Case Report

Complete Diphallia – Our Technique to Avoid Complications

Vidyanand Deshpande

Diphallia is a rare congenital anomaly with the incidence of 1 in 5–6 million live births. We are reporting a case of complete diphallia associated with accessory scrotum and undescended testis in a 2-year-old boy. We did amputation of the left phallus, urethral end-to-side anastomosis between the spatulated end of proximal left penile urethra and side of proximal part of right penile urethra, excision of accessory scrotum, and left-sided orchidopexy. Avoiding dissection in the posterior urethra leads to an acceptable outcome.

KEYWORDS: Diphallia, orchidopexy, undescended testis

INTRODUCTION

Diphallia is a rare congenital anomaly with the incidence of 1 in 5–6 million live births.[1] We are reporting a case of complete diphallia associated with accessory scrotum and undescended testis in a 2-year-old boy. Our approach in the surgical management of this condition and short review of literature has been described.

CASE REPORT

A 2-year-old male child was referred to us with abnormal genitalia. He had two well-formed penises. The left-sided penis was slightly smaller than the right-sided one [Figure 1a]. The right testis was descended in the scrotum, and the left testis was palpable in the left inguinal area. There was associated small, atretic, empty scrotal sac, lateral and inferior to the left penis. The child was fully continent and passed urine in good streams through both the penises.

Micturating cystourethrogram (MCUG) showed single bladder, no reflux, and two different urethras opening in a single bladder neck [Figure 2a]. Magnetic resonance imaging was suggestive of two complete phalli with two separate posterior urethras.

Preoperatively, cystourethroscopy was done through both the meati, and the findings of MCUG were confirmed. Verumontanum was seen in the right-sided urethra.

We proceeded with surgery and did amputation of the left phallus, urethral end-to-side anastomosis in anterior urethra, and excision of accessory scrotum and left-sided orchidopexy [Figure 1b]. A wide end-to-side anastomosis was done between spatulated end of proximal left penile urethra and side of proximal part of right penile urethra with 6-0 polyglactin sutures. Two, 5 Fr-sized catheters were placed in bladder to drain the urine postoperatively, one through the right urethra and another across the anastomosis. These catheters were removed on the 10th postoperative day and postoperative dye study showed a patent Y-shaped urethra with intact anastomosis in anterior urethra [Figure 2b].

The patient is fully continent after a follow-up of 10 months and passing urine in good stream.

DISCUSSION

Diphallia is a rare congenital anomaly with the incidence of 1 in 5–6 million live births. The first case was reported in 1609. Since then, just over 100 cases have been reported in the literature.[2] Most of the reported cases of diphallia are associated with urogenital and anorectal malformations. Associated urogenital anomalies include hypospadias and epispadias in either or both the phalli, exstrophy bladder, duplication of bladder, and renal agenesis.[3] Caudal duplication syndrome, imperforate anus, duplication, and triplication of colon has also been described.[4]
There are many theories to explain the embryology of diphallus. Explanation of diphallus seems to be either “separation” of the pubic tubercles, in which each phallus has one corporal body and urethra, or “cleavage” of the pubic tubercle in which each phallus has two corporal cavernous bodies and urethras.[9]

Schneider has classified diphallus in the following three groups: diphallus of glans alone, bifid diphallus, and complete diphallus.[9] Vilanova described a fourth category called pseudodiphallia.[3]

Treatment of diphallia consists of excision of one phallus along with its urethra and surgical correction of associated anomalies.[1,5] Kundal et al. described a case of isolated complete diphallia in a 3-year-old boy with two completely separate phalluses, unequal in size, one with hypospadias and the other with normal meatus. It was associated with a soft-tissue mass in the scrotum. They amputated the smaller phallus, did urethral anastomosis and phalloplasty. Outcome in terms of continence and site of urethral anastomosis has not been described.[6]

In another series of six cases of diphallia by Mirshemirani et al., four had associated bladder anomalies. One patient had complete diphallia without bladder involvement and with rectourethral fistula. This case was operated for fistula first and diphallia later. Outcome’ as described was good.[5]

According to Schneider’s classification, our case has complete diphallus. Uniqueness of our case lies in associated anomalies of the accessory scrotum and left-sided palpable undescended testis. We believe that in our case, the position of left phallus prevented the descent of left testis.

We did end-to-side anastomosis between anterior parts of urethras, in the proximal penile part. This gave structurally and functionally acceptable results. Since our dissection did not involve the posterior urethra, there was no risk of loss of continence.

**CONCLUSION**

Diphallia is a rare anomaly, presenting with many variations, and hence, each case needs an individualized approach. Wide end-to-side urethral anastomosis in the anterior urethra (proximal penile part) and avoiding dissection in the posterior urethra leads to an acceptable outcome.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**

1. Torres Medina E, Sanchez Puente JC, Aragon Tovar A. Diphallia, report of one case and review of literature. Rev Mex Urol 2009;69:32-5.
2. Sharma KK, Jain SK, Purohit A. Concealed diphallus, a case report and review of the literature. JIAPS 2000;5:18-21.
3. Bhat HS, Sukumar S, Nair TB, Saheed CS. Successful surgical correction of true diphallia, scrotal duplication, and associated hypospadias. J Pediatr Surg 2006;41:E13-4.
4. Dhua AK, Sinha S, Ratan S, Aggarwal S. Duplication of peno-scroto-testicular unit-A rare form of caudal duplication syndrome. APSP J Case Rep 2013;4:45.
5. Keckler S, Stephany HA, Spilde TL, Snyder CL. Isolated diphallia: Case report and literature review. Eur J Pediatr Surg 2009;19:254-5.
6. Kundal VK, Gajdhar M, Shukla AK, Kundal R. A rare case of isolated complete diphallia and review of the literature. BMJ Case Rep 2013;2013. pii: bcr2012008117.
7. Mirshemirani AR, Sadeghyian N, Mohajerzadeh L, Molayee H, Ghaffari P. Diphallus: Report on six cases and review of the literature. Iran J Pediatr 2010;20:353-7.