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Case Report

Double External Genitalia in a 2 days Infant, A so Rare Case: A Case Report

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Abstract

A normally developed scrotum could be associated with ectopic scrotal tissue which may be detected in the perineum, groin, or elsewhere but usually without containing testes within. Congenital scrotal anomalies are uncommon. There are four categories of scrotal anomalies, including penoscrotal transposition, bifid scrotum, accessory scrotum, and ectopic scrotum. Scrotum shows multiple congenital anomalies which could be explained by embryological abnormalities in migration of labioscrotal swelling. Patients with ectopic scrotum should undergo upper urinary tract imaging with ultrasonography. This imaging study showed that our patient had no genitourinary or other anomalies. Accessory scrotum is the presence of scrotal skin that is outside its normal location and does not contain a testicle. It is usually treated by simple excision after verification that it contains no testicle.

Our case was a 2 days old male with double external genitalia that both of them is working and has separated vessels and uretras. We start examination and ultrasonography for anomalies that all of them was normal and schedule surgery for him in 6-12 months years old of his life.

When facing cases of this kinds of cases, physicians should move away from considering surgical excision and biopsy as exclusive first-line management. Instead, we place emphasize and raise awareness about the option of conservative management if imaging shows no abnormalities. Surgery and plastic surgery can avoid from future problems.

Keywords: circumcision; extracorporeal testicular ectopia; accessory scrotumpubic scrotum; genitourinary anomalies

Introduction

Congenital scrotal anomalies are uncommon. There are four categories of scrotal anomalies, including penoscrotal transposition, bifid scrotum, accessory scrotum, and ectopic scrotum [1]. Scrotum shows multiple congenital anomalies which could be explained by embryological abnormalities in migration of labioscrotal swelling [2]. Accessory scrotum is a rare congenital anomaly [3]. There are four categories of scrotal anomalies, including penoscrotal transposition, bifid scrotum, accessory scrotum, and ectopic scrotum [3]. Ectopic scrotum, the anomalous position of one hemiscrotum along the inguinal canal, is extremely uncommon [4]. The location of ectopic scrotum can be suprainguinal, infrainguinal, or perineal, and the suprainguinal type is the most common [5]. Different surgical methods of correcting ectopic

scrotum, such as rotation flap, inverted Y incision, Z-plasty, or excision of the ectopic hemiscrotum, have been used, but none have produced optimal cosmetic results for all types [6].

A normally developed scrotum could be associated with ectopic scrotal tissue which may be detected in the perineum, groin, or elsewhere but usually without containing testes within [7,8].

Case presentation:

Our case was a neonate who had an accessory scrotum with a rare position in the pubic area discovered at delivery. Prenatal and maternal medical histories were unremarkable, and his karyotype was 46 XY. He had no history of urinary difficulties or urinary tract infection. Our case was a 2 days old male with double external genitalia that both of them is working and has separated vessels and uretras (figure1). We start examination and ultrasonography for anomalies that all of them was normal and in this report, we describe color Doppler ultrasound, Ultrasound scan showed moderate hydronephrosis of the left kidney measuring about 19.9 mm with turbidity within. Consequently, voiding cystourethrogram was done showing dilatation of the posterior urethra but with a normal caliber of the anterior urethra excluding the diagnosis of a posterior urethral valve. Additionally, adequate capacity, shouldering, and smooth outline with mild residual urine of bladder were detected Due to the reassuring nature of these findings, a conservative approach was taken, with the patient referred for regular follow-up and schedule surgery for him in 6-12 months years old of his life.

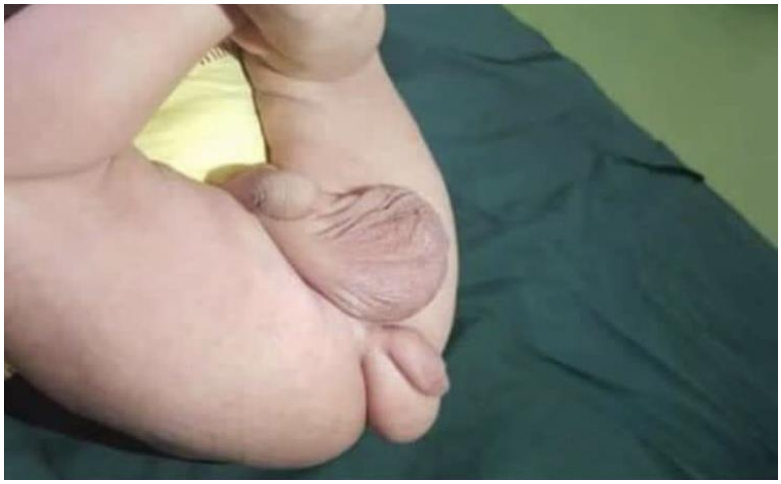

Figure 1: Double external genitalia

Discussion:

Scrotal anomalies which are extremely rare include four types: bifid scrotum, penoscrotal transposition, ectopic scrotum, and accessory scrotum. Over the literature, there are 52 patients of an accessory scrotum. A perineal lipoma was associated in 37 patients (71%), whereas perineal lipoblastoma was detected in three patients (6%) and an isolated accessory scrotum was observed in twelve patients without lipoma or lipoblastoma [10,11]. Accessory scrotum could be associated with other abnormalities as a perineal lipoma, hypospadias, diphallia, defects of scrotal position, anorectal malformation, and the VACTERL (vertebral, anal, cardiac, tracheoesophageal, renal, and limb anomalies) association [12]. Patients with ectopic scrotum should undergo upper urinary tract imaging with ultrasonography [13]. Ectopic scrotum must be differentiated from accessory scrotum. In general, ectopic scrotum contains a testicle and requires repositioning or excising the scrotum with preservation of the testicle [14]. Accessory scrotum is the presence of scrotal skin that is outside its normal location and does not contain a testicle [15]. It is usually treated by simple excision after verification that it contains no testicle [16]. Doppler ultrasound is the preferred method for evaluation, diagnosis and follow-up of this condition, and it can help avoid the need for surgery. Radiologists and urologists and surgeons must be aware of this condition when evaluating any scrotal mass [17]. Surgery and plastic surgery can avoid from future problems [18]. When facing cases of this kinds of cases, physicians should move away from considering surgical excision and biopsy as exclusive first-line management. Instead, we place emphasize and raise awareness about the option of conservative management if imaging shows no abnormalities [19].

Conclusion:

The accessory scrotum can be dilemma before a specialist referral. This mandates early discussion in multidisciplinary team including the relevant specialists, beside radiography to determine the precise anatomy, diagnosis, and any associated anomalies [9]. Patients with ectopic scrotum should undergo upper urinary tract imaging with ultrasonography [13]. This imaging study showed that our patient had no genitourinary or other anomalies. Surgery and plastic surgery can avoid from future problems [18] but when facing cases of this kinds of cases, physicians should move away from considering surgical excision and biopsy as exclusive first-line management. Instead, we place emphasize and raise awareness about the option of conservative management if imaging shows no abnormalities [19]. The awareness of such variation is important for all surgeons whose interest is related to the testicular blood vessels or, generally, blood vessels of the retroperitoneum [20]. Male sex determination is mainly regulated by two testicular hormones. Testosterone, synthesized by Leydig cells, maintains the Wolffian ducts. While AMH secreted by immature Sertoli cells is responsible for the regression of Mullerian ducts [21]. function of the two hormones could lead to various genitourinary malformation, including TTE and PMDS [22].

Declarations:

Ethical Approval and Consent to participate:

The content of this manuscript is in accordance with the declaration of Helsinki for Ethics. No committee approval was required. Oral and written consent to participate was granted by her mother.

Consent for publication:

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal."

Availability of supporting data

It is available.

Competing interests:

The author declares that they have no competing financial interests and nothing to disclose.

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Authors' contributions:

Ahmad Reza Shahraki is the surgeon of patient and writes this paper. Elahe Shahraki collects Data's, Hamide mirshekari and Mohammad Reza Shahraki edit paper and Elham Shahraki reviews paper.

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