar-hypertrophy, an unstable, low hanging penis, reduced sensitivity in the shaft or in the glans, or – in the worst case – a shortening of the penis because of the forming of scar-tissue. Another complication is *scrotalization*, which occurs when scrotal skin with hair follicles changes position and becomes part of the shaft of the penis.^{10,13} This complication, as well as necrotic wounds, seem to occur more often when performing the VY-plasty, making (double) Z-plasty the preferred technique.

One of the methods to broaden the penis is by injecting autologous fat tissue in between the superficial and deep fascia. This has the result of not only making the penis a little broader, but because of the increased weight, often also a bit longer.¹⁴ Complication for this technique are the irregular shaft due to post necrotic fat lumps, reabsorption, edema of the corona, wound infection, and problems with intromission caused by excess fat deposits on the shaft.^{13,15} Another possibility is transplanting deepithelialised skin with matching subcutaneous fat. These transplants are harvested from the groin area or from the buttock region and are inserted just above Buck's fascia. The next phase is to tuck both dartos fascia and the presenting skin over the transplanted area. This operation technique avoids necrosis and re-absorption, allowing symmetry.¹⁰

In the Netherlands it is, as a result of the aforementioned problems, considered good practice not to perform thickening operations.

New techniques are constantly being developed and refined. Perovic and Djordjevic (2000) used an extraordinary operation technique on experimental basis, whereby the penis is not only optionally lengthened, but is actually lengthened.¹¹ The entire penis becomes disassembled. The technique starts with the dissection of the corpus spongiosum (the spongy body around the urethra) on the ventral side as well as the neurovascular bundle at the dorsal side. Following this, the glans and the cavernous bodies are separated, leaving a small space between the glans and the ends of the corpora cavernosa. A small piece of rib cartilage is inserted inside this cavity. However, the achieved result largely depends on the flexibility of the neurovascular bundle. After this extension, the urethra, the spongy body and the nerve-bundle are reattached. A risk of

absorption remains, which is why Perovic et al. experimented with silicon extensions.¹⁶ The disadvantage of using the latter is the enhanced risk of infection. This operation technique can be combined with the cleaving of the fundiform and suspensory ligaments. To avoid a curvature, it is important to optimize the stretching of the neurovascular bundle. Using penile stretch-apparatus during the first post-operative month can avoid this complication. According to Perovic et al., the aforementioned technique offered 13 men (out of 19) an enhanced penis-length by 2-3 cm. The remaining men achieved an additional 3-4 cm in length. The follow-up, after 3.3 years, indicated that not a single patient had suffered from erosion, inflammation, or infection in the operated area. Furthermore, the urethra and neurovascular bundle showed no signs of damage as a result of the operation technique. Last, none of the patients reported erectile dysfunction. It is unfortunate that the article does not indicate the reported level of satisfaction from a postoperative perspective.¹¹

CONCLUSIONS

There is a large market for penis-enhancing products. Penis lengthening pills, stretch apparatus, vacuum pumps, silicone injections, and lengthening and thickening operations are widely available. Research has shown that surgery is the only proven treatment, in which the division of the penile suspensory ligament is a simple and commonly used technique.

Before any operation is performed, the urologist and the psychologist-sexologist need to find out whether the patient is a suitable candidate for surgery. The urologist is also responsible for detailed explanation of risks and complications of any operation, preferably using random pictures of previous operations and the patients' complications.

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Surgery for an ‘acute erection angle’, when counseling fails

Helena M. Nugteren¹, Astrid L. Pascal²,
Willibrord C. M. Weijmar Schultz²,
Mels F. van Driel¹

1. University of Groningen,
University Medical Center Groningen,
Department of Urology, Groningen, the Netherlands

2. University of Groningen,
University Medical Center Groningen,
Department of Obstetrics and Gynecology, Groningen,
the Netherlands

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INTRODUCTION

A hyper-erect penis is a rare anomaly of the penile erectile deficit angle. This phenomenon was first introduced by Daniel Yachia as the 'hyper-erection'.¹ However, because this term suggests a cause-effect relationship with the intensity of sexual arousal, which is most definitely not the case, the authors of this article have decided it to be more appropriate to use the term 'acute erection angle'.

An acute erection angle may be caused by severe shortness of the penile suspensory ligament (PSL), severe dorsal curvature at the penopubic level or a combination of both. This article describes the clinical history and subsequent treatment of two men who have had sexual problems due to their acute erection angle. This ground-knowledge is particularly provided for urologists and sexologists in order to offer clear information on the great variations in erection angles and, in case of coital difficulties, on the possible surgical treatment options.

Case 1

A 40-year old Caucasian male and his 36-year-old wife were referred to the urologist because of coital difficulties. They were in a 7-year relationship. Since the start of the relationship the female had experienced pain in the distal anterior vaginal wall during intercourse. In addition, she had irritating vaginal flatulency. The couple reported that coitus with female on top position was not physically possible, although neither the male nor the female had encountered severe coital difficulties in former relationships. On photographic examination an acute erection angle was diagnosed (figure 1a). There were no signs of Peyronie's disease. The patient and his partner were counseled about the possible surgical treatment options: partial release of the suspensory ligament, a ventral

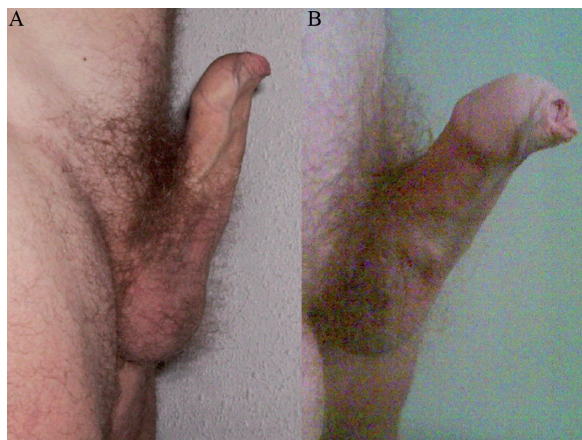


Figure 1. An acute erection angle. Homemade photograph (A) and at 18 months follow up after corporoplasty (B)

corporoplasty according to Nesbit or, if necessary, a combination of both. The potential drawbacks of both procedures were discussed extensively, especially shortening after a Nesbit procedure and instability in erection after ligament release. Together with the patient we decided to start surgery with a ventral corporoplasty according to Nesbit. During surgery including its intermittent artificial erection tests it became clear that this was sufficient. At 18 months follow up the erect penis showed an erection angle of approximately 45 degrees from vertical (figure 1b).

Case II

A 41-year old Caucasian male was referred to the urologist for coital difficulties since the beginning of his sexual career. He had experienced various relationships. At the time of the intake he was in a stable heterosexual relationship, lasting 12 months. The couple complained about not being able to have intercourse in various positions, especially that with the female on top. At home photographs showed an acute erection angle in combination with a 25 degree congenital dorsal curvature. There were no signs of Peyronie's disease. After counseling the patient chose for partial release of the PSL and not for correction of the curvature. The surgical technique in this case included identifying the PSL proper via a transverse infrapubic incision. Once identified, the ligament was released gradually until a 45-degree erection angle was reached, as shown by intermittent artificial erection tests (figure 2b). To prevent re-attaching the pubic bone and the cavernous bodies were separated by interposing some suprapubic fat and two folded GentaFleece® collagen fleeces.

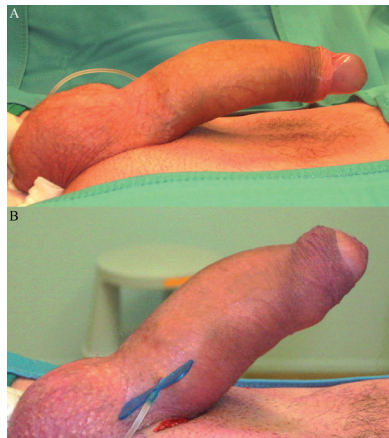


Figure 2. Induction after an acute erection angle with Saline (A) and the result after releasing the suspensory ligament (B)

Following surgery both males were advised to wear boxer-shorts type underwear, not to wear the penis upwards toward the abdomen, not to wear tight trousers, and to delay sexual intercourse for 6 weeks. Unfortunately, we were not able to realize MRI's of their erect penis, neither preoperatively nor postoperatively.

Both males had a 9-month follow up assessment of the functional status of the penis; including residual deformities and ability to have sexual intercourse. Both were satisfied with the surgical outcome. 'Good' surgical outcome was defined as correction of the penile anomaly, no instability and normal sexual intercourse without pain, including 'female on top' position.

DISCUSSION

On entering the keyword `erection angle` in search engine Google, 1.700.000 hits are exposed. However, by contrast, textbooks of urology and sexology provide only very limited information about erection angle dysfunction and do not include any description of the problems with regards of the erection angle. Furthermore, a PubMed search on `erection angle` provides only one relevant reference.²

The penile suspensory ligaments support and maintain the erect penis in an upright position during sexual intercourse. The suspensory apparatus of the penis consists of the fundiform ligament, the suspensory ligament proper and the arcuate subpubic ligament.³ The fundiform ligament is superficial and not adherent to the tunica albuginea, whilst the suspensory ligament proper bridges between the symphysis pubis and the tunica albuginea of the corpora cavernosa; in its course it circumscribes the dorsal vein of the penis. The arcuate subpubic ligament runs a similar course to the suspensory ligament. It is a slightly denser structure and lies further posterior. Functionally, the ligaments maintain the base of the penis in front of the pubis and acts as a major point of support for the erect penis during sexual intercourse.⁴

Abnormalities of the suspensory apparatus can be congenital, often presenting with a ventral curvature of the penis, or acquired secondary to penile trauma.⁴ In males with a long heavy penis or a loose suspensory ligament, the angle usually will not be sharp, even with full rigidity.⁵ When the ligament is too short, it will decrease the angle between the penis and the abdominal wall, making penetration difficult, if not impossible. The more skywards the penis points, the less flexible the erection is and the more careful one has to be when using it, especially with the female on top position. It's possible to fracture the penis, so a male with an acute erection angle should not attempt to get too acrobatic during intercourse. In addition, deep upward thrusts can put uncomfortable pressure on the anterior vaginal wall, causing pain and/or the urge to urinate.

Undoubtedly, the entity of 'acute erection angle' is uncommon. The diagnosis can be made by at home photography. However, the degree of curvature measured using at home photography will be underestimated as compared with intracavernous injection.⁶ Office photographs after intracavernous injection are more accurately to determine the degree of curvature.⁷ For several reasons it is advisable to evaluate sexual functioning by a validated questionnaire as the IIEF.

According to Yachia the initial surgical approach of a an acute erection angle is through an infrapubic incision.^{1,8} A 4-5 cm long horizontal incision is made along the lower edge of the symphysis and carried down through the suprapubic fat until the space between the penile base and the ligament area is reached. Since one of the reasons is shortness of the suspensory ligament proper, its partial release will decrease the erectile angle down from vertical. If partial release is not sufficient, checked by artificial saline erection, a complete transection may be done. This however may result in instability of the erect penis, but according to Yachia this can be corrected by corporopexy. By this approach an artificial erection is created after the ligament is cut. With the penis in an erect state the corporopexy sutures are applied and, keeping the penis in 60 to 90 degree erection angle, the 2/0 braided polyester sutures are tied.¹

An acute erection angle can also be treated by ventral corporoplasty according to Nesbit. In our first case corporoplasty was sufficient. This of course needs another approach than the infrapubic one; a degloving procedure was performed.

If one prefers to combine ventral corporoplasty with PSL release, the single-incision perineal approach described by Lupu and Gillespie may be a good alternative.⁹ With this approach the ventral corporoplasty is performed, the resulting erection angle checked, and then the ligament release is done accordingly. It should be stressed that this approach necessitates sufficient experience.

The first Kinsey report was concerned principally with sexual behavior but also reported on the topic of erection angle.¹⁰ The authors wrote: *'In any age group there is considerable variation in the angle at which the erect penis is carried on the standing male. The average position, calculated from all ages, is very slightly above the horizontal, but there are approximately 15 to 20 percent of the cases where the angle is about 45° above the horizontal, and 8 to 10 percent of the males who carry the erect penis nearly vertically, more or tightly against the belly.'* A profound strength of the Kinsey report was its large database, but a weakness (for the study of erections) was the fact that the data were self-reported. Sparling's reanalysis of the Kinsey data plus documented photo data from a sample of 81 males between the age of 21 and 67 years showed more erection angles in the lower ranges (table 1).²

Table 1. Erection angles comparing self-reported and photo documented data

Angle in 0 down from vertical	Kinsey sample, n=1357	Sparling sample, n=81
0-30°	10.7 %	4.9 %
30-60°	27.6 %	29.6 %
60-85°	30.3 %	30.9 %
85-95°	24.1 %	9.9 %
95-120°	7.1 %	19.8 %
120-180°	0.2 %	4.9 %

CONCLUSIONS

In modern society, with its huge access to internet information, couples may complain of very specific sexual concerns. In a social climate that emphasizes the importance of sexual 'performance', one is less reticent in presenting a problem such as coital difficulties due to an acute erection angle. Counseling a couple with complaints of sexual inadequacy which has resulted specifically from this abnormality should be based on objective reassuring information about anatomical and physiological facts. In addition, psychological reasons for painful intercourse should be explored. Furthermore, health care professionals are to offer sex-educational information on more enhanced functional penile positions for sexual intercourse in different positions. However, in our experience taking a passive attitude, such as 'leaving it to nature' or leaving the couple to grapple/experiment with a set of instructions, does not always work out well. Hence, when counseling has failed to provide relief, simple and safe surgical techniques can yield adequate results.

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Short intermezzo
Myths and facts about the penis

5

In many ancient cultures, the phallus was the symbol of immortality, of eternal rejuvenated life. The Etruscans placed a phallus on every grave.¹ As an externally visible biological characteristic, the phallus has received a heavy religious and moral load in the course of history. So no matter what, studying the phallus always leads us towards the study of religion. Phallic culture was a striking feature in the religion of the ancient Greeks. Herms stood “everywhere” in ancient Greece, not only in front of houses, temples, and city gates but also in front of an acropolis, at markets, and in gymnasiums. These squared pillars, with only a bearded male head on top and, halfway up, an erect phallus, were crowned with green branches and anointed with olive oil. During worship, people would place their hand on the herm’s head, and take hold of his beard and then his phallus. Herms were believed to provide protection from thieves and robbers; their main function was to ward off the “evil eye”.^{2,3}

At the Piazza della Signora in Florence a statue of Neptune is situated in the middle of an ornamental pond, with bronze fauns sitting around the edge. The naked fauns all have a penis in erection. Whereas the bronze bodies of the fauns show the usual oxidation color, their phalluses are yellow-copper tinted, owing to countless touching by hands. Florentine women believe that by doing this, they can increase their chance of pregnancy. But while this touching takes place in secret in Florence, it occurred openly and without qualms in ancient Greece.

Dionysian festivals illustrated the religious meaning of the phallus. Large phalluses were carried in their processions.⁴ Dionysus was the god of intoxication, of the exaltation caused by wine, the blood of the earth. He was the god of passion, enthusiasm, the god of wild abandonment, which characterized these nightly autumn festivals. These were enormous city festivals that were important state occasions. They attracted onlookers from miles around. Not only were countless phallus images carried in the procession but the participants also tied large artificial penises onto themselves. Apart from this, it was the Greeks in particular who drew a sharp dividing line between the phallus with its symbolic meaning and the same organ as an anatomical part. The phallus was only used symbolically and in rituals.

ETYMOLOGY

In the ancient Greek and Latin languages, there was already a wide variety of names for the male genital organ.⁴ Only a few have been preserved. It is often difficult to discover why one name continued to be used, while others did not. The majority of words were metaphors in relation to length, cylindrical shape, or vertical position. Sometimes the stem of a plant, the shaft of a spear, or the blade of a sword served as a model. But also the vertical warp of fabric (Greek *stêma*) and the bronze beak of a war-galley that was intended to ram other ships during sea battles (Greek *embolon*) were used as examples. Ultimately, it became “penis.” The precise origin of the word “penis” remains unclear.

Some language experts believe that the word was derived from the Latin verb “pendere” (to hang). In that case, the vulgar Dutch noun “lul” is not such a bad translation, because the verb “lullen” originally meant “hanging down.” The status of words that originally had a neutral meaning can rise and fall on account of all sorts of factors, from obscene to scientific, and from decent and descriptive to vulgar. Another example of such Dutch word is “kloot” (ball/testicle), which was used by the Dutch poet Joost van den Vondel (1587-1679) in literary language, but is used almost exclusively today as a vulgar expression.

THE PENIS IN ANIMALS

There are many different types of “penis”: the *aedeagus* of flies, mites, and butterflies; the protuberances that some frogs have near the anus; the organ with which the ordinary honey bee copulates (which breaks off – and costs him his life – but also makes sure that no other drones can mate with the queen); the embolus of the gold spider; the anal fin of fish; the double-jointed penises of snakes; the proboscis of the dragonfly.³ These are all penises that are used to deposit male sperm cells within the female. Phalluses vary from small protuberances to the penis of a whale, which, although it is usually safely tucked away in his body, can reach a length of 1.8 meters.

The first surgical implantation of a penile prosthesis took place in 1936. To reconstruct a penis amputated during trauma, the plastic surgeon Bogoras used a piece of rib cartilage.⁵ He used cartilage because he had observed that many male mammals had a penile bone. Possessors of such a bone, called a *baculum* or *os penis*, include the whale, dolphin, walrus, otter, bear, martin, badger, squirrel, wolf, dog, and monkey. In some species of animal – the spider monkey, for example – the females also have an *os clitoris*.

In 1951, W.R. Bett wrote an extensive article about the *os penis*.⁶ In the whale, the bone is about two meters long, with a circumference of forty centimeters at the base. As species ascended the evolutionary ladder, the bone became smaller. In the walrus, it is only fifty-five centimeters and, in the monkey, one or two centimeters. So far, there is no evidence that *Homo sapiens* ever had such a bone.

The position and form of the *os penis* differ from one species to another. In the dog it forms a canal for the urethra. In the bear and wolf, it is indispensable for mating. The *os penis* appears in many different shapes: in the raccoon it is s-shaped, while in the bat it is forked. The squirrel’s penis has a sharp hook, which, according to some experts, is used to break through the hymen; others believe that it is used to remove so-called mating plugs (figure 1). A mating plug consists of sticky residual sperm, which temporarily seals the vagina of the female squirrel to prevent other males from mating with her.

The *os penis* of the otter is characterized by its extreme hardness. Healed penile fractures have been observed in these animals. When male otters fight, they aim their powerful jaws and sharp teeth at the penis of their challenger. Sometimes they manage to break the other otter’s penis!

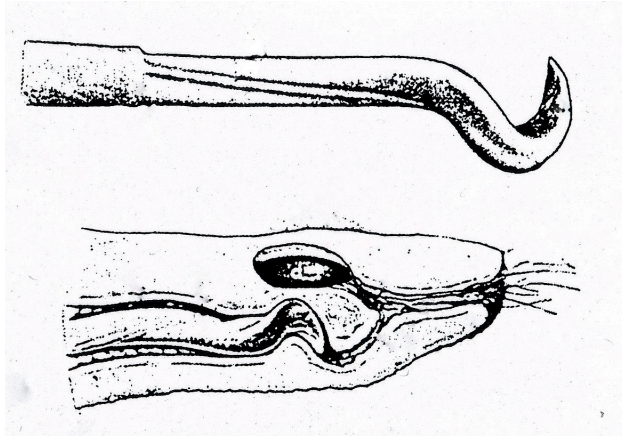


Figure 1. Penis of a squirrel

Many mammals have keratinized epidermal spines overlying tactile receptors in the *glans dermis*, including chimpanzees and mice. Penile spine growth is androgen-dependent: Primates lose spines upon castration and treatment with testosterone restores spine formation. McLean and colleagues reported on human-specific genomic deletion of the Androgen Receptor gene that eliminates an ancestral mammalian regulatory enhancer that drives expression in developing penile spines and sensory vibrissae.⁷ This is why humans fail to form penile spines. Ablation of the spines decreases tactile sensitivity and increases the duration of intromission. According to McLean and colleagues, the loss of penile spines in human evolution was linked to monogamous, pair-bonded social systems.

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**Chronic genital lymph edema:
perineal skin flap plus graft reconstruction
after excision**

Helena M. Nugteren¹, Peter H. Robinson²,
Rien J.M. Nijman¹, and Mels F. van Driel¹

1. University of Groningen,
University Medical Center Groningen,
Department of Urology, Groningen, the Netherlands

2. University of Groningen,
University Medical Center Groningen,
Department of Plastic Surgery, Groningen, the Netherlands

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INTRODUCTION

Lymph edema is caused by stasis of lymph in subcutaneous tissues. It may cause problems with hygiene, urinary incontinence, inability to have sexual intercourse, and even immobility.¹ The most effective treatment is surgical excision of the affected tissue, followed by wound closure. In males with genital lymph edema, the wounds can be closed using split-thickness skin grafts, or skin flaps from the perineum and the most posterior part of the scrotum. We treated six patients using a combination of these two techniques.

PATIENTS AND METHODS

Between 1996 and 2010, we treated six men with chronic lymph edema of the penis and/or scrotum. Two patients had a previous history that led to the diagnosis of secondary lymph edema. In two patients, the legs were also affected. Five patients complained that the edema had increased to such an extent that intercourse was no longer possible. To exclude any possible secondary causes, four patients underwent MRI of the genitals, lower pelvis, and abdomen. Other than the lymph edema, no abnormalities were found.

Case reports

Case 1

A 40-year-old man had been suffering from genital edema for three years (figure 1). It had started in the penile shaft. He had regular erections, but coitus was no longer possible. The man denied that he entertained the possibility of self-mutilation. Owing to severe depression, the patient had sought treatment from a psychiatrist for more than a year. Further inquiries revealed that the psychiatrist also considered self-mutilation very unlikely. After the surgery, the initial result was very good (figure 2). Within six months, hypertrophic scarring occurred, particularly on the penis. Nevertheless, the patient was very satisfied and did not wish to undergo any further surgery.

Case 2

A 23-year-old man had been suffering from lymph edema since the age of six, initially only in his legs, but, since the age of 13, also in the scrotum and penis. At the age of 18, attempts had been made to improve the situation with lymphatico-venous shunts. For psychological reasons, the young man entered a special, supervised home-accommodation project. Physical examination revealed such gross edema that the penis was totally invisible and micturition took place via a sort of groove (figure 3). During surgery, 26.7 kg of tissue was excised. Both the short-term and long-term results were satisfactory (figure 4).



Figure 1. Preoperative front view of case 1

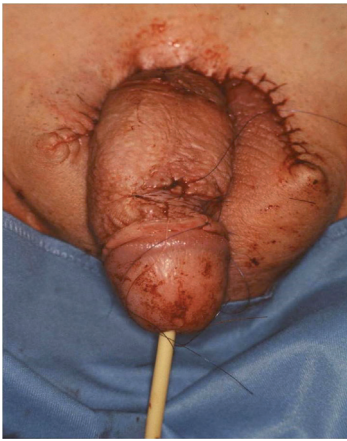


Figure 2. Case 1 direct postoperatively

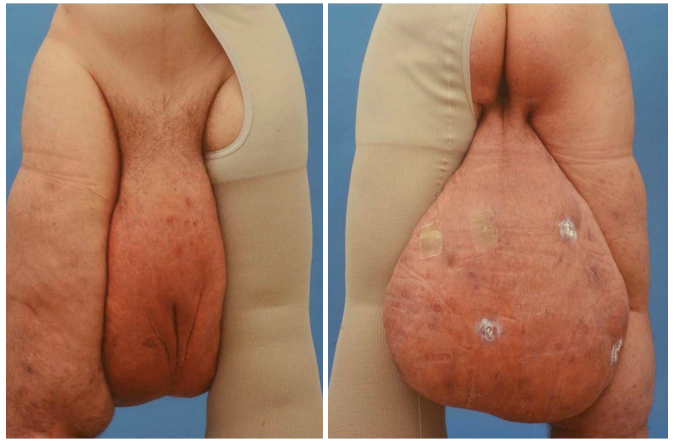


Figure 3. Frontal and dorsal view case 2: massive oedema of the legs and the genitals

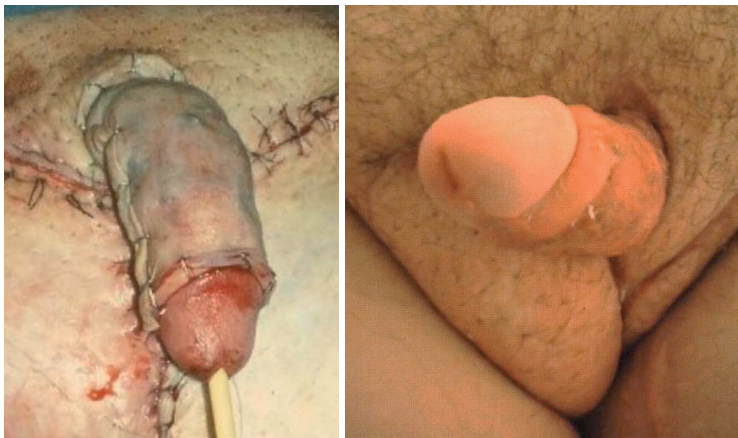


Figure 4. Case 2 direct postoperatively and at three-month follow-up

Case 3

A 50-year-old patient had been suffering from genital lymph edema for two years, for which no obvious cause had been found. According to the patient, the edema had developed in a relatively short time, that is, over a period of a few weeks. Erections still occurred, but the patient experienced them as painful. Furthermore, he was ashamed of himself in front of his wife, coitus had ground to a halt, and his urine erupted as a spray. Physical examination revealed gross genital lymph edema with signs of fungal infection in the groin (figure 5). Surgery took place after successful treatment of the fungal infection. The patient was satisfied with the postoperative result. One and a half years later, second-stage surgery was performed to correct hypertrophic scar formation.

Case 4

A 35-year-old patient had been suffering from edema since the age of 22, initially in the legs, but later extending to the penis and scrotum (figure 6). At the age of 15, he had undergone partial bladder resection and bilateral pelvic lymph node dissection because of leiomyosarcoma. Without success, a total of six attempts had been made to improve the situation with lymphatico-venous shunts. The patient said that he was not concerned about his legs, but that he was dissatisfied with the inability to have normal sexual intercourse and normal micturition. Postoperatively, he was very satisfied with the result.

Case 5

A 59-year-old patient had been suffering from genital lymph edema for more than nine years. According to the patient, the edema developed after an erysipelas infection. Three years before presentation at our clinic, he had already undergone surgery elsewhere. An excision of the pathologic skin using liposuction and a split-thickness skin graft had been carried out. Unfortunately, the edema recurred (figure 7). We operated on this patient, and during surgery 4.1 kg of tissue was excised. Both the short-term and long-term results were satisfactory (figure 8).

Case 6

A 63-year-old patient had been suffering from scrotal edema and a hidden penis for 4 years. The patient was autistic and mentally retarded. The cause of the edema was unknown. He had problems walking and urinating. Surgery was complicated by a bacterial infection under the skin graft, but with intravenous antibiotics the wound healed without any further problems. The long-term result was satisfactory (figure 9).



Figure 5. Case 3 with signs of fungal infection in the groins



Figure 6. Case 4 with scrotal scars of lymphatico-venous shunt surgery



Figure 7. Case 5 recurrence after surgery elsewhere

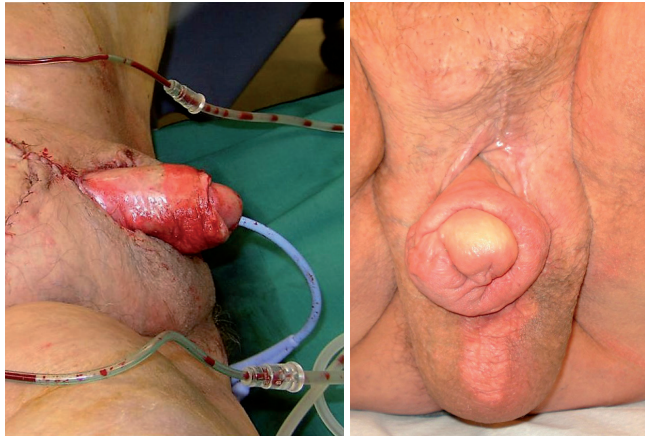


Figure 8. Case 5 direct postoperatively and at thirty-month follow-up



Figure 9. Case 6 preoperative view and at sixteen-month follow-up

SURGICAL TECHNIQUE

Intravenous administration of broad-spectrum antibiotics was started preoperatively. Surgery was performed with the patient in the lithotomy position. The patient was cross-matched in advance, in view of possible blood loss and lymphorrhagia. A Foley catheter was inserted into the bladder. Penis, scrotum, and both legs above the knee were shaved and disinfected. An incision was made around the affected penile and scrotal skin, which was then removed together with the subcutaneous tissue layer. The most posterior part of the scrotal skin and the skin of the perineum were normal in all the patients; these were therefore spared from excision and used for wound closure. The spermatic cords, testes with tunica vaginalis, and Buck's penile fascia remained *in situ*; dissection was performed distally up to the level of the coronary sulcus under the glans. In cases where a split-thickness skin graft was necessary, mineral oil was applied to the thigh. Split-thickness skin grafts, 0.8 mm thick, were taken using a compressed air dermatome. The donor site was covered with alginate. The spermatic cords were sutured together to prevent a bifid scrotum before applying the grafts. The skin graft for the scrotum was meshed (1:1.5) and sutured in such a way that apposition was achieved on the spermatic cords and testes. In contrast, the skin graft for the penis was not meshed. Instead, it was fixed with loose sutures (or surgical staples) to the coronary sulcus and the ventral raphe. The skin grafts were covered with a layer of fatty gauze, saline gauze, and an elastic foam rubber outer layer to provide light compression.

POSTOPERATIVE COURSE

The urinary catheter remained *in situ* for at least one week. After seven days, the bandage was removed. Previous to that, the patient had been prescribed bed-rest. As soon as it became clear that the skin grafts had taken, the patient was discharged from the hospital. The patient was advised to refrain from coitus for two months. Erections, however, were not opposed: these served as natural tissue expanders.

RESULTS

The skin grafts healed successfully in all six patients. Wound healing on the perineal side of the scrotum was uneventful in all cases. One patient underwent secondary surgery for excision of hypertrophic scar tissue. All patients were satisfied with the cosmetic and functional result. During follow-up (7 to 65 months), edema had not recurred in any of them.

DISCUSSION

According to books in Sanskrit, treatment for lymph edema of the penis and scrotum already started in 600 BC.² World-wide, the most common cause of this condition is filariasis.³ Other causes of acquired (secondary) chronic lymph edema are lymphogranuloma venereum (LGV), chancroid, erysipelas, tuberculosis, leprosy, syphilis, cancer of the lower pelvis, radiotherapy, surgical interventions and trauma. When the cause is not clear, which happens in a number of cases, the condition is referred to as primary (or congenital) chronic lymph edema. In such cases, the lymph vessels have intrinsic abnormalities: aplasia, hypoplasia, and hyperplasia have been mentioned in the literature.⁴

The management of genital lymph edema should focus on treating the underlying disease, and the presence of neoplastic processes should be excluded.⁵

Three options are available for the treatment of chronic genital lymph edema: i) conservative, ii) the creation of lymphatico-venous shunts, and iii) excision of the affected tissue followed by wound closure.¹ Doxycycline against LGV infection and diethyl carbamazepine, albendazole, ivermectin, or doxycycline against *Bancroftian filariasis* can cure early stages of the disease.⁶⁻⁹ Other conservative treatments comprise elevation of the affected part(s), the administration of diuretics, and compression bandages, with or without manual lymphatic drainage-based decongestive therapy.¹⁰ As reflected in our cases (especially case 2), a conservative approach is generally of little use for severe genital lymph edema. The same applies to lymphatico-venous shunts, which probably have the greatest chance of success in cases of secondary lymphoedema.¹¹ The most widely used solution is excision of the affected tissue.¹²

In most cases, the skin is thickened and ulcerated. It is our experience that a cleavage plane can easily be found, since lymph vessels are situated in the dermis and the underlying fascia of Dartos. After excision, the skin of the penis and scrotum must obviously be replaced. We successfully used perineal skin and that of the most posterior part of the scrotum to create a neo-scrotum with no local recurrence, as previously reported.¹³ It is likely that this part of the scrotal skin will not be affected, because its lymphatic drainage includes the perirectal lymph vessels. An important advantage of using posterior scrotal skin flaps is that the resulting coverage is “true to nature.” The remaining scrotal contents can be covered with split-thickness skin grafts if there is a shortage of tissue. Meshed grafts lead to the best cosmetic results on the scrotum, but it is better to use relatively thick non-meshed skin grafts for the penis. Split-thickness skin grafts are hairless and, in contrast to full-thickness skin grafts, usually take more easily. In addition, the risk of recurrence is minimized because split-thickness skin grafts do not contain any reticular dermis, that is, they contain very few lymph vessels.

CONCLUSION

The results of surgical treatment for chronic lymph edema of the penis and scrotum are generally satisfactory. In our experience, closing the scrotal defects with skin from the posterior part of the scrotum did not lead to any recurrence. Covering the rest of the scrotal contents with meshed split-thickness skin grafts and covering the penis with non-meshed split skin grafts led to satisfactory cosmetic and functional results. In our opinion, this specific method of surgical treatment for male genital edema should be the therapy of choice. However, because scrotal edema is rare outside of filariasis endemic regions, it should be done in clinics with sufficient experience.

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The association between Peyronie's and Dupuytren's disease

Helena M. Nugteren¹, Rien J.M. Nijman¹,
Igle Jan de Jong¹, Mels F. van Driel¹

1. University of Groningen,
University Medical Center Groningen,
Department of Urology, Groningen, the Netherlands

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INTRODUCTION

Peyronie's disease (PD) is a localized, connective tissue disorder characterized by the formation of fibrous tissue plaques within the tunica albuginea of the penis, causing a penile deformity, and a subsequent degree of erectile dysfunction and penile pain. Although its aetiology has not been elucidated, PD probably results from the presence of a predisposing genetic susceptibility combined with a trauma to the erect penis.^{1,2} PD appears to be more common in northern European Caucasians; it's uncommon in African-American men and very rare in Asians.³ Diabetes mellitus also seem to be a potential risk factor for PD.⁴

The very first cross-sectional study giving the proposed incidence and prevalence rates of PD was published in 1991 by investigators from Minnesota, USA.⁵ The reported prevalence rate of 0.38 % was consistent with earlier reports. It was estimated that there were more than 423,000 men with PD in the USA at that time, and that 32,000 new cases occurred annually. Mean patient age at diagnosis was 53 years (range 19 to 83). The first PD prevalence rates to be reported in a European cross-sectional study came from Germany. In a survey of 4432 men (aged 30 to 80 years) in Cologne, 3.2 % reported a palpable plaque in the penis.⁴ It is now believed that the actual prevalence rate may be closer to 8 % or higher.⁶ The true prevalence rate of PD may be even higher than that of Dupuytren's disease (DD) because men are likely to underreport a condition that causes embarrassment.

The association between PD and DD was first recognized in 1828 and reported by Abernathy.⁷ According to the literature there is a three to fifteen percent chance that a man with DD will have PD.^{8,9,10} DD is a fibro-proliferative condition of the palmar fascias in the hand, typically resulting in progressive contracture of one or more fingers.¹¹ DD is thought to be the most common hereditary connective tissue disorder in Caucasians.¹² The prevalence of DD in different geographical locations is extremely variable (0.2% to 56%), and it's not clear whether this is genetic, environmental, or a combination of both. Population structure, the prevalence of associated diseases and the diagnostic criteria of DD, makes the understanding of epidemiology quite difficult.¹³ The literature concerning coexisting DD in patients presenting with PD shows wide ranges varying from 0.01 to 58.8 percent.^{5,9,14-24}

The aim of this study is to investigate the coexistence of DD in a consecutive series of patients with PD and their clinical characteristics, presenting at the outpatient urological clinic of the University Medical Centre Groningen, The Netherlands.

METHODS

From January 1988 until December 2009 all patients presenting with PD were examined on DD by one of the investigators (MFvD). The investigator checked both hands for nodules and finger contractures. The diagnosis PD was made by history and examination, i.e. the presence of a palpable plaque in the penis. The investigator measured the size of the plaque using a ruler and noted the size, together with the location of the plaque, in the patients' chart. The direction and severity of the curvature were based on homemade photographs. All patients were asked for the presence of pain, a decreased rigidity and problems with intercourse. The patients were also asked for a positive family history for PD and DD.

RESULTS

The sample consisted of 415 Caucasian male subjects with PD. The mean age of the patients was 60 ± 12 years and the mean duration of the disease was 18 ± 29 months. A total of 252 patients (60.7%) were referred by their general practitioner, 99 (24.0%) by another urologist and 30 (7.2%) by other medical specialties. Eighty-nine of the patients (22.1%) also had DD. In 13 patients (3.1%) there was no information about the presence of DD (table 1).

A total of 28 patients (6.7%) reported to have one or more close relatives (mother, father, brother, sister or grandmother) with DD. Five patients reported to have a father and two patients reported to have a brother with PD.

At the first visit 171 patients (44.9%) experienced a decreased rigidity at erection and 124 patients (34.1%) had pain during erection. A dorsal curvature was present in 238 patients (57.3%) and a ventral curvature in 31 patients (7.5%). There was a problem with intromission of the penis in the vagina in 214 patients (59.6%), problems with coital-movements occurred in 118 patients (33.3%) and 53 (16.8%) of the partners experienced pain during intercourse.

Ninety three patients (22.4%) underwent surgery; a penoplication according to Nesbitt (n=88) or plaque excision or incision with grafting (n=5). Twelve patients (12.9%) reported a complication or an uneventful outcome; erectile dysfunction (n=3), curvature recurrence (n=3), wound infection (n=2), paraphimosis (n=1), urethral stricture due to circumcision (n=1), skin surplus after circumcision (n=1) and anorgasmia (n=1).

Table 1. Clinical characteristics of the study population (415 men)

	N (%), mean \pm SD
Age	60 \pm 12
Duration of PD (months)	18 \pm 29
Coexistence of DD	89 (22.1%)
Referral	
General practitioner	252 (60.7%)
Urologist	99 (24.0%)
Other	30 (7.2%)
Unknown	34 (8.2%)
Curvature	
Dorsal	149 (35.9%)
Dorsal left	67 (16.1%)
Dorsal right	22 (5.3%)
Ventral	18 (4.3%)
Ventral left	11 (2.7%)
Ventral right	2 (0.5%)
Left	60 (14.5%)
Right	13 (3.1%)
Left and right	1 (0.2%)
None	9 (2.2%)
Unknown	63 (15.2%)
Stable relationship	319 (89.4%)
Pain partner during intercourse	53 (16.8%)
Pain in erection	124 (34.1%)
Decreased rigidity	171 (44.9%)
Intromission problems	214 (59.6%)
Problems coital movements	118 (33.3%)
Shortening of penis	73 (21.7%)
Operation	93 (22.4%)
Penoplication according to Nesbit	88 (21.2%)
Plaque excision or incision with grafting	5 (1.2%)
Complications	12 (12.9%)
Curvature recurrence	3 (25%)
Decreased rigidity	3 (25%)
Wound infection	2 (16.7%)
Paraphimosis	1 (8.3%)
Urethral stricture due to circumcision	1 (8.3%)
Skin surplus after circumcision	1 (8.3%)
Anorgasmia	1 (8.3%)
Family with DD	28 (6.7%)
Father	12 (2.9%)
Mother	13 (3.1%)
Brother	5 (1.2%)
Sister	3 (0.7%)
Grandmother	4 (1.0%)
Grandfather	0 (0%)
Family with PD	7 (1.4%)
Brother	2 (0.5%)
Father	5 (1.2%)

DISCUSSION

In order to evaluate the coexistence of DD in patients with PD, 415 consecutive patients were examined. This study is the largest series which examined the coexistence of DD in patients with PD. In 22% both diseases were present. Previous reports concerning coexisting DD in patients presenting with PD shows ranges varying from 0.01 to 58.8 percent, a positive family history for PD in 1 to 4 percent and a positive family history for DD in 9,8 percent (table 2).^{5,9,14-24} The series from the UK, Italy, Australia and Serbia show the highest percentage of PD patients with coexisting DD.^{9, 15, 17, 19, 24} The two reports focusing on PD patients < 40 years show a lower percentage of coexisting DD.^{21, 23} Only Williams and Thomas specified their method of finding DD in PD patients.¹⁵ Twenty-five had a follow up period of two months up to thirteen years. Seventeen of them were examined particularly for `Dupuytren's contracture` in view of the known incidence with PD. Eventually, DD was diagnosed in ten patients.

Table 2. Studies on Dupuytren's disease in patients with Peyronie's disease

Authors	Country	Year	Number pts	Plus DD	Positive family history for PD	Positive family history for DD
Smith ¹⁴	USA	1966	26 (dead)	2 (7.7%)	-	-
Williams ¹⁵	UK	1968	17	10 (58.8%)	-	-
Chilton ¹⁶	UK	1982	408	63 (15.4%)	8 (1.9%)	-
Lindsay ⁵	USA	1991	101	4 (4 %)	-	-
Ralph ⁹	UK	1997	51	15 (34%)	-	-
Carrieri ¹⁷	Italy	1998	134	28 (21%)	6 (4%)	-
Perimenis ¹⁸	Greece	2001	134	3 (2.2%)	-	-
Johnson ¹⁹	Australia	2002	294	73 (25 %)	-	-
Kadioglu ²⁰	Turkey	2002	307	3 (0.01%)	3 (1%)	-
Levine ²¹	USA	2003	30 < 40 yrs	1 (3 %)	-	-
Bjecic ²²	Serbia	2006	82	32 (39%)	-	8 (9.8%)
Deveci ²³	USA	2007	296 over all 32 < 40 yrs 264 > 40 yrs	11 (3.7%) 0 11 (4.2%)	-	-
Rhoden ²⁴	Brazil	2010	83	5 (6%)	-	-
This series	Netherlands	2010	415	89 (22.1%)	7 (1.4%)	28 (6.7%)

The wide variation of patients with PD having coexisting DD may have several reasons. First, the gene expression profiles of DD and PD may differ throughout the world. Second, in some articles the authors clearly discuss about `Dupuytren`s contracture` and not about DD. Patients with discrete nodules in their hands may not be diagnosed as DD and one may presume that not in all series patients have been examined by an experienced clinician. The third explanation for the wide spread may be the examination of different age groups, because the risk of acquiring PD as well as DD increases with advancing age. A Turkish study showed that out of 231 men with PD, only 8.2% presented under the age of 40.²⁵ A comparable percentage (9.9%) was also observed in the Minnesota study.⁵

Hindochoa et al. report a mean age of onset in familial DD of 49 years and 55 in non-familial DD.¹³ This may explain the very low percentage of coexisting DD in the study of Levine et al.²¹ Finally, the varying or unknown follow up periods in the different studies may explain the wide ranges of coexisting DD in PD.

In 1982 Nyberg and co-workers documented the familial transmission of PD as an autosomal dominant trait in three pedigrees.¹⁰ The occurrence of DD in 7 of their 9 (78%) affected individuals, which was a significant increase over the average 0% reported in sporadic cases, suggested that both of these fibrosing disorders should be pleiotropic effects of the same genes in these families. Similarly, the histocompatibility B7 cross-reacting antigens were present in 90% of their PD patients.

Ziegelbaum and co-workers reported on identical twins with PD and the HLA-B40 antigen.²⁶ Family studies were also undertaken by Bias and co-workers when three patients reported similarly affected first-degree relatives.²⁷ One kindred showed father-to-son transmission of PD with DD in three generations. Pedigree analysis of the three families suggested that PD also is a male-limited, autosomal dominant trait. Antigens of the HLA-B7 cross-reacting group occurred in all three kindred's; however, the data ruled out close linkage of the disease and HLA.

CONCLUSIONS

DD is thought to be the most common hereditary connective tissue disorder in Caucasians. However, the true prevalence rate of PD may be even higher than that of DD due to the fact that men are likely to underreport a condition that causes embarrassment. This single-center study in Dutch PD patients showed coexisting DD in twenty percent. Given the increased recognition of PD, as well as the emerging treatment which is currently being used for DD and may become approved worldwide for PD (collagenase), the association between these two disorders will gain greater importance.

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**General discussion
and
future perspectives**

8

GENERAL DISCUSSION

Today, penile and scrotal abnormalities have become important as a medical topic. As we discussed in chapter 1, this is not only because they occur more frequently but because of a combination of and interaction between medical and especially cultural changes. Moreover, it was concluded that: *“This high level of acceleration may also have some disadvantages; particularly in healthcare, hypes need to be prevented. For urologists, of course, it is important to remain in touch with innovative techniques both in the domain of human sexuality as well as in its problems. Given the primary medical ethical amendment, however, of doing no harm (primum non nocere), it is also important to take note of the limitations of these medical technical developments and of his/her profession. There will always be patients with penile abnormalities who are ‘surgically incurable,’ or who are better off visiting a sexologist or psychotherapist. In all cases iatrogenic damage, big or small, emotional or financial, must be prevented.”*

It was the awareness of this delicate balance that brought us to our first overarching research question: Is surgery a good, preferably the best, treatment option in case of penile or scrotal deformity?

This main question was differentiated into three sub-questions that refer to three morphological qualities and accompanying diagnoses:

- *size*: the small penis
- *angle*: acute erection angle
- *expansion*: genital lymph edema.

Because treatment decisions are closely linked to a specific context, that is, diagnosis, our answer to the overarching question will be put in perspective here, using the international literature per anomaly.

Our second research aim was to investigate the association observed between Peyronie’s and Dupuytren’s disease. Both questions will be discussed here separately, and, afterwards, some implications or lessons learned – based on our limited study – will be discussed for the urosexology of tomorrow.

I. Is surgery a good, preferably the best, treatment option for:

The small penis

In chapters two and three we concluded that surgery is the only proven treatment for penile enlargement. This treatment is relatively simple and safe, and leads to the highest levels of patient satisfaction, if there was a correct indication, that is, it was a matter of functional complaints. This type of intervention is not suitable for patients with small penis syndrome or for patients who already achieve a penis length of no less than 7.5 cm in extension. These findings are in concordance with other, international findings. Li and colleagues noted that, despite an adequate penile length gain, overall only 35% of the patients were satisfied with the outcome after cutting the suspensory ligament. Satisfaction rates were low in patients with dysmorphophobia (27%), while higher in patients with an organic cause for their shortness of penis.¹

This confirms the idea that some patients have unrealistic expectations, and any length gain would not be enough in these patients' view. Furthermore, in men with functional complaints, surgical treatment is the last resort and should be implemented only when all other conservative measures have proved unsuccessful.

From a clinical perspective this means that two conditions must be met before surgery can be used as a good, possibly even the best treatment option:

- an indication of functional complaints
- an informed consent at a multidisciplinary level, counseling sessions with a psychologist-sexologist, and detailed explanation of the risks and complications of any operation.

Acute erection angle

In chapter four, we found that, when counseling has failed to provide relief, simple and safe surgical techniques to divide the penile suspensory ligament can yield adequate results in patients with an acute erection angle. In our case report, both males, who were treated for their acute erection angle, were satisfied with their surgical outcome.

Before operating, it is important that a couple with complaints of sexual inadequacy, which has resulted specifically from this abnormality, receive objective reassuring information about anatomical and physiological facts. Furthermore, healthcare professionals should offer sex-education information on more enhanced, functional penile positions for sexual intercourse in different positions.

Moreover, to the best of our knowledge, there is no comparative literature concerning the operation for this indication. That said, another anomaly of the erection angle, the lateral deviation of the erect straight penis (LDESP), has been recently described by Shaer.² This latter is described as “a penis that points laterally when erect, despite being straight, yet can be redirected forward by manual correction, without the use of force.” Surgery was performed in patients with LDESP, in cases where counseling had

failed and if the angle of the erect penis was easily manually corrected. During surgery an erection was induced and the skin on the contra lateral side of the LDESP was “pinched” to determine the extent of skin reduction required to correct the angle. The skin was then excised, and the incision was closed in two layers. The author reported a satisfaction rate of 73.3% after surgery.

Genital lymph edema

As reflected in our cases in chapter seven, a conservative approach is generally of little use for severe genital lymph edema. The study showed that excision of the affected tissue and closing the scrotal defects with skin from the posterior part of the scrotum using split skin led to satisfactory cosmetic and functional results. However, scrotal edema is rare outside of filariasis-endemic regions, and therefore any intervention should be done in clinics with sufficient experience.

Another surgical treatment for secondary male genital lymph edema consists of a physiologic operation, in which new channels are created to increase the transport capacity of lymphatic fluid.³ Up until now, this treatment has not been very successful, and control of the edema has been considered difficult to maintain over a long period. However, the development of super-microsurgery makes it possible to create anastomosis of very small vessels. Using this microscopic anastomosis technique, new methods for lymphaticovenular anastomosis (LVA) are being developed.⁴⁻⁷ In LVA surgery, a lymphatic vessel is anastomosed to a venule or small vein in an intima-to-intima coaptation manner so as to bypass congested lymph into venous circulation.⁸ However, LVA has a potential risk of thrombosis of the anastomosis, because venous blood can be refluxed into the anastomosis site.^{9,10} Because of this thrombosis, Yamamoto and Koshima describe a technique in which a non-obstructed lymphatic vessel is used as a recipient vessel for anastomosis of an obstructed lymphatic vessel.⁸ They describe a case of a male with a cellulite-induced lymph edema of his lower leg, who was treated with super-microsurgical superficial-to-deep lymphaticolymphatic anastomosis (LLA). At six months post-operative, there was a slight volume reduction in the affected lower leg. However, the superficial-to-deep LLA method can only be used in limited cases. It cannot, for example, be used for the treatment of secondary lymph edema after lymph node dissection or radiation, because both the superficial and the deep lymphatic systems are obstructed.

Theoretically, lymphovenous or lymphaticolymphatic micro-anastomoses are promising yet technically complex. Application to scrotal lymph edema has not yet been defined, and further studies are required before widespread application of these treatments.

II. Is there an association between Dupuytren's and Peyronie's disease?

In order to evaluate the coexistence of DD in patients with PD, 415 consecutive patients were examined in our study described in chapter six. This study is the largest series, which has examined the coexistence of DD in patients with PD. Our conclusion was that, in 22%, both diseases were present.

These findings are in concordance with those of Qian and colleagues, who demonstrated that the pattern of alterations in the expression of certain gene families in PD and DD was similar. This suggests that they share a common pathophysiology. A recent genome-wide association study performed by Dolmans and colleagues revealed nine susceptibility loci for DD.¹¹ Another research by this author with 111 PD patients showed that the WNT2 is a susceptibility locus for PD; this is one of the loci associated with DD. The WNT2 association was even more significant after removal of 15 patients with co-morbid DD, which proves that the association is caused by PD and not by DD. This study provides evidence for a partly shared genetic susceptibility between PD and DD.¹²

The recognition of Peyronie's disease is growing, and there is an increase in the various treatments currently being used for Dupuytren's disease. The association between these two disorders will most likely gain in importance, as they may be amenable to the same therapeutic regimens.

CONCLUSIONS ON A META-LEVEL

In this thesis, the results of clinical studies, case reports, and literature studies on the diagnostics and treatment of anatomical abnormalities of the male genitals have been described. We have tried to provide an answer to questions such as: which anatomical and scrotal abnormalities are relatively common, and whether, in that case, operating is a responsible or even preferable approach. In short, we have tried to survey a part of the urological field, while keeping in mind the medical ideal of maximum rapprochement while maintaining distance. On a meta-level this leads to some interesting and cross-curricular insights:

1. Basically, urology can contribute to the quality of the sexual life of patients with deformed genitalia. It seems that the ethical principles of doing no harm (*primum non nocere*) is sufficiently met.
2. The decision as to whether this contribution is beneficent, however, is only partly a urological matter. This kind of decision belongs to the domain of co-creation with patients and professional colleagues. After all, this is a situation in which:
 - there are many different stakeholders, such as the patient, partner, practitioner, and society;
 - different angles are conceivable, such as medical, ethical, aesthetic, and financial; and there are always pros and cons, since no surgery is, after all, “for free,” nor does it escape the placebo and/or nocebo effect.

FUTURE PERSPECTIVES

What this thesis makes clear is that urological sexology is not an isolated phenomenon, an island in the urological, surgical, or even medical sea. Our conclusions, that dysmorphic penile problems can be resolved by surgery and that the side effects are acceptable, do not mean that we have found a panacea for all these kinds of problems. The fact of the matter is that our reassuring conclusions are only the starting point in a far wider healthcare discussion. In order to structure this discussion, the model for multi-level implementation suggested by Joore's thesis might prove helpful.¹³

Level 1

Urological surgery as a technical solution; the criteria are safety and quality in terms of technical feasibility, functional results, and acceptable side effects, preferably seen in the light of an evidence base.

Level 2

The practical use: the criteria are practicality for the patients (duration, understandability/meaning, burden) and for the professionals (surgeon, nurses, etc., in terms of work satisfaction, time consumption on an individual basis, etc.).

Level 3

Managerial use by team leaders and coordinators; the criteria are not only quality of care but also cost/benefit ratio on a group level, functionality in multi-disciplinary collaboration, etc.

Level 4

Governance level: the criteria are consistency with organizational values; in other words, does the whole chain (operation, treatment, etc.) help us reach our aims as a medical center?

The caesura between 1 and 2 versus 3 and 4 reflects the current discussion in healthcare as to whether to focus on the content of the managerial and/or political dimensions. Here the roles of "practical physicians and nurses" meet the roles of "managerial team leaders." From a bio-psycho-social perspective, these dimensions can be distinguished yet not separated. It is, therefore, important to note that, until now, research has only focused on "respondents" from the first two levels. Future research should certainly also include team leaders (level 3), who have their own opinions about the use of surgeons and nurses (level 2), let alone representatives of the board of directors of the hospital involved, who have to negotiate with the financial-political system about the scale of healthcare provided.

All in all, it is the combination of all these aspects that makes a multidisciplinary (including healthcare management), stepped-care approach preferable, one in which:

1. On a multidisciplinary level, where which intervention is medically-technically feasible must be checked, along with whether there are multiple options: What are the pros and cons of the interventions, and what are their far-reaching consequences? Questions that can be asked during this discussion are:
 - a) What is the exact dilemma? Is there, for example, an indication for invasive urological intervention, or is there dysmorphic suffering?
 - b) What basic moral principal is at stake: 1) autonomy, 2) beneficence, 3) non-maleficence (*primum non nocere*), or 4) justice?
2. Since there are differences within but also between treatments, all these points need to be worked out per moral principal, in other words, what are the pros and cons?
3. This should then be worked out between the principals themselves. For example, one of the consequences of not honoring the question that the patient has is that his autonomy might be harmed. This stepped-care approach makes clear which criteria collide, and one can then set about prioritizing.
4. The outcome of this consideration – of course, within legal and social frameworks – is the stepping-stone to a dialogue between patient and practitioner.
5. Doctor and patient should arrive at a shared plan of action. When there is a simple anomaly, which is easy to correct, the shared plan of action can take place in the form of an informed consent. The doctor will propose a treatment, and the patient decides if he will undergo this treatment. However, more often than not there will be a more complicated dilemma, and then “shared decision-making” is the standard. The urologist will go through all the options together with the patient, examining which treatment is a realistic solution to his problems, and if its consequences are in proportion to the expected result.
6. At the same time, a continuous dialogue should be held with representatives of the management layer or preferably even higher, the board of directors. In order to guarantee safety and quality in the short term, this kind of surgery should be well organized. In the long term, the question of whether this kind of surgery has added value to the hospital should be discussed. Only if this is also the case, does urogenital surgery of penile or scrotal dismorphic problems have a sound future.

In short, the urological-surgical act should always be contextually assessed; a rigid never/always approach or a simple prescription of medicine has no place here.

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Summary

9

In this thesis the findings and results of clinical studies, case reports, and literature reviews on diagnosis and treatment of the abnormalities of penile and scrotal anatomy have been reported. We tried to give an answer to the questions of “what is normal?” and “when to operate?”

The first aim of the introduction, **chapter 1**, was to provide general information about the short penis, the acute erection angle, Peyronie’s disease, and genital lymph edema. The four anomalies are put into a urological and sexological perspective. The second aim was to describe the two main research questions: 1) Is surgery a good, preferably the best, treatment option in case of penile or scrotal deformity? 2) Is there a common pathway to Peyronie’s and Dupuytren’s disease?

The erect penis has been, in many cultures and throughout the millennia, the ultimate symbol of masculine qualities. Because of this symbolism, men are likely to feel insecure or embarrassed if their penis is less than average size. A large body of research indicates that 45% of interviewed males would prefer a larger penis. When there is a fear-fuelled subjective perception that a penis is considerably smaller than average, it is called “small penis syndrome.”

Chapter 2 described our experience in the management of men with complaint of a small penis. In eighteen years we collected 60 men with this complaint. For 44 of the men, counseling was sufficient. Sixteen of the 60 men underwent penis-enhancing surgery; only nine of them were satisfied with the result. In addition, we found that six of the operated men had a penis length of 7.5 cm or more in extension. It appeared that only two of these men were pleased with the operation’s results. We concluded in this chapter that those men, who had already achieved a penis-length of 7.5 cm in erection, experienced only limited benefit from penis-enhancing surgery. This particular patient category should therefore be dissuaded from surgery. Men with small penis syndrome do, without fail, benefit from several counseling sessions with a psychologist-sexologist. Depression and personality disorders are legitimate reasons for not proceeding with surgical treatment.

Besides lengthening and thickening operations, penis-lengthening pills, stretch apparatus, vacuum pumps, and silicone injections are also available for men who worry about their penis size. Surgery, however, is thus far the only proven scientific method for penile enlargement.

In **chapter 3** we considered patient selection, outcome evaluation, and techniques applied for penile enlargement. The most widely used technique for lengthening is cutting the fundiform and suspensory ligaments of the penis, in combination with removal of suprapubic fat tissue. Obviously, this technique only provides an optical lengthening; a visual length gain of 2 cm, in flaccid state, is considered an adequate result.

One of the methods for broadening the penis is by injecting autologous fat tissue in between the superficial and deep fascia. Because of the increased weight, it can also make the penis a bit longer. Another possibility is transplanting deepithelialized skin with

matching subcutaneous fat. However, both techniques have a high complication rate, and in the Netherlands it is considered good practice not to perform thickening operations.

An experimental and extraordinary operation technique has been developed by a famous Serbian urologist, whereby the entire penis is disassembled. The technique starts with the dissection of the corpus spongiosum and the neurovascular bundle. Following this, the glans and the cavernous bodies are separated, leaving a small space between the glans and the ends of the corpora cavernosa. A small piece of rib cartilage is inserted inside this cavity. This operation technique can be combined with the cleaving of the ligaments. According to this urologist, this technique offers an enhanced penis length of 2-3 cm.

During erection, the penis increases in volume, rigidity, and angle. Textbooks of urology and sexology provide only very limited information about erection angle dysfunction. In some men, this angle is too tight against their belly, causing problems with intercourse. This may be caused by severe shortness of the penile suspensory ligaments (PSL), severe dorsal curvature at the penopubic level, or a combination of both. The surgical treatment options are: partial release of the suspensory ligaments, a ventral corporoplasty according to Nesbit, or, if necessary, a combination of both.

In **chapter 4** two cases were reported of this so-called “acute erection angle,” and the pertinent literature was discussed. In the first patient a corporoplasty, according to Nesbit, was sufficient to lower the erection angle. In the other patient the ligament was released and, to prevent re-attaching, the pubic bone and the cavernous bodies were separated by interposing some suprapubic fat and two folded collagen fleeces. At nine-month follow-up, both patients were satisfied with the surgical outcome. We concluded this chapter with the statement that, if counseling has failed to provide relief, simple and safe surgical techniques can yield adequate results.

Chapter 5 was a short intermezzo in which the symbolic meaning of the penis and the etymology of the phallus were described. We also provided a short outline of the characteristics of the penis in animals.

Genital lymph edema is a debilitating disease of the male genitals. It is caused by stasis of lymph in subcutaneous tissues. It may cause problems with hygiene, urinary incontinence, inability to have sexual intercourse, and even immobility. The most effective treatment is surgical excision of the affected tissue, followed by wound closure using split-thickness skin grafts, or skin flaps from the perineum and the most posterior part of the scrotum.

In **chapter 6** we described the surgical treatment of six patients with genital lymph edema and the results. In all the men, a satisfactory cosmetic and functional result was established. In our opinion, this specific method of surgical treatment for male genital edema should be the therapy of choice.

In **chapter 7** another form of penile deformity was described. Peyronie's disease (PD) is a localized, connective tissue disorder characterized by the formation of fibrous tissue plaques within the tunica albuginea of the penis, causing a curvature, and a subsequent degree of erectile dysfunction and penile pain. According to the literature there is an association between PD and Dupuytren's disease (DD). DD is a fibro-proliferative condition of the palmar fascias in the hand, typically resulting in progressive contracture of one or more fingers. In this chapter we investigated the coexistence of DD in a consecutive series of patients with PD. We found that, in 22% of 415 investigated PD patients, there was a coexisting DD. Other studies concerning coexisting DD in patients presenting with PD show ranges varying from 0.01 to 58.8%. This wide variation may have several reasons. First, the gene expression profiles of DD and PD may differ throughout the world. Second, patients with discrete nodules in their hands may not be diagnosed as DD. The third explanation for the wide spread may be the examination of different age groups, because the risk of acquiring PD as well as DD increases with advancing age.

Given the increased recognition of PD, as well as the emerging treatments currently being developed for DD, the association between these two disorders should grow in importance.

With all anatomic penile and scrotal disorders, it is important for the urologist to provide objective and reassuring information about anatomical and physiological facts. If there is an indication for an operation, the urologist should discuss all available options together with the patient, culminating in clarity on whether an operative intervention can or cannot bring relief for the problem experienced.

If there are complaints of sexual inadequacy, sexological counseling is mandatory before any operative intervention.

Nederlandse samenvatting

10

In dit proefschrift worden de resultaten beschreven van klinische studies, case reports en literatuurstudies over de diagnostiek en behandeling van enkele anatomische afwijkingen van het mannelijk genitaal. We hebben geprobeerd inzicht te geven in een aantal anatomische peniele en scrotale afwijkingen en antwoord te geven op de vragen: wanneer is opereren een verantwoorde of zelfs preferente aanpak, en wanneer gaat het de grens van ultimum refugium voorbij?

Het eerste doel van de inleiding, **hoofdstuk 1**, is het geven van algemene informatie over de korte penis, de 'acute erection angle', de ziekte van Peyronie en genitaal lymfoedeem. De vier anatomische afwijkingen worden in een urologisch en seksuologisch perspectief geplaatst. Het tweede doel van de inleiding is het omschrijven van de twee belangrijkste onderzoeksvragen: 1) is chirurgie een geschikte behandeloptie in geval van peniele of scrotale afwijkingen? 2) is er een associatie tussen de ziekte van Peyronie en de ziekte van Dupuytren?

De erecte penis is door de eeuwen heen en in vele culturen het ultieme symbool van mannelijkheid geworden. Vanwege deze symboliek zijn mannen onzeker of beschaamd als, in hun ogen, hun penisgrootte onder het gemiddelde is. Uit wetenschappelijk onderzoek is gebleken dat 45% van de ondervraagde mannen liever een grotere penis zou willen hebben. Als een man een angstige en subjectieve perceptie heeft dat zijn penis aanzienlijk kleiner is dan gemiddeld, dan wordt dit het 'kleine penis syndroom' genoemd.

Hoofdstuk 2 beschrijft onze ervaring met de behandeling van mannen die klagen over een te kleine penis. In een tijdsperiode van achttien jaar zijn er 60 mannen verzameld met deze klacht. Voor 44 mannen was (psychosociale) begeleiding voldoende. Zestien van de 60 mannen hadden een penisverlengende operatie ondergaan; slechts negen van hen waren tevreden met het resultaat. Bovendien vonden we dat zes van deze geopereerde mannen een penislengte van 7,5 cm of meer in extensie hadden, waarbij slechts twee van deze mannen tevreden waren met het resultaat. De conclusie van dit hoofdstuk is dat mannen met een penislengte van 7,5 cm of meer in erectie slechts in beperkte mate profiteren van penisverlengende chirurgie. Deze patiëntencategorie moet daarom worden ontmoedigd om een operatie te ondergaan. Voor mannen met het 'kleine penis syndroom' is het raadzaam om een aantal sessies met een psycholoog-seksuoloog te ondergaan. Depressie en persoonlijkheidsstoornissen zijn legitieme redenen voor het niet uitvoeren van een chirurgische behandeling.

Voor mannen die zich zorgen maken over de grootte van hun penis zijn er, naast penisverlengende en -verdikkende operaties, ook penisverlengende pillen, strekapparaten, vacuümpompen en siliconeninjecties op de markt. Een operatieve behandeling is tot nu toe de enige wetenschappelijk bewezen methode om een penis te verlengen.

In **hoofdstuk 3** wordt de patiëntselectie bij penisverlengende operaties besproken en worden de resultaten en technieken die worden toegepast geëvalueerd. De meest

gebruikte techniek voor verlenging van de penis is het klieven van het ligamentum fundiforme en suspensorium, in combinatie met het verwijderen van suprapubisch vetweefsel. Deze techniek zorgt uiteraard alleen voor een optische verlenging, waarbij een visuele lengtewinst van 2 cm in niet-erecte toestand wordt beschouwd als een afdoend resultaat.

Een van de methoden om de penis te verdikken is door het injecteren van autoloog vetweefsel tussen de oppervlakkige en de diepe fascie van de penis. Een bijkomend voordeel is dat door het toegenomen gewicht van de penis, deze ook wat langer lijkt. Een andere mogelijkheid is het transplanteren van huid inclusief onderhuids vetweefsel (ontdaan van de epitheel laag). Beide technieken hebben echter een hoge kans op complicaties en in Nederland worden penisverdikkende operaties dan ook niet uitgevoerd.

Een beroemde uroloog uit Servië heeft een experimentele en vooral buitengewone operatietechniek voor penisverlenging ontwikkeld, waarbij de penis als het ware wordt gedemonteerd. De techniek begint met het vrijprepareren van het corpus spongiosum en de neurovasculaire bundel. Vervolgens worden de glans en de corpora cavernosa van elkaar gescheiden, waardoor een kleine ruimte tussen de glans en het uiteinde van de corpora cavernosa ontstaat. Vervolgens wordt een stukje autoloog kraakbeen in deze holte geplaatst. Deze operatie kan gecombineerd worden met het splijten van de ligamenten. Volgens de Servische uroloog kan bovenstaande operatietechniek zorgen voor een penisverlenging van 2 tot 3 cm.

Handboeken over urologie en seksuologie geven slechts zeer beperkt informatie over de hoek van een erecte penis. Tijdens een erectie neemt de penis toe in volume en stevigheid en verandert de hoek ten opzichte van de buik. Het kan echter voorkomen dat deze hoek te scherp is in de richting van de buik, wat problemen kan geven tijdens geslachtsgemeenschap. Deze scherpe hoek kan veroorzaakt worden door een te kort ligamentum suspensorium en fundiforme, door ernstige dorsale kromming van de penis ter hoogte van het os pubis of een combinatie van beide. De chirurgische behandelingen zijn het gedeeltelijk klieven van het ligament, ventrale corporoplasty volgens Nesbit of een combinatie van beide.

In **hoofdstuk 4** wordt relevante literatuur geëvalueerd en worden twee casussen besproken van deze zogenoemde 'acute erection angle'. Bij de eerste patiënt was een corporoplasty volgens Nesbit voldoende om de hoek van erectie te vergroten. Bij de andere patiënt werd een adequaat resultaat verkregen na het klieven van het ligament, waarbij ook suprapubisch vet met twee stukjes 'collagen fleecce' werd ingebracht ter hoogte van het gekliefd ligament om opnieuw aangroeien van os pubis en corpora te voorkomen. Na negen maanden follow-up bleek dat de patiënten tevreden waren met de chirurgische uitkomst. De casussen laten zien dat eenvoudige chirurgische technieken een adequate oplossing kunnen bieden voor mannen met een 'acute erection angle'.

Hoofdstuk 5 is een kort intermezzo waarin de symbolische betekenis en de etymologie van de penis is beschreven. We geven tevens een korte beschrijving van de karakteristieken van penissen bij een aantal dieren.

Genitaal lymfoedeem is een invaliderende ziekte van het mannelijk geslachtsdeel. Het wordt veroorzaakt door stagnatie van lymfevocht in subcutane weefsels. Het oedeem kan leiden tot problemen met hygiëne, urine-incontinentie, het onvermogen om geslachtsgemeenschap te hebben, en zelfs immobiliteit. De meest effectieve behandeling is chirurgische excisie van het aangetaste weefsel, gevolgd door wondsluiting met behulp van split-thickness huidtransplantaties of gebruik van huidflappen gecreëerd vanuit het perineum en het achterste deel van het scrotum.

In **hoofdstuk 6** wordt de chirurgische behandeling van zes patiënten met genitaal lymfoedeem beschreven. Alle mannen hebben postoperatief een bevredigend cosmetisch en functioneel resultaat. Naar onze mening is bovenstaande chirurgische behandeling voor mannelijk genitaal lymfoedeem de therapie van keuze.

Hoofdstuk 7 beschrijft een andere anatomische afwijking van de penis. De ziekte van Peyronie (PD) is een gelokaliseerde bindweefselaandoening gekenmerkt door de vorming van een streng bindweefsel in de tunica albuginea van de penis. Hierdoor ontstaat een kromming van de penis tijdens erectie en ontstaat op langere termijn ook erectiele dysfunctie en peniele pijn. Volgens de literatuur is er een associatie tussen PD en de ziekte van Dupuytren (DD). DD is een progressieve aandoening waarbij de vingers, door het fibroseren van de fascia palmaris, in een flexiestand komen te staan.

In dit hoofdstuk hebben we onderzocht hoeveel patiënten met PD ook DD hebben. We vonden dat in 22 procent van 415 onderzochte PD-patiënten ook DD aanwezig was. Bij andere studies varieerde de prevalentie van DD bij patiënten met PD tussen 0,01 en 58,8 procent. Dit is een erg grote spreiding, die op meerdere manieren verklaard zou kunnen worden. Allereerst kan over de hele wereld het genexpressieprofiel van DD en PD verschillen. Ten tweede kunnen discrete knobbeltjes in de handen van patiënten niet gediagnosticeerd worden als DD. De derde mogelijke verklaring voor deze grote spreiding is dat in de studies verschillende leeftijdsgroepen zijn onderzocht, aangezien het risico op het verwerven van PD en DD stijgt met de leeftijd.

Gezien de huidige, steeds groter wordende erkenning voor PD en de nieuwe behandelingen die momenteel worden ontwikkeld voor DD, zal de associatie tussen deze twee ziektebeelden steeds belangrijker worden.

Bij alle anatomische peniele en scrotale aandoeningen is het belangrijk dat de uroloog objectieve informatie geeft over de anatomie en fysiologie van penis en/of scrotum. Als er een indicatie bestaat voor een operatie, is het van belang dat de uroloog alle beschikbare opties met de patiënt bespreekt en vooral duidelijkheid verschaft of een operatieve ingreep een oplossing voor het probleem is.

Als er klachten zijn van seksuele tekortkoming, is seksuologische counseling sterk aan te raden voordat er een operatieve ingreep plaatsvindt.

Curriculum Vitae

Helena Madeline Nugteren werd geboren op 7 maart 1983 te Delfzijl. In 2001 behaalde ze haar VWO diploma aan het Ommelander College te Appingedam. Een jaar later werd ze ingeloot voor de studie geneeskunde aan de Universiteit van Maastricht. Als vijfdejaars student ging ze vier maanden naar de Filipijnen om hier haar keuzecoschap te volgen op de afdeling chirurgie in het University of the East Ramon Magsaysay Memorial Medical Center te Manilla en in de provincie Samar voor Community Healthcare. Terug in Nederland begon ze haar wetenschapsstage bij de afdeling urologie in het Universitair Medisch Centrum Groningen. Tijdens deze stage kwam, onder de bezielende leiding van dr. M.F. van Driel, de eerste publicatie tot stand betreffende mannen die klagen over een te kleine penis.



In oktober 2008 ontving Helena haar artsenbul en verhuisde ze van Maastricht naar haar geboorteplaats. Ze deed vervolgens werkervaring op als ANIOS urologie in het Refaja Ziekenhuis te Stadskanaal, het Ommelander Ziekenhuis te Winschoten en het Universitair Medisch Centrum Groningen. In deze periode verschenen er meer publicaties over afwijkingen van het mannelijk genitaal en kwam het huidige promotietraject tot stand.

In 2011 werd ze toegelaten tot de opleiding urologie. De vooropleiding heelkunde vond plaats in het Scheper Ziekenhuis te Emmen (opleider dr. M. van den Berg), waarna het academische gedeelte volgde op de afdeling urologie in het Universitair Medisch Centrum Groningen (opleiders prof. dr. J.M. Nijman en dr. M.F. van Driel). Momenteel doorloopt ze het perifere gedeelte van de opleiding op de afdeling urologie in het Martini Ziekenhuis te Groningen (opleider dr. L.F.A. Wymenga). De verwachting is dat in juni 2017 de opleiding zal worden afgerond.

In 2014 is Helena getrouwd met Herman Hamstra en in 2016 is hun zoon, Leander, geboren.

Publications and presentations

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Nugteren HM, Balkema GT, Pascal AL, Weimar Schultz WCM, Nijman JM, van Driel MF

European Society for Sexual Medicine (ESSM), Lyon 2009, Poster presentation
Physical therapy for premature ejaculation and erectile dysfunction
Nugteren HM, Weijmar Schultz WCM, Nijman JM, van Driel MF

European Society for Sexual Medicine (ESSM), Lyon 2009, Poster presentation
Surgery for an 'acute erection angle' when counselling fails
Nugteren HM, Pascal AL, Weijmar Schultz WCM, Nijman JM, van Driel MF

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Physical therapy for premature ejaculation and erectile dysfunction
Nugteren HM, Weijmar Schultz WCM, Nijman JM, van Driel MF

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