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Scrotoschisis: A rare congenital urologic anomaly

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Abstract Scrotoschisis is a congenital extrusion of the testis through a defect in the scrotum. The condition is extremely rare, with only a few cases reported in the literature. We present 2 full term neonates one of whom had bilateral and the other right scrotoschisis and a contralateral communicating hydrocele. They presented at the ages of 7 and 22 days, as referral from rural health centers. They both had successful repair. Testes were in the scrotum after 1 year and 6 months follow-up, respectively. Long term follows up and further evaluations are, therefore, required.

Key words Scrotoschisis; congenital anomaly; testis; scrotum; testis extrusion

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INTRODUCTION

Congenital extrusion of one or both testes outside the scrotum is one of the rarest testicular anomalies; only a few cases have been reported in the literature [1]. There are inadequate data to support any specific etiology, but theories include gubernacular malfunction, scrotal ischemia, or local mesodermal abnormality [2]. No associated

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anomalies were reported in patients with this condition in the literature. We herein present 2 neonates, one with a bilateral scrotoschisis and testicular exstrophy and the other with a right testicular exstrophy and a contralateral hydrocele.

CASE REPORT

CASE 1

The first neonate presented to our accident and emergency unit, at 7 days of age, as a referred case from a rural health center, with bilateral extruded testes since birth.

He was a product of full term unsupervised uneventful pregnancy, delivered via spontaneous vertex delivery at home; no trauma to the genitalia. No consanguinity in the parents.

The patient was a healthy looking baby, weighing 2900g. He was not febrile or pale.

Examination of the external genitalia revealed a completely exstrophied right testes while the left was partially exstrophied. Both were covered by granulation tissue and were extruded through a defect, at the bottom of the scrotum traversing the median raphe (Fig. 1). The defect was lined by fibrous tissue which was attached to the scrotal skin and testicles. There was no evidence of infection. No other anomalies were noted in this patient.

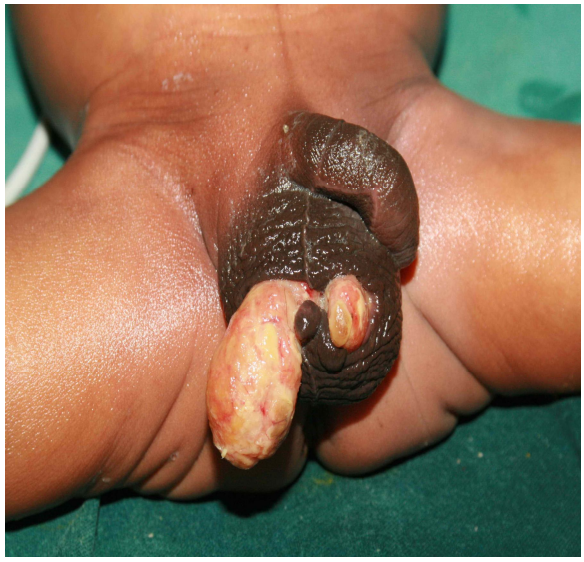

Fig. 1. Exposed testes in a 7 month old neonate.

Full blood count & differentials, urea, creatinine and electrolytes were within normal limits; wound swab microscopy and culture were negative. The testis was fixed to the dartos, after the granulation tissue was lysed which exposed apparently healthy testes. The wound was closed in layers. Post-operative period was uneventful and baby was discharged after 4 days on admission. The patient was followed up for about a year and testis was within the scrotum during this period.

CASE 2

This 22-day old neonate also presented through our emergency unit with right extruded testis noticed at birth. He was also referred from a rural health center and was seen at other peripheral hospitals before

reaching our center, which led to delay in presentation. He was a full term baby; pregnancy was also unsupervised and no significant maternal illness was reported. However, the mother took traditional medications in the second trimester of the pregnancy. Labour and delivery were at home and uneventful. He had been on oral antibiotics and dressing prior to presentation. At presentation, his weight was 3200g, he was afebrile and not pale. The right testis was extruded through a defect in the upper part of the scrotum close to the root of the penis (Fig. 2).

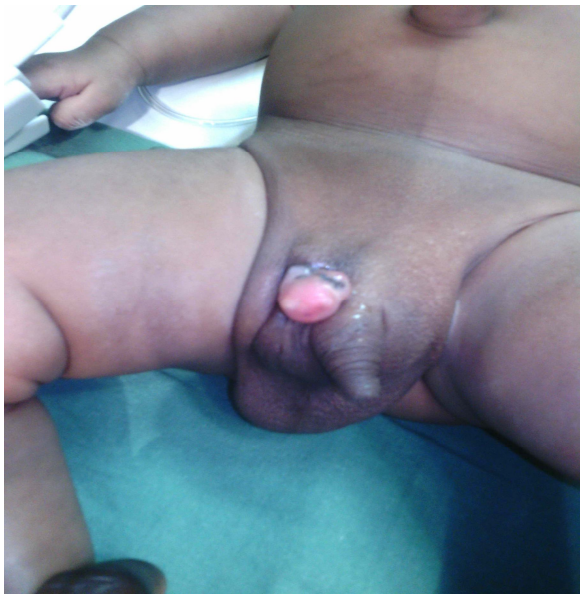

Fig. 2. Exposed right testis in the upper part of the scrotum

The testis was covered by a thick granulation tissue, with minimal purulent discharge. The defect was surrounded by a ring of thick scrotal skin adherent to the testis. The right hemiscrotum was empty, but well developed. The left testis was in the scrotum and a communicating hydrocele was noted on that side.

Laboratory investigations- full blood count & differentials, urea, creatinine and electrolytes - were within normal limits; wound swab microscopy and culture were also negative. The granulation tissue was lysed exposing healthy testis; subdartos pouch orchidopexy was done. The testis was able to reach the bottom of the scrotum in spite of the position of the exstrophy. The wound was closed in layers. The post-operative period was uneventful, and he was discharged after 5 days. The testis was in the scrotum after follow up for about 6 month, after which he was lost to follow up.

DISCUSSION

Scrotoschisis is a congenital defect of the scrotum which is associated with extracorporeal testicular ectopia. In this condition, the testis is in the normal descent pathway, but a defect in the scrotal skin wall causes extrusion of the testis. However, the exact cause of the pathology is still unclear [1]. There are some theories for its etiopathogenesis. A congenital defect disrupts the integrity of the scrotal wall can cause an ectopic extracorporeal position of the testis. In addition to this main reason, however, neither the theory of hyperactive

or mis-directed phagocytic action of gubernaculum testis, nor a localized infective or ischemic process has been demonstrated [2]. Gongaware *et al*. [3] suggested that the failure of differentiation of scrotal mesenchyme leaving a defect where gubernaculum was covered only by a thin layer of epithelium. The lack of supporting structure may results in rupture or avascular necrosis leading to a defect in the scrotal wall. According to Chun *et al*. [4], experimental studies in rats showed that excision of future scrotal skin inhibited gubernacular migration, leading to ipsilateral testicular ectopia. This suggests that a normally developed scrotum is required to guide gubernacular migration before testicular descent. They further suggested that since, in scrotoschisis, the defect is medial; the scrotal skin required for gubernacular descent may arise more laterally or posteriorly.

Additionally, early amnion rupture or the adhesion/band spectrum has been suggested as a possible factor in scrotoschisis. Aberrant amnion bands, strands, or sheets may cause disruption of morphogenesis in the abdominal wall [5]. External mechanical compression due to arthrogryposis is also suggested as a cause of scrotoschisis by Lais *et al*. [6]. At present, the occurrence of meconial periorchitis is the best theory available. According to this theory, the proposed pathophysiology is the late rupture of the scrotal skin secondary to an inflammatory reaction caused by exposure to meconium extruded from an intestinal segment and delivered to the scrotum due to the patent peritoneo-vaginal conduit during fetal life [7]. Meconium residues have been described in the scrotal wall for three of the infants reported and one case of contralateral meconium periorchitis, diagnosed at 4 months of age as a paratesticular calcified mass [5]. In our patients, however, no evidence of residual meconium or meconium peri-orchitis was seen, at presentation or during follow up. Also, the well- developed right hemiscrotum and spermatic cord, in our second patient makes us believe that the defect occurred after testicular descent was completed.

Scrotoschisis does not usually present as an emergency except in case of torsion [8]. However, immediate repair is warranted to avoid these complications. Simple orchidofuniculysis and orchidopexy are satisfactory treatment and the immediate and short term outcome is good, as in our cases [2,9,10]. Follow up to puberty is therefore required to determine the long term outcome.

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