CASE REPORT

Congenital Extrusion of Testis (Scrotoschisis)
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ABSTRACT
A one-day, full term newborn, born to a healthy mother presented with exposed right testicle out of right hemiscrotum since birth. Physical examination showed normal looking testicle and spermatic cord, which was stained with meconium. All baseline investigations and ultrasound of abdomen were within normal limits. There was no visible associated anomaly. Scrotum was explored and viable testis was repositioned. Postoperative recovery was uneventful. At three months follow-up, testicle was good in size and normal in position.

Key Words: Extracorporeal testicular ectopia. Scrotoschisis. Scrotal anomaly.

INTRODUCTION
Malposition of the testis outside of the line of its descent is supposed to be ectopic testis. Testicular ectopia is a well-accepted pathology of abnormal testicular descent.1 Common ectopic site is superficial inguinal pouch, while ectopic testis has also been found at perineum, femoral canal and contralateral inguinocrural region. When ectopic testis is extracorporeal and eviscerated through the wall of scrotum, the condition is labeled as scrotoschisis.2 This is very rare congenital anomaly and only 10 cases have been reported in English literature till now.3 We are reporting this case due to its rarity.

CASE REPORT
A one-day male newborn presented with extrusion of right testicle through right hemiscrotum since birth. He was born of an uneventful pregnancy with spontaneous vaginal delivery at 37 weeks of gestation. On physical examination, there was superficial inflammation around the wound as well as testis. There was a yellowish cystic swelling on surface of testis and greenish yellow tinges over the cord and testis as shown in Figure 1. There was no history of trauma during delivery and no other visible anomaly was seen on physical examination. Ultrasound of abdomen and pelvis was normal. All base line investigations were within the normal limits.

Under general anesthesia, scrotum was explored and viability of testis confirmed by applying hot sponges. Testis was repositioned in sub-dartous pouch after washing with normal saline and excision of cystic swelling and meconium tinted superficial tissues. Postoperative recovery was uneventful. Patient is on regular follow-up and doing well.

DISCUSSION
Testicular development starts along the mesodermal ridge of posterior abdominal wall. Both testes reach at the respective hemiscrotum during the 28 - 40 weeks of gestation. Mechanism of testicular descent is complex phenomenon including the interaction of endocrine, paracrine, growth and mechanical force.

Testis, which descends away from its normal pathway, is considered as ectopic testis, while extracorporeal ectopia is a rare congenital anomaly with normal descent of testis. When a testis extrudes through the scrotum, the condition is called scrotoschisis. The etiology of scrotoschisis is not known but multiple theories have been postulated. Chun et al. suggested that future scrotal skin in fetus helps the gubernaculum

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Figure 1: Right testicle extruded through the scrotum.
as guideline for descent of testis in scrotum. This suggests that any defect in future scrotal skin may lead to scrotoschisis. He further mentioned that defect in scrotoschisis is medial, while scrotal skin required for gubernaculum descent arises more laterally and posteriorly. Gongaware et al. reported that failure of differentiation of scrotal mesenchyme leaving defect, where gubernaculum was covered by a thin layer of epithelium, lack of supporting structures results in rupture or avascular necrosis leading to scrotal defect. Aberrant amniotic bands, strands or cords may cause disruption in formation of abdominal wall, hence an early amniotic rupture or adhesion is supposed to be a causative factor for scrotoschisis.

Presence of meconium periorchitis in scrotoschisis has been accepted as the best available theory. It has been suggested that late rupture of scrotal skin is secondary to inflammatory reaction caused by meconium, which has been extruded from intestinal rupture during fetal life. The passage of meconium from intestinal rupture to scrotum is through a patent Peritoneal-Vaginal Conduit (PVC). Meconium residue has been reported in four cases; three in scrotal wall and one as contralateral para testicular mass. While in this case, meconium staining was seen on scrotal wall, cord as well as on testis in the form of small cystic swelling.

Anatomical position of patent peritoneal-vaginal conduit (anteromedial to cord and testis) may be related to rupture of scrotal wall craniomedially. Most of cases had no intestinal abnormalities, which can be explained by fetal cicaterization of original intestinal lesion without sequela, while jejunal atresia has been reported in one case. In this case, there was no associated anomaly. Most of the reported cases were normal and healthy with unilateral involvement, which was the same as in this case. Testis in scrotoschisis is unfixed and prone to torsion, which was not in this case but Ameh et al. had reported torsion.

Eviscerated testis should be moistened with normal saline to prevent desiccation and torsion. Operative treatment is scrotal exploration, washing of testis with normal saline, excision of necrotic or meconium staining tissues and fixation of testis in sub-dartous pouch. Immediate result is excellent, while long-term result is awaited.

In conclusion, extracorporeal ectopia is a rare congenital anomaly affecting the normal healthy male. The defect can be repaired with conventional transscrotal orchidopexy with good prognosis.

REFERENCES