

Penile Duplication and Two Anal Openings; Report of a Very Rare Case

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Abstract

Background: Penile duplication (diphallus) is an extremely rare disorder. It is almost always associated with other malformations like double bladder, exstrophy of the cloacae, imperforate anus, duplication of the rectosigmoid and vertebral deformities. Meanwhile anal canal duplication, the most distal and least common duplication of the digestive tube and is a very rare congenital malformation.

Case Presentation: A 21 days old Egyptian neonate is reported with complete penile duplication and two scrotums with each one carrying two palpable testes. Both penises have normal shaft with normally located meatus. Clear urine voids from both meati spontaneously. The child had also a fold of redundant skin about 4×5 cm at the anal region in which two separate anal openings are present. In rectal examination we found two normal anuses passing stool spontaneously. Ascending (voiding) cystourethrography revealed two penises with two separate meatuses and one bladder from which the two urethras go out separately. Intravenous pyelogram (IVP) revealed two normal kidneys and ureters. Barium study revealed duplication of rectum and colon, otherwise normal GIT.

Conclusion: In our review of the literature, we did not come across any other case of this variety of the penile duplication and congenital presence of two anuses. Unfortunately the patient expired before any surgical correction.

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Key Words: Diphallia; Double Anus; Penile Duplication; Malformation

Introduction

Penile duplication is an extremely rare disorder with only approximately 1000 cases of diphallia recorded since the first, reported by Johannes Jacob Wecker in 1609 [1]. This occurs when the baby is born with 2 penises and it is seen in 1 out of 5,000,000 male births. It is extremely rare and

only about 100 cases have been reported up to date. Generally, a child that is born with penile duplication will also have other congenital defects, including spina bifida. Babies born with this condition are at an increased risk of infant death because of the defects and infections that are associated with it. Penile duplication develops around 23-25 days of gestation because the genital

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tubercle fails to fuse properly [2]. Treatment should always be individualized. The malformations that are potentially life-threatening should be solved first [4]. Intestinal anomalies are frequently associated with complete diphallia and imperforate anus [5].

Anal canal duplication, the most distal and least common duplication of the digestive tube, is a very rare congenital malformation [6]. It can be confused with other types of anorectal pathology including hemorrhoids, fistula-in-ano, and perirectal abscess. Anal canal duplications are usually located posteriorly presenting as a 1 to 2 mm perineal opening just behind the anus in the midline [7,8].

Case Presentation

History:

This 21 days old Egyptian neonate with duplication of penis and two separate anal openings was presented to our pediatric outpatient clinic by his parents. The patient is a product of normal vaginal delivery to a young Egyptian couple belonging to same extended family. Mother had a smooth delivery and was not exposed to any harm during pregnancy.

Physical examination:

Weight 4.3 Kg, no dysmorphic features and no associated other congenital anomalies, feeding well. Abdominal, chest and heart examinations normal. There was a true duplication of penis, both equal in size (5 cm in length) with normal located meatuses, both voiding clear urine at the same time. They are attached to two scrotums each carrying two palpable separate normal testes separated from each other by a smooth skin fold of about 2×2 cm free from any skin rouge (Fig. 1). Urine voiding was forcible and passed from both penises at the same time during micturation. It was clear and not mixed with fecal matter, indicating no associated fistulae. There was a 4×5 cm redundant skin fold over the perineum in which two separate anal openings passed feces simultaneously. It was possible to pass two separate thermometers in each anal opening

freely (Fig. 2). Rectal examination revealed two anuses with doubled rectum and descending colon.

Laboratory findings:

All routine laboratory results including hormonal assays were within normal ranges for the age. Abdominal and cranial sonar as well as echocardiography were normal. Voiding cystourethrogram revealed two penises with single bladder. Each penis was attached to the bladder by a separate opening. Intravenous pyelogram (IVP) revealed two normal kidneys and two ureters. No fistulae were detected. Barium meal revealed normal upper GIT pattern. Barium enema revealed double colon, rectum and anus. Urinary and GIT imaging showed no fistulae between the two systems. Chromosomal study: 46 XY.



Fig. 1: Shows two penises and two scrotums



Fig. 2: Two thermometers freely entered into the two anal openings

Discussion

Penile duplication (synonyma: diphallus, bifid penis, penis duplex) is a rare anomaly with an incidence of 1 in 5,500,000. It is almost always associated with other malformations like double bladder, exstrophy of the cloacae, imperforate anus, duplication of the rectosigmoid and vertebral deformities. The duplication may involve whole or only part of the penis. Urine may pass by one or both penises. Mirshemirani A.R [4] in their report found that the scrotum may be normal or bifid, and they report 5 patents had bifid scrotum and only one had normal type. In the other hand Priyadarshi [10] had reported a case of bifid scrotum.

It is suggested that the anomaly results from failure of fusion of mesodermal bands. Incomplete diphallus is treated by excision of the duplicated non communicating glans. Complete diphallus is best treated by excision of the less well developed penile structure and its urethra [9].

Anal canal duplication represents an extremely rare intestinal congenital anomaly of unknown origin [11]. There are not many reports in the English literature, with just a few from each institution [12]. Usually evidenced within 2 years of age, nearly 45% of reported cases present associated malformations such as presacral mass, anorectal malformation and genitourinary anomalies [11]. The treatment of choice in children is complete excision (perineal/posterior sagittal approach), even if asymptomatic. Morbidity is minimal, and outcome is excellent [13].

In the current case we found a true duplication of the penis with normal shaft and normally located meatuses. They voided clear forcible urine simultaneously. They were attached to two scrotums adherent to each other. Both were separated from each other by a 2×2 cm smooth skin fold free from any skin rouge, each scrotum carrying two palpable testes. Two separately functioning anuses opened in a redundant skin fold. Rectal examination and barium study revealed two recti, two colons, two anal openings and normal perineum. Chromosomal study revealed 46 XY male pattern neonate. Koga et al in a retrospective study of anal canal duplication treated from 1988 to 2009 found ten cases, all females [12].

In our exhaustive review of the literature, we did not come across any other case of this variety of the penile duplication and congenital presence of two anuses with duplication of rectum and colon. Association of these anomalies is very rare. We decided to correct the anomaly surgically after 1 year to leave one penis and one anus. Associated anomalies could also be repaired surgically [14]. Unfortunately after five months of meticulous follow up the baby expired from severe gastroenteritis with uncontrolled vomiting and diarrhea.

Conclusion

In our exhaustive review of the literature, we did not come across any other case of this variety of the penile duplication and congenital presence of two anuses. Penile duplication is a rare anomaly. Thorough investigation is mandatory in all cases to reveal associated congenital malformations that can be potentially life threatening and require immediate surgical correction. Treatment should always be individualized according to the degree of penile duplication and the extent of the concomitant anomalies.

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