

Congenital Isolated Penile Torsion: A Case Report in 2 Brothers

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Faisal Ahmed¹
Saleh Al-wageeh²
Ebrahim Al-shami¹
Khalil Al-naggar¹
Mohammad Reza Askarpour³
Mohammed Naji⁴

¹Urology Research Center, Al-Thora General Hospital, Department of Urology, Ibb University of Medical Science, Ibb, Yemen; ²Department of General Surgery, Ibb University of Medical Science, Ibb, Yemen; ³Department of Urology, Shiraz University of Medical Sciences, Shiraz, Iran; ⁴Department of Urology, Dar Al-Shifa Hospital, Ibb, Yemen

Abstract: Penile torsion is a congenital anomaly rarely detected in isolation and mostly accompanied by hypospadias and chordee. It could also happen iatrogenically after penile circumcision. In previous literature, the incidence rate of isolated penile torsion has been reported between 1.7 and 27%. The diagnosis and treatment of penile torsion have been illustrated in the previous published articles. However, there are few published studies on the familial predominance of penile torsion. Additionally, the clinical characteristics and potential hereditary components of penile torsion are not well known. Therefore, we studied a 12-year-old boy with a severe 180-degree penile torsion, which surgically corrected and his one-year-old brother, who had mild penile torsion and surgically corrected during circumcision.

Keywords: congenital anomaly, familial, penile torsion, surgery

Plain Language Summary

Penile torsion is a congenital anomaly which is rarely detected in isolation, and mostly accompanying with hypospadias and chordee. The incidence rate of isolated penile torsion has been reported between 1.7 and 27%. based on the degree of glanular angulation, the severity of penile torsion is classified into mild; less than 45 degree, moderate; between 45-90 degree, or severe; more than 90 degrees. There are few published studies focused on the familial predominance of penile torsion. Additionally, the clinical characteristics and potential hereditary components are not well known. We present a severe penile torsion 180 degrees in a 12 years old boy, which surgically corrected and mild penile torsion in one-year old his brother. The surgical procedure includes degloving the penis, Bucks fascia resection, some incision in the base of the penis, plication of tunica, and dorsal dartos flap. Three months after the operation, the patient had a successful correction of penile rotation with no residual rotation.

Introduction

Penile rotation firstly was defined by Horton and Devine in 1973. It is a 3D rotation of corporal bodies of the penis in an abnormal way.¹ Due to limited data on this subject, the actual incidence of this anomaly is unknown; therefore, it may remain undetected until either circumcision or the foreskin retractility. The overall rate of penile torsion was reported between 1.7% to 27% in previous literature when the rotation of corporal bodies is more than 90 degrees; this prevalence rate could decrease to 0.7% of cases.^{2,3} It could occur in isolation or in accompany with hypospadias, chordee, or even after penile circumcision.³ Based on the degree of glanular angulation, the severity of penile torsion is classified into mild; less than <45 degrees, moderate; between 45–90 degrees, or severe; more than 90 degrees.

Correspondence: Faisal Ahmed
Al-Thora General Hospital, Alodine
Street, Ibb 71234-44251, Yemen
Tel/Fax +9674407128
Email fmaaa2006@yahoo.com

However, penile torsion degrees in the majority of cases (87%) are between 10 and 20 degrees.² Cosmetic, functional, and sexual activity are the main reasons for surgical penile corrections.¹ Treatment of penile torsion is a surgical exploration including the development of the penis, Buck's fascia resection, some incision in the base of the penis, and cutting angular ellipses of corporeal tissue followed by plication of tunica, dorsal dartos flap, and suturing the lateral aspect of the corpus cavernosum to the pubic periosteum.⁴ In middle age, the functional outcomes of isolated penile torsion are not completely demarcated. However, in a study, about 12% of men visiting infertility/sexual dysfunction clinics in Egypt had penile torsion.⁵ Considering the small number of studies on potential hereditary component of congenital penile torsion, we presented our case series.

Case Report

Two brothers, aged 1 and 12 years old, were examined by a urologist. Patients' parents were involved with cosmetic and sexual problems of the older child while younger brother needed circumcision. The patients were evaluated physically and in terms of history. Associated anomalies of penile layers such as hypospadias were absent in both cases. However, the older boy had a history of penile torsion at birth.

The first brother was diagnosed with an isolated 180-degree clockwise penile rotation (Figure 1). As previously described, the problem had been surgically corrected (Figure 2). The surgical procedure includes degloving the penis, Buck's fascia resection, some incision in the base of the penis, plication of tunica, and dorsal dartos flap. Three



Figure 1 Showing penile torsion (showing the meatus on dorsal side).



Figure 2 Showing correction of penile torsion.

months after the operation, the patient had a successful correction of penile rotation with no residual rotation. The second brother (younger) was visited and during a physical examination, an isolated 20-degree torsion was detected, which was surgically corrected during circumcision.

Discussion

The difference between penile curvature and penile torsion in the published articles has not clarified so that both terms have been frequently used for both bending of the penis and rotation of the penis. Many authors have referred to the bending of the penis as curvature, while rotation has been often used when there was a torsion on the penile axis.⁵ Moreover, few published reports have recognized a potential hereditary trace in penile rotation etiology, such as Paxson and associations. They have observed congenital torsion of the penis in three neonates with a previous history of penile torsion in their fathers.⁶

Isolated congenital penile torsion might be familial, father to son. There are a few studies reporting this malformation in brothers, such as Gunduz et al as well as the present case report.³ Also, a strong association has been

found between maternal age and parity number and the incidence of penile torsion.⁷ On the other side, the etiology of isolated penile rotation is not known yet.⁸

Several studies have suggested the unequal growth of the corpora cavernosa, eccentric fusion of the ectodermal/endodermal folds, and false attachment of the ectoderm (skin and dartos fascia), as possible etiologies.^{1,8} For example, Bauer et al assumed that the groin area's fetal heel is the mechanism leading to the deformed Buck's fascia. However, this theory could not explain the counter-clockwise torsion.⁸

Rotation more than 60 degrees is rare. The shaft of the penis is usually rotated to the left, in a counter-clockwise; however, despite having a severe torsion, the rotation in our cases was in the clockwise direction.² Although radiologic imaging, such as cavernosography and magnetic resonance imaging (MRI) is highly beneficial to show how the corporal bodies are wrapped around each other,⁹ unfortunately, due to regional conflict, MRI was out of our reach. The reasons for surgery in the penile rotation could be cosmetic, parents' psychological concerns, and functional sexual activity.⁹ However, surgical correction has been clinically indicated only in 4% of patients.¹⁰

The latest available surgical procedures include the release of skin adhesions and realignment of the skin after evaluating torsion severity and resection of Buck's fascia and the tunica albuginea based on the severity.³ However, multiple studies have shown that in the short term, the dorsal dartos flap rotation technique could provide excellent outcomes.^{8,10} For instance, Gunduz and associations performed the degloving and realignment procedure, which is a minimal and noninvasive method compared to other techniques. However, in moderate or severe penile torsion such as our case, this technique could not be strong enough to hold the expanding and rotating force of an erection and may lead to under correction or recurrence.³

Modified Nesbit procedure is another procedure used in severe penile torsion and includes the excision of angled ellipses from the corporal tissue and plication. It is also accompanied by complications, such as neurovascular injury, loss of penile sensitivity, lymphedema, ugly scarring at the base of the penis, penile shortening, and hemorrhage.^{1,10,11}

Bhat et al performed surgery on 116 patients with hypospadias and penile torsion. Penile degloving was per-

formed on 11%, mobilization of the urethral plate and the corpus spongiosum were performed in 28%, mobilization of the proximal urethra was performed in 40%, and mobilization of the urethral plate into the glans was performed in 21% of the cases.⁴

Conclusion

Penile torsion remains a debatable condition since there is no consensus on its complete definition and genetic inheritance. Therefore, we suggest that if one sibling is diagnosed with penile torsion, the other male siblings should be thoroughly examined in this regard.

Consent for Publication

Written informed consent for publication of this case was obtained from the patient parents.

Ethical Consideration

The parental written informed consent was obtained for publication of the case details including the publication of the images.

Institutional Approval

The operation is done following standard protocols and is reported in a way to hide patients' identities completely. Thus, no institutional approval was required.

Disclosure

The authors declare that they have no competing interests.

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