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Scrotoschisis in a Central African Newborn

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Abstract

Scrotoschisis, a rare congenital disease, is defined by an extracorporeal ectopic testis eviscerated as a result of a scrotal wall defect. We report a case in a Central African newborn, born via vaginal delivery, to non-consanguineous parents, admitted at three days of life from a level II health center. The pregnancy was poorly monitored without ultrasound. On admission, the temperature was 40˚C, he was polypneic and tachycardic. There was a congenital defect of the scrotal wall with exteriorization of the left testis and the right testis was normal. The blood count showed pancytopenia and the blood culture isolated amiklin-sensitive Klebseilla pneumonia. The diagnosis of scrotoschisis associated with sepsis was made. A tri-antibiotic therapy associated with general resuscitation followed by testicular transposition and orchidopexy performed on the eleventh day of life did not prevent the fatal outcome three hours after the surgery. This observation confirms the rarity of the malformation and the benefit of an early management.

Keywords

Scrotoschisis, Neonatal Sepsis, Bangui

1. Introduction

Scrotoschisis is an extremely rare congenital urogenital malformation associating a defect of the scrotal wall with exteriorization of one or both testis [1] [2]. Its prevalence remains poorly known. Yaokren *et al.* reported 23 cases in 2016, while Sidibe *et al.* in 2017 reported 17 [1] [2]. Based on literature reviews, few cases have been described in resource-limited countries. Its etiology also remains unclear even if many etiopathogenic hypotheses have been made: meconium pe-

ri-orchitis, phagocytosis of the gubernaculum testis, obstetrical trauma or lack of labioscrotal padding [2]. Based on literature reviews, about thirty cases have been reported worldwide and unilateral forms are the most frequent. The treatment of scrotoschisis is surgical in all cases and the outcome is generally favorable; however, complications such as testicular torsion and sepsis could occur. We report a case of unilateral left scrotoschisis in a Central African newborn with a fatal outcome. The purpose was to report the rarity of this malformation.

2. Case Presentation

A full-term male newborn was born on November 14, 2020. He weighs 1790 gr and heights 50 cm. He was born at 39 weeks of gestational age. The father was 24-year-old and the mother, a 16-year-old para 1 and gravida 1. There is no history of consanguinity. The pregnancy was without anomaly. Three antenatal cares were performed, all by midwives, without obstetrical ultrasound. Toxoplasmosis, rubella and syphilis serologies were all negative. The birth was vaginal delivery in a level II maternity. The adaptation of the newborn to the life was good. Three days after birth, the newborn was transferred to the pediatric teaching hospital of Bangui for a urogenital malformation. The investigation of the parents and first cousins regarding a history of malformative pathologies was negative in all cases.

Upon admission, the temperature was 40˚C, polypnea, tachycardia, and a congenital defect of the scrotal wall with exteriorization of the left testis (**Figure 1**). There was no necrosis or ischemia on the testis and the defect. The controlateral

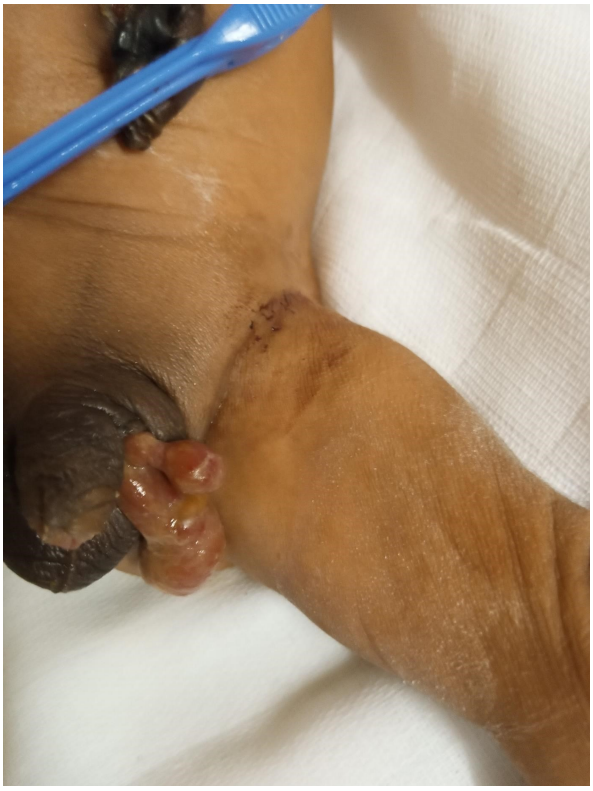

Figure 1. Picture of scrotoschisis before surgery.

Figure 2. Picture of scrotoschisis after surgery.

non-extrophied testis was normal and no other organ malformation was noted.

Complete blood cell count showed pancytopenia with leukopenia at 1500/mm^3 along with granulocytic predominance (55%); normocytic normochromic anaemia with haemoglobin at 9 g/dl and thrombocytopenia at 800/mm^3. The morphological workup for other malformations was not performed.

A scrotoschisis complicated by sepsis was presumed. A blood culture was taken and a probabilistic treatment was started then readjusted after the culture isolated Klebsiella pneumonia sensitive to amikacin. He was treated by cefotaxime and ampicillin in intravenous at 50 mg/kg/8hours during 7 days as well as gentalline intravenous 3 mg/kg/day during 3 days; then switched to amikacin at 15 mg/Kg/day from 4th day to 7th. He received Glucose 8% at a rate of 100 ml/kg/24h at 3rd day, with an increase of 20 ml per day until 170 ml/kg/day. A blood transfusion was done at 9th day at a rate of 15 ml/kg in order to prepare him for the surgical procedure. The newborn underwent the surgery procedure the eleventh day of life (**Figure 2**). During the procedure, an extrophied left perineal transverse testicular ectopy of normal morphological appearance was noted. There was no persistence of the peritoneovaginal canal. He presented a distress respiratory requiring resuscitation. The newborn died 3 hours post-surgery, probably due to complications of the general anesthesia.

3. Discussion

Scrotoschisis, a congenital malformation that associates a defect of the scrotum with extracorporeal testicular ectopia, is rare [2] [3]. The first published case was by Von der Leyer1 in 1963 [4]. As of 2017, a total of 10 cases have been published worldwide [2]. Sidibe *et al.* published the 17th case [1]. We report the first

Central African case. Scrotoschisis is most commonly seen in full term newborns of normal weight for age [3] [4] [5]. This was not the case in this observation where the newborn had a low-birth-weight.

The pathogenesis of scrotoschisis is not well known. However, several theories have been put forward [1]. The one following an infectious or ischemic process [2] and the traumatic theory of evisceration occurring during delivery or after cesarean section [6] [7]. In the present case, the delivery, although vaginal, was normal [5] [8]. The theory of a congenital anomaly that disrupts the integrity of the scrotal wall responsible for the extracorporeal position of the testis has also been put forward without consensus, as well as that of hyperactivity or misdirected phagocytic action of the gubernaculum testis.

An experimental study in rats by Chun *et al.* [6] showed that excision of the future scrotal skin inhibited gubernacular migration, leading to ipsilateral testicular ectopy. This shows that normal scrotal development is necessary to guide gubernacular migration before testicular descent. The most accepted theory is that of meconium peritonitis. In this case, it would be 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 during fetal life through the peritoneovaginal duct. Considering this hypothesis of meconium exposure, histological examination of the excrescence reveals the presence of calcified meconium that would have migrated through the peritoneovaginal canal [1] [8] [9] [10]. Scrotoschisis is often unilateral but bilateral forms have been described [10]. Currently, there is no plausible explanation as to whether it is unilateral or bilateral [11]. Extrophied, often necrotic or atrophied testes may have normal morphology and/or histology. The most plausible explanation for the necrosis is that of a torsion of the spermatic cord associated with the malformation [12].

Treatment of scrotoschisis is not an emergency if there is no associated torsion [12] [13]. However, repair within hours of birth can prevent complications. Surgical treatment consists of scrotal exploration and placement with fixation of the testis in the scrotum followed by orchidopexy. The prognosis depends on the associated malformations and the condition of the extrophied testis. The outcome is generally favorable, but sometimes serious complications are reported, such as testicular torsion and irreversible ischemia [11] [14]. However, our case is the first death in the literature review. According to Lejus *et al.*, the physiology of the newborn is characterized by an immaturity of most of major vital functions. This results in limited adaptation capacities and modifications of the pharmacokinetic and pharmacodynamic behavior of all the anesthetic agents [15]. The anesthetic management of neonatal surgical emergencies also requires knowledge of the pathophysiological implications of the associated surgical defect. Analysis of a series of more than 22,000 anesthesias shows that neonates constitute the population at highest risk of intraoperative events. Respiratory complications (airway obstruction, laryngospasm, bronchospasm, apnea) are the most frequent, consistent with our case where the neonate presented a respira-

tory distress a few hours before his death [16].

4. Conclusion

Scrotoschisis is a rare pathology. Clinical examination, possibly helped by intraoperative exploration, is essential for better management. Its treatment, surgical in all cases, should not be rushed or delayed. Its outcome, often favorable, may be marked by complications.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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