

Nonsurgical factors in the success of hypospadias repair

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Accepted for publication 10 January 2005

KEYWORDS

hypospadias, psychology, sexuality, outcome

INTRODUCTION

Hypospadias is a common congenital disorder affecting ≈ 1 in 300 live male births. Classically it is a triad consisting of a malpositioning of the urethral meatus, a ventral curvature of the penis and an abnormal distribution of foreskin, giving a 'hooded' appearance [1]. The resulting defect varies, with 80% having a distal malpositioning of the meatus, requiring a single-stage repair, and 20% having a more severe, proximal malpositioning, which may require two or three operations to repair. The more severe the initial problem the higher the complication rate, and the more operations the patient is likely to require.

There are many reports describing the surgical management of hypospadias. The results may be measured easily in terms of complications. The appearance and the acceptability to the patient are just starting to be considered [2,3]. The difficulty is that the patients are children and it may not be their opinions that are sought, but only those of their parents [2]. Although the many operations described suggest that none is perfect, the results now are sufficiently good that nonsurgical factors, and particularly the psychological aspects, should be considered in assessing the outcomes [4]. However, there is little published that describes what psychological problems need addressing (if any), what interventions have proved useful or how often they are offered.

Adult patients who are now available for outcome analysis had their surgery in the 1970s; much of the evidence cited in the present review comes from surgery that is even older. Techniques have changed and surgical results have improved. However, against this must be set the increase in patient expectations that has come with greater education. In this review, publications

on the nonsurgical aspects of hypospadias repair are reviewed and compared with the practices of surgeons in the UK, as assessed by a questionnaire.

THE TIMING OF SURGERY

Early practice guidelines from the American Academy of Pediatrics recommended that children with hypospadias should be operated on after 4 years of age [5]. It was felt that the parental separation from the child would be detrimental to the child's development, and surgery should best be delayed until after the phase of separation anxiety, thought to end at ≈ 4 years old. This recommendation has changed for two reasons; first, surgical and anaesthetic techniques for infants have improved [6], and second, there was a large move within paediatrics as a whole to encourage parents to stay with their child. This allowed the admission of young children with no increase in anxiety as a result of separation [7]. More recently, hypospadias has been repaired as a day-case procedure, avoiding the concerns of separation anxiety.

Four papers focused on the timing of elective surgery in hypospadias and agreed that an early operation minimizes the potential psychological damage caused by genital surgery [6,8–10]. Surgery when aged 6–15 months avoids five particularly sensitive phases of psychosocial development, the disruption of which is thought to predispose to psychological problems in later life (Table 1) [11]. This period was derived on the basis of theory rather than research, a point noted by the authors. Indeed, little research, least of all prospective or controlled, has been done to verify these theories [12]. Mondaini *et al.* [13] studied 40 hypospadias patients and compared them to over 10 000 unaffected controls; the age at which surgery took place was not associated with abnormal psychological adjustment later in life.

Usefully, the timing of the development of body image also corresponds to the Freudian 'phallic stage' of development and the Oedipal Complex, where the child is supposed to

experience rivalry with the parent of the same sex and develop an attachment with the parent of the opposite sex. The guidelines from the American Academy of Pediatrics now recommend operations for hypospadias before 30 months old, to minimize the psychological impact on body image and gender identity [14].

BODY IMAGE

BEFORE SURGERY

Impending surgery for hypospadias induces anxieties in both parents and children that are not seen in families with other surgical conditions of similar severity. Genital awareness is thought to develop at 3–5 years old, earlier if there are older male siblings [8]. This is when the child begins to widen his social circle in nursery and play-groups, and has the opportunity to compare genitals [9].

In severe hypospadias the genitalia may be ambiguous [15]. If the parents are anxious about the 'maleness' of the child, there may be an adverse affect on body image [9]. A preoperative assessment of children aged 2–6 years with hypospadias found that parents were indeed concerned about the 'maleness' of their child, which mirrored findings in the children themselves, who showed predominantly genital-centred anxieties, as measured on the Robertson Auditory Projective Test [16]. The assessment by Robertson and Walker also compared the anxieties of the parents of children with cleft palate with those who had hypospadias. They found that the parents of the hypospadias group had anxieties based on the future potency of the child, whereas the children themselves, and both the parents and children in the cleft palate group, had anxieties based more on the present operation. The authors felt that the main difference between the groups was the presence of a hidden 'guilty secret' which prevented the parents from discussing the operation within the family.

Ironically, there are many men who have a hypospadiac meatus for which they have

TABLE 1 The ages of main psychosexual milestones in infancy and early childhood

Age	Psychological event	Other high risk periods
Infancy	Maternal bonding	18–24 months 4–6 years
18 months	Rapprochement Stranger anxiety Separation anxiety	
3–4 years	Development of genital body image	
2–7 years	Cognitive development	
5 years	Oedipal/phallic awareness Castration anxiety	

TABLE 2 The number of patients in three age groups with a stretched penile length in different centiles; adapted from [3]

Age group, years (n)	No. of patients		
	<10th centile	Average	>90th centile
9–12 (16)	1	12	3
13–15 (10)	2	8	0
16–18 (7)	4	3	0

never had surgery and of which, in many cases, they are unaware. In a prospective analysis of 500 men presenting for prostate surgery, 65 had a hypospadiac meatus (but no chordee); 60% of patients and 55% of their partners were unaware of the abnormality [17].

AFTER SURGERY

Early operations concentrated on functional correction, believing that 'minor' cosmetic abnormalities, such as a coronal hypospadias, could be ignored. Surgeons felt that the cosmetic appearance was only of concern to the parents, not the patients [18]. Nowadays it is felt that poor cosmetic results are not accepted by patients [1] and have an effect on genital body image [19]. Recent papers specifically assessing patient satisfaction found that the meatal position was important to the patient and was reflected in their level of satisfaction [3,20]. This finding was replicated by a further study which found that the cosmetic appearance, together with satisfactory sexual function, were correlated with overall satisfaction, with micturition being less important [21].

There is little research into what makes a good cosmetic result, although it was reported that the surgeon's view has little correlation with

that of the patient [3]. In that study, the surgeon based a good cosmetic outcome on those variables that were surgically corrected, such as meatal position. Patients tended to place emphasis on other factors that are not currently operable. Mureau *et al.* [3] identified eight features of importance in judging the outcome of hypospadias surgery:

Surgically correctable:

- meatal position
- glans shape
- scars
- scrotum
- general appearance.

Uncorrectable:

- volume of the glans
- penile size
- penile thickness.

Much disagreement among surgeons and patients centres on the uncorrectable features.

Studies of the long-term effects of hypospadias surgery on body image have found that patients were more embarrassed than controls about their penis, and had more

sexual inhibitions as a result [22]. Genital body image in hypospadias patients was negatively correlated with the initial severity of the hypospadias [13,19], and positively with the terminal position of the meatus [20]. Importantly, one study found the better the genital body image, the better the psychological function [22]. Therefore, in children who have had an operation for hypospadias a good cosmetic result is most important for future psycho-sexual functioning. It is not known whether this is because having surgery raises an expectation of a perfect outcome, or whether there is an intrinsic desire to have a perfect penis.

Size may also be a cause of dissatisfaction. The hypospadiac penis is often said to be short. In part this may be because of the circumcised appearance, especially in societies where infant circumcision is unusual. However, where a formal measurement has been made, a fifth of hypospadiac penises were below the 10th centile. The finding was most marked in adolescents, with four of seven being below the 10th centile (Table 2) [3].

Penile size is a source of considerable anxiety in many adolescents. Limited research is available on the relationship of penile size to sexual satisfaction. Men with micropenis and with epispadias report intercourse that is satisfactory to themselves, although the opinions of their partners has not been investigated [23]. An investigation of women with several sexual partners suggested that intercourse with an uncircumcised penis gives greater pleasure than a circumcised one [24].

PSYCHOLOGICAL OUTCOME

EARLY OUTCOME

An early paper assessing boys at 2–6 years old found an increase in initial withdrawal behaviour followed by an increase in aggressive behaviour [25]. This study used a 'unblinded' 'formal observation' approach with no controls, but it replicated the clinical feeling at the time that patients who were operated on at a later stage experienced more distress [8]. Some studies agree with this preliminary research, showing more behavioural problems than in controls [26], more inferiority [12], and an increase in shyness and enuresis [27]. However, two recent studies were unable to show any

emotional or behavioural differences between patients and controls [22,28], although Sandberg *et al.* [28] noted that higher rates of hospital admissions were associated with an increase in emotional problems.

LATE EFFECTS

The studies assessing late effects of hypospadias surgery divide their findings into three groups: psychological, psychosocial and psychosexual. Although there have been several studies evaluating the psychological sequelae of hypospadias, most comprise few patients and have poor controls, often comparing the surgical patient with a community control. Where papers assessed behavioural changes after surgery the sample patients were frequently those who had been operated on when aged 3–6 years, and it could be argued that the resulting behaviour was a consequence of operating during a psychologically vulnerable age. Up to 20% of patients with severe hypospadias and genital ambiguity felt that their psychological well-being had been impaired, and 10% had evidence of mild depression [29].

However, with those less severely affected there is some variation in findings. One study, using open interviews, found that patients who had been operated on for hypospadias showed more neurotic symptoms, e.g. depression and anxiety, used immature defence mechanisms and had poor relationships as a result [27]. In the same patient group, a second study, using Rorschach's test, found that patients who had been operated on for hypospadias had more neurotic symptoms, less self-esteem and less capacity for relations, which was felt to prove the existence of the castration complex [30]. The converse was found in another study, again using an unblinded interview, which found no psychological problems in adulthood [31].

Research into psychosocial factors is similarly divided. Those studies which have found psychosocial deficits showed that some patients who have been operated on for hypospadias have less capacity to form relationships or felt that their relationships were affected by the disorder [20,27]. One study found that the marriage rate was 20% lower than in a community population [32]. In addition, one study suggested that adults who had had a hypospadias repair were employed in less competitive and less

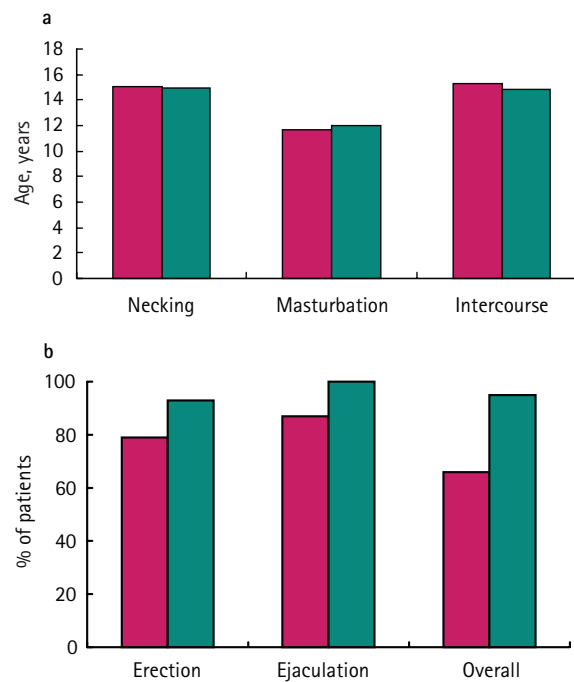


FIG. 1.
a, The age of sexual milestones in men operated in childhood for hypospadias (red bars) compared with age-matched men operated for hernia (green bars). Data adapted from [19]; **b**, The percentage of men operated in childhood for hypospadias (red bars) compared with age-matched men operated for therapeutic circumcision (green bars) who were satisfied with aspects of sexuality. The data were collected in Finland, where circumcision in infancy is only for medical indications. Adapted from [33].

responsible jobs than surgical controls [27]. There are several other studies which have not found such a correlation, and suggest that the hypospadias patient is psychosocially unimpaired in the long term [19,22,31,33].

Psychosexual research has also given rise to conflicting results. Mureau *et al.* [19] assessed sexual inhibition in men operated in childhood for hypospadias and age-matched men operated for hernia; 24% and 2%, 61% and 71% and 15% and 28%, respectively, were considered 'inhibited', 'not inhibited' or 'don't know'. In other reports, up to 19% of hypospadias patients had more sexual difficulties and inhibitions, and anticipated more ridicule, than surgical controls. They were older at the first sexual intercourse and they had fewer partners as a result [20,34].

However, importantly in the study of Bracka [20], those men who were satisfied with their penile appearance had a near-normal age of sexual debut (15.6 years), the delay being mainly in those who were dissatisfied (debut at 19.0 years). Even in those who were dissatisfied, sexual debut frequently occurred before the 'final' surgery was completed. In contrast, some studies found that patients had a normal adult sex life (Fig. 1a,b) [19,31,33,34].

Some studies have focused on the development of general and gender-role behaviour. An early study reported that compared with matched controls, patients with hypospadias were less secure in their maleness and had a tendency to take a more feminine sex role, although had similar sexual orientation to controls [35]. Other research groups reported more behavioural problems and lower social competency than in nonclinical controls, but found better adjustment than a psychiatric clinical control group. Boys with hypospadias showed more cross-gender behaviour than the psychiatric controls [26], although this was associated with hospitalization rather than severity of the condition. In a larger sample the same research team subsequently reported fewer men exhibiting masculine behaviour, although once again the number of hospitalizations for surgery were correlated with gender-atypical behaviour rather than severity of the hypospadias [36]. The significance of the data in that study is very dependent on the selection of the subjects. Apparently the reasoning for the gender-atypical behaviour was the hypo-androgenization associated with hypospadias rather than the surgery itself. In most patients with hypospadias the pituitary and testicular axis is normal, and so the presence of hypo-androgenization must imply some selection bias [20].

FIG. 2. An artificial erection showing persistent chordee in a man operated for hypospadias in childhood.



FIG. 3. Clinical photograph of the penis of a man who had undergone several operations in childhood for hypospadias. He has the features of a 'hypospadias cripple', with an eccentric, patulous and misplaced meatus, extensive scarring and no normal skin.



RISK FACTORS

Several risk factors for a poor psychological outcome can be conjectured:

- Surgery outside the optimum age bracket.
- Severity of the hypospadias.
- Number of operations.
- Child's/parent's unfavourable view of hypospadias.

Most have not been rigorously tested and rely on psychodynamic theories. Probably the most important are the severity of the hypospadias and the number of operations needed for correction. These two factors are understandably difficult to differentiate, as the one will frequently lead to the other. Poor surgical results (Figs 2 and 3) are associated with a delayed age of sexual debut.

TREATMENTS

Most authors cited have agreed that psychological support would be beneficial for this group of patients. This rarely occurs and one study found that two-thirds of patients had received no guidance, 60% stating that they had never even heard of the term 'hypospadias'! [20]. Studies, especially of severely affected patients, emphasize 'the importance of follow-up in adolescence and adult life with adequate counselling when necessary' [29]. However, to date there are no studies that have assessed what therapies are useful, for how long they should run, or which patients are suitable; all are questions worth investigating.

One case study reported a prepubescent boy who was described as having subjectively

experienced the appropriate hypospadias surgery as repeated abuse, and had subsequently become a sexual offender [37]. A cognitive behavioural model was used to break a pattern of offending. The authors argue that a psychodynamic model would have been more appropriate if the boy had engaged with the treatment consistently. Treatment for his offending failed, although earlier treatment (implying during surgery and before offending) may have altered the outcome. The difficulty with this view is that the huge majority of men born with hypospadias are not criminals and therapy aimed at preventing such behaviour in all patients would not be practical or useful.

Work has been carried out in clients with eating disorders or body dysmorphic disorder, and shows some success in challenging and changing distorted views on body image using a cognitive-behavioural model [38,39]. This is an area of potential development for the psychological treatment of hypospadias. However, again it would not be practical to provide all hypospadias patients with individual cognitive therapy. It would be useful to establish the risk factors for a poor psychological outcome and so be able to target psychological treatments effectively.

It is also reasonable to question whether psychological treatment for all patients would be beneficial. Bracka [20] suggested that all children should have regular follow-up to identify surgical and psychological problems at an early stage. Possibly a regular visit to hospital for an inspection of the genitalia (which the child may think are normal) might generate a psychological problem that previously had not existed. Work in our unit with adolescents born with exstrophy has suggested that patients wish to be considered normal, and that facilities provided by adults to help in fact serve only to emphasize the abnormalities from which they wish to escape [40].

SURVEY

The psychological factors in hypospadias might be considered sufficiently well documented that surgeons would be aware of them and adjust their practices accordingly. After all, the operations are, in many cases, cosmetic in intent, so that the psychological outcome ought to be very important [4];

sadly, at least in the UK, this appears not to be the case.

In a mailing to 193 urology centres (71% response rate) we identified 34 surgeons in 30 hospitals who regularly carried out hypospadias surgery. Ten were paediatric urologists, 20 were general urologists and four were plastic surgeons. The mean age at which the hypospadias was repaired was strongly related to the speciality and experience of the surgeon, at 21, 36 and 40 months (range 12–120) for the three groups of surgeons, respectively; and 21, 30 and 51 months for those with a case load of more than seven, two to four, or 'rarely' each year. Paediatric surgeons and those with a greater case load usually operated within the optimum age range. Most surgeons apparently were aware of the relevant psychological factors, but those who operated late misinterpreted the data which were cited as a reason to justify their practice. Some surgeons who operated late were under the illusion that the penis grows significantly between 18 and 36 months.

Of the surgeons, 60% had no concerns about psychological problems in the children under their care. Many of those who did have such concerns thought that the immediate peri-operative period was the most important, often more for the parents than the child. The support used was mainly from nurses and play therapists; only 18% of current support was from a psychologist (or similar psychosocial professional). When it was realized that psychological help was needed, 80% had an idea about how to refer, with 56% referring to a paediatric psychiatrist or psychologist.

The opinions expressed did not always seem to be based on the medical evidence, and were sometimes self-contradictory. Some surgeons felt that having the child in nappies was helpful, while others thought it to be a disadvantage. Penile size and anaesthetic risk were used to support surgery at all ages. The contradictions were even more marked when surgeons were citing the psychological reasons for their choice. It would appear that surgeons use an often spurious psychological reason to support their personal prejudice.

There appeared to be little support for the notion that all children had psychological problems as a result of hypospadias and its surgery. Of more concern, the views did not

always accord with the available evidence. Where surgeons did have concerns, most made referrals only when a specific problem occurred.

CONCLUSIONS

Which nonsurgical factors are important in successful hypospadias treatment? There is good evidence that a better surgical result does produce a happier adult. However, there is also evidence that the patients and surgeons do not agree on that which constitutes a good result. For the surgeon, successful relocation of the meatus and correcting chordee may constitute success. For the patient with hypospadias, the goal is a normal penis. This is a significant challenge when some of the features of hypospadias are not surgically correctable, with a particular difficulty in the increasing number of societies which do not routinely circumcise.

Although psychological evidence is limited, several consistent findings suggest that in the long term some patients show a greater tendency towards low mood and low self-esteem. Whilst patients are later in initiating intercourse and feel that their relationships have been affected (often expecting more ridicule from their partners) most ultimately experience a satisfying sexual relationship. However, importantly, most of these studies were focused on patients who would have been operated at the standard age (i.e. 3–6 years) which is different from current accepted practice.

Several risk factors for a poorer psychological outcome can be conjectured. These include the severity of hypospadias, the timing of the operation and individual ways of dealing with hypospadias, e.g. negative cognitive schema associated with physical appearance. In our survey, practising surgeons identified the main psychological issues as parental anxiety, anticipatory anxiety and some anxieties about future sexual function. However, so far there is little rigorous evidence that these really are risk factors for postoperative psychological distress.

Given these hypotheses, it should be possible to consider carefully that which surgeons could be doing to facilitate improved psychosocial outcomes in both the short- and long-term. It is not clear at what point or what type of psychological support would be

most helpful or to whom it should be offered. This is an area that needs further research.

Although we still lack proof that surgery is less traumatic, in the broadest sense, in infants aged < 30 months, the theoretical basis for early surgery is sound. Those surgeons, particularly those with no specialist practice, should pay attention to the established psychological aspects and operate within the recommended age limits.

CONFLICT OF INTEREST

None declared.

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