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Epididymo-orchitis in an extremely preterm infant

Scott Morris [a,b,*], Rebecca Reeves [b], Patrick Ziesing [c], Sanjeev Khurana [d]

[a] School of Medicine, Flinders University, Bedford Park, South Australia 5042, Australia
[b] Neonatal Unit, Flinders Medical Centre, Bedford Park, South Australia 5042, Australia
[c] Medical Imaging, Flinders Medical Centre, Bedford Park, South Australia 5042, Australia
[d] Department of Surgical Services, Women's and Children's Hospital, 72 King William Street, North Adelaide 5006, Australia

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ABSTRACT

Epididymo-orchitis (EO) is a rare condition in the neonatal period. An underlying uropathy is variably associated with EO in published cases and more frequently seen in older children. In this case report, a male baby born at 26 weeks gestation had confirmed congenital *Escherichia coli* septicemia. The *E. coli* was sensitive to gentamicin, and he was treated with 10 days of intravenous gentamicin and cefotaxime, with normalization of markers of infection. He did not have a urinary catheter at any stage. He developed recurrent *E. coli* septicemia 19 days after ceasing antibiotics, in association with a tender scrotal swelling. A urine culture could not be obtained prior to commencing intravenous antibiotics. Surgical exploration revealed a right pyocele and a viable right testis. He was treated with 2 weeks of intravenous piperacillin and tazobactam and a further 2 weeks of oral amoxycillin and clavulanic acid. Urological investigations were normal. No further episodes of EO occurred, and follow-up showed normal testicular growth. This case adds weight to existing literature which suggests hematogenous spread as the most frequent cause of neonatal EO, without urinary tract abnormality. EO as cause of recurrent sepsis in the neonate after apparently adequate treatment of antecedent blood born infection is highlighted. Surgical exploration confirmed diagnosis, and evacuation of the pyocele assisted resolution of systemic sepsis and decompressed the testis.

Epididymo-orchitis (EO) is a rare condition in the neonatal period [1,2]. In pre-pubertal children, genitourinary abnormalities characterized by urinary tract infection and a pathological connection between the urinary tract and genital duct system are frequently associated with EO [1]. In the single center retrospective case series of Seigel et al., 8/17 (47%) of children under the age of 11 years presenting with EO over a 10 year period had an underlying uropathy [1]. Seigel et al., also noted an underlying urinary tract abnormality in 3/4 (75%) infants under the age of 1 year including 1 neonate, suggesting that uropathy is more likely to be associated with EO with younger age [1]. Chiang et al., also described 3 cases of EO in infants less than 1 month of age over a 10 year period in a single center retrospective case review, and of these cases 1/3 (33%) had an underlying uropathy and urinary tract infection [2].

However, other published isolated case reports of EO in the neonatal period suggest that uropathy and/or urinary tract infection are uncommon antecedents of EO [3–9]. In the majority of these case reports hematogenous spread is the hypothesized causative mechanism of EO during this period of life [3–9]. The number of reported cases of neonatal EO is however very small, and hence information regarding pathogenesis and the frequency of associated uropathy in this age group is limited. Consequently recommendations for surgical management of acute presentations and subsequent urological investigation are necessarily extrapolated from experience in older children.

In this article we document the first reported case of EO in an extremely preterm infant. EO occurred after a well-defined episode of congenital *Escherichia coli* sepsis. No abnormalities of the urinary tract were found. The case adds weight to existing literature which suggests hematogenous spread to an otherwise normal urinary tract is the most frequent cause of neonatal EO, and highlights a propensity for antibiotic treatment failure.

1. Case report

This male baby was born at 26 weeks gestation via caesarean section for fetal distress associated with maternal fever following

* Corresponding author.
E-mail address: scott.morris@sa.gov.au (S. Morris).

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prolonged preterm rupture of membranes. At birth he was neutropenic with an elevated band neutrophil ratio, and blood cultures confirmed congenital sepsis due to *E. coli*. Cerebrospinal fluid cultures were negative. Placental swabs also grew *E. coli* and placental histopathology showed funisitis and chorioamnionitis. He was treated for 10 days with antibiotics including gentamicin and cefotaxime. A blood culture at the conclusion of treatment was negative and hematology and CRP were normal. He did not have a urinary catheter inserted at any stage.

At 29 days of age (19 days after ceasing antibiotics) he developed cardio-respiratory instability and physical examination noted lethargy, pallor, mottled skin and a soft, distended abdomen. Abdominal x-rays did not suggest necrotizing enterocolitis. He was stabilized on a ventilator and antibiotics (piperacillin and tazobactam) commenced. Full blood examination showed neutropenia and an elevated band neutrophil ratio. A urine culture was not able to be obtained prior to commencing antibiotics. Blood culture was positive for *E. coli*. Cerebrospinal fluid cultures were negative.

After several days of antibiotic treatment he was noted to have an erythematous, firm and tender swelling of his right scrotum. Ultrasound demonstrated a small complex collection of echogenic material around the right testis consistent with a pyocele, and testicular hyperemia (Fig. 1). Needle aspiration of the tunica returned no fluid. Surgical exploration excluded torsion and confirmed a right pyocele with inspissated pus in the tunica. The right testis was noted to be compressed at surgery but viable. Histopathology showed an inflammatory exudate with numerous polymorphs and a negative gram stain for microorganisms. Cultures of the pyocele material were negative. After 2 weeks of intravenous antibiotics he was changed to oral amoxycillin and clavulanic acid a further 2 weeks. Recovery was thereafter uncomplicated. Follow-up renal imaging showed mild dilation of the left renal pelvis on ultrasound not considered to be significant. A micturating cystourethrogram was normal. Over a 3 year period of follow-up no subsequent episodes of EO occurred and testicular growth was normal.

2. Discussion

This is the first documented case of EO in an extremely preterm baby that we are aware of. The clinical course is consistent with congenital *E. coli* septicemia resulting in seeding of the epididymis or testis, with incomplete treatment resulting in the subsequent pyocele and late onset septicemia. This case has similarities to that described by Di Renzo et al., in a 33 week gestation baby with congenital *Klebsiella pneumoniae* septicemia, who was treated with appropriate antibiotics and then developed bilateral *K. pneumoniae* pyoceles on day 14 of life in the absence of urinary tract infection or underlying uropathy [7]. Malkin et al., also described a term gestation infant born normally without a set-up for sepsis who developed *E. coli* septicemia on day 6 in the absence of urinary tract infection or uropathy, with EO and a pyocele occurring 7 days later. Fromme et al., reported a term baby with negative blood cultures at birth who became septicemic on day 11 with *E. coli* and then developed pyoceles 5 days later [6]. These cases suggest that either testicular tissue can be a relative sanctuary site for coliforms or that hematogenous testicular seeding in the neonate has a propensity for early subclinical pyocele formation preventing clearance of bacteria by systemic antibiotics leading to treatment failure. In our case, we could not exclude a urinary tract infection in association with the EO. However a urinary tract infection is unlikely to have been present as there was no debris visible in the upper tracts or bladder on ultrasound at the time of the presentation, and no anatomical abnormalities were detected on subsequent investigation.

Other reported cases of early neonatal EO in the literature have not had clear antecedent episodes of congenital septicemia, although bacteremia and seeding the testis has been implicated by the absence of a urinary tract focus or predisposing anatomical anomaly. Chiang et al., described a 35 week baby born vaginally at term without a set-up for sepsis, who presented on day 6 with *E. coli* septicemia and testicular swelling due to pyoceles [5]. Urinary tract infection was excluded and a normal urinary tract was subsequently identified [5]. Goirand et al., described a 33 week gestation baby who was well at birth and with negative blood cultures, and who developed Streptococcus agalactiae septicemia and EO on day 19, also without urinary tract infection or underlying uropathy [9].

Urological anomalies appear to be uncommon in neonatal EO based on our literature review. Kabiri et al., described a term gestation infant who developed *Pseudomonas aeruginosa* septicemia and EO on day 7 of life [8]. Although a urine culture was negative, an underlying hydro-ureteronephrosis, ureteral duplication and ureterocele were implicated in pathogenesis. Seigel et al., also reported a newborn case of EO in their retrospective case series with a urinary tract infection and vasal ectopia [1]. Raveenthiran et al., described anorectal malformations as a major risk factor for EO in infancy [10]. However, in the absence of anorectal malformation, the approach to urological investigation in the neonatal and preterm age group after a single episode of EO could reasonably be restricted to renal ultrasound.

Neonatal EO requires differentiation from other causes of acute scrotum in the neonate, in particular testicular torsion and incarcerated inguinal hernia. The presentation of a tender testicular swelling in a septic infant suggests EO, and ultrasound with color

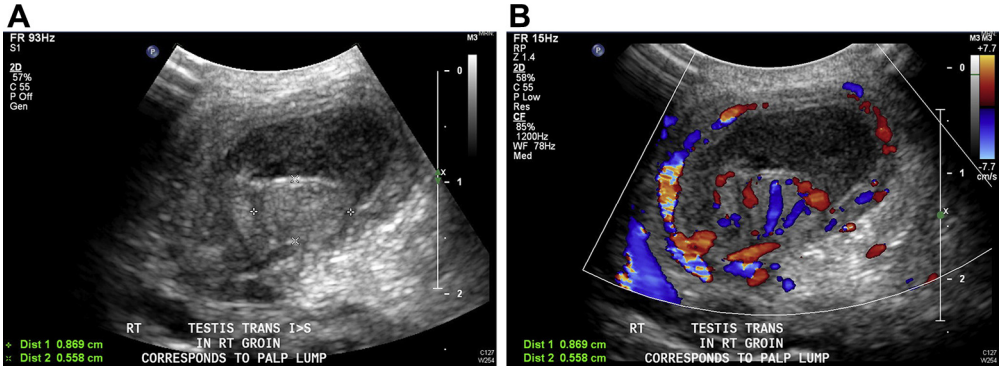

Fig. 1. Ultrasound of the testis showing A) a normal sized testis surrounded by an echogenic fluid collection, and B) Hyperemia of the tunica and testis demonstrated by color Doppler.

Doppler is characteristic in showing altered echo-texture of the testis, increased blood flow and a collection in the tunica vaginalis. Non-surgical management of EO in older children has been advocated where presentation is characteristic, although routine diagnostic exploration is also proposed due to inaccuracy of clinical assessment [11,12]. In the case of neonatal EO however, routine surgical exploration may be prudent not only for diagnosis, but also to assist a microbiological diagnosis, guide antibiotic therapy, to evacuate a pyocele and hasten resolution of systemic sepsis. The long term follow-up of our case documents normal testicular volume in childhood. At the time of surgery the testis was compressed by the pyocele, and we suggest that early surgical decompression may also improve long term testicular viability.

3. Conclusion

Neonatal EO can result from hematogenous seeding following congenital septicemia. This case and other literature reports suggest that either the testis is a relative sanctuary site for bacteria or that a propensity for early subclinical pyocele formation during septicemia results in treatment failure and later recurrent sepsis. Clinical and ultrasound findings of EO are characteristic, but surgical exploration is suggested for both diagnosis and treatment. Although underlying uropathy is more common in younger children with EO in the literature, this case and a literature review suggest that uropathy with neonatal and preterm EO is unlikely and extensive urological investigations are not warranted.

Conflicts of interest

None.

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