



Psychosocial, Sexual Function, and Fertility in Hypospadias

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27.1 Introduction

Hypospadias has several implications for a child which extend beyond those of a structural abnormality, due to the myriad functions of the penis [1]. These include abnormal appearance of genitalia, spraying of urinary stream, potential inability to void standing, the psychological impact of genital abnormality, and multiple corrective operations as a child [2]. The goal of reconstructive surgery in hypospadias is to achieve a cosmetically and functionally normal penis. This includes a normal position as well as the shape of the meatus, glans, and shaft, ability to stand and void with a stream directed forwards and normal erections without penile curvature that enables satisfactory sexual intercourse [3]. Despite repair, a large proportion continues to experience negative genital perception and desire corrective surgery as adults [4].

Long-term follow-up studies on body image, psychological and sexual function are riddled with problems such as lack of periodic review, loss to follow-up, the use of diverse techniques, and lack of a control group. These limitations make comparison difficult [5]. Many long-term studies may not be applicable today due to improvements in surgical technique and better

outcomes. Most reports on psychosexual outcomes are retrospective and based on questionnaires administered to patients. Drawbacks of this approach include loss to follow-up, recall bias, representation bias (those who had better psychosexual outcomes may be over-represented as they may be more willing to participate) [6] and misunderstanding of questions. Information obtained from interviews with patients may not be completely accurate as the patients may not be forthcoming with sensitive personal information. However, mailed questionnaires allow patients to respond to delicate questions in the privacy of their homes and are more likely to elicit accurate replies [7]. These aspects emphasize the need for long-term follow-up to build trust with the patient and to understand the sequelae of hypospadias repairs carried out in childhood [2].

We aim to summarise the available evidence on psychological, sexual, and fertility outcomes after hypospadias repair.

27.2 Psychosocial and Sexual Development

27.2.1 Psychosexual Development and Gender Identity

Psychosexual development refers to the attainment of psychological and sexual maturity that enables people to have satisfactory relationships and sexual lives [8]. The term psychosexual

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development is used because it is believed that sexual development is not merely a physical process but is influenced by psychological factors. It encompasses multiple aspects such as gender identity, gender-related behavior, body image, and sexual habits [9]. These can be accurately assessed only after puberty which is the earliest when psychosexual maturity is achieved [10]. Studies differ in their reports of the effect of hypospadias on psychosexual development. Some suggest that boys with hypospadias had a more uncertain gender identity and more feminine traits than controls [11–13]. However, contrary results were reported in other series [14] where gender-appropriate behavior was seen in spite of hypospadias. In adolescent boys, it was found that masculine gender role was predicted by younger age at surgery [9].

27.2.1.1 Age at Surgery and Personality

Blotchsky reported that genitourinary surgery in childhood resulted in more emotional disturbances, compared to other forms of surgery [15]. In addition to personal stress, these patients also face the stress of parental anxiety and multiple operations in childhood [6]. Bracka believed that hospitalization between 18 months and 3 years of age was more stressful and preferred to perform a two-stage repair after 3 years of age [16]. However, contemporary reports have shown that adults treated for hypospadias were comfortable with surgery as a child as there was less recollection and mental trauma [3, 17]. About 10% reported decisional regret [17]. Men that were interviewed said that they did not often receive adequate information on follow-up and were reluctant to seek medical attention for problems. They were often anxious about intimate relationships and sexual performance due to which they grew up into reserved adults [13, 17, 18]. This indicates that periodic follow-up with psychological counseling may be indicated [19].

27.2.1.2 Behavior and Relationships

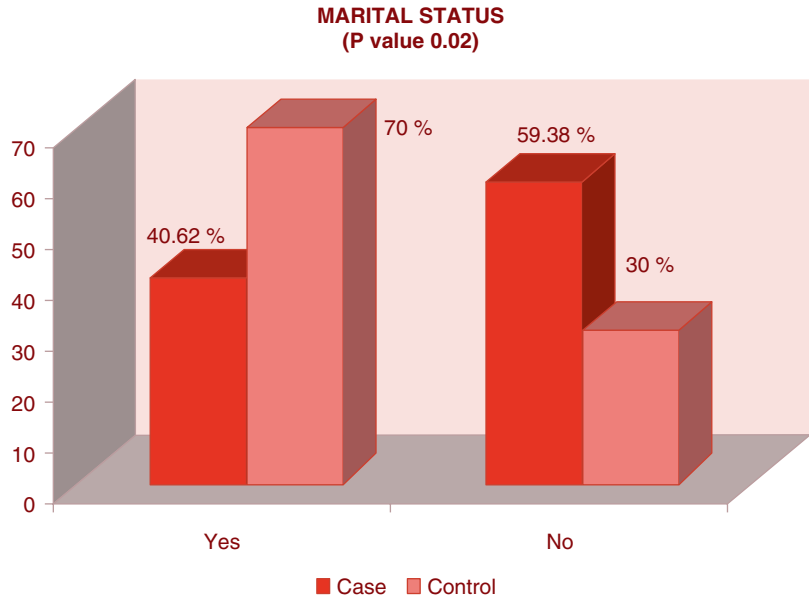
There was a higher prevalence of behavioral problems and worse psychosocial outcomes especially in those with proximal hypospadias,

older age at surgery, and a greater number of operations, in some studies [12, 17, 20, 21]. However, others reported that there was no difference in psychosexual outcome between patients and the general population [5, 6, 8, 22]. In spite of a poorer genital image, the psychosexual satisfaction levels were similar between hypospadiacs and the general population [8]. Their education levels and professional achievement were also similar [6, 19] contrary to a few earlier reports [23]. The rates of stable relationships/marriage and biological children among those with hypospadias were no different from those in the general population in most studies [24, 25]. In a study from India, Bhat et al reported that the rate of marriage was 40.62% in those with hypospadias and 70% in controls (Fig. 27.1). This was higher than those reported by Bubanj (37.23% in cases and 33.33% in controls) [26] and Kenawi (36.6% in those treated for hypospadias) [27]. This may be attributed to earlier marriage in keeping with societal customs in India. The age at marriage was similar in the two groups (26 years in cases vs. 24.71 years in controls).

27.2.1.3 Sexual Activity and Quality of Life

Some authors have reported delayed sexual milestones, fewer partners, and less frequent sexual activity in those with hypospadias [28], especially if the genital image was poor [16, 23, 29–31]. Avellan et al showed that sexual debut was delayed in those who underwent hypospadias repair later than 13 years of age [32]. However, these findings have not been echoed by others. Milestones such as falling in love, erotic kiss, fondling, masturbation, age at ejaculation, and intercourse were not delayed in boys with hypospadias. Sexual behavior including libido, frequency of masturbation, number of partners, and intercourse, were also similar. However, they were more inhibited in seeking sexual contact due to decreased confidence of sexual performance, were reluctant to discuss topics related to sex, and feared the ridicule of genital appearance. This was more prevalent in proximal than distal hypospadias [5, 22, 30, 33, 34]. A trend towards better psychosocial function was seen with better

Fig. 27.1 Rates of marriage in cases and controls [Courtesy Dr. Amilal Bhat]



body/ genital perception, counseling, and higher levels of education [6, 22, 35]. Up to 50% avoided situations involving exposure of genitalia and did not prefer to change clothes in public or use communal showers and washrooms [3, 19, 36]. However, more recent series have reported that the majority of those with hypospadias had a more positive body image and did not fear adverse genital appraisal by a partner [3]. In a study on health-related quality of life (HRQoL) in young adults who had undergone hypospadias repair, it was shown that low genital perception and decreased orgasmic function directly correlated with lower HRQoL. There is a need for timely diagnosis and psychological support in these individuals [37]. Up to 50% of the patients and their parents were anxious about sexual activity and fertility in the future. This further reinforces the need for periodic counseling and prolonged follow-up [17].

27.2.1.4 Objective Assessment of Psychosexual Function

Multiple objective tools for assessment of psychosocial well-being have been used for assessment after hypospadias repair. These include Psychological general well-being index (PGWBI) [13], Minnesota Multiphasic Personality Inventory

Test [22], TNO AZL Child Quality of Life Questionnaire, Child Behaviour Checklist [38], Gender Role Questionnaire [9], Child Behaviour Checklist, Youth Self Report, Self-Perception profile for adolescents [21], Psychological General Well-Being Index, Body-Esteem Scale for Adolescents and Adults [39] and Global Sexual Functioning questionnaire [10]. However, there is a need for a unique validated tool to assess psychosexual outcomes with elements such as genital image, genital ridicule, social function, behavioral problems, self-confidence, inhibition to establishing sexual contact, and the various aspects of sexual behavior.

The salient features of psychosexual development in hypospadias are summarized in Table 27.1.

27.2.2 Genital Perception

27.2.2.1 Associated Anomalies

Congenital anomalies associated with hypospadias include micropenis, penoscrotal transposition, undescended testes, inguinal hernia, ectopic kidney, and disorders of sexual differentiation [28, 40]. These are more prevalent in proximal hypospadias [13]. Reported non-urogenital

Table 27.1 Features of psychosexual development after hypospadias repair

Psychological, scholastic, and professional development is normal after hypospadias repair [14, 18]
There is a reluctance to discuss sexual issues, avoidance of intimate relationships [13, 30]
They are socially withdrawn, inhibited in seeking sexual partners [5, 30]
There is fear of genital ridicule, avoidance undressing in public [3, 19]
Sexual milestones are normal: Erotic kiss, masturbation, ejaculation, intercourse [5, 34]
Proximal hypospadias: Low genital image, delayed sexual milestones [30, 33]
There is a need for prolonged follow-up and periodic counseling [17, 19]

abnormalities are congenital heart disease, musculoskeletal anomalies, anorectal malformation, cleft palate, cerebral palsy, and psychological disorders [33, 40]. These may negatively affect genital perception and this, in turn, has been reported to influence psychological and sexual outcomes [29].

Satisfaction with Cosmesis

Older studies that reported on the cosmetic outcomes of operative techniques such as Denis Browne, Ombrédanne, van der Meulen, Broadbent, and Mustarde which are not in common use today [27, 41]. Most patients had undergone two or more operations in these series. Most (38–80%) recognized that the appearance of the repaired penis differed from normal. This was more pronounced for older techniques such as Denis-Browne, van der Meulen, and Browne dorsal meatoplasty. Cultural differences such as circumcision being less prevalent in the population also contribute to this difference [4]. Patients were often dissatisfied with the appearance of the penis due to scars, fistulae, chordee, penile torsion, flat shape of glans, and redundant distal penile skin. The reported rates of dissatisfaction with cosmesis range from 13 to 53.5% [27, 31] and were higher than that in controls [42]. A third considered their penis to be small and 40–60% wished that they had a larger penis [4]. Objective measure-

ment of penile length showed smaller length and girth of the penis after hypospadias repair when compared to controls [30, 43]. Penile size was the single most important aspect that influenced genital perception [4]. Up to 44% requested revision surgery for poor flow, spraying of stream, fistulae, residual chordee, or poor overall cosmesis [19, 36].

More recent studies reported improved cosmetic outcomes (80–90%) with techniques such as TIP, Mathieu, MAGPI, preputial flap, and staged repairs [4]. In redo operations, the satisfaction rate was 60% [44]. Aho et al reported that dissatisfaction was higher with Denis Browne than Mathieu repair (46% vs 13%) [24]. The use of Testosterone pre-operatively did not affect genital perception or penile length [45]. Those with proximal hypospadias were less satisfied with appearance and size and were likely to be embarrassed by the same. They were also more likely to have poor flow and were thrice as likely to seek corrective surgery [24, 31, 39].

27.2.2.2 Meatal Position

Reports of the influence of the position of the meatus after repair, on body image were inconsistent. Bracka et al reported that more than two-third patients felt that a terminal meatus led to a better body image [19]. However, in other series, patients were not too concerned by a slightly proximal meatus [36] and the position of meatus and chordee did not influence satisfaction [8].

27.2.2.3 Age and Genital Perception

Flynn et al believed that children with a successful operation were satisfied till adolescence after which, growth of the neourethra failed to keep up with that of the penis [41]. However, it is more likely that pre-pubertal children have a more positive genital perception as the penis has not acquired functions other than micturition and that with the onset of sexual activity and heightened body image, adolescents have higher expectations [4]. Even after surgical correction, many patients faced ridicule from peers for the abnormal appearance of the penis [5].

Patients were more satisfied with cosmesis if they had been treated before the age of three, were younger than 12 years of age, and had more knowledge about hypospadias [5, 9]. Those who had complications after hypospadias repair and a greater number of operations were more likely to be dissatisfied [35, 46].

27.2.2.4 Patients Versus Surgeons

There is often a mismatch between the perception of results by patients and surgeons. The patient compares results with his peers whereas the surgeon uses prevailing surgical standards for a particular severity of hypospadias [4]. Mureau et al, in the first such study, showed that there was no correlation between the rates of satisfaction of surgeons and patients. Surgeons tended to rate cosmesis after hypospadias repair higher than their patients [5]. However, other studies showed that that surgeons rated penile appearance as abnormal more frequently than patients or their parents [17, 47].

Validated scores for genital perception include the following:

- Genital perception score [5]
- Hypospadias Objective Score Evaluation [48]
- Pediatric Penile Perception Score and Penile Perception Score [49]
- Hypospadias Objective Penile Evaluation [50]
- Glans Meatus Shaft Score [51]

Objective records of cosmetic outcomes post-op and on follow-up and the use of validated questionnaires may improve the quality of reporting and enable objective comparison [4, 44].

27.2.2.5 Perception of Genitalia by Women and Healthy Men

In a novel study, Ruppen-Greef et al showed 105 women standard photographs of ten adults circumcised and an equal number of repaired hypospadiac genitalia. Women considered general appearance, public hair, and penile skin to be important to cosmesis. Penile length, position, and shape of meatus were not important. Distal hypospadias was graded similar to a circumcised

penis while proximal forms were considered to be abnormal in appearance. Similar results were found in a study with 70 men with normal genitalia where the appearance of distal hypospadias was considered no different from that of a circumcised penis. Older age and higher sexual interest led to more favorable genital perception [52, 53].

27.2.2.6 Untreated Hypospadias

Scholmer et al. reported that untreated hypospadias, especially if proximal led to a more negative genital image, greater difficulty in voiding, and a more pronounced chordee [54].

27.2.2.7 Follow-up

The majority wished that they had a longer follow-up because of reasons such as anxiety arising from poor genital image, prior operations, and their complications and lack of understanding of their problem. There is often a reluctance to seek medical attention due to shame, ignorance, fear, or resignation [19]. This emphasizes the need for long-term periodic follow-up till adulthood in these patients [13].

The salient features of genital perception in hypospadias are summarized in Table 27.2.

Table 27.2 Genital perception in hypospadias

Most are satisfied with cosmesis with TIP, MAGPI, preputial tube [4]
More dissatisfaction seen with older repairs like Denis Browne and Ombrédanne [27, 41]
Genital perception is better in the young [5, 38]
Children face ridicule from peers even after surgical correction [5]
Negative genital perception is associated with disorders of sexual differentiation [29]
Penile length may be important for genital perception [4]
Patients request revision surgery for poor flow, spraying of stream, fistulae, residual chordee, cosmesis [19, 36]
There is a mismatch between genital perception of patients and surgeons [4, 5, 47]
Repaired distal hypospadias and circumcision appear similar for laypersons [52]
Patients are reluctant to seek medical attention due to shame, ignorance, or fear [19]

27.2.3 Erectile Function and Sexual Satisfaction

27.2.3.1 Factors Affecting Intercourse

Most studies on sexual function in hypospadias are on adolescents and data in adults is scarce. They focus on the short-term cosmetic and functional results. However, the repercussions of corrective surgery are felt a decade or two later in adolescence, with the onset of sexual activity. Patients may idealize their performance, be lost to follow-up and those who did not participate in interviews, maybe more dissatisfied with sexual life due to which the results may not be generalizable. Problems with erections in hypospadias may be due to correctable or non-correctable problems. The size of the penis cannot be corrected whereas chordee, scars, and torsion may be corrected [1]. Difficulty with sexual intercourse may be due to residual chordee, painful scars, decreased glans turgidity, or small size of penis [19, 27, 36]. Decreased glans sensation is rare [19].

27.2.3.2 Sexual Behavior

Erectile dysfunction may be seen in up to a third to half of the patients with proximal hypospadias [33, 35]. Libido, ability to have penetrative inter-

course and achieve orgasm were unaffected in most reviews, [19, 27, 55] even after prior failed repairs necessitating staged reconstruction with buccal mucosal grafts [7]. In other published series, there was greater difficulty with erections, intercourse, and ejaculation with decreased satisfaction, especially in proximal hypospadias [13, 26, 31]. In a study from India, Bhat et al observed that the incidence of erectile dysfunction among hypospadiacs was greater than that in controls (Fig. 27.2).

Patients tended to masturbate less, have shorter foreplay, less frequent intercourse, and fewer partners [26]. In contrast, Majstorovic et al showed that desire, arousal, frequency of intercourse, erections, and orgasm was higher in proximal hypospadias than controls [10]. Rynjah et al reported delayed sexual milestones, lower orgasmic function, and weak ejaculation in patients with proximal hypospadias who underwent transverse preputial island tube reconstruction when compared to controls [56]. Problems with achieving orgasm may be due to shame from negative genital perception and lesser arousal [57]. However, it was also noted that the IIEF is inconsistently answered in adolescents and these results need to be interpreted with caution [58].

Fig. 27.2 Shim scores among patients and controls. [Courtesy Dr. Amilal Bhat]

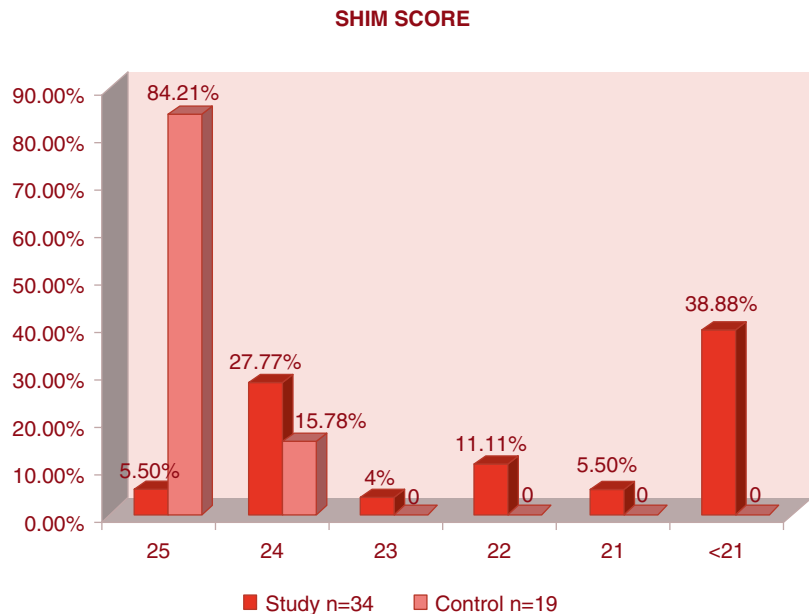
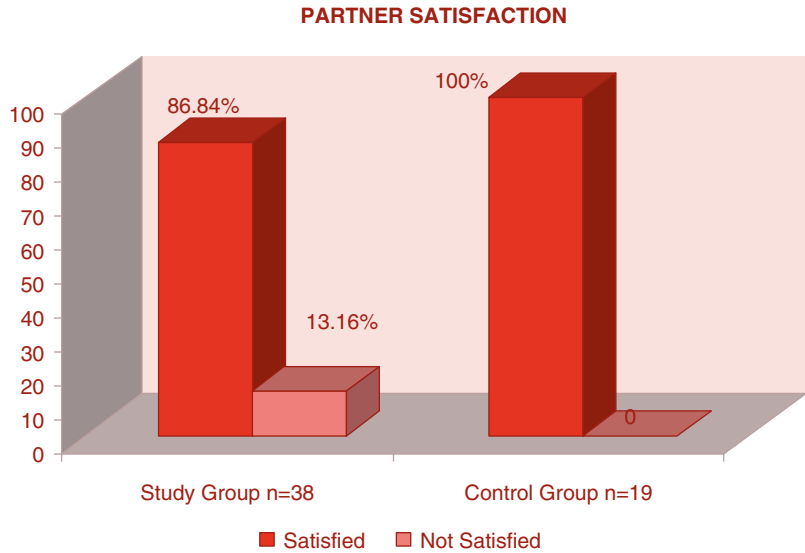


Fig. 27.3 Partner satisfaction among cases and controls. [Courtesy Dr. Amilal Bhat]



The extent of flexibility in the erect penis was more important than the degree of chordee for satisfactory intercourse [19]. A few studies reported that the size of the penis correlated with the ability to have normal intercourse [31, 57] while others showed that the overall cosmesis and not the penile length, determined sexual satisfaction [13]. Those who were dissatisfied with surgical outcomes reported poor genital image and lower sexual satisfaction. Patients, therefore, require long-term follow-up and appropriate counselling starting early in life [24]. The number of operations and occurrence of complications were not reported to influence erectile function, ejaculation, or sexual satisfaction in some series [7, 30, 35]. Bhat et al. reported a higher incidence of partner dissatisfaction among those with hypospadias due to the small size of the penis and poor erections (Fig. 27.3) which emphasizes the need for counselling of the partner in addition to that of the patient.

27.2.3.3 Ejaculation

Most series report dribbling on ejaculation: 33% overall [19] and up to 75% in proximal hypospadias [34], the need to milk out the ejaculate in up to 43% [59] or anejaculation in up to 20% [13], especially in proximal hypospadias [30]. These may be due to inadequate spongiosal and muscular support to the reconstructed skin tube, failed

urethral peristalsis, abnormal bladder neck, large utricle, as well as complications such as stricture/diverticulum [7, 19, 31, 57]. Split ejaculation from urethrocutaneous fistulae, was another unique problem reported in the hypospadias group [17].

27.2.4 Hormonal Status and Fertility

Literature on hormonal levels and fertility in hypospadias is scant [1]. Associated genital anomalies (17–30%) include micropenis, penoscrotal transposition, undescended testes, inguinal hernia, and disorders of sexual differentiation. These are more prevalent in proximal hypospadias and may affect hormonal levels and fertility to varying degrees [40, 60]. Association with undescended testes, disorders of sexual differentiation, and reduced anogenital distance suggest androgen deficiency as a possibility during embryogenesis. Endocrine-disrupting chemicals were shown to influence the development of hypospadias in animals. However, this is not proven in humans. The constellation of hypospadias, undescended testes, subfertility, and testicular cancer is believed to be a result of testicular dysgenesis [61]. Defects in androgen signaling such as decreased receptor concentration and mutation in the second Zinc finger element of the

DNA binding domain, have been described in patients with ambiguous genitalia associated with hypospadias [33].

Testicular volume, Testosterone, and semen parameters were normal unless there were associated undescended testes or chromosomal abnormalities [19, 62]. In those with proximal hypospadias, hypogonadism was noted in 15% [63]. FSH and LH were high while Testosterone and semen quality were low in hypospadiacs when compared to controls [64]. However, FSH was found to be elevated only in the presence of undescended testes in another study. The confounder was that undescended testes were more commonly noted with proximal hypospadias [65]. Dihydrotestosterone was also noted to be lower among those with hypospadias. The presence of raised LH with normal Testosterone may indicate relative androgen insensitivity [66]. The incidence of oligozoospermia in hypospadias ranged from 4 to 29% [19, 27].

Fertility in hypospadias may be affected by multiple factors. These include testicular dysgenesis, mechanical factors that affect semen delivery (stricture, diverticulum, non-terminal meatus, micropenis, lack of spongiosal support resulting in dribbling on ejaculation), and reduced semen quality in proximal hypospadias [67, 68]. Fertility and paternity were both found to be lower in hypospadias, especially if proximal, even when those with undescended testes were excluded [13, 69]. This suggests that hypospadias in isolation, even without hormonal and familial factors, predisposes to lower fertility. Anatomical or surgical factors may be responsible for this finding [69]. In a population register-based study, it was noted that both hypospadias and undescended testes were associated with a 21% decrease in paternity compared to the general population. The decreased paternity rates were higher in proximal hypospadias (54%). Assisted reproduction techniques were used by 0.4% of those with hypospadias which was comparable to the general population [70]. Similarly, Asklund reported a lower paternity rate in hypospadiacs (24%) when compared with the general population (29%) [62]. Patients who underwent reoperation for obstructive causes had lower paternity (0%)

when compared with those who had reoperation for non-obstructive causes and controls (80%) [71]. Bhat et al showed that paternity was similar among hypospadiacs and controls (50% in each group) which was in keeping with other published reports [13, 29].

The salient features of sexual function and fertility in hypospadias are summarized in Table 27.3.

27.3 Conclusions

Long-term follow-up studies after hypospadias repair have several drawbacks. These include their retrospective nature, small sample size, loss to follow-up, use of non-validated questionnaires with the attendant biases, lack of valid controls, and obsolete surgical techniques with late age at repair. Typically, they involved multiple operations with higher complication rates. Currently, most children undergo surgery at a much younger age (1–2 years) and the implications of this are unknown. With the advances in surgical technique, it is expected that the cosmetic, functional, and psychosexual outcomes will also improve. However, after the repair, there is a lag period of more than a decade for the subjects to become sexually active. Proximal hypospadias, especially if associated with undescended testes may be associated with low testosterone levels, decreased semen quality and subfertility [13, 64]. The development and use of validated questionnaires, appro-

Table 27.3 Sexual activity and fertility in hypospadias

Chordee, pain, scars, soft glans, and short penis hinder intercourse [19, 27]
Normal desire, erections, intercourse, and satisfaction seen in distal hypospadias [10]
Erectile dysfunction, difficult and infrequent intercourse is seen in proximal hypospadias [56]
Lower sexual satisfaction if genital perception is poor [24]
Dribbling of ejaculate may be due to poor spongiosal/muscular support or urethral stricture [7, 19]
Testicular volume, testosterone, and semen are normal unless testes are undescended [19, 62]
Fertility and paternity lower in proximal hypospadias [13, 69]

appropriate psychological counseling after corrective surgery, and extended follow-up till adulthood may offer the necessary support to these patients and also improve our understanding of this often neglected aspect of hypospadias [42, 72].

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