

**Clinical Study** 

# Phlebectasia and Phlebothrombosis of the Penile Veins in Children after Non-Medical Circumcision

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## Keywords

Male Circumcision Complications of Surgery Penile Phlebectasia Penile Vein malformations

# Abbreviations

 DPV - Dorsal Penile Vein
MC - Male circumcision
PMD - Penile Mondor's Disease
VM - Venous malformations

# Abstract

**Background:** Abnormally enlarged, tortuous and dysplastic penile venous vasculature (phlebectasia) are rare in children. The prevalence of this anomaly and its possible etiologic correlation with circumcision is examined in this study.

**Patients and methods:** This is a prospective comparative cohort study of 830 children, aged 6 months to 12 years, enrolled between June 2021 and December 2023. Among them, 420 were circumcised (group A) and 410 were intact (group B). All of them were physically examined and investigated with Doppler ultrasound for any abnormality of the penile venous vasculature. The two groups were compared for any difference in the incidence of different anomalies of the penile veins and penile deviation or chordee. Data were analyzed by Mann–Whitney U-test and a P-value less than 0.05 was considered significant.

**Results:** Phlebectasia (21/420) and phlebothrombosis (3/420) were more frequently seen in the circumcised group, but they were rare in the intact group (2/410). These venous abnormalities were commonly seen in the dorsum of the penis (15/24). In circumcised patients, the phlebectasia was associated with penile deviation to the left side in 13 cases and a ventral penile chordee in 1 case. But, in uncircumcised children, the prominent dorsal penile vein was not associated with any chordee. An associated dilation of the deep dorsal penile vein was confirmed in 15 of the 24 cases.

**Conclusion:** Circumcision wound and the subsequent scar may increase the incidence of abnormal penile venous vasculature which is commonly associated with penile deviation.

### INTRODUCTION

Unlike many other veins of the human body, penile venous drainage is specially designed. The usual pattern of 'veins outnumbering arteries' is reversed in the penis. In contrast to paired dorsal arteries, dorsal penile veins (DPV) are unpaired and single which are arranged in deep and superficial layers. Superficial DPV starts distally and it mainly drains the skin of the penis and prepuce. This vein, as it courses proximally in the midline within the subcutaneous tissue, receives numerous tributaries from the skin of the penile shaft. Deep DPV lies between the Buck's fascia and the tunica albuginea. It receives drainage from the distal two-thirds of the corpora cavernosa via emissary veins and the corpus spongiosum via circumflex veins.<sup>(1)</sup> The superficial DPV ultimately drains into the left and right superficial external pudendal veins, a tributary of the great saphenous vein. Deep DPV drains into the prostatic plexus. Recently, another small pair of dorsal veins has been found that lie just deep to the deep dorsal vein, but above the tunica albuginea.<sup>(2)</sup> The unique venous anatomy is intended to facilitate penile erection; the engorged corporal tissue compresses penile veins and venules to maintain penile erection.

Variations in the appearance of the superficial veins of the penis are most innocuous and do not require any medical attention. Several factors like age, ethnicity, tumescent state, genital pigmentation and hormonal effects influence the clinical visibility of penile veins. 'Veiny penis' (penile phlebectasia) is the term used to refer to the normally visible, dilated, painless superficial veins of the penis. It is commonly seen in elderly fairskinned men. On the other hand, penile varicose veins are similar to phlebectasia but the veins are torturous. Penile vein thrombosis is yet another painful condition with visibly engorged veins. All the 3 entities closely mimic each other.

In phlebectasia the swollen and twisted veins may occur due to inherent weakness of the venous wall or due to increased pressure within the veins. Although phlebectasia may not pose any serious medical risk, it can cause discomfort during erection and is unsightly.

Male circumcision (MC) is associated with a wide spectrum of complications, which may be early or late. However, abnormal penile venous vasculature as a complication of MC was not reported before. The association between post-circumcision abnormal DPV and penile deviation (lateral chordee) has also not been studied. It appears to be secondary to the distorted elastic penile tissue and altered anatomy that follows MC.<sup>(3)</sup> This paper is intended to examine this unusual complication of MC.

#### PATIENTS AND METHODS

Eight hundred and thirty healthy children with normal penile size were included in the study. Children who had major systemic diseases or intellectual disabilities were excluded. Baby boys below 3 months of age or those above 12 years and those with any un-related penile anomalies like microphallus or webbed penis were also excluded.

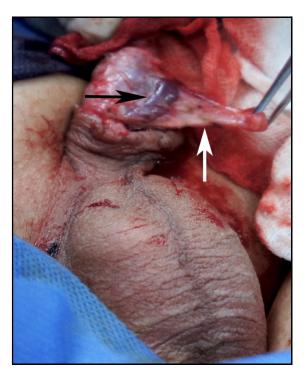
This prospective cohort study was approved by the ethical committee for human experimentation at the authors' universities. Also, this study was carried out following the ethical standards of the hospital's institutional ethics committee on human experimentation. Parents of all study subjects signed a written informed consent for photographing their children and they also consented to the usage of the photographs and data of their children for publication, but with masked identities.

A total of 830 children were examined prospectively between June 2021 and December 2023. They are divided into two groups: Group A consisted of 420 children aged 4 months to 12 years who underwent non-medical MC. They were evaluated at 2 to 5 months (mean 3 months) after non-medical MC. Group B consisted of 410 agematched intact (uncircumcised) children, who sought medical help for unrelated conditions like hernia or hydrocele, but without any detectable congenital anomalies of the penis.

Diagnosing superficial phlebectasia of the penis is mainly clinical. The examination commenced sequentially for the circumcision scar, coronal sulcus, frenular remnants, penile shaft, glans, and urinary meatus. Assessment of the visibly dilated veins was done in both flaccid and erect states. In those who required a surgical operation, erection was induced artificially by intra-corporal injection of saline. Any hardness or tenderness elicited was also recorded. The length and the diameter of visible veins were measured by a digital caliper (Fixtec<sup>™</sup>, Model FHVC0151) with a resolution of 0.01 to 0.1 mm. The degree of penile deviation was measured according to the Sarkis-Sadasivam method<sup>(4)</sup> by using a sterile protractor. Deviation of the meatus from the midline after aligning the protractor with the penile shaft was taken as the reference point for measuring the penile deviation.

Dilation of the DPV was assessed by colored Doppler ultrasound. Primary hyper-coagulability was excluded by investigating the bleeding and coagulation profiles. For all circum-cised children, the age at the time of MC, the technique of operation, the duration of bandage and any detectable bleeding or other early compli-cations were recorded. For those with early post-MC bleeding, the method of hemostasis was also noted.

In children with dilated DPV who deserved surgical intervention (n=21; Group A) the procedure was done through a circumferential or semicircumcision incision. Partial penile degloving (n=8), or more extensive degloving maneuver (n=13) was done to expose serpiginous tortuous superficial DPV which is sandwiched between the skin and the Buck's fascia. The fibrotic tissue of the dysplastic Buck's fascia (Fig.1) that is responsible for lateral deviation was excised and penile alignment was restored with absorbable sutures. The deep dorsal veins were inspected for any dilatation or varicosity, and its recognition was enhanced by an opening made on the Buck's fascia and the corpora cavernosa was milked analogous to the squeezing of a balloon.<sup>(5)</sup> The superficial DPV was stripped thoroughly and ligated with 6-0 nylon suture. The remaining healthy Buck's fascia (recognized as fleshy sheet of fascia) was approximated using undyed 6-0 Vicryl<sup>™</sup> sutures. (Fig.1) All the patients were operated under general anesthesia as day-case. The postoperative course of those who underwent ligation of superficial DPV was uneventful except for minimal lymphatic edema of the preputial remnant in 2 cases and it resolved spontaneously.



**Fig 1.** Prominent dilated dorsal vein (black arrow) along with dissected dysplastic Buck's fascia (white arrow). (NB: The dorsal penile vein appearing on the ventral side is a photographic illusion caused by the rotation of the penile shaft during operative manipulation.)



**Fig 2.** (A) Prominent dorsal penile vein (DPV) in a circumcised 2-year-old boy; (B) Transversely abnormal DPV giving the appearance of a double coronal sulcus; (C) A case of ventral prominent veins; (D) Prominent vein confined only to the inner preputial layer (retracted view)

The venous status and any residual deviation were assessed before skin closure by artificial erection induced by using saline infusion into the corporal bodies. Finally, the wound was approximated while an assistant surgeon consistently stretched the skin of the penile shaft.

Cases diagnosed with phlebothrombosis (n=3; Group A) were managed conservatively for 2 weeks and they had uneventful outcomes. Two cases diagnosed in the control group (n=2; Group B) were simply followed up by reassuring the parents without any therapeutic intervention. The follow-up period of those with abnormally dilated DPV in both groups ranged from 6 to 22 months (mean  $7.7 \pm 4.9$  months; n = 26).

Discrete data were analyzed by Mann–Whitney Utest. Data with continuous values are expressed as mean  $\pm$  standard deviation. Student's paired t-test was applied for inferential purposes after logarithmic transformation. Statistical significance was set at *P* < 0.05.

## RESULTS

In 16 patients of the group-A, the superficial DPV was distended longitudinally in the midline for

about 2cm in length (range 18- 32 mm) reaching the preputial remnant or only confined to the inner preputial layer. Transversely abnormal DPV was rare (n=3; Group A) and it gave an appearance of a 'double coronal sulcus'. Ventral phlebectasia was seen in 2 patients (Fig 2). The overlying skin was tethered and occasionally erythematous.

Penile deviation to the left side was diagnosed in 13 cases and ventral chordee in one case. (Fig 3) An associated suprapubic depression (deficiency of the suspensory ligament) was noted in 3 cases (Fig 3C). Penile phlebothrombosis with a hard, cord-like, thrombosed vein was felt in 3 patients; one of them also had a bluish discoloration of the skin around the dilated vein; in one case it was painful on palpation. (Fig 4)

Detailed history of MC procedure revealed that all the 24 cases of the Group-A were circumcised during the first month of life and the prepuce in all of them was cut by guillotine method using a bone-cutting forceps. Nine cases had thermal cauterization for hemostasis and in 4 cases a monopolar diathermy was used. Post-MC bandage was left *in situ* for 2 days in 12 cases. Post-MC hematoma developed in 4 cases. Post-procedure infections occurring in 5 cases were managed with



**Fig 3.** (*A*) Prominent dorsal penile vein (DPV) with chordee; (B) Prominent DPV with a left sided penile deviation; (C) Prominent DPV with a suprapubic depression.

systemic antibiotics and local ointment. Phlebectasia was recognized by the family after a mean period of 3 months after circumcision (range 2-5 months). All the patients of both groups had normal coagulation profiles. Dilation of the deep DPV was confirmed in 15 of the 24 cases with dilated super-ficial DPV in the group A; but in none of group B.

Statistical analysis showed a significant correlation between circumcision status and abnormally prominent penile veins. Frequency of phlebectasia (5% vs 0.5%; P<0.001), penile deviation (3.57% vs 0%; P<0.004) and phlebothrombosis (0.71% vs 0%; P<0.256) were all more frequent with group-A than with group-B. (Table 1)

#### DISCUSSION

Visibly dilated penile veins are not rare in adults as they are seen in 24% of individuals.<sup>(6)</sup> However, abnormally dilated, tortuous, serpiginous penile veins in children are rare and are scantly reported in the literature. Prominently visible penile veins may be due to phlebectasia, varicosity of the DPV or thrombophlebitis (Mondor's disease of the penis). Acquired phlebothrombosis of the DPV may be due to distorted local anatomy as in circumcision, coagulation disorder or disorders of hemoconcentration (e.g. leukemia). Sometimes it is considered as sequelae of hamartomatous embryonic remnants <sup>(7)</sup> and it may also complicate penile surgeries like circumcision.

Penile phlebectasia appears as asymptomatic, abnormal dilatation and enlargement of the penile veins. When complicated by phlebothrombosis it appears either as a bluish raised and serpiginous tract or as a bluish soft nodule. They may be single or multiple, and the lesions usually measure 0.5 to 1 cm in diameter. The thrombosed area may be tender. The dilated vein usually disappears under slight pressure but refills spontaneously.

of the penis			
Variables	Group A*	Group B*	P-
	(n=420)	(n=410)	value
Dilated DPV	24 (5.5)	2 (0.5)	<0.001
Longitudinal DPV	16 (3.8)	2 (0.5)	0.002
Transverse DPV	3 (0.7)	0 (0.0)	0.256
Dilated VPV	2 (0.5)	0 (0.0)	0.490
Phlebothrombosis	3 (0.7)	0 (0.0)	0.256
Lateral chordee	13 (3.1)	1 (0.2)	0.004
Ventral chordee	1 (0.2)	0 (0.0)	1.000

Table 1: Comparison of venous abnormalities		
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\* Values in parenthesis are percentage

DPV - Dorsal penile vein; VPV - Ventral penile vein



**Fig 4.** (A) Prominent dorsal vein in penile phlebothrombosis; (B) Bluish discoloration associated with thrombosis.



Fig 5. Dialed tortuous vein in an uncircumcised child

Phlebectasia is common in adults and the elderly, possibly secondary to hormonal and physiologic stimuli.<sup>(8)</sup> It has no significant functional impact on erection or intercourse; but may cause problems with body image and cosmesis. Rarely superadded vascular occlusion by thrombosis may result in erythema, bruising or painful swelling. Minor bleeding into the subcutaneous plane may also complicate these lesions.

Penile phlebectasia should be differentiated from rarities like varicose vein of the penis, penile hemangioma, and penile Mondor's disease (PMD). PMD is a rare condition characterized by thrombosis of the superficial DPV.<sup>(9)</sup> It is common in adolescents and is often underdiagnosed. It is characterized by sudden onset; hard, painful, rope like palpable structure on the penile dorsum; and is well appreciable during erection. Penile color duplex ultrasound is an important tool in diagnosing and monitoring this condition. Lack of blood flow and non-compressibility of the dorsal vein indicate thrombosis.<sup>(9)</sup> The PMD is a self-limiting condition. However, anticoagulants such as lowmolecular-weight heparin can be used to treat thrombophlebitis. Persistent thrombus may rarely remain visible as a hyper-echoic linear shadow on the dorsum of the penis.<sup>(10)</sup>

Penile hemangioma may be superficial, deep or combined. They may be in proliferating or involuting phase. Infantile hemangiomas are not inherited but may be formed secondary to mutation in a primitive stem cell which is responsible for the developing blood vessels.<sup>(11)</sup> Venous malformations (VM) are the commonest vascular anomalies and can be found anywhere in the body, but those of the external genitalia are quite rare. Although they may be small, they may cause significant psychological impact and functional impairment. Management of penile hemangioma is usually challenging.<sup>(12)</sup> Owing to rarity, treatment has not been standardized.<sup>(13)</sup> Usually VM slowly worsen over time and never regress spontaneously. They clinically appear as bluish soft vascular masses.<sup>(14)</sup> Most of the VM are often asymptomatic. Sclerotherapy and laser have become viable options for treating small lesions. Although penile VM often relapses or demands multiple treatments, the prognosis is generally favorable. Percutaneous sclerotherapy of penile VM in children under the guidance of digital subtraction angiography is safe and effective, without affecting the cosmetic appearance and function of the penis.<sup>(15)</sup>

Proper clinical examination and history can differentiate abnormally prominent DPV from the other lesions. In this study, penile phlebectasia is characteristically more common on the dorsal aspect and runs longitudinally in the midline of the dorsum of the penis reaching to the preputial remnant of circumcised individuals. Transversely dilated DPV are rare and give the appearance of a double coronal sulcus; ventral dilated veins are extremely rare. Although clinical appearance is enough for diagnosis, colored Doppler ultrasonography may be used for precise understanding of the anatomical and physiological features. Rarely, direct phlebography may be indicated, to confirm the clinical diagnosis. For a more comprehensive study of the venous system, magnetic resonance angiography (MRA) is beneficial, and it may be particularly helpful in PMD.<sup>(16)</sup> However, in our cases, Doppler ultrasound was sufficient for the detection of both the dilated superficial DPV and any anomalies that may affect the deep venous vasculature.

Penile curvature (lateral chordee) and torsion are generally not rare. Its prevalence is 0.5 to 10% during adolescence or young adulthood.<sup>(17)</sup> The exact etiology of this deviation is not well known, but it may be genetic. Pathogenic mechanism of lateral chordee or torsion includes abnormal skin and dartos fascia attachment, asymmetry of the tunica albuginea of the corpora cavernosa, abnormal bony fixation of the corporal bodies or dysplastic Buck's fascia.<sup>(17)</sup>

Male circumcision (MC) is usually practiced for several reasons, such as social, religious, cultural, or rarely for medical indications like phimosis. This unnecessary procedure is largely performed for secular reasons in the Western world, which is sustained by a variety of rationalizations such as aesthetic value. Although MC is considered as one of the oldest and the most common surgical procedures practiced globally, recently the rate of MC has been declining across several countries.<sup>(18)</sup> This declining rate may reflect the changes in demographic patterns and parental beliefs raised by studies in psychology and ontogeny.

In this prospective comparative cohort study of 24 cases of pediatric penile phlebectasia were diagnosed 2-3 months after neonatal MC was done for non-medical reasons. There is significant correlation between MC and the occurrence of penile phlebectasia. Families reported that there had been no such abnormal veins noticed before MC. Even the physician who performed the MC had not noted any pre-operative phlebectasia. Although this could be a subjective correlation, but the high frequency of phlebectasia in circumcision group as compared to the control group is highly significant. New onset penile torsion in 13 cases and chordee in 1 patient in association with abnormal veins in 69% of children is yet another point that supports our hypothesis.

We propose that post-circumcision bleeding and the use of aggressive hemostatic techniques may have a pathogenic link to the onset of post-MC phlebectasia by distorting the Buck's fascia. Histological comparison of the penile scars shows extensive fibrosis, abundant amounts of collagen and absence or paucity of normal smooth muscle cells more often with using electro-coagulation than with using ligatures.<sup>(19)</sup> None of the patients in this series had diabetes, hormonal insufficiency or other sort of trauma except circumcision.

Venous stripping was superior to venous ligation in terms of cosmetic look and it also entailed a careful relief of fibrotic tissues.

Thus far, there have been no scientific studies that support regeneration of penile veins following their surgical excision. Therefore, not only the surgical outcome is promising, but also morphologic complications are minimal.<sup>(3)</sup> When postcircumcision bleeding occurs, it is safer to apply compression or applying fine stitches than using any mode of electro-coagulation. This is essential to avoid any deleterious effect on the penile vasculature; to preserve erectile capability and to avoid infection especially, in the delicate penile tissue of neonates.<sup>(19)</sup> Early detection and management of penile phlebectasia may have a positive impact on affected children not only in terms of general psychological wellbeing but also on their sexual life. Abnormal penile vasculature may contribute to male sexual dysfunction.<sup>(20)</sup>

## CONCLUSION

Post-MC penile phlebectasia with or without phlebothrombosis is infrequently reported. Its actual incidence could be greater than what is accounted. Due to the embarrassing nature of genital symptoms, patients may not be reporting to physicians of dilated veins. Distorted Buck's fascia following circumcision could be the common etiology of both penile phlebectasia and penile curvature.

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