The Scrotoschisis about a Case in the Pediatric Surgery Department of the Donka National Hospital

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Abstract

Introduction: Scrotoschisis is a very rare congenital defect of the scrotum characterized by the exteriorization of one or two testes. We report a case of right scrotoschisis in a newborn as well as a review of the literature for an approach of probable etiology. Patient and Observation: A newborn baby of 8 hours of life, weighing 3200 g was referred to our department for a right scrotal defect with exteriorization of the testis associated with fluid swelling of the left bursa. The 18-year-old mother, primiparous and primigeste followed all the prenatal consultations with eutocic delivery. After clinical investigation the diagnosis of right scrotosisis and left hydrocele was retained. Surgical treatment was carried out by primary closure after orchidopexy and exploration of the contralateral bursa, the content of which was calcified meconium bathed in a yellowish liquid. The post-operative consequences were simple. Conclusion: Scrotoschisis is an extremely rare birth defect whose etiology is poorly understood. However, it could be the cause of meconium peritonitis secondary to a scarred fatal intestinal perforation.

Keywords

Scrotoschisis, Congenital Anomaly, Etiology

1. Introduction

Scrotoschisis is a congenital anomaly of the scrotum associated with the exteriorization of one or both testes. This very rare anomaly was first described by Von der Leyen in 1963 [1]. Only 10 cases were reported in the literature until 2016 [2]. All available information comes from clinical cases [3]. The exact me-
chanism behind this rare birth defect is unknown [1]. Several theories have been put forward to explain its occurrence, the most accepted nowadays is meconium peri orchitis. Late rupture of the scrotal skin secondary to an inflammatory reaction is caused by exposure to meconium through a permeable peritoneovaginal duct [4]. We report here a rare case of unilateral scrotoschisis in a newborn baby as well as a review of the literature on the probable etiology of this malformation.

2. Patient and Observation

A newborn, 8 hours old, weighing 3200 g was referred and admitted to our department for a right scrotal defect leading to exteriorization of the testicle. The anomaly was noticed by the midwife who delivered it to us immediately. The 18-year-old mother, primiparae and primigeste, followed all the antenatal consultations and the pregnancy had proceeded normally to term with a eutocic delivery. There was no blood relationship between the two parents.

The examination found a newborn baby awake, toned, feverish to the touch with a temperature of 39.6°C, an oxygen saturation of 95%, a heart rate of 152 beats per minute, a respiratory rate of 36 cycles per minute, a good staining of the legs and conjunctiva.

The external genitalia were in place and male in appearance; exploration showed a defect in the upper part of the anteromedial aspect of the right he-scrotum. The spermatic cord, epididymis and testis thickened and covered with fibrin deposits had eviscerated through this defect measuring 4 × 2 cm on its vertical and transverse axis, involving the scrotal structures. Over a distance of 2 cm surrounding the defect, the scrotal wall consisted of a thin membrane instead of skin. The testis was well vascularized and of normal size, shape and appearance. The left bursa was swollen with fluid content (Figure 1). The examination extended to the other devices did not note any particularity.

Laboratory examinations were within normal limits. Conditioning of the newborn was started with rehydration made of Ringer lactate combined with 30% hypertonic glucose serum at a rate of 80 ml/kg per 24 hours. A treatment consisting of paracetamol infusion 15 mg/kgp, Cefotaxime injection 50 mg/kgp, Metronidazol infusion 10 mg/kgp and gentamycin 3 mg/kgp. After normalization of the parameters, the newborn was taken to the operating room under

![Figure 1. Right scrotoschisia: high defect in the anterior part near the median raphe.](image-url)
general anesthesia. The exploratory puncture of the left scrotum had brought back a yellowish fluid.

After washing the defect abundantly, the exteriorized elements and the perineum with physiological serum, we proceeded to open the inter scrotal septum and the left vagina giving issue to the same liquid from the puncture, mixed with lumps resembling a stercoral fluid that we took it for bacteriological examination (Figure 2).

Reintroduction of the left testicle into the vagina followed by its closure with 3/0 vicryl, repair of the inter scrotal septum with 3/0 vicryl overlock. The inguinal openings were free. Dissection and detachment of the Dartos from the right hemi scrotum leaving a compartment allowing the reintroduction of the right testicle then orchidopexy. Skin closure in separate points of 3/0 vicryl on a glove slide serving as a drain (Figure 3). The suites were simple.

The consequences were simple, the examination of the sample liquid did not reveal a germ and the culture was negative. The newborn was released after 9 days of hospitalization with healing of the first-line operative wound and we obtained the informed consent of the parents for the publication of the case. The patient is regularly followed since his discharge at the rate of once a week for 21 days then once every 2 weeks for 1 month and the patient is seen for control every month. The last examination showed that the two testes are of normal size and consistencies located at the bottom of the bursae.
3. Discussion

Scrotoschisis is an extremely rare congenital anomaly, it is the first case encountered in Guinea. The etiology is not well known, the examination of what is known about the testicular descent and the formation of the scrotum suggests some hypotheses. Scrotal development begins with the cloacal division into the urogenital sinus and anorectal canal. Labioscrotal folds form on either side of the urogenital folds, the future labia majora in girls and the scrotum in boys. Testicular descent involves hormonal effects, intra-abdominal pressure and the action of the gubernaculum to guide the testis into the inguinal region [1]. As the factors governing the descent of the testes are not clear, several theories explaining scrotoschisis have been put forward: failure of labial fold fusion, overactive phagocytosis of Von Der Leyen’s guvernaculum, failure of development of cremasteric fibers or Gupta’s mesenchyme, the infectious process or localized Heyns ischemia, and iatrogenic scrotal laceration during cesarean section [5]. The meconium peri orchitis at the origin of Scrotoschisis suggested by Kajori and DeMaria is the most accepted theory today. The late rupture of the scrotal skin is thought to be secondary to an inflammatory reaction caused by the meconium released during intestinal perforation during fetal life. The passage of this meconium from the perforated intestine to the scrotum takes place through the peritoneo-vaginal canal [6]. VS is a rare entity associated with cured meconium peritonitis [7]. A case with an association between jejunal atresia and scrotoschisis has been reported, and the absence of intestinal abnormalities in most newborns can be explained by fetal scarring of the original intestinal lesion without sequelae, a recognized phenomenon [3]. Schaffer et al. [8] also opened the septum and explored the hydrocele and found lumps of meconium bathing in the liquid. The lumps corresponding to calcification of the meconium found in the vaginal area of the left bursa, the appearance of the exteriorized testicle, its normal size and consistency in our case could corroborate this hypothesis. The defect is always high on the anterior wall of the scrotum, close to the median raphe [9]. Wet, warm compresses should be applied to the testicles while awaiting surgery to prevent dehydration of the organ. The surgeon should avoid twisting the exteriorized testicle, as it is not attached to any anatomical structure except the cord.

4. Conclusion

Scrotoschisis, also called testicular exstrophy or extracorporeal ectopia, is a very rare congenital anomaly. The etiology is not well known, but it could be related to spontaneous fetal intestinal perforations healed with meconium peritonitis. The result of the surgical treatment is good with normal growth of the testicles but the long-term follow-up data is not yet available.

Conflicts of Interest

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