24 Management of Penoscrotal Transposition with or without Hypospadias

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24.1 Introduction

The surgical strategy in severe hypospadias and penoscrotal transposition aims to achieve orthoplasty (correction of chordee), urethroplasty, spongioplasty, glanuloplasty, meatoplasty, scrotoplasty, and skin coverage with circumcision or prepucial reconstruction. Penoscrotal transposition is a rare anomaly. Broman described the first case of penoscrotal transposition in 1911 and the accurate description of the proper anatomical relationship between the penis and the scrotum. Classically, the scrotum is viewed as being improperly positioned in reference to the penis. The penis appears to arise from the centre of the scrotum or is enveloped by scrotal tissues in less severe forms. The penis is positioned behind the scrotum in the complete transposition. Many a times, it may be associated with severe hypospadias with varying degrees of transposition. The presentation can often be variable ranging from location of the penis through bifid scrotum to the most severe variant where the penis emerges through the perineum [1]. Scrotoplasty in these cases depends on the severity of its transposition. It can be done in a single stage or three stages with correction of hypospadias or as a separate surgical procedure [2].

24.1.1 Definitions

Scrotum is located in a cephalic/dorsal position with respect to the penis in complete penoscrotal transposition. A less severe form is defined by a bifid scrotum where the two halves of the scrotum meet above the penis. A minor degree of scrotal tissue transposition is called a shawl scrotum (doughnut scrotum). As scrotal tissue is above the root of the penis, it could be bilaterally symmetrical on both sides of the penile shaft or unilaterally. Regression of the scrotum caudally, ending with a vast distance between penis and scrotum, is a new entity described recently [3].

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24.1.2 Incidence

True incidence is not known as significant transposition is reported as case reports only, and the less severe cases go unreported. National Institute of Health in the US reported less than 200,000 cases of major and minor cases in the office registry of rare congenital diseases. The recently reported incidence was about 0.7% in the patients referred for phimosis and other congenital diseases [3]. These reports show that more attention needs to be paid to diagnosis and reporting this anomaly to know the exact incidence.

24.2 Embryology

The embryological basis of penoscrotal transposition is not fully elucidated. The aspect of sexual differentiation and the mechanism controlling the position of genitalia are still poorly understood. The external position of male gonads represents one of the most important differences between both sexes. The labioscrotal swellings migrate inferiomedially and fuse in the midline caudal to the penis during normal development in a male embryo under the influence of dihydrotestosterone. The abnormal positioning of the genital tubercle in relation to the labioscrotal swellings may result in partially or wholly failed migration of labioscrotal folds caudally during the 4th–5th weeks of gestation. Intrinsically associated abnormality affects the development of the corporal bodies and the urethral groove and folds. That explains the frequent occurrence of the other genital abnormalities like hypospadias.

It has also been suggested that scrotal anomalies like penoscrotal transposition may result from early division and/or abnormal migration of the labioscrotal swelling. Labioscrotal folds fail to migrate caudally or migrate, and they remain fused anterior or lateral to the genital tubercle leading to a different grade of penoscrotal transposition. Lamm and Kaplan suggested that abnormal migration or unilateral failure might result in the ectopic scrotum or unilateral penoscrotal transposition. Early division of labioscrotal swelling and subsequent abnormal migration might also result in an accessory scrotum [4].

There is evidence of 5-alpha-reductase type 2 deficiency in penoscrotal transposition. The 5-alpha reductase type 2 deficiency being an autosomal recessive sex-limited condition, prevents conversion testosterone to dihydrotestosterone. Some of the authors believe that gubernaculum abnormality is the underlying cause for the development of this anomaly. A genetic basis for this abnormality with deletion of a critical region on chromosome 13 has been seen in these patients. A familial basis is reported in about 13% of cases, with an X-linked inheritance pattern in few cases [5]. Though the condition may present as an isolated anomaly, it is often associated with various genitourinary abnormalities like chordee, hypospadias, anorectal malformations, hernias, renal agenesis, or dysplasia. Hypospadias and chordee are seen in about 80% of cases [6]. Parida et al. had noted significant renal anomalies in the form of agenesis, horseshoe kidney, ectopic and dysplastic kidney, obstructive uropathy and hydronephrosis [7]. The penoscrotal transposition has been reported in association with VACTERL anomalies [8]. Gastrointestinal abnormalities, craniofacial, and central nervous system abnormalities are often seen in about 30% of cases. Mental retardation and growth deficiency have been noticed in 60% of patients. About 20% of patients have underlying cardiovascular abnormalities [8]. Sometimes the diagnosis can present a challenge, especially during intrauterine life when abnormal appearances of the external genitalia are diagnosed on routine prenatal ultrasonography. The appearance may also have superimposing features resembling pseudohermaphroditism, penoscrotal hypospadias, micropenis, and intrauterine penile amputation. Cases have been reported showing isolated renal dysplastic anomalies [1].

24.3 Classification

1. **Congenital**: Congenital disability with complete penoscrotal transposition to minor defect.

2. **Acquired** (secondary severe infection and scarring): Extensive scarring and debridement after Fournier's gangrene may pull the penis below the scrotum causing penoscrotal transposition. Repositioning of such scrotum is more complicated and challenging than congenital penoscrotal transposition.
3. **Iatrogenic**: Sometimes penis may have to be located below the scrotum to bridge the long urethral defect and achieve continuity of the urethra. Sometimes it is a post-circumcision webbed penis.

24.4 Congenital

Fahmy [9] proposed a new classification for these congenital penoscrotal transpositional anomalies:

Penoscrotal transportation is divided into.

1. (A)■ Incomplete.

■ Complete (extreme).
(B) Minor, which is subdivided into:
■ Bilateral (symmetrical).
* Unilateral.

2. Central scrotalization of the median raphe.
3. Wide penoscrotal distance or caudal penoscrotal transposition.

24.5 Central Scrotalization

The scrotum is attached to the penis by a longer-than-usual stretch of skin, and the webbing of the scrotum varies from mild scrotalization to a completely buried penis. The theory of penoscrotal transposition is the scrotal skin migration towards the ventral penile area. The penoscrotal angle loss may cause sexual problems during adulthood for these children. Abnormal genital appearance is the cause of anxiety in these children and their families. Usually, the diagnosis is made early, but sometimes it is delayed to adult age, and they present with urinary stream abnormalities or genital pain and dysfunction. This is classified as:

24.5.1 I. Primary Webbing [10]

A. Simple

Grade 1 web extends up to proximal 1/3 of shaft of the penis (Fig. 24.1a).

Grade 2 web extend sup to the middle 1/3 of the shaft of the penis (Fig. 24.1b).

Grade 3 web extends up to distal 1/3 of the shaft of the penis (Fig. 24.1c).

Grade 4 Buried penis when the penis remains hidden and embedded under the suprapubic area, this condition is called a buried penis (Fig. 24.1d).

B. Compound

Type 1. Web with pre-penile scrotum.
Type 2. Web with penile curvature.
Type 3. Broad web.

24.5.2 II. Secondary Webbed Penis

Post-circumcision in obese children or concealed penis.

24.6 New classification

We classified the penoscrotal transposition and graded it with scrotal location to the penis.

A. Cranial

This can be graded according to the severity into

Grade S. Scrotalization of penis mild, moderate, and severe (Fig. 24.1a–d).
Grade 1. The scrotum covers the penis superiorly, and only there is no bifurcation of the scrotum (Fig. 24.2a–c).
Grade 2. Scrotum covering of the penis with the partial bifurcation of the scrotum (Fig. 24.2d–h).
Grade 3. Complete bifurcation of the scrotum (Fig. 24.2i).

Fig. 24.1 Grading of penoscrotal webbing. (**a**) Grade 1. (**b**) Grade 2. (**c**) Grade 3. (**d**) Grade 4

Grade 4. Complete bifurcation of the scrotum with superiorization of the scrotum (Fig. 24.2j, k).

Grade 5. No bifurcation of the scrotum but complete superiorization of the scrotum (Fig. 24.2l–n).

B. Caudal

Grade 1 Fig. 24.3a.
Grade 2 Fig. 24.3b.
Grade 3 Fig. 24.3c, d.

Fig. 24.2 Showing grading of penoscrotal transposition cranial. (**a**). mild to (**n**). most severe

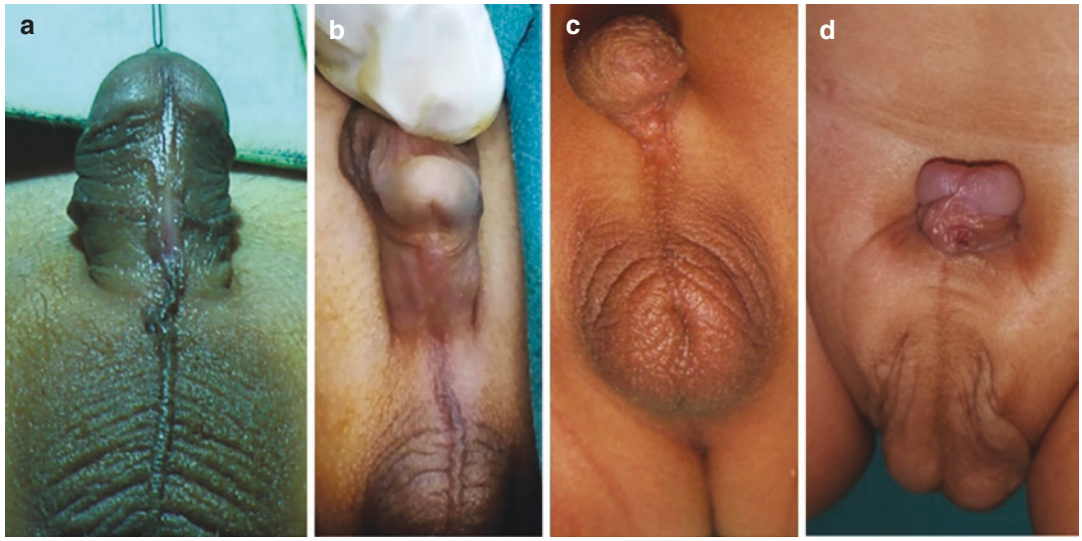

Fig. 24.3 Grading of penoscrotal transposition caudal

24.7 Management

The management of penoscrotal transposition may be divided into.

24.7.1 I. Surgery for Penoscrotal Web

Simple minor degree (Grade 1&2) of penile webbing may not require surgery. But severe and complex penoscrotal webbing may require surgery**.** Unfortunately, there is no uniform opinion and universal consensus on the timing of surgery. Still, it is crucial to perform reconstructive surgery for penoscrotal web before achieving gender identity, so surgery is recommended at the same age as hypospadias repair (6–18 months).

Surgical approach: The surgical correction objective is to reconstruct a normal-looking scrotum with an adequate penoscrotal angle in a single or two stage, depending on the severity. Various scrotoplasty to achieve these goals are:

Heineke–Mikulicz principle: A transverse incision is made centred at the expected penoscrotal angle. The horizontal incision must not be extended too laterally as it may cause a resultant narrowing at the base of the penis. Then the skin flaps are sutured a two-layer vertically, closing the defect, including subcutaneous sutures before the skin closure.

V-Y Scrotoplasty. The V-Y scrotoplasty technique allows tissues lengthening by an inverted V-shaped flap. The centre of the V should be located at the maximal skin tension on the ventral shaft, and the Y leg—ends at the new penoscrotal angle. Traction sutures and skin hooks are used to facilitate the design and raise of the skin flaps. The created defect is closed by forming a 'Y' configuration in two layers with a deep dermal plication stitch to relieve excess tension.

Z-Scrotoplasty: A vertical incision is given proximal to penoscrotal junction, and two wings of the Z are created, having the parallel angles 60 degrees on the proximal and distal ends of the incision. The two flaps created by these three incisions are approximated in such a way as to elongate the coverage over the ventral shaft and to create the penoscrotal angle. All three incisions lengths should be equal to avoid asymmetry from twisting penile skin. Wide based flaps are developed to maintain the blood supply of the flap and prevent flap necrosis. Multiple Z-plasty incisions may be needed sequentially to augment the repair in severe cases, and excess skin of the flaps raised may be resected along the median raphe.

Hanna and Bonitz [11] reported the results of these three techniques in different grades of the non-circumcised webbed penis with acceptable results. The complications rates were 5.3% in the Heineke–Mikulicz scrotoplasty, 7.8% in the V-Y technique, and 2.9% in Z-plasty patients while Negm and Nagla 2020 [12] had a complication rate of 4.54% in Heineke–Mikulicz patients and 11.1% in multiple Z-plasty. Another technique was described by Chen et al. [13] for webbed penis correction in adults using a longitudinal median incision and the separation of the scrotal and ventral penile dartos with longitudinal closure. However, the technique has a limitation in that it only corrects the web at the dartos fascia level, so it does not resolve the problem if there is deficient ventral skin.

24.7.2 II. Management of Penoscrotal Transposition (PST)

The management should start with a multidisciplinary approach taking care of other associated anomalies and parents' involvement in the treatment plan. Early treatment is ideal for preventing the psychological trauma to the child and parents, but this should not be done at the cost of evading through evaluation protocol. Patients with severe penoscrotal transposition should be evaluated for other associated VACTERL anomalies. A staged repair is often needed if the pathology is associated with hypospadias or other anorectal malformations. Objectives in complete PST management are locating the penis in normal position, chordee correction, urethroplasty, glanuloplasty, and scrotoplasty. There may not be hypospadias in the severe form of penoscrotal transposition, but the urethra is usually short, so these patients also require urethroplasty with scrotal correction transposition. Repair may be staged or can be done in a

single stage, provided the goals mentioned above are achieved. The repair may be staged in two or three in perineal hypospadias and bifid scrotum cases. Orthoplasty is done in the first stage, followed by urethroplasty or scrotoplasty in the second, and scrotoplasty or urethroplasty in the third stage. In two-stage orthoplasty and/or scrotoplasty in the first stage, and urethroplasty glanuloplasty and scrotoplasty in the second stage. The principle to be followed in all types of scrotoplasty is to preserve the blood supply and lymphatics of the penis to prevent complications like penile skin necrosis and lymphoedema.

24.8 Surgical technique:

The common surgical method includes rotating two scrotal flaps, joining them in the midline, and vertical skin closure. Other complex surgical procedures require reorienting the scrotum inferiorly with limited rotation flaps, inguinal based groin flaps, and transposition of the penis superiorly in a planned two-stage approach. The urethroplasty and transposition of the scrotum can be done along with the Koyanagi procedure and tubularized incised plate urethroplasty in a single-stage procedure. The two-stage repair is preferred in severe proximal hypospadias with penoscrotal transposition due to its lower complication rates. Prior reconstruction of transposition may give more satisfying results since the scrotum is placed in its proper anatomical position before urethroplasty. The penile shaft also becomes more prominent with an acceptable appearance cosmetically that, in turn, affects the outcome after hypospadias surgery. Sometimes the dimpling of the skin in the midline of the scrotum may occur, resulting in suboptimal aesthetic results.

24.8.1 Modified Glenn–Anderson Technique

The incisions are given around the root of the penis on both sides to elevate the two halves of the rotational scrotal flaps keeping the dorsal penile skin connected to the mons pubis skin. It is ensured that the designed incisions do not meet in the midline, leaving a bridge of skin about 5–10 mm separating the two incisions and connecting the penile skin to the skin of mons pubis (Fig. 24.4a). No circular incision is given around the root of the penis as in other techniques, and penile skin remains connected and drained to the mons pubis skin. Dartos based two scrotal wings are thus created, mobilizing the flaps. The scrotal raphe is incised along approximately half its length, leaving the mobilized urethra entirely free. Tubularized incised plate urethroplasty/flap procedure is done for hypospadias (Fig. 24.4b). It is ensured to have a complete mobilization and fixation of the testes in all cases. The two scrotal wings are rotated inferior-medially and sutured with 4–0 absorbable sutures (Fig. 24.4c, d). A complete closure of the scrotum in layers, shows almost normal appearing scrotum (Fig. 24.4e).

The technique can be used even in two-stage urethroplasty. An inverted U-shaped incision is given, and skin is mobilized to prepare the midline skin urethral plate (Fig. 24.5a, b, c). The tubularization of the urethral plate is done to reconstruct the neourethra and then covered with dartos fascia. Scrotal flaps are raised sparing dorsal midline penile skin (Fig. 24.5d) and penile skin flaps are sutured together to complete the scrotoplasty, and penile skin closure is done (Fig. 24.5e, f). Continuity of the dorsal penile skin is maintained with the suprapubic skin to prevent oedema (Fig. 24.5f).

24.8.2 Single-stage Correction of Hypospadias and Scrotoplasty

Scrotoplasty is feasible with hypospadias repair in a single stage. Hypospadias repair is decided according to the hypospadias type, severity of the curvature, urethral plate, spongiosum development, and the urethral plate's width. The penoscrotal defect may be incomplete in penoscrotal and scrotal hypospadias and complete in perineal hypospadias (Figs. 24.6a and 24.8a). Scrotoplasty can be done after tubularized urethral plate urethroplasty in scrotal and perineal hypospadias and the penoscrotal defect. And in the skin

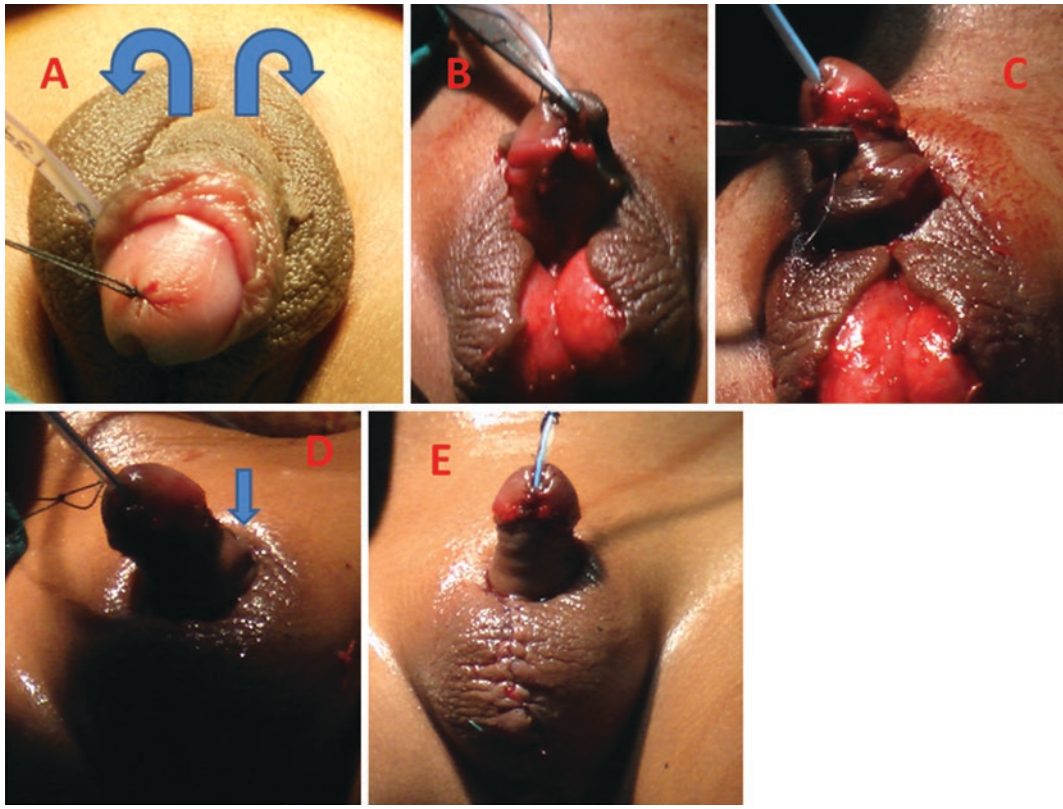

Fig. 24.4 Scrotoplasty in penoscrotal hypospadias. (**a**) Penoscrotal hypospadias lines showing the incision leaving midline skin attached to the penile shaft for raising the scrotal flaps; (**b**) Inner preputial flap urethroplasty completed; (**c**) Glanuloplasty and penile skin closure; and (**d** and **e**). Scrotoplasty completed and an arrow showing the continuity skin to the penile shaft

flap urethroplasty, penile degloving is done, and curvature is corrected (Fig. 24.6b, c). The inner preputial flap is marked with stay sutures, and the flap is mobilized (Fig. 24.6d, e). The inner preputial flap is tubularized to reconstruct the neourethra (Figs. 24.6f and 24.8b, c). Then the urethroplasty and glanuloplasty are done (Fig. 24.7a). After urethral plate tubularization with spongioplasty in TIPU and neourethral reconstruction in flap urethroplasty, scrotal wings are raised, and midline skin over the scrotum is excised. Both scrotal sacs are sutured in the midline to create the midline septum. Then skin closure is done starting from the perineum to the penoscrotal junction. The final reconstructed scrotum shows an almost normal appearance (Fig. 24.7b). Scrotoplasty is feasible even in perineal and perineoscrotal hypospadias (Fig. 24.8a–d). Scrotoplasty should be done in incised plate tubularized urethroplasty in scrotal or perineoscrotal hypospadias. Prepucioplasty can also be done with scrotoplasty in such cases (Fig. 24.9a, b). A partial or complete penoscrotal defect may often be seen in mid-penile hypospadias and proximal penile hypospadias. Scrotoplasty is feasible in single-stage flap urethroplasty in such cases (Fig. 24.10a, b).

Fig. 24.5 Showing scrotoplasty in second-stage urethroplasty. (**a**). Perineoscrotal meatus. (**b** & **c**). Tubularization of the midline skin urethral plate. (**d**). Mobilization of the scrotal flaps. (**e** & **f**) Completed Scrotoplasty with arrow continuity of dorsal penile skin

24.9 Discussion

An ideal approach for proximal hypospadias and bifid scrotum still remains to be decided. Commonly such complex, severe cases are treated in a multistage repair. A few techniques are described in the literature for two-stage repair. The objective of managing penoscrotal transposition with hypospadias or de-novo is creating phallus in the normal position with the urethra at the tip of the glans. The aim of the techniques and their various modifications are to preserve the blood and lymphatics supply of the penis and closure of scrotum cosmetically in the midline. Surgical management has come a long way. Though Broman described the first case of penoscrotal transposition in 1911, details of the case were not mentioned. Appleby published the first detailed description of it in 1923 [14]. Initial repairs were based on creating scrotal-based rotational flaps. Mcilvoy and Harris [15] first performed surgery for scrotal transposition and moved the penis in more cranial position by creating a subcutaneous tunnel. This was followed by an era of scrotal-based flaps popularized by Forshall and Rickham [16], wherein scrotal flaps

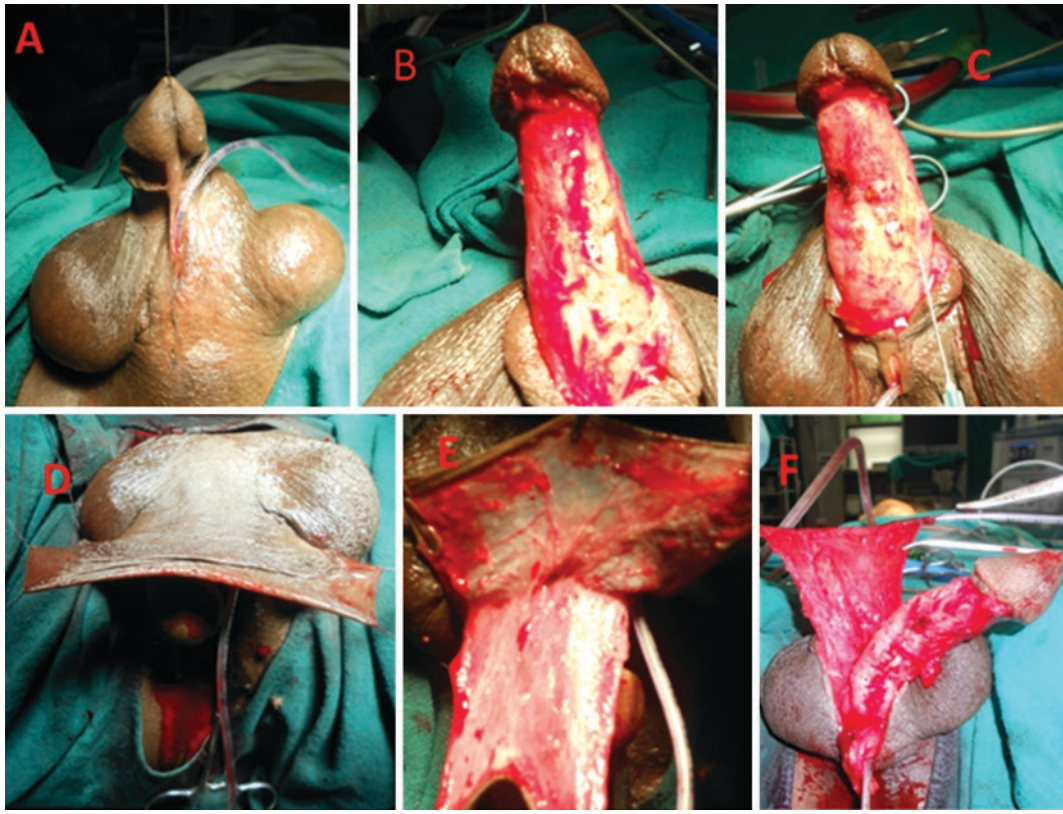

Fig. 24.6 (**a**–**f**) Scrotoplasty in scrotal hypospadias with bifid scrotum. (**a**) Scrotal hypospadias with bifid scrotum; (**b**) Degloving giving circumferential circumcoronal incision; (**c**) Complete correction of chordee on Gittes test; (**d**, **e**). Raising of the inner preputial flap; (**f**). Tubularized inner preputial flap to reconstruct the neourethra

are moved medially and caudally. Glenn and Anderson also used the same method. Dresner [17] later modified the technique in 1982 and is common in practice as modified Glenn and Anderson technique. Kolligian et al. [18] corrected it by transferring the penis after creating a buttonhole in the skin of the pubic area. However, the cosmetic results were suboptimal, as this correction was associated with chordee due to the non-releasing of soft tissue bands. Of late, based on the principle of preserving blood supply, various types of repairs based on release incisions have been in vogue like M-plasty or W-plasty with acceptable results in different reported case series. When associated with hypospadias, as seen in many patients, a tubularized urethral plate urethroplasty is the most common repair done in a single stage or sometimes in two stages. Lately, Bhat et al. [19] reported single-stage urethroplasty in both flap and TIPU with scrotoplasty in severe hypospadias. Koyanagi [20] repair has been another option in these patients with associ-

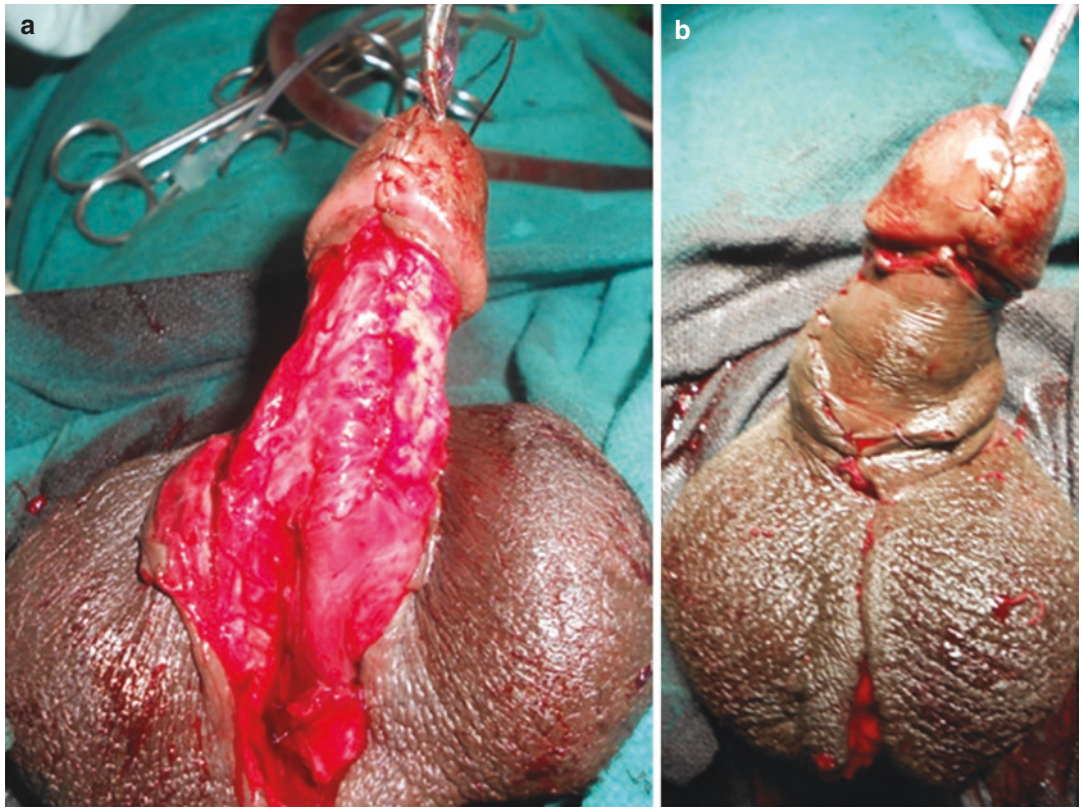

Fig. 24.7 (**a, b**) Urethroplasty and scrotoplasty

ated hypospadias. The modifications in scrotal skin closure are Z-plasty, multiple Z-plasties, V-Y-based rotational flaps, and Singapore flap repairs. Bladder mucosa or buccal mucosa has also been used during urethroplasty with satisfying results. Z-plasty involves creating two triangular flaps of equal dimensions created using an angle of 60 degrees which theoretically can lengthen a contracted scar by about 75%. These flaps are transposed to improve the functional and cosmetic appearance of scars.

Lately, pudendal-based thigh flap repairs have been used in cases with deficient penile skin and tethering of penile skin with quite satisfying cosmetic and functional results.

Complications after surgery include urethral fistula, flap necrosis, penile oedema, and rarely a testicular injury. Though Glenn–Anderson [21] popularized the circular incision at the root of the penis, it is likely to compromise lymphatic drainage leading to lymphoedema partially. In addition, the patient's transposition correction by the Glenn–Anderson technique witnessed gross oedema that persists for long periods. Even after resolution it leaves the penile skin dusky and darkly pigmented scrotal skin.

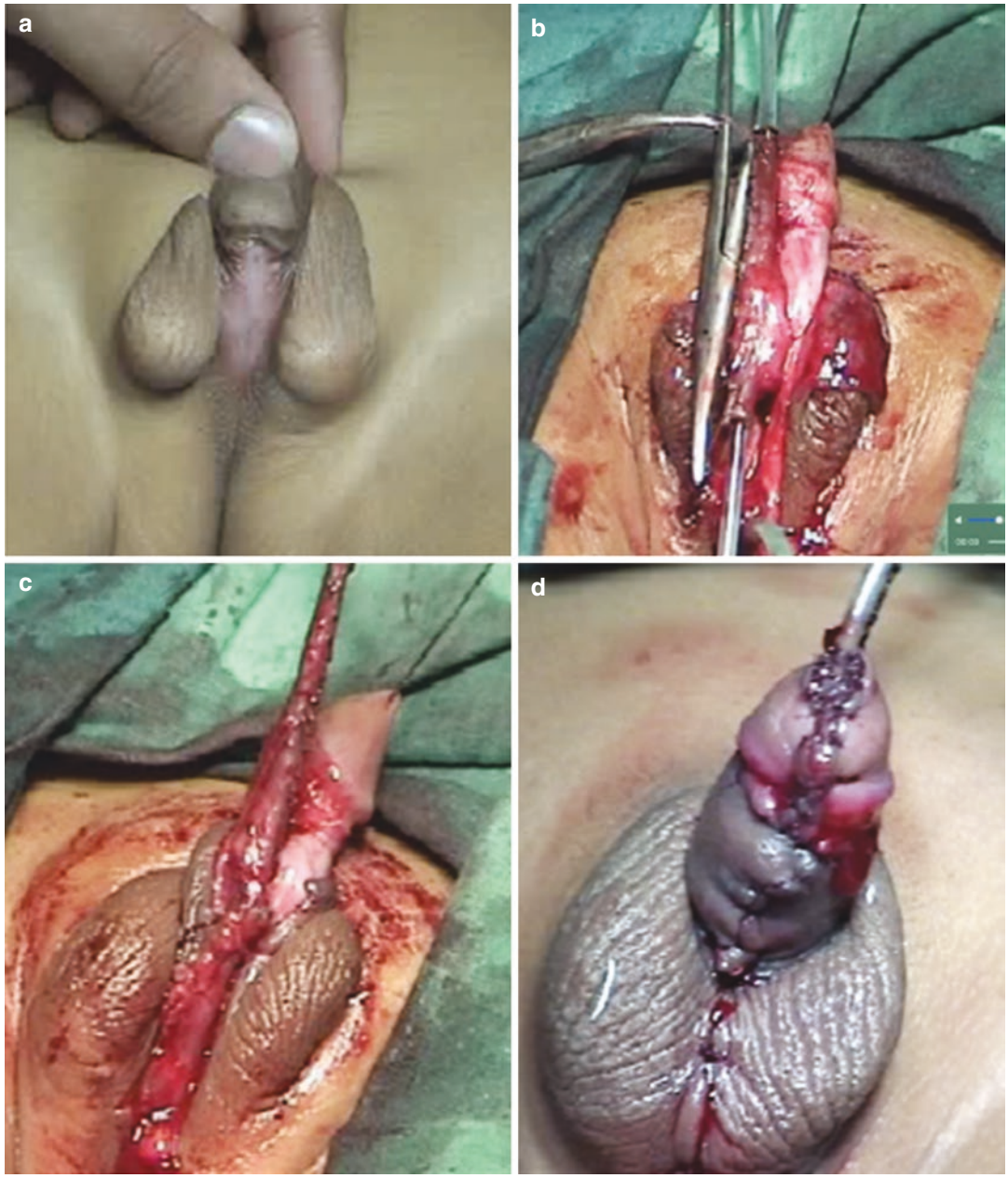

Fig. 24.8 Scrotoplasty in perineoscrotal hypospadias with bifid scrotum. (**a**) Perineoscrotal hypospadias with bifid scrotum; (**b** & **c**) Tubularized inner preputial flap to reconstruct the neourethra. (**d**). Scrotoplasty

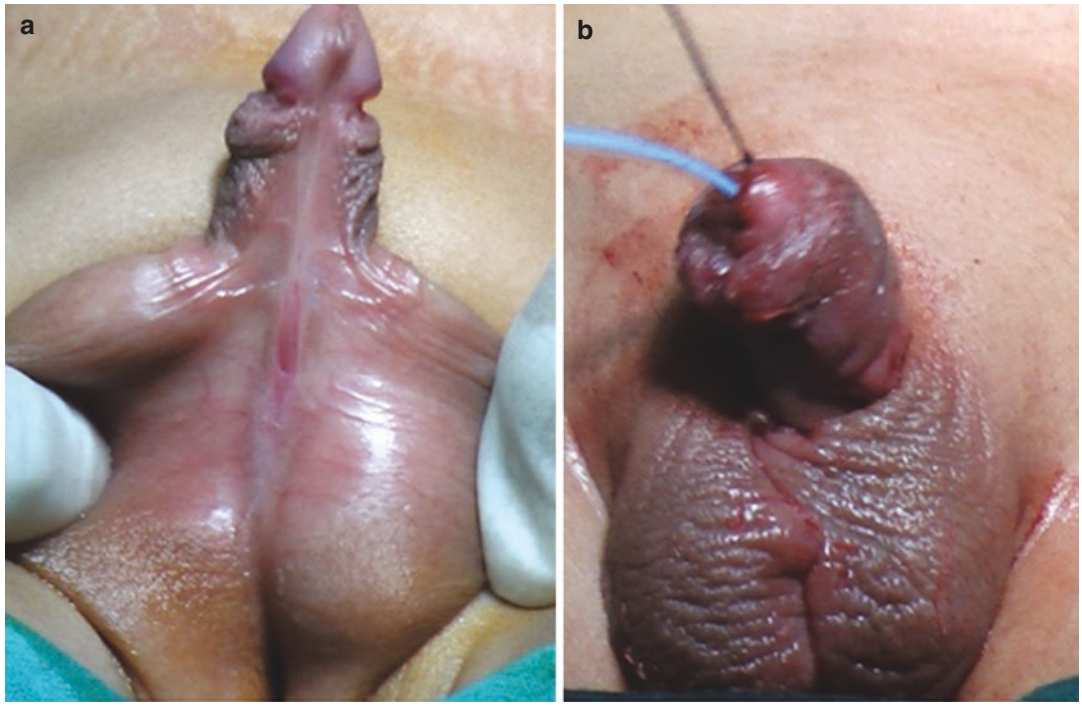

Fig. 24.9 (**a** & **b**) Scrotoplasty and prepucioplasty in scrotal hypospadias with incomplete bifurcation

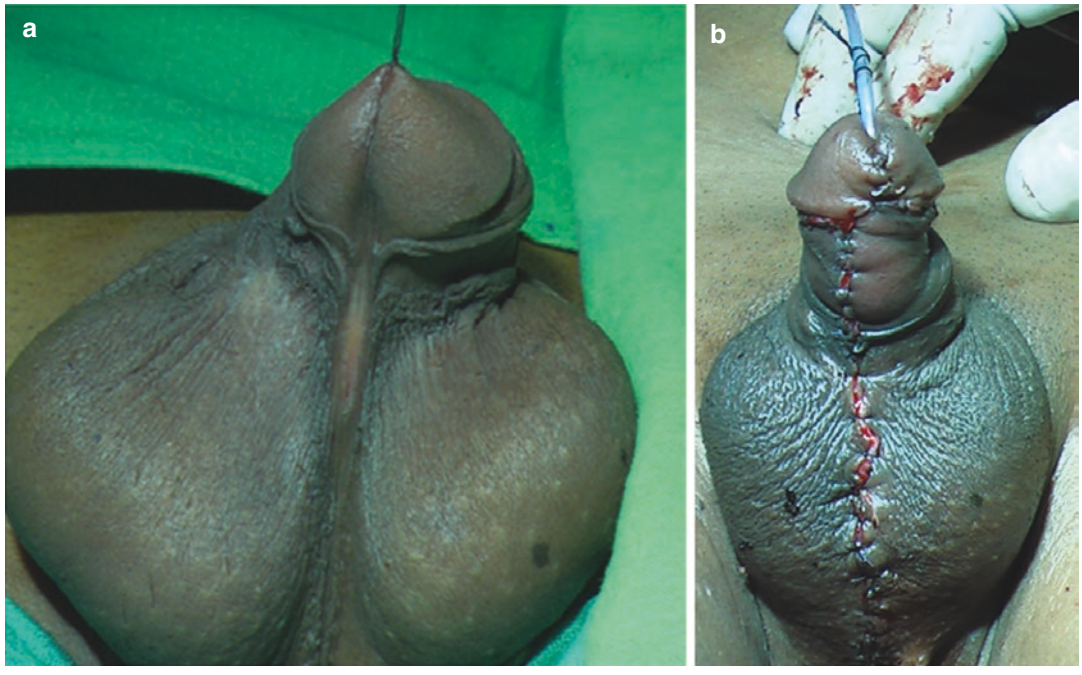

Fig. 24.10 (**a** & **b**) Scrotoplasty in scrotal hypospadias with bifid scrotum

24.10 Conclusion

Penoscrotal transposition is a rare anomaly characterized by the scrotum's positional aberration in relation to penis and is often associated with hypospadias. The scrotal defect may be cranial or caudal in varying degrees and partial or complete. Minor blemishes of scrotalization and perineal migration may not require treatment. Scrotoplasty is an integral part of hypospadias repair in modern-day Hypospadiology, which is often missed. It may be combined with single-stage hypospadias repair and the first or second stage in two hypospadias repair stages. Modified Glen Anderson technique is the most familiar technique, but it may compromise lymphatic drainage leading to lymphoedema. So, the blood supply and lymphatics of the penis should be preserved to prevent these complications.

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